Acceptability and feasibility of fabric orthoses for movement control in multiple sclerosis

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Acceptability and feasibility of fabric orthoses for movement control in multiple sclerosis.

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A thesis submitted in partial fulfilment of the requirements of Sheffield Hallam University for the degree of Doctor of Philosophy

Academic supervisors: Sionnadh McLean, Jon Wheat and Hilary Piercy

Candidate declaration

I hereby declare that:

1. I have not been enrolled for another award of the University, or other academic or professional organisation, whilst undertaking my research degree.

2. None of the material contained in the thesis has been used in any other submission for an academic award.

3. I am aware of and understand the University's policy on plagiarism and certify that this thesis is my own work. The use of all published or other sources of material consulted have been properly and fully acknowledged.

4. The work undertaken towards the thesis has been conducted in accordance with the SHU Principles of Integrity in Research and the SHU Research Ethics Policy.

5. The word count of the thesis is 68,500.

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Date of submission	31st July 2019
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Summary of PhD thesis

Acceptability and feasibility of fabric orthoses for movement control in multiple sclerosis

Multiple sclerosis (MS) is a common neurological disorder, characterised by slowed, diminished sensory feedback and weakness leading to reduced mobility. Fabric orthoses are suggested to improve movement by enhancing sensory feedback and providing a flexible exoskeleton. At the outset of this doctoral programme, only low quality evidence existed, acceptability was suggested to be poor and therapists commonly believed that fabric orthoses needed several weeks' wear before any benefits might be seen. There was no published research evaluating use in MS.

This thesis aimed to investigate the feasibility of fabric orthoses in MS. A series of inter-linked research projects were conducted, starting with systematic reviews investigating fabric orthoses, compression garments and joint supports. A qualitative investigation was conducted with people with MS who were long-term fabric orthosis users. Latterly, the project focussed on orthotic shorts as a means of improving walking and, following a small pilot study, a mixed methods feasibility study was conducted evaluating acceptability and potential efficacy.

The programme of research generated new knowledge. Specifically, fabric orthoses were shown to have an important influence on confidence. Acceptability was shown to be affected by how fabric orthoses are introduced and the support provided in the early stages of use. The recently proposed Theoretical Framework of Acceptability was used for analysis, demonstrating for the first time how that framework might be interpreted and its value in evaluating and improving acceptability of healthcare interventions. Orthotic shorts were demonstrated to be acceptable to people with MS. Changes in walking ability were measurable after 30 minutes' wear and included improved speed, decreased variability of footfall and improved adaptability of trunk and pelvic movement. A suggested mechanism of effect has been proposed and a proof of concept randomised controlled trial designed. Overall, the doctoral programme has demonstrated potential for use of fabric orthoses in MS.

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Chapter 1: Introduction, aims and outline of the thesis

Summary

This chapter sets the scene for this PhD thesis, which aimed to investigate the acceptability and feasibility of using fabric orthoses in people with multiple sclerosis (PwMS).The nature of multiple sclerosis (MS) is explored and justification for investigating fabric orthoses is explained, as well as a justification for the more specific focus on orthotic shorts in this research programme. Finally, aims, objectives and an overview of the thesis are presented.

1.1 Introduction

Multiple sclerosis (MS) is a chronic, progressive neurological disease characterised by plaques of demyelination and transection of axons in the central nervous system. It is one of the world's most common neurological disorders and is a leading cause of non-traumatic disability in young adults (Browne et al., 2014). MS is more common in women with a UK prevalence of 286 per 100 000 in women and 113 per 100 000 in men (McKenzie et al., 2014). MS is associated with high costs to society in terms of lost employment, care needs and drug costs (Mccrone, Heslin, Knapp, Bull & Thompson, 2008).

MS is characterised by uncertainty in terms of the longer-term prognosis, the day-to-day variability caused by symptoms such as fatigue and heat sensitivity and, for those with relapsing-remitting MS, the seemingly random relapses (Dennison, McCloy Smith, Bradbury & Galea, 2016). The feeling of living with an uncertain future and an unreliable body is exacerbated by difficulty with sensory feedback, movement control and symptoms such as visual disturbance, bladder and bowel disturbance, pain and cognitive impairment (Vickers, 2009).

Plaques of demyelination can occur throughout the central nervous system (CNS) but especially in the optic nerves, spinal cord, brainstem, cerebellum, periventricular white matter and cortical grey matter (Popescu & Lucchinetti, 2012). Therefore, difficulties with motor control are caused by a multitude of inter-related factors that, taken together, result in movement that is unpredictable, difficult to control and extremely tiring. Firstly, muscle weakness is common in MS (Hoang, Gandevia & Herbert, 2014), probably due to cortical, subcortical or spinal lesions to motor neurones but likely exacerbated by decreased physical activity (Petajan & White, 1999). Secondly, sensation is slowed and diminished, affecting coordination of voluntary movement and balance (Cameron, Horak, Herndon & Bourdette, 2008a). Vestibular impairment is common and PwMS appear to use multiple sensory systems to balance, as each individual system becomes unreliable (Cattaneo & Jonsdottir, 2009). Thirdly, due to lesions in the cerebellum, brainstem and cortex, the centres governing integration and control of movement are affected. Cognitive ability can be directly affected (Johansson et al., 2007) and indirectly affected due to the increased cognitive load associated with compensation for sensorimotor problems (Etemadi, 2017; Prosperini et al., 2015; Wajda, Roeing, Mcauley, Motl & Sosnoff, 2015). Finally, fatigue is a common and important symptom (Hemmett, Holmes, Barnes & Russell, 2004) and may contribute towards and be exacerbated by the sensorimotor challenges. For example, fatigue severity is related to the degree to which cognition is required for walking tasks (Garg et al., 2016; Gunn, Newell, Haas, Marsden & Freeman, 2013; Hamilton et al., 2009).

The challenges of uncertainty and unreliability are extremely relevant to walking difficulties. An estimated 76% of PwMS complain of mobility problems (Jones et al., 2013) and 50-70% report that walking is the most challenging aspect of MS that they face (LaRocca, 2011; Pike, Jones, Rajagopalan, Piercy & Anderson, 2012). Walking ability may predict employment status and the need for caregiver support and healthcare resources (Pike et al., 2012). PwMS walk more slowly, take fewer steps, have a shorter step length, a more prolonged double support phase and a wider base of support than non-neurologically impaired people (Givon, Zeilig & Achiron, 2009). Walking pattern is variable from one step to the next, even in PwMS who are not aware of any impairment (Flegel, Knox & Nickel, 2012; Spain et al., 2012) and gait variability might in turn relate to risk of falling (Allali et al., 2016a; Socie at al., 2013a).

Currently, the main intervention for improving motor control in PwMS is exercise, which has been shown to be effective in improving muscle power, walking ability, balance, fear of falling and quality of life (Carter et al., 2014; Kasser et al., 2015; Learmonth, Paul, Miller, Mattison & Mcfadyen, 2012; Rietberg, Brooks, Uitdehaag & Kwakkel, 2005). However, adherence to exercise is known to be challenging in MS (Carter et al., 2014) therefore, any interventions that might support exercise adherence or directly improve motor function are worthy of investigation.

Fabric orthoses are garments, made from elasticated fabrics but designed to support and control the body. They were first used in children with cerebral palsy (Nicholson, Morton, Attfield & Rennie, 2001) but have been developed for use with adults more recently (Watson, Crosby & Matthews, 2007). Watson et al. (2007) suggested that fabric orthoses might improve body posture, dampen down involuntary movement; provide a motivational effect via a novel intervention; enhance sensory feedback and provide a flexible "exoskeleton" (p754) for people with weakness. The suggestion that such orthoses might have a sensory and supportive effect implies that they may be capable of improving movement control in MS. In 2014, when this doctoral programme commenced, there was no published research investigating the use of fabric orthoses in MS. A small qualitative study, with practitioners who frequently used fabric orthoses suggested that fabric orthoses had a positive impact upon PwMS (Hassan & Snowdon, 2015). Acceptability has been stated to be a key challenge with fabric orthoses (Coghill & Simkiss, 2010), as found in early studies in children with cerebral palsy (for example, Blair, Balluntyne, Housman & Chauvel, 1995; Knox, 2003).

The Medical Research Council guidelines for Developing and Evaluating Complex Interventions (Craig et al., 2008) were first published in 2000 and have been influential in shaping a systematic, phased approach to research and development of novel interventions (Skivington, Matthews, Craig, Simpson & Moore, 2018). The Guidelines suggest that early work should develop an intervention through identifying an evidence base and developing theory (see Figure 1.1). Early studies need to focus on pilot and feasibility work, which may in turn develop theories around mechanism of effect. Development work should be iterative rather than linear, so that stages of evaluation and implementation might in turn influence development of the intervention or lead to additional feasibility testing. These principles have shaped this doctoral programme, which consists of development, feasibility and pilot work in the context of determining the potential for further evaluation of the use of fabric orthoses in MS.

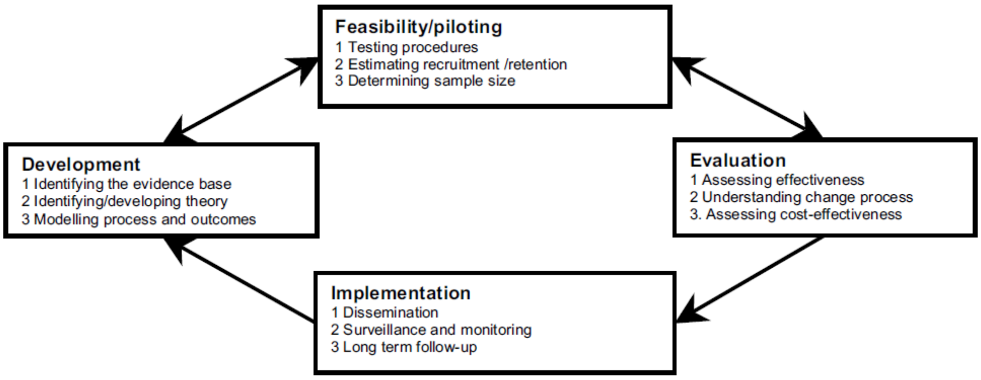


Figure 1.1: Key elements of the development and evaluation process for a complex intervention (from Craig et al., 2008)

The wider aim of this doctoral programme was to develop an understanding of the potential for using fabric orthoses in MS and factors that might influence acceptability. However, to conduct a novel investigation, it was necessary to focus down on one specific type of orthosis and one specific movement problem. The decision to focus on the use of orthotic shorts for improving walking was a collaborative decision with DM Orthotics, the orthotics company who supported this programme of study. Their orthotic shorts had not been systematically investigated previously and they believed the shorts could improve walking. Previous research supported the possibility that hip and pelvic instability might be important in the control of walking (Huisinga, Mancini, St. George & Horak, 2013) and that orthotic shorts might provide stabilisation and improved feedback around the hip (Cameron, Adams & Maher, 2008b; Maguire, Sieben, Frank & Romkes, 2010). In addition, it seemed likely that any challenges with acceptability might be particularly relevant to shorts, because they could interfere with movement, might be difficult to put on and take off and might interfere with toileting.

1.2 Aims and Objectives

The aim of this thesis was to investigate the acceptability and feasibility of using fabric orthoses with PwMS.

Objectives were to:

1. Identify and evaluate the existing evidence into the use of fabric orthoses
2. Determine influences on acceptability of fabric orthoses
3. Determine the feasibility of using orthotic shorts to improve walking in PwMS
4. Develop a theory explaining the possible means of effect of fabric orthoses, specifically orthotic shorts
5. Design a future randomised controlled trial to investigate the effectiveness of orthotic shorts in MS.

1.3 Outline of the thesis

The programme of research reported in this thesis consisted of:

1. Systematic reviews, which:
    i. Mapped previous research into fabric orthoses, compression garments and joint supports
    ii. Investigated the effectiveness of fabric orthoses in adults with neurological conditions
2. The Fabric Orthosis Interpretative Phenomenological Analysis (FabO IPA) study - a qualitative study that investigated the experiences of PwMS who were long-term users of fabric orthoses
3. A single case methods pilot used to evaluate outcome measures for walking and balance in PwMS, in preparation for a feasibility study
4. The Orthotic Shorts Feasibility in MS (OSFeaMS) study - a mixed methods crossover study investigating the acceptability and feasibility of orthotic shorts for walking in PwMS.

The programme of research enabled both an exploration of fabric orthoses in general and a more in-depth investigation of orthotic shorts. The first systematic review considered not just fabric orthoses but also compression garments and joint supports, because these are similar interventions more thoroughly investigated than fabric orthoses, thus enabling additional insight into possible mechanisms of effect. Reviews confirmed the lack of previous evidence for fabric orthoses, in particular a marked absence of qualitative studies, despite the controversies around acceptability. The FabO IPA study enabled exploration of acceptability from the perspective of PwMS who use fabric orthoses on a regular, ongoing basis. This enabled important insight into users' experiences and various factors that might influence acceptability. Following the FabO IPA study, the research programme focussed on walking ability and orthotic shorts. In developing the protocol for the OSFeaMS study, a methods pilot was conducted with the help of one of the participants from the FabO IPA study. This informed the choice of outcome measures for the OSFeaMS study, which utilised a convergent parallel, mixed method approach to explore acceptability and potential efficacy of the orthotic shorts.

Chapter 2 reports both systematic reviews, firstly the mapping review and secondly the systematic review into fabric orthoses for adults with neurological conditions. Chapter 3 reports the FabO IPA study and explains the relevance of the findings to understanding acceptability. Chapter 4 reports a review of outcome measures for assessing walking ability in MS and explains how the single case methods pilot influenced measurement choices for the OSFeaMS study. Chapter 5 explains the methodological approach to the OSFeaMS study and the methods used. In Chapter 6, the quantitative results of the OSFeaMS study are reported and discussed, providing insight into the potential efficacy of the orthotic shorts. Chapter 7 reports the qualitative findings of the OSFeaMS study, including acceptability and feedback on study design. Chapter 8 integrates the quantitative and qualitative findings predominantly around understanding the impact of orthotic shorts on walking ability and the design of future research studies. Chapter 9 provides an overview of how the thesis as a whole met the objectives set, the original contributions to knowledge within the work and additional priorities for future research.

1.4 Conclusion

This first chapter has justified the focus of the thesis on PwMS, explained key features of motor control in MS and explained the potential for fabric orthoses for improving motor control in PwMS. An outline of the thesis studies and structure has been provided. The next chapter describes the systematic reviews conducted in the early stages of the work.

Chapter 2: Literature review

Summary

This chapter presents two systematic reviews. One is a systematic mapping review of primary research investigating fabric orthoses, compression garments and joint supports. The second is a systematic review of parallel-group randomised controlled trials (RCTs) and crossover trials investigating the effectiveness of fabric orthoses in adults with neurological disorders. The findings of each are discussed. The systematic mapping review was published as Snowdon, Sier, Potia, Wheat & McLean (2018; Appendix 1).

2.1 Introduction

At the outset of this doctoral programme, I was aware that very little previous research had been conducted into fabric orthoses in multiple sclerosis. Therefore, fabric orthoses and related interventions were examined more broadly, to understand what types of research approaches had been used, what effects had been investigated and to determine whether a systematic review specifically on fabric orthoses was justified. The related interventions explored were compression garments and Neoprene joint supports. These are manufactured from elastic, synthetic fibres. Compression garments are used by healthy athletes with the belief that they might improve performance and recovery from exercise (MacRae, Cotter & Laing, 2011; Skins International, 2015). Neoprene joint supports are orthoses used for weak, painful joints (Tiggelen, Coorevits & Witvrouw, 2008) and are hypothesized to improve proprioception (Webster, Nussbaum & Madigan, 2017). Research into compression garments had investigated several effects that might be of relevance to clinical populations. For example, research suggested that compression garments may be effective for improving circulation (MacRae et al., 2011), kinematics (de Britto et al., 2017), coordination (Hasan, Davids, Chow & Kerr, 2017), balance (Michael, Dogramaci, Steel, & Graham, 2014), proprioception (Cameron et al., 2008b) and recovery from exercise (Brown et al., 2017). These variables are relevant to mobility, movement and function across a range of pathologies. Similarly, there had been extensive research investigating the impact of Neoprene joint supports on pain, quality of life and proprioception (Duivenvoorden et al., 2015; Ghai, Driller & Ghai, 2017).

Craig et al. (2014) were conducting a review into effectiveness of fabric orthoses in children and previous reviews of fabric orthosis use in children had been published (Attard & Rithalia, 2004; Coghill & Simkiss, 2010) with little additional primary research made available since those reviews. Thus, including research into children in a systematic review did not seem valuable within this doctoral programme. Searches confirmed the absence of any previous systematic review on fabric orthoses in neurologically impaired adults but a number of such primary studies were identified. Thus, the second part of this chapter reports a systematic review of the effectiveness of fabric orthoses in neurologically impaired adults.

2.2 Methods - systematic mapping review

Systematic mapping reviews explore the characteristics of a body of research to determine where gaps exist. They are particularly useful where research is disparate (Booth, Papaioannou & Sutton, 2012). They can suggest research questions for additional reviews or primary research and can contextualise more in-depth reviews within the broader literature (Clapton, Rutter & Sharif, 2009).

This systematic mapping review was conducted according to guidelines published by the Social Care Institute for Excellence (Clapton et al., 2009). As is usual within a mapping review, quality assessment was not conducted because the aim was to describe a body of evidence, rather than to draw conclusions about findings (Booth et al., 2012).

2.2.1 Search strategy

Searches for the systematic mapping review were conducted twice, first in December 2015 and secondly in June 2017. The December 2015 search was instrumental in informing the thesis and many of the key findings were apparent from that first review. However, the search was updated in seeking publication. The second search is reported here and in Snowdon et al. (2018).

One reviewer (NS) conducted searches on MEDLINE, CINAHL and Sports Discuss from inception to June 2017, as shown in Table 2.1. The first searches in December 2015 used a wider range of databases, including AMED, EMBASE, Scopus and Web of Science; however, MEDLINE, CINAHL and Sports Discuss retrieved the same relevant studies but with fewer conference presentations and fewer irrelevant studies. The search was designed to locate studies on compression garments, fabric orthoses and joint supports without specifying specific participant groups, study designs or outcomes. A review of citations highlighted some missing studies investigating fabric orthoses and haemodynamics. Altering the search strategy to identify these studies greatly increased the number of irrelevant records; therefore, additional studies were identified from citations. Citations of all included studies were searched. All studies citing the following influential systematic reviews and primary studies were identified using Scopus: Birmingham, Kramer, Inglis & Mooney (1998); Blair et al. (1995); Born, Sperlich, & Holmberg (2013); Coghill & Simkiss (2010); Gracies et al. (2000); Hammond, Jones & Prior (2016); Kirkley et al. (1999); MacRae et al. (2011).

Table 2.1: Search strategy for systematic mapping review

Search	Concept	Search terms	Limited to
1	Intervention – type of material	Elastane OR spandex OR Lycra OR Elastomer* OR Theratog* OR compression OR Neoprene	All text
2	Intervention – nature of garment	Orthos* OR orthotic* OR shorts OR garment* OR splint* OR brace OR sock* OR stockings	Title and abstract
		Orthotic devices (MEDLINE); Orthoses and Orthoses design (CINAHL)	Subject headings
3	Search 1 AND Search 2		

2.2.2 Study selection

Studies were selected according to the eligibility criteria below. Title screening was carried out by one reviewer (NS). A second reviewer (DS) examined the excluded titles to ensure relevant studies were not excluded. Full-text screening was performed independently by two reviewers (NS and DS).

The inclusion criteria were as follows:

- Primary research of any design, including case descriptions

- Interventions were garments or orthoses incorporating elastomeric fabrics and without inelastic components
- Investigation relevant to mobility, active movement or any type of function.

The exclusion criteria were as follows:

- Reviews and editorials, because the aim was to map primary research only
- Abstracts and conference proceedings, because these frequently lacked sufficient detail on study procedures and measures
- Studies primarily investigating oedema, lymphoedema or venous disease, because there is a well-established role for compression in these conditions and, therefore, mapping this research was unlikely to provide novel insights
- Studies on wound healing, thrombus prevention, acute postsurgical management and orthostatic hypotension, because these were felt to have limited relevance to other conditions
- Studies on interventions designed only for use in therapy sessions, such as the Adeli suit and Therasuit, which would be impractical for regular, daily wear due to the inclusion of large numbers of elastic cords on the surface of the garments.

There were no disagreements on the included texts, so the third reviewer (SMcL) was not used.

2.2.3 Coding and mapping process

Included studies were coded by one of three reviewers (NS, DS and TP) via a standardised online form that had been piloted by these reviewers. Data extracted were author(s), country, year of publication, study design, participants, intervention classification (defined below), outcome measures and whether pressure beneath the garment was measured.

Interventions were classified as compression garments if compression was described as a feature of the garment; as a joint support if only one joint or body area was covered by the orthosis and as a fabric orthosis if the garment was not described as a compression garment but where the orthosis covered more than just one body area.

Orthoses are defined as externally applied devices used to modify the structural or functional characteristics of the neuromusculoskeletal system (Ponton, 1997). All three interventions could be described as orthoses; however, terminology was chosen to reflect descriptions used in the studies.

The mapping process was undertaken by one reviewer (NS) and involved organising all of the studies into themes for ease of description of the body of evidence. Initially, studies were grouped using their titles and outcome measures. A pattern was identified that themed studies according to their main aim of investigation. Once themed, new codes were created that were felt to best describe each theme and these codes were added to the study database. The final description of the studies was checked and agreed by other reviewers (SMcL, DS, TP and JW).

2.3 Results - systematic mapping review

2.3.1 Study selection

The search found 5408 records, of which 1133 were duplicates; therefore, 4275 records were screened by title. A total of 504 studies appeared to be relevant and had their full-text reports obtained and screened. Of these, 153 did not meet the eligibility criteria. Figure 2.1 provides a Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) (Moher, Liberati, Tetzlaff & Altman, 2010) flow chart of study selection, including reasons for exclusion. A total of 351 studies were included in this review.

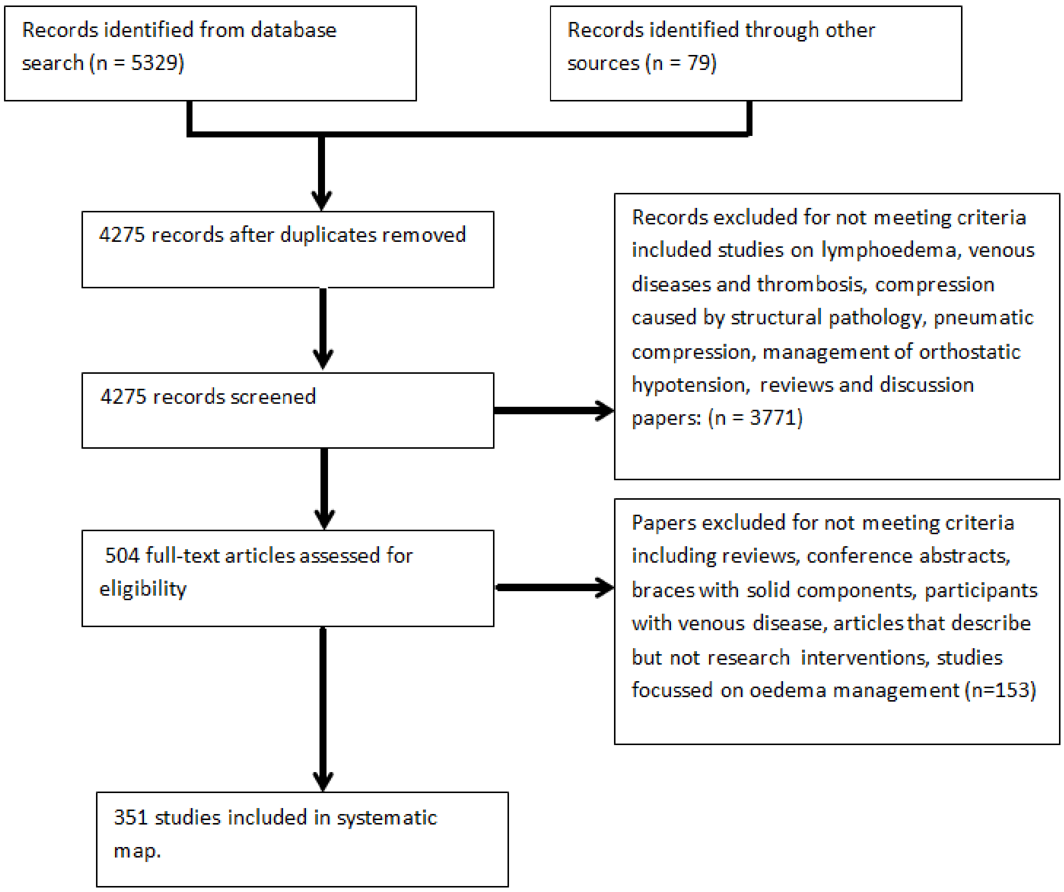


Figure 2.1: PRISMA flow chart showing study selection for systematic mapping review

2.3.2 Study characteristics

Interventions and study participants

Two hundred and thirty-six studies (67%) investigated compression garments, 64 (18%) investigated joint supports and 42 (12%) investigated fabric orthoses. Nine studies did not fit this intervention classification: two investigated close-fitting clothing without specifying them as compressive; three investigated heat-dissipating clothing; and four investigated orthokinetic cuffs, which were intended to alter muscle tone via differential sensations of elasticity over agonist and antagonist muscles. The number of compression garment studies grew significantly between 2006 and 2014, with a slight decrease in 2015 and 2016. The number of studies into joint supports and fabric orthoses remained steady, with between none and six studies published each year for joint supports and between one and four published each year on fabric orthoses.

Two hundred and thirty-three studies (66%) investigated healthy participants. Studies on healthy participants made up 83% of the compression garment studies, 44% of the joint support studies and 7% of the fabric orthosis studies. Table 2.2 summarises the garments investigated, participant group, study aims and study designs used.

Study designs

The most common study designs were crossover trials (225 studies, 64%) and RCTs (38 studies, 11%). These more robust scientific methods were more common in compression garment and joint support research than in fabric orthosis research, in which other, less robust approaches were used, such as case descriptions and single-case experimental designs. Crossover studies made up 72% of the studies into compression garments but only 19% of the fabric orthosis studies. Only three studies used qualitative methods (May et al., 2006; Stone, 2014; Miller, van Wijck, Lamont, Preston & Hair, 2016).

Table 2.2: Overview of studies in review organised by interventions researched

	Garments used	Population	Aim of investigation	Study designs
Compression garments (236 studies)	Below-knee socks (86); waist-to-ankle leggings (50); thigh-length stockings (24); shorts (19); calf sleeves (11); whole-body garments (10); waist-to-foot tights (10); thigh sleeves (4); long-sleeved tops (10); arm sleeves (7); gloves (6); groin-to-ankle leg sleeves (4); waist-to-mid-calf leggings (1); abdominal binding (1)	No condition or pathology (197); anterior cruciate ligament reconstruction (1); ankle sprain (3); pregnancy or postnatal (6); leg symptoms with prolonged standing (5); arthritis (5); spinal cord injury (4); groin injury (2); stroke (1); cerebral palsy (1); runner with clotting disorder (1); Parkinson disease (1); developmental delay (1); sternal instability post-surgery (1); no participants (1)	Sport performance (59); cardiovascular and respiratory (59); sport recovery (41); investigating pressures applied (18); relief or prevention of pain (15); sport performance and recovery (13); thermal effects (8); muscle activity and reflex excitability (non-sport) (5); posture and movement control (non-sport) (5); proprioception (3); psychological effects (2); acceptability and compliance (3); side effects (3)	Crossover study (170); randomised controlled trial (26); non-randomised trial (15: nine used one leg in each person for intervention and one for control, one was a preference trial, two were pre–post trials); matched-pair trial (5); observational study (6); measurement study (5); developing and testing predictive models for the biomechanical effect of pressure on the body (4); single-case experimental design (2); survey (1); qualitative study (1)
Joint supports (64 studies)	Knee (14); lumbar (14); thumb (7); wrist (5); shoulder (3); finger (2); ankle (2); elbow (1); various supports in study (2)	No pathology (28); knee OA (8); thumb OA (5); knee pain or instability (7); low back pain (3); one study each on RA, hamstring injury, cerebral palsy, Charcot–Marie–Tooth disease, ankle instability, shoulder instability, joint hypermobility, finger contracture, tennis elbow, club hand, wrist trauma	Proprioception (19); posture and movement control (non-sport) (17); relief or prevention of pain (11); muscle activity and reflex excitability (non-sport) (8); side effects (4); cardiovascular and respiratory (1); investigating pressures applied (1); novel splint designs (3)	Crossover study (40); randomised controlled trial (7); case description (10); observational (3); repeated-measures design (2); single-case experimental design (2)
Fabric orthoses (42 studies)	Whole-body suit (11); shorts (8); arm sleeve (5); below-knee socks (5); waist-high leggings (4); various-length gloves (4); short body suit (shoulders to thighs) (2); hip orthosis (1); trunk support (1); short-sleeved shirt (1); shoulder support (2)	Cerebral palsy (15); stroke (7); no pathology (3); multiple neurological pathologies (3); neuropathic scoliosis (2); brachial plexus palsy (2); Duchenne muscular dystrophy (2); multiple sclerosis (2); one study each on traumatic shoulder subluxation, developmental coordination disorder, post-hip fracture, post-meningitis, RA, hip OA hip, spina bifida, groin injury	Posture and movement control (non-sport) (32); relief or prevention of pain (6); muscle activity and reflex excitability (3); proprioception (1)	Case description (12); pre–post trial (9); single-case experimental design (8); crossover study (8); randomised controlled trial (4); qualitative study (2); notes audit (1)

(Numbers in cells do not add up to totals because some studies contain different research designs and assess multiple garments/orthoses and populations; RA = rheumatoid arthritis; OA =osteoarthritis.)

Theming of studies according to main aim of investigation

Overall, the themes closely matched the measures used in the studies. Measures differed significantly between sporting and non-sporting contexts; therefore, whether the study was conducted in a sporting context was used to define some themes.

Studies were themed as sport performance (59 studies); sport recovery (41 studies); both sport performance and recovery (13 studies); cardiovascular and respiratory function (60 studies); posture and movement control in non-sporting contexts (62 studies); muscle activity and reflex excitability in non-sporting contexts (16 studies); impact on proprioception (23 studies); relief or prevention of pain (30 studies) and assessing pressure applied (19 studies). Smaller themes were thermal effects (12 studies); side effects of orthoses (7 studies); development of novel designs (3 studies); acceptability and compliance (3 studies) and psychological effects (3 studies).

Studies in the sport performance and recovery themes investigated only compression garments. There was a greater range of interventions used in the other themes. For example, proprioception had been investigated in both compression garments and joint supports; posture and movement control and muscle activity had been investigated across compression garments, joint supports and fabric orthoses using similar measures. Tables further describing themes, measures and study designs are included in Snowdon et al. (2018, Appendix 1).

Comparators used in effectiveness studies

In the crossover studies, the experimental intervention was compared to some sort of placebo or sham intervention in only 15 studies (7%), to a garment or orthosis of a different size in 31 studies (14%), to no garment/orthosis in 133 studies (59%) and to an active comparator in 39 studies (17%). In the RCTs, the experimental intervention was compared to some sort of placebo or sham intervention in only seven studies (18%), to a garment or orthosis of a different size in four studies (11%), to no garment/orthosis in 17 studies (45%) and to an active comparator in ten studies (26%).

Pressure measurement

In the joint support and fabric orthosis studies, pressure beneath the orthosis was measured in only one joint support study (Cholewicki, Lee, Reeves & Morrisette, 2010). Within the compression garment studies, pressure beneath the garment was measured in 30% of studies and varied between 10 and 40 mmHg. The number of studies measuring pressure changed little between 2006 and 2016.

2.4 Discussion - systematic mapping review

This review found extensive research into compression garments, joint supports and fabric orthoses. There were several common measures and uses, in particular, measurement of kinematics. There was limited use of placebo comparators in the effectiveness studies and pressure exerted by the intervention was not routinely reported. Compression garment research was characterised by robust research designs, investigating relatively short-term effects in healthy people. Joint support studies mainly investigated short-term effects in healthy people and those with musculoskeletal conditions. Fabric orthosis research was dominated by study designs with a high risk of bias. There were very few qualitative investigations.

2.4.1 Discussion of main findings regarding research design

Determining the effectiveness of all these interventions is complicated by difficulty blinding participants as to which intervention is experimental. For compression garments in sports, provision of a placebo may be impossible because many athletes are familiar with the look and feel of compression garments. Recent studies have investigated the placebo effect by manipulating participants' expectations (Mothes, Leukel, Seelig & Fuchs, 2017) or by asking participants about their beliefs (Brophy-Williams, Driller, Kitic, Fell & Halson, 2017). These approaches help to distinguish placebo effects from physiological effects. The impact of the placebo effect can be reduced by minimising use of self-report measures and including some measures for which change cannot be explained purely by a change in voluntary effort.

The importance of clear description of interventions was stressed in the TIDieR guidelines (Hoffman et al., 2014). The fact that only 30% of the compression garment studies measured pressure is a cause for concern. The proportion of studies in which pressure was measured has not increased significantly recently, despite this being strongly recommended by MacRae et al. (2011). Although Beliard et al. (2015) argued that pressure applied is not relevant to effectiveness, this claim was based upon a review of the effectiveness of a range of garments and may be complicated by the heterogeneity of the studies included and other features of garment design, such as whether pressure profiles were graduated (higher distally) or progressive (higher proximally). A number of primary studies demonstrated that physiological effects vary according to pressure applied (Bochmann, Seibel, Haase & Hietschold, 2005; Miyamoto, Hirata, Mitsukawa, Yanai & Kawakami, 2011; Coza, Dunn, Anderson & Nigg, 2012). In addition, it has been demonstrated that pressure applied is hard to predict from garment size and varies with posture and activity (de Godoy, Braile, Perez & de Godoy, 2010; Brophy-Williams, Driller, Shing, Fell & Halson, 2014a; Hill et al., 2015). Pressure applied is likely to have a bearing on comfort and acceptability of fabric orthoses. Measuring pressure between the garment and the body is recommended to describe these interventions clearly.

2.4.2 Potential effects of compression garments and joint supports - implications for fabric orthoses and future systematic reviews

No quality assessment was performed in this mapping review and publication bias is likely, therefore, no conclusions can be drawn regarding effectiveness. Nevertheless, in order to use the extensive compression garment and joint support research to inform fabric orthosis research and design, it is important to consider effects that may be relevant to clinical populations. In addition, such an analysis is important to prioritise ideas for future systematic reviews. These findings are summarised in the section below, supported where possible by other systematic reviews.

A large proportion of the studies investigating compression garments and sport performance found no evidence of effectiveness. Systematic reviews support this impression, concluding that there is moderate evidence that compression garments do not improve sport performance (Born et al., 2013; da Silva et al., 2018). In addition, da Silva et al. (2018) concluded that there was no effect on cardiovascular parameters during exercise, such as oxygen uptake or cardiac output.

Many studies investigating compression garments and recovery from exercise found the garments were effective in accelerating recovery. Although studies used garments before, during and after exercise, effects seemed more positive for after exercise, particularly where exercise was intensive or over and above what participants were accustomed to. These findings have been confirmed in systematic reviews (Brown et al., 2017; Marqués-Jiménez, Calleja-González, Arratibel, Delextrat & Terrados, 2016). The mechanism for this effect has been suggested to be improved circulation. This has not been specifically investigated but is supported by findings on muscle oxygenation. Six studies in the mapping review investigated muscle oxygenation and four found improved oxygenation beneath compression garments of 5-16 mmHg, mostly at rest (Book, Prince, Villar, Hughson & Peterson, 2016; Bringard, Denis, Belluye & Perrey, 2006; Coza et al., 2012; Dermont, Morizot, Bouhaddi, & Ménétrier, 2015). These authors suggested this effect was due to vasodilation of arterioles in response to pressure. No previous systematic reviews were found investigating muscle oxygenation, so the finding is uncertain. Other potential mechanisms for improved circulation do not appear to be supported by the research. For example, venous return does not appear to improve with compression during exercise in healthy people and, indeed, pressures over 37 mmHg might decrease blood flow at rest (Sperlich, Born, Kaskinoro, Kalliokoski & Laaksonen, 2013; Wahl, Bloch, Mester, Born & Sperlich, 2012).

Four studies investigated whether compression improved muscle strength during resistance exercise and no effect was seen in any of these studies (Gustafson, 1998; Kraemer et al., 1998a; Martorelli et al., 2015; Pereira et al., 2014). Some of the compression garments and sports performance studies that demonstrated benefit investigated repeated performance of relatively high-powered actions, such as repeated vertical jumps or intermittent sprinting (Ali, Creasy & Edge, 2011; Born et al., 2014; Higgins et al., 2009; Kraemer et al., 1996; Kraemer et al., 1998b; Rugg and Sternlicht, 2013; Sear, Hoare, Scanlan, Abt & Dascombe, 2010). This suggests that compression might decrease the impact of or development of fatigue in such activities. No previous systematic review focussed specifically on the effect of compression garments on strength or power and this may be a useful future systematic review.

Of particular relevance to neurological impairment, three studies investigated the impact of compression on reflex excitability, using the H-reflex in the calf muscle. One study investigated healthy people wearing compression socks (Espeit, Pavailler& Lapole, 2017) and two studies investigated orthokinetic compression garments exerting pressures below 32 mmHg in stroke (Ibuki, Bach, Rogers & Bernhardt, 2010a & 2010b). No change in the H-reflex was seen in any of these studies but previous research had demonstrated that the H-reflex reduced with compression between 36 and 41 mmHg in healthy people and people with neurological impairment and there was no effect with lower levels of compression (Robichaud, Agostinucci & Van der Linden, 1992; Robichaud & Agostinucci, 1996). The mechanism through which pressure might inhibit reflex excitability has been suggested to be via inhibitory sensory feedback. Such inhibition could conceivably be useful in management of spasticity or tremor but might be impractical if it is only effective at pressures over 36 mmHg.

Most published studies investigating the impact on proprioception demonstrated positive effects. The suggestion that joint supports improve proprioception was supported by a recent systematic review (Ghai et al., 2017). Whether proprioception is influenced by compression garments and whether improved proprioception carries over into improved balance, coordination or control is unclear. However, there are examples of such effects. These include improved kick coordination in healthy people in compression garments, particularly in less-skilled people (Hasan et al., 2017), improved steadiness in stance in compression leggings (Michael et al., 2014), improved control of shoulder movement in a compression top (Tsuruike and Ellenbecker, 2013), enhanced postural stability in compression socks in people with ankle sprains (Genthon et al., 2010) and improved balance and gait kinematics in people with knee osteoarthritis wearing joint supports (Bryk et al., 2011; Chuang et al., 2007; Schween et al.,2015).

Overall, the compression garment and joint support research suggests that there may be relevant effects for improving proprioception, coordination, recovery from exercise and circulation in muscles at rest. Positive benefits during exercise appear less likely. Future systematic reviews may be helpful for investigating the effect on muscle oxygenation, proprioception in compression garments and the impact of compression garments on strength and power. In addition, a narrative review exploring potential effects and their mechanisms would be valuable. MacRae et al. published such a review in 2011 but the research has expanded significantly since it was conducted.

2.4.3 Key gaps in primary research

The mapping review identified key gaps in primary research. Firstly, fabric orthosis research utilised less robust study designs. The majority of the fabric orthosis research investigated the longer-term effects of the orthoses as therapeutic interventions, despite an orthotic impact never having been established. It could be argued that if no short-term orthotic effect exists, orthoses are unlikely to have a longer-term therapeutic effect. For other orthotic interventions, notably Functional Electrical Stimulation (FES) for weak dorsiflexion, effects are investigated for both the immediate orthotic and longer-term effect (Street, Swain & Taylor, 2017). Street et al. (2017) defined "direct orthotic effect" as the immediate change seen in movement as soon as an orthosis is applied; "training effect" refers to changes over time caused by wearing the orthosis (assessed without the orthosis) and "total orthotic effect" is the combined effect of wearing the orthosis for a "training" period plus actually wearing the orthosis during testing. These definitions appear to be well accepted in the wider orthotic literature but have not been used in previous fabric orthosis studies. Adopting this terminology should improve clarity around the aim of fabric orthosis studies.

The impact of fabric orthoses on proprioception has only been investigated in one study (Gracies et al., 2000), which used a method with unknown reliability. Although, as discussed earlier, improvement in proprioception could theoretically improve function, this effect should not be assumed. Proprioception studies usually test proprioception at a cognitive level and in relatively artificial contexts. This may not be functionally relevant. In this mapping review, only four out of the 23 proprioception studies assessed variables in addition to proprioception and none of these demonstrated relationships between improved proprioception and improved function (Baltaci et al., 2011; Barrett, 2003; Marchini, Lauermann, Minetto, Massazza & Maffiuletti, 2014; Webster et al., 2017). Future research is recommended assessing both proprioception and function in fabric orthoses and investigating the relationships between these variables to determine whether fabric orthoses do indeed alter proprioception and, if so, whether this has any functional relevance.

Qualitative research has a role in explicating user perspectives, and this would enable in-depth understanding of the acceptability and feasibility of these interventions. Early research into fabric orthoses (e.g. Blair et al., 1995) collected feedback using "user comments" and unvalidated questionnaires, and found very low levels of acceptability, with participants finding orthoses to be hot, uncomfortable and difficult to get on and off. More recent studies used properly designed qualitative interviews and these enabled the issues around acceptability to be better understood (Stone, 2014; Miller et al., 2016). Further qualitative investigations are required to fully understand acceptability of fabric orthoses generally and of each specific type of fabric orthosis.

2.4.4 Strengths and limitations

This review followed guidelines on mapping review methods to identify and describe research on similar interventions from a range of disciplines and perspectives. Excluding studies on oedema and venous disorders may have limited the review, as there may have been studies relevant to mobility, movement and function in this wider body of literature. Additionally, some of the studies used ambiguous terminology to describe the interventions, such as sleeves, pantyhose and tights. The diversity of terms used to describe these interventions may have prevented some studies from being identified. The accuracy of data extraction was checked by one reviewer (NS), who referred repeatedly back to the original studies during the theming/categorising process; however, using multiple reviewers to monitor consistency of data extraction would have been more robust. Although usual for a mapping review, the lack of critique means that conclusions about the quality of this research evidence cannot be made.

2.5 Methods – effectiveness systematic review

2.5.1 Search strategy and study selection

The search and initial study selection, described in Sections 2.2.1 and 2.2.2, identified 42 studies investigating fabric orthoses. Those studies were included in this systematic review if:

- participants included adults with any neurological impairment
- robust designs were used to investigate efficacy or effectiveness, specifically either RCTs or crossover trials.

Single case experimental designs (SCEDs) were considered but excluded. Whilst a well-designed SCED study can demonstrate effectiveness in individuals (Tate et al., 2008) and they have an important role in investigating interventions in rare conditions (Guyatt et al., 1990), they provide very limited evidence because they cannot demonstrate lack of effectiveness and findings can often not be generalised to a wider group.

Studies were screened and selected by one reviewer (NS).

2.5.2 Data extraction

One reviewer (NS) extracted data relating to study design, sample size, participant group, intervention, outcome measures and results.

2.5.3 Appraisal of risk of bias

Appraisal was undertaken independently by two reviewers (NS and TP) who completed risk of bias (RoB) proformas. TP had worked on previous systematic reviews, so had experience of review methods and RoB appraisal. Reviewers recorded their judgement (high, low or unclear RoB) for each domain and their reasoning behind each judgement. Once all the studies were independently assessed, completed proformas were compared and decisions discussed. Consensus was reached through discussion.

RCTs were assessed using the original version of the Cochrane RoB tool (Higgins, Altman, & Sterne, 2011). Crossover studies were assessed using the method recommended by Ding et al. (2015), which adds specific criteria to the Cochrane RoB tool to make the appraisal more specific to crossover studies. The criteria added are "is a crossover design appropriate?", "is the treatment order randomised?", "have the authors checked whether there is a carry-over effect?" and "are data for every period of the study available?" In appraising the crossover trials, allocation concealment and assessor blinding were considered to relate to allocation to the order of treatment.

2.5.4 Data synthesis

Studies identified differed in terms of the interventions, measures and conditions investigated. Therefore, meta-analysis was inappropriate and a narrative synthesis approach was used according to the guidance published by Popay et al. (2006). Popay et al. (2006) suggested using a framework involving four elements. These are (1) developing a theory of how the intervention works, why and for whom; (2) developing a preliminary synthesis of findings of included studies; (3) exploring relationships within and between studies and (4) assessing the robustness of the synthesis. In the current review, the first element was addressed by re-reading each study to determine theories informing use of the interventions. Authors stated justification for testing the intervention was considered and these were compared to the measures they chose to use, the period over which the intervention was assessed and the discussion of their findings. A preliminary synthesis of the findings of the studies was achieved by tabulating the study characteristics and study findings. Studies were compared and grouped according to interventions, participants, the stated aims of the interventions and the outcome measures used and the information from these elements was used to explore relationships between studies.

An overall assessment of the strength of the evidence was produced using the terminology and definitions suggested in the GRADE guidelines (Guyatt et al., 2011a). These classify quality of evidence using four categories: high, moderate, low and very low. An evidence profile was created for each of the variables measured in the included studies that appeared to be important, meaning either that it had been assessed in multiple studies or had clear clinical relevance. The evidence profiles were informed by the Cochrane RoB assessment. This was reviewed to determine a RoB at the level of the variable rather than the study (Guyatt et al., 2011a). GRADE guidelines suggest that in addition to the RoB assessment, evidence is judged according to the consistency of the findings across studies, the directness of the evidence, the precision of the results and the likelihood of publication bias (Guyatt et al., 2011a). In this review, this was simplified to only consistency and precision. Directness did not seem relevant because there were no issues with the generalisability of the samples and, whilst variables were measured at different levels of the International Classification of Functioning, Disability and Health (ICF; World Health Organisation, 2001), none might be considered as “surrogate markers” (Guyatt et al., 2011b). Publication bias was not considered because of difficulty determining how likely this may have been for individual studies. Summary statements for quality of evidence were obtained by starting with a rating of high quality and grading down two levels for high risk of bias, one level for unclear risk of bias and additional levels if the evidence available was considered inconsistent or imprecise.

Relationships were explored within and between studies by exploring how study findings related to the underlying theories described and the quality of the evidence. Finally, the robustness of the synthesis was reviewed by critically reflecting upon the process and outcome of the review.

2.6 Results – effectiveness systematic review

2.6.1 Study selection

Studies were selected from the first search, performed in December 2015 and papers were appraised and reviewed at that point. The later search in June 2017 did not locate any additional studies that would have met the inclusion criteria. Stone (2014) was reviewed using the author's thesis and personal communication. The remaining five papers were reviewed from the published articles alone.

Study selection is shown in Figure 2.2. Six studies were identified investigating the impact of fabric orthoses in adults with neurological impairments.

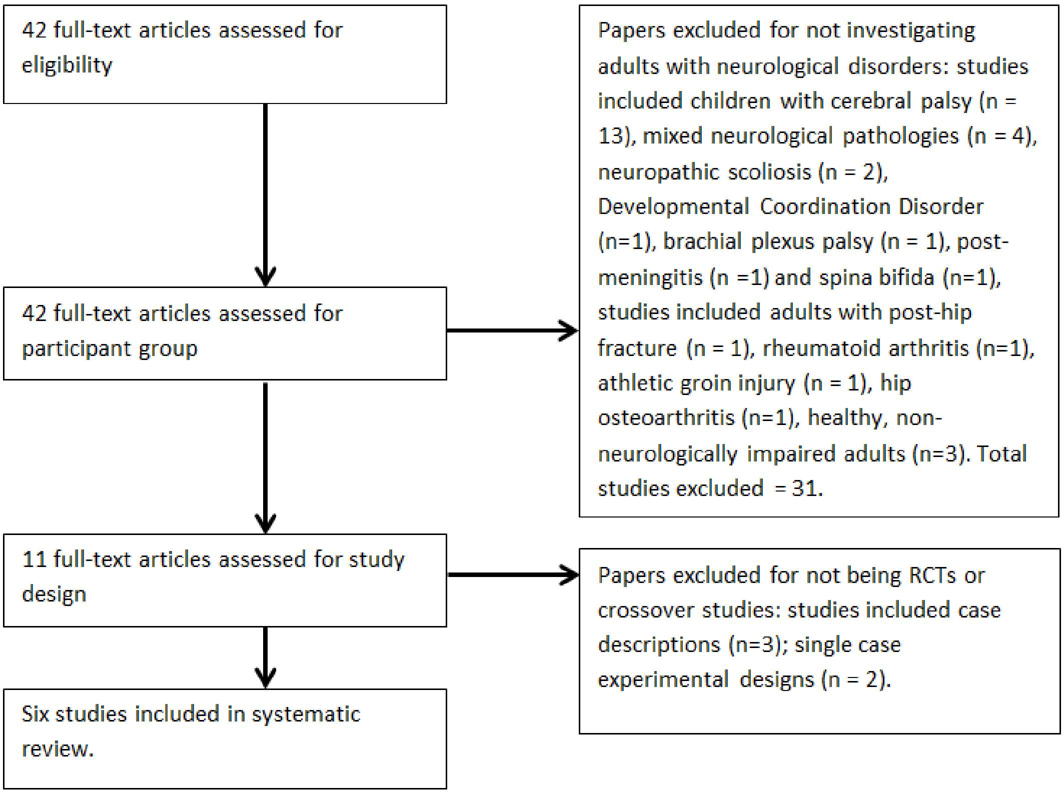


Figure 2.2: Study selection for systematic review

2.6.2 Study description

The six studies included two RCTs and four crossover studies (Table 2.3). Four studies investigated stroke survivors (Gracies et al., 2000; Ibuki et al., 2010a & 2010b; Maguire et al., 2010); one investigated PwMS (Miller et al., 2016) and one investigated people with focal spasticity, meaning spasticity that mostly affected one part of the body as opposed to the whole of a limb (Stone, 2014). Stone (2014) included people with a range of underlying conditions, including 18 stroke survivors and one person with MS. Three studies investigated upper limb sleeves with or without gloves (Gracies et al., 2000; Miller et al., 2016; Stone, 2014), three investigated socks (Ibuki et al., 2010a & 2010b; Stone, 2014) and one investigated TheraTogs^{TM}, which were a pair of shorts with additional straps to promote specific support for hip abduction (Maguire et al., 2010).

The two RCTs (Miller et al., 2016; Stone, 2014) were both designed as mixed methods feasibility studies and were the only studies to assess outcome measures at all three levels of the ICF (World Health Organisation, 2001). The two RCTs differed from the crossover studies in the period of time over which the intervention was used. Both RCTs investigated their interventions over a number of weeks, whereas all the crossover studies assessed either immediate effects or short-term effects.

The majority of studies investigated the impact of the intervention on spasticity (Gracies et al., 2000; Ibuki et al., 2010a & 2010b; Stone, 2014). One study investigated the impact of the intervention on upper limb cerebellar tremor and ataxia (Miller et al., 2016), whereas Maguire et al. (2010) investigated the effect of the intervention on neurological muscle weakness and walking ability following stroke.

Overall, it can be seen that the research is extremely heterogeneous in terms of the interventions, measures used and participants included. The only area of research with findings from more than one study involves the impact of fabric orthoses on spasticity following stroke. However, these studies differ regarding whether they have investigated long-term or short-term effects and upper limb or lower limb spasticity, both of which potentially influence their findings.

Table 2.3: Studies investigating the use of elastic garments and fabric orthoses in neurologically impaired adults

Author and year	n	Study design	Participants	Orthosis	Procedure	Measures			
						Body functions and structures	Activity	Participation and quality of life	Other
Gracies et al. 2000	16	Crossover trial	Stroke	Lycra sleeve axilla to wrist, over-lapping Lycra glove with boning mid-forearm to palm; Second Skin.	Assessed before and after 3 hour wear period on different days; change in measures compared between the day the splint was worn and the day the splint was not worn.	Limb circumference, posture, Tardieu spasticity scale, active ROM, proprioception, line bisection	Participant rated effect on function – walking, toileting, eating, dressing, ease of movement		Participant rated effect on comfort and confidence
Ibuki et al. 2010a	13	Crossover trial	Stroke	Orthokinetic Compression Garments, two different levels of compression	Shoes only, Dynamic Foot Orthosis, range of motion walker and two Orthokinetic Compression Garments assessed on same day in standing in random order.	Soleus H-reflex in standing			
Ibuki et al. 2010b	15	Crossover trial	Stroke	Orthokinetic Compression Garments	Shoes only, tone-reducing AFO, hinged AFO, Orthokinetic Compression Garments and combination assessed on same day in standing in random order.	Soleus H-reflex in standing			
Maguire et al. 2010	13	Crossover trial	Stroke	Shorts with hip abductor strapping, TheraTogs^{TM}	Taping, TheraTogs^{TM}, walking with and without stick assessed in walking on the same day in random order.	Hip abductor activity in walking (EMG)	Walking speed and symmetry		
Miller et al. 2016	21	RCT/ mixed methods study	Multiple sclerosis	Full-length sleeve, wrist crease to axilla; Jobskin UK.	Compared compressive sleeve with non-compressive sleeve worn eight hours a day for nine weeks.	FAHN A – tremor amplitude at rest	FAHN B – pouring water and drawing, FAHN C – self-report measure of ADL, ARAT, 9HPT	Canadian Occupational Performance Measure	Psychological Impact of Assistive Devices Scale
Stone, 2014	25	RCT/mixed methods study	Spasticity, stroke (*n* = 18), MS (*n* = 1)	Sleeve with glove (*n* = 22) or sock (*n* = 3), DM Orthotics.	Orthoses used following Botox for 6 weeks, assessed at baseline, 6 and 12 weeks.	VAS for pain, ArmA (self-report), LASIS (self-report)	10-metre walk test for participants provided with sock	EQ-5D	Goal Attainment Scale

(AFO = Ankle foot orthosis; ARAT = Action Research Arm Test; ArmA = Arm Activity measure; Botox = Botulinum toxin; EMG = electromyography; FAHN = Fahn-Tolosa-Marin Tremor Rating Scale; LASIS = Leeds Arm Spasticity Impact Score MS = multiple sclerosis; RCT = randomised controlled trial; ROM = range of movement; VAS = Visual Analogue Scale; 9HPT = Nine-hole Peg Test.)

2.6.3 Quality of evidence

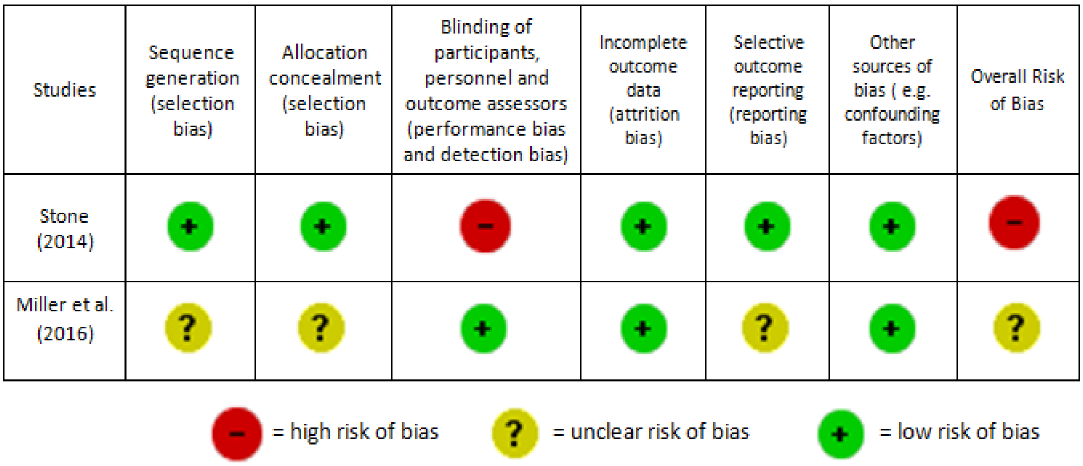
Risk of bias appraisal

The results of the RoB appraisals are shown in Tables 2.4 and 2.5. Most studies were found to be at high risk of bias and none was judged at low risk of bias. The most problematic areas were blinding and allocation concealment. Blinding was judged to result in high risk of bias in two studies. In Stone (2014), this was because the primary outcome measure was a self-report measure, the participants were not blinded to group allocation and the control group received usual care alone. This may have resulted in assessor and performance bias. In Gracies et al. (2000), there was no attempt to blind the assessor, many measures were assessor-rated and assessments were performed when the orthosis was worn and visible to the assessor. Allocation concealment was not done in any of the crossover studies and was unclear in one of the RCTs. Allocation concealment is important because without it, randomised allocation to groups in RCTs appears not to work, groups are more likely to be unequal at baseline and trials are more likely to produce positive findings (Moher et al., 1998). It could be argued that this problem is less likely to happen in a crossover study where all participants receive all interventions and this may be why it was overlooked in these studies. Nevertheless, Ding et al. (2015) consider allocation concealment important in a crossover study, as an integral part of the blinding process.

Selective outcome reporting was considered to be present in three of the studies because there was no published protocol and authors did not clearly state that they had reported all of the variables that had been assessed. The Miller et al. (2016) RCT was considered at unclear risk of selection bias. Both of these issues relate to the level of detail included within the study reports and may result from journal word count limitations or the authors' lack of awareness of the standards implied by the RoB assessment tools.

A further issue across all crossover studies was the lack of assessment of carry-over. Ding et al. (2015) require researchers to re-assess each participant at baseline prior to testing in the second or subsequent conditions. Gracies et al. (2000) assessed baseline on each day but did not consider whether baseline was similar on the two days. The other crossover studies did not reassess baseline between the different conditions, so researchers were unable to confirm whether changes from the first condition had carried over into the next.

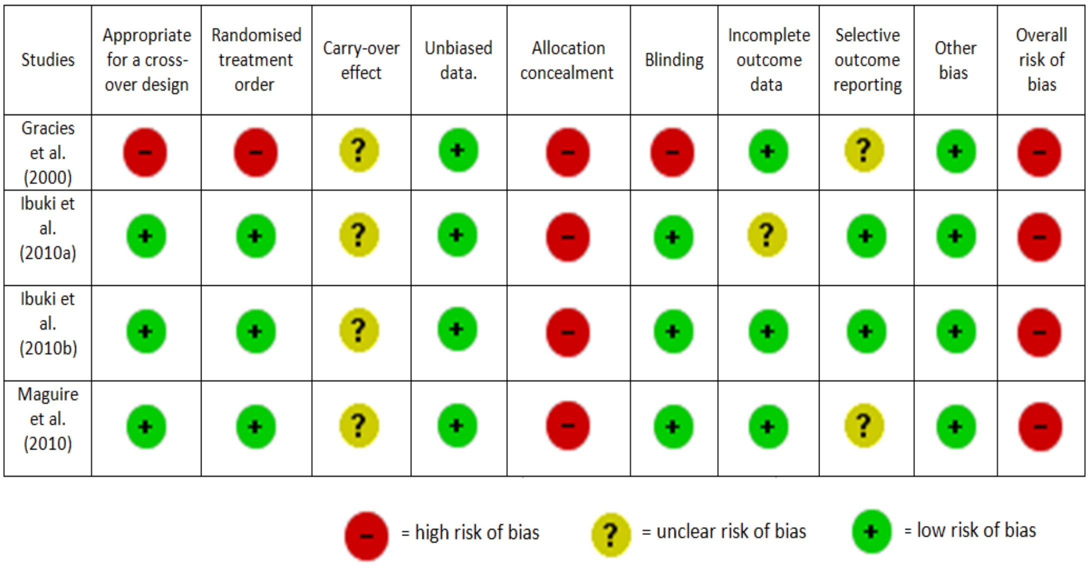
Table 2.4: Completed Cochrane Risk of Bias assessment for RCTs



Studies	Sequence generation (selection bias)	Allocation concealment (selection bias)	Blinding of participants, personnel and outcome assessors (performance bias and detection bias)	Incomplete outcome data (attrition bias)	Selective outcome reporting (reporting bias)	Other sources of bias (e.g. confounding factors)	Overall Risk of Bias
Stone (2014)	+	+	−	+	+	+	−
Miller et al. (2016)	?	?	+	+	?	+	?

− = high risk of bias ? = unclear risk of bias + = low risk of bias

Table 2.5: Completed Risk of Bias tool for crossover studies (Ding et al., 2015)



Studies	Appropriate for a cross-over design	Randomised treatment order	Carry-over effect	Unbiased data.	Allocation concealment	Blinding	Incomplete outcome data	Selective outcome reporting	Other bias	Overall risk of bias
Gracies et al. (2000)	−	−	?	+	−	−	+	?	+	−
Ibuki et al. (2010a)	+	+	?	+	−	+	?	+	+	−
Ibuki et al. (2010b)	+	+	?	+	−	+	+	+	+	−
Maguire et al. (2010)	+	+	?	+	−	+	+	?	+	−

− = high risk of bias ? = unclear risk of bias + = low risk of bias

Evidence profile using GRADE guidelines

Table 2.6 shows an overview of the findings for each study and Table 2.7 presents an overall evidence profile for the variables investigated in this systematic review.

Table 2.6: An overview of study findings from the systematic review

Author and year	What appeared to respond to intervention?	What appeared not to respond to the intervention?
Gracies et al. 2000	Resting posture at wrist with less flexion, decreased limb circumference, spasticity in wrist and finger flexors as assessed using range at which muscles exhibited stretch reflex, passive range of shoulder movement – all above measures showed statistically significant differences; participant perceived confidence in the arm better with intervention	Spasticity at the shoulder and elbow, proprioception, line bisection, participant reported function – all showed no significant difference, active range of finger movement significantly decreased with glove on
Ibuki et al. 2010a	One participant showed a potentially important decrease in soleus H-reflex	Soleus H-reflex in standing - no significant change at group level
Ibuki et al. 2010b	Two participants showed a potentially important decrease in soleus H-reflex wearing the orthokinetic sock and an AFO with a tone reducing footplate	Soleus H-reflex in standing - no significant change at group level
Stone, 2014	GAS - statistically significantly better in the intervention group with a very large effect size, caring for and using the affected arm (ArmA and LASIS; *n* = 22) - no significant difference between groups but small to moderate effect sizes favouring the intervention at 6 weeks, 10MWT - improved at six weeks (*n* = 3)	Pain – no significant difference between groups but large effect size at 6 weeks, quality of life (EQ-5D) – no significant difference between groups and very small effect size
Miller et al. 2016	FAHN Subset A (tremor at rest) - statistically significantly improvement in the intervention group, total FAHN score - statistically significantly improvement in the intervention group	FAHN - total and subset scores no significant difference between treatment and placebo, 9HPT - no significant difference between groups and no within-group change over time, COPM - placebo was significantly more effective than the intervention, maybe related to differences in severity of the two groups; PIADS - no statistically significant difference between the groups and no perceived impact upon quality of life
Maguire et al. 2010	Hip abductor activity in walking (EMG) - significantly higher in TheraTogs^{TM} compared to walking stick with a moderate effect size, Muscle activity in tensor fascia lata (EMG) - significantly higher in TheraTogs^{TM} step length symmetry - slightly improved in TheraTogs^{TM}	Walking speed - better with TheraTogs^{TM} but not statistically significant and differences appear small, step time symmetry – spends less time on hemiplegic leg with TheraTogs^{TM} but not statistically significant

(AFO = Ankle foot orthosis; ARAT = Action Research Arm Test; ArmA = Arm Activity measure; COPM = Canadian Occupational Performance Measure; EMG = electromyography; FAHN = Fahn-Tolosa-Marin Tremor Rating Scale; GAS = Goal Attainment Scale; LASIS = Leeds Arm Spasticity Impact Score; PIADS = Psychological Impact of assistive devices Scale; 9HPT = Nine-hole Peg Test; 10MWT = 10-metre Walk Test.)

Table 2.7: Evidence profile for important variables investigated with systematic review, based upon the GRADE guidelines (Guyatt et al., 2011a)

Variable	Condition	Studies	Risk of bias	Inconsistency	Imprecision	Summary of findings
Self-reported function	Upper limb spasticity	1 crossover study and 1 RCT	High on RoB + crossover used a non-validated measure	Findings inconsistent, better quality study showed more effectiveness. RCT showed improvements on several self-report measures, crossover study suggested no difference in function	Findings imprecise - confidence intervals not provided, statistically significant effect shown in the RCT with a large effect size	There is low quality evidence that fabric orthoses might improve self-reported function in people with spasticity affecting the upper limb
	Cerebellar tremor in multiple sclerosis	1 RCT	Unclear on RoB + Groups unequal at baseline	Only one study available	Only one study available. Findings statistically significant with a moderate effect size favouring placebo	There is low quality evidence that a placebo orthotic sleeve is more effective at improving self-reported function than an orthotic sleeve in cerebellar tremor
Reflex excitability	Spasticity following stroke	3 crossover studies	High on RoB	Findings inconsistent, better quality studies showed no effect but applied orthosis for less time than the poorer quality study, which showed some effect. Better quality studies used a more objective measure of reflex excitability	Three studies available, unclear whether Ibuki et al. (2010a and 2010b) used same sample in both studies, no sample size calculations, effect sizes or confidence intervals to judge precision of findings	There is very low quality evidence that an orthotic glove might decrease reflex excitability, if it directly influences joint alignment and is worn for at least 3 hours. There is low quality evidence that an orthotic sock is ineffective in producing any immediate decrease in reflex excitability
Upper limb cerebellar tremor	Multiple sclerosis	1 RCT	Unclear on RoB + Groups unequal at baseline	Only one study available	Only one study available, no significant difference between groups but no sample size calculation performed and groups unequal at baseline, effect sizes not presented for this variable	There is low quality evidence that an orthotic sleeve is ineffective at improving cerebellar tremor in multiple sclerosis
Active voluntary movement	Upper limb spasticity	1 crossover study	High on RoB + included people with no spasticity	Only one study available	Only one study available, no sample size calculation, no effect sizes presented.	There is very low quality evidence that an orthotic glove is ineffective at improving wrist and finger active extension and decreases active range of finger flexion
	Hip abductor weakness following stroke	1 crossover study	High on RoB	Only one study available	Only one study available, no sample size calculation, statistically significant different in hip abductor activity seen with moderate effect size, statistics and effects sizes not reported for walking speed or symmetry.	There is low quality evidence that TheraTogs^{TM} shorts can increase hip abductor activity in walking compared to no walking stick or with a walking stick

(RoB = risk of bias; RCT = randomised controlled trial)

Variables chosen for the evidence profile were self-reported function, reflex excitability, cerebellar tremor and active voluntary movement (Table 2.7). Self-reported function and reflex excitability were assessed in more than two studies. Active voluntary movement was reported in two studies. Cerebellar tremor was only investigated in one study (Miller et al., 2016) but it was felt that this was a clinically important variable. Variables of passive range of movement and proprioception were considered for inclusion in the evidence profile but these were only assessed by Gracies et al. (2000). Because the Gracies et al. (2000) study was at high risk of bias for multiple domains, it was felt that there was insufficient evidence available to make any judgements regarding the impact of fabric orthoses on these variables.

In the following sections, the conclusions formed using the evidence profile will be examined in relationship to one another and to the theoretical perspectives discussed in the studies.

2.6.4 The impact of fabric orthoses on spasticity

Four of the studies in this review examined the effectiveness of fabric orthoses with people with spasticity. Three variables in the evidence profiles were relevant to spasticity. These were self-reported function, reflex excitability and active voluntary movement.

Self-reported function was assessed by Gracies et al. (2000) and Stone (2014), who both focussed on the upper limb following stroke. Gracies et al. (2000) showed no change in function but used an unvalidated measure and three-hour intervention period, which could be too short for participants to explore any changes in function. Stone (2014) used several validated measures. She found large effect sizes and statistically significant change; however, there was a lack of blinding. Thus, there is only low quality evidence that fabric orthoses might improve self-reported function in stroke survivors with spasticity in the upper limb and it is important to note that Stone (2014) used fabric orthoses as an adjunct to Botulinum Toxin injections (Botox).

Reflex excitability was assessed by Gracies et al. (2000) and Ibuki et al. (2010a & 2010b). The Ibuki et al. (2010) studies were of better quality but all were at high risk of bias. It could be argued that the lack of allocation concealment is unimportant in the

Ibuki studies but they did not attempt to demonstrate lack of carry-over effect, which may have had an important influence due to the short period of time during which the different interventions were tested. Because the Gracies et al. (2000) and the Ibuki studies used different interventions, on different body parts for different lengths of time, their findings cannot be combined into one synthesis. The evidence profiles (Table2.7) suggest that there is very low quality evidence that an orthotic glove worn for three hours might decrease reflex excitability and there is low quality evidence that an orthotic sock is ineffective in producing any immediate decrease in reflex excitability.

Only the Gracies et al. (2000) study investigated active voluntary movement in spasticity. They suggested that there was no improvement in wrist and finger extension with the orthosis and that there was a loss of active flexion. However, in addition to the significant quality issues described in Table 2.5, this study appeared to include stroke survivors who did not have spasticity whose otherwise relatively unimpaired active finger flexion was reduced by the glove. Therefore, there is very low quality evidence that an orthotic glove is ineffective at improving wrist and finger active extension and decreases active range of finger flexion.

Each study applied the fabric orthoses for different durations: Stone (2014) for 6 weeks of regular wear, Gracies et al. (2000) for 3 hours and Ibuki et al. (2010a & 2010b) only minutes before testing took place. The reasons for these different approaches can be seen in their discussion of the theories around spasticity and around how the orthoses might work. All discussed that spasticity might be decreased via biomechanical or neurophysiological mechanisms. Gracies et al. (2000) explained that lengthening a muscle could inhibit spasticity and they implied that there had to be a biomechanical effect before any neurophysiological effect was possible. They suggested that rigid splints might be equally effective if they could be tolerated. Their use of a 3-hour wear period before assessment was because they assumed the orthosis would need to apply a stretch for some time before there might be any benefit. Stone (2014) explained neuroplasticity and muscle plasticity in her explanation of how orthoses might influence spasticity. She considered muscle lengthening, low-level stretch and optimal positioning to be important in spasticity management but suggested that for spasticity to improve, movement patterns need to adapt over time. She considered that a flexible support would promote motor learning because it allowed movement and sensory feedback. This plasticity hypothesis is in keeping with the decision to trial the orthoses over an extended period. In contrast, Ibuki et al. (2010a and 2010b) argued that neurophysiological and biomechanical mechanisms of decreasing spasticity are separate. Their work was prompted by wanting to determine whether orthotic features believed to have a specific neurophysiological effect really worked. They measured the neurophysiological effect directly using an H-reflex to determine if reflex excitability changed and hypothesized that if the α-motor neurone were inhibited by the fabric orthosis, then this effect would be seen immediately, as soon as the orthosis was applied.

Because these three research groups have taken such different approaches, it is difficult to combine their findings. In a way, the findings of Ibuki et al. (2010a & 2010b) and Gracies et al. (2000) agree that the biomechanical effect of the orthoses may be more important than any neurophysiological effect. Gracies et al. (2000) argued that the fact that they found less of an effect on spasticity in the forearm supinators compared to the hand and wrist flexors indicated that the orthoses only worked where the Lycra contained flexible plastic “boning”. However, there are other potential reasons for such a finding, such as the relative accuracy of reflex testing at the wrist and forearm. Similarly, whilst Ibuki et al. (2010a & 2010b) showed no immediate change in reflex excitability, the basis of their assumption that any neurophysiological changes would be immediate is not clear.

Overall, only low quality, contradictory evidence has been published investigating effectiveness of fabric orthoses for managing spasticity and no clear clinical recommendations can be drawn from this evidence. The lack of consistency may be related to the diversity of theories used to understand spasticity and fabric orthoses. The large difference in self-reported function seen in the Stone (2014) study suggests there may be potential for fabric orthoses to be used as an alternative for rigid splints as an adjunct to Botox injection but further research that makes use of objective measurement alongside self-reported function would be required to develop theory and influence practice.

2.6.5 The impact of fabric orthoses on cerebellar tremor

Only Miller et al. (2016) investigated the impact of an orthotic sleeve on cerebellar tremor and ataxia. The variables of self-reported function and tremor severity have been examined within the evidence profiles. This study was found to be at unclear risk of bias due to a relative lack of reporting of randomisation, allocation concealment and selective outcome reporting and these are relevant to both of these variables. However, the study’s findings were that the placebo intervention was more effective than the orthotic sleeve for self-reported function and that the sleeve was ineffective for improving severity of cerebellar tremor, both of which might be suggested to be opposite to what might have been found if bias really were an issue in this study. There were some within-group improvements in tremor severity; any between-group effects may have been masked by a lack of baseline similarity. Furthermore, the qualitative data suggested that the placebo sleeve might have influenced tremor. The pressure applied by the two sleeves was not measured so it is unclear how different they were.

The authors suggested that the orthoses might work via both a proprioceptive and a biomechanical mechanism. A potential role for the orthoses in inhibiting reflex excitability was considered relevant. Neither explanation is in keeping with the decision to have participants wear the sleeve for nine weeks before assessment. The intervention period suggests that the authors were expecting a training effect but this was not discussed.

Overall, there is low quality evidence that fabric orthoses may not be effective for cerebellar tremor and ataxia but significant methodological issues around the choice of the placebo comparator and baseline similarity could have obscured potential treatment effects.

2.6.6 The impact of fabric orthoses on neurological weakness

Two studies investigated the impact of a fabric orthosis on muscle weakness following stroke but in very different functions and with very different hypotheses. Gracies et al. (2000) investigated muscle weakness as one variable in their study looking at inhibiting spasticity with an orthotic glove and this has been described in Section 2.6.4. Maguire et al. (2010) investigated TheratogsTM orthotic shorts for walking following stroke.

Maguire et al. (2010) had a very different theoretical approach to the other studies in this review. They explained that walking aids used during stroke rehabilitation might inhibit muscle activity around the hip and therefore limit the recovery of hip abductor activity and normal balance mechanisms. Their theoretical discussion is the only one reviewed that did not mention a sensory or proprioceptive mechanism. Instead, they hypothesized that the Theratogs^{TM} shorts would support the hip without inhibiting muscle activity and this appeared to be what was found in the study, albeit with some uncertainty around the effect on walking pattern.

Similar to the other crossover studies, Maguire et al. (2010) were at high risk of bias due to lack of allocation concealment and a potential carry-over effect from one condition to the next. Although, all their measures were objective a possible carry-over effect might have influenced their results. Overall, it can be concluded that there is low quality evidence that TheraTogs^{TM} shorts can increase hip abductor activity in walking in stroke survivors compared to no walking stick or with a walking stick.

2.7 Discussion – effectiveness systematic review

2.7.1 Summary of main findings

This review has shown that there remains very limited research into fabric orthoses in adults with neurological problems. The studies that have been performed are at high or unclear risk of bias and no application of the orthoses has been tested by more than one research group. Theories suggested explaining potential effects varied across the studies reviewed, with no one explanation being common to all six studies, even where those studies were investigating the same problem. Individual GRADE guidelines explain that a rating of “low quality of evidence” indicates “our confidence in the estimate is limited. The true effect may be substantially different from the estimate of the effect” (Balshem et al., 2011a, p404). This is a fitting summary of the findings of this systematic review.

All of the studies reviewed can be considered "pre-efficacy" or early efficacy studies (Robey and Schultz, 1998) in that they have relatively small sample sizes, test new hypotheses and specify objectives relating to testing feasibility or safety of their interventions.

2.7.2 Discussion of main findings

Because of the cautious nature of the findings suggested above, the clinical findings will not be discussed further here. However, there are some key lessons for future research within this systematic review. These include the need to design and write-up research with due attention to accepted quality standards such as (1) full description of interventions, (2) full description of randomisation processes, (3) inclusion of a clear statement that all planned measures have been analysed and (4) assessment of whether carry-over has occurred in a crossover study, prior to commencing any subsequent conditions. In addition, the review confirms the difficulties of using a placebo comparator in fabric orthosis studies and reminds us of the need to include objective measures alongside self-report measures, particularly because of the difficulties inherent in blinding participants in fabric orthosis studies. It illustrates the importance of theory in clarifying the logic within a research study and aligning measures, intervention period and intervention design to that theory.

This review has not been updated since it was first performed in December 2015. Citation alerts and a database search for more recent studies identified two new single group pre-test post-test studies in adults with learning disabilities (Finlayson, Crockett, Shanmugam & Stansfield, 2018) and degenerative cerebellar ataxia (Serrao et al., 2017) but no additional studies that would have met the inclusion criteria for this review.

2.7.3 Strengths and limitations of the systematic review

Review and appraisal of the published papers was not supported by contact with the researchers' themselves. This might have clarified some of the methodological issues that appeared unclear in the papers.

Use of both RoB tools was challenging in that the initial judgements made on a number of domains differed between the reviewers. For the RCTs, there was initial disagreement on allocation concealment, selective outcome reporting and "other bias". For the crossover studies, there was low initial agreement on allocation concealment, unbiased data, blinding and selective outcome reporting. Challenges related to lack of clear descriptions in the study reports of allocation concealment, lack of protocols available to assess whether there had been any selective outcome reporting and uncertainty as to where baseline similarity fitted within the tool. Challenges were resolved with discussion but similar challenges were reported in other reviews, suggesting low reliability of the RoB tools (Armijo-Olivo et al., 2014; Hartling et al., 2009; Jargensen et al., 2016). For example, in systematic reviews of the effectiveness of compression garments for recovery from exercise, different reviews included contradictory judgements around allocation concealment for the same primary studies (Marqués-Jiménez, et al., 2016; Hill, Howatson, van Someren, Leeder & Pedlar, 2014). The Cochrane RoB tool has been updated since our review was conducted. The new version (RoB 2.0) has a section specifically designed for crossover studies, clarifies the assessment of allocation concealment, has assigned less importance to a published protocol to assess selective outcome reporting and offers additional guidance around deciding on the potential impact of blinding on the study results (Higgins et al., 2016). Although, these changes make the tool easier to use, they are unlikely to alter the overall judgement of RoB for the studies in this review.

The narrative synthesis guidelines were extremely valuable in ensuring that the evidence synthesis was performed systematically with sufficient depth to highlight common problems and investigate underlying theory. The small number of studies, their low quality and heterogeneity have meant that there is insufficient evidence to draw clinically relevant conclusions. The GRADE guidelines were valuable in providing clear definition of the quality of the evidence but they have been applied in this review to variables investigated by only one study in one context and this is not the true purpose of the GRADE guidelines (Guyatt et al. (2011a).

2.8 Implications for the thesis

The systematic mapping review has directly contributed to knowledge by comparing and contrasting research investigating compression garments, joint supports and fabric orthoses, suggesting gaps in primary research and systematic reviews. In addition, both reviews have drawn attention to key research design issues that needed to be heeded in the remainder of the doctoral programme. Specifically, they drew attention to the lack of qualitative research in the field, which is extremely important given the controversies around acceptability of fabric orthoses. This inspired the first primary study in this thesis, a qualitative study investigating the perceptions of PwMS who were long-term users of fabric orthoses. In addition, the reviews demonstrated the importance of crossover designs and enabled familiarity with the relevant quality criteria for designing a crossover study. Finally, the reviews highlighted the need to describe an intervention clearly, including pressure applied.

2.9 Conclusion

Chapter 2 has provided an overview of the previous research into fabric orthoses and the related interventions of joint supports and compression garments. Both reviews demonstrated the low quality of research investigating fabric orthoses with a predominance of small-scale feasibility or "pre-efficacy" studies and insufficient use of qualitative methods.

The systematic review demonstrated that fabric orthoses have most commonly been researched in adults with neurological impairment for the management of spasticity and that applications in spasticity vary, partly due to different underlying theories of change. In terms of the effect on spasticity, there is: (1) low quality evidence that fabric orthoses might improve self-reported upper-limb function when used as an adjunct to Botox; (2) very low quality evidence that an orthotic glove might decrease reflex excitability, if it directly influences joint alignment and is worn for at least 3 hours and (3) low quality evidence that an orthotic sock applying less than 32 mmHg in pressure is ineffective in immediately decreasing reflex excitability. There is low quality evidence that fabric orthoses may be ineffective for cerebellar tremor and low quality evidence that Theratogs^{TM} shorts can increase hip abductor activity in walking in stroke survivors compared to no walking stick or with a walking stick.

The subsequent chapter in this thesis presents the qualitative investigation performed into the perceptions and experiences of PwMS who were long-term users of fabric orthoses.

Chapter 3: An Interpretative Phenomenological Analysis study investigating experiences of fabric orthoses in long-term users with MS - the FabO IPA study

Summary

This chapter reports the qualitative study conducted to gain initial insights into users' experiences of living with fabric orthoses. The chosen methodological approach is discussed before presenting methods, results and a discussion of the relevance of the key findings to the wider thesis.

3.1 Introduction

This study was crucial to the thesis because, at the time it was first designed, there had been no previous qualitative work published on users' experiences of fabric orthoses. The study aimed to explore the experiences of long-term users to provide a well-informed view of the advantages and disadvantages and provide a solid base from which to further explore acceptability. There was a risk that working with only regular, long-term users would bias the study towards an overly positive viewpoint. However, it was felt that this was justified given the previous research had been weighted towards describing disadvantages.

3.2 Aims and objectives

This study aimed to explore the experiences of people with multiple sclerosis (PwMS) of using fabric orthoses.

Objectives were:

1. To understand how it feels to use a fabric orthosis and what meaning a fabric orthosis holds for users
2. To explore the first experience of using a fabric orthosis
3. To understand the advantages and disadvantages of using a fabric orthosis

3.3 Methodological approach

Qualitative research approaches are the natural choice for analysing experiences and perceptions, "giving voice" to people whose experiences can inform our perspective. Within qualitative research there are a number of specific traditions (Creswell, 2012). Each tradition is characterised by a philosophical stance, methodological guidelines arising from that philosophy and typical methods for data collection and analysis. In addition, there is a growth in the use of "generic qualitative methods" in which the tools and techniques of qualitative research are applied without reference to a specific tradition (Caelli, Ray & Mill, 2003). The first step in study design is to choose a qualitative approach that fits the aims of the study.

Phenomenology is one of the five key traditions of qualitative research (Creswell, 2012). It is both a philosophy and a research approach (Giorgi, 1997). As a philosophy, phenomenology is the study of the essence of conscious experience (Finlay, 2011). Phenomenology was considered as an approach for this study partly because it recognises the importance of the body as something that we experience and as the conveyor of our experiences (Merleau-Ponty, 2002), which fitted the study aim. It was important to discover not just how the orthoses felt to the users but also how the orthoses impacted upon the users' perceptions of themselves and their movement and function.

Phenomenology can be either descriptive or interpretative (Giorgi & Harlow, 2014). Descriptive phenomenology aims to fully describe a phenomenon that is existential in nature (Finlay, 2011). In other words, descriptive phenomenology takes experiences that are essential or common human experiences and sets out to describe the essence of that experience. Classic examples are phenomena such as bereavement, fatherhood and marriage. Husserl (1913/1962, cited in Finlay, 2011) proposed the idea of getting back to *"the things themselves*" meaning that, in descriptive phenomenology, the phenomenon itself is the focus, rather than an individual's experience of it (Crotty, 1996). This seemed not to match the aims of this study. The experience of using a fabric orthosis is not existential; rather it is an experience specific to a small number of people and probably experienced very differently depending upon individual ability, needs and symptoms. It seemed inappropriate to understand what it was like to wear a fabric orthosis without understanding the individual wearer and how MS had affected them. It felt unrealistic to attempt to separate the phenomenon of "fabric orthosis" from the individuals who experienced the phenomenon.

Interpretative phenomenological analysis (IPA) is a specific approach to interpretative phenomenology (Finlay, 2011). Interpretative phenomenology seeks not just to understand what a phenomenon is but also to understand the meaning that the phenomenon holds for people (Smith, Larkin & Flowers, 2009). This approach allied closely with the aims of this study. The meaning that a clinical intervention holds for a patient is probably more likely to determine whether that intervention is acceptable to them than the experience itself, taken at face value. We need to be able to "see" an intervention as they see it, in order to be able to understand their perspectives and how they "make sense" of the experience. The double hermeneutic approach is a key feature of IPA (Smith et al., 2009), meaning that the researcher attempts to find the meaning in the meaning that the individuals themselves attach to their experiences. Exploring meaning is relevant to the emotional importance of an intervention and the participants' explanations and interpretations of what an orthosis might be doing to their body.

A further strength of IPA for this study is that it focusses on each individual prior to searching for themes across a group (Finlay, 2011). This enables the individual and their circumstances to be considered in depth, before any search for commonalities across individuals. This promotes a more in-depth analysis. This approach was felt to be particularly appropriate for a relatively inexperienced qualitative researcher and for a topic for which the sample size was likely to be small.

3.4 Methods

3.4.1 Research design

This was a qualitative study utilising an Interpretative Phenomenological Analysis. The study protocol was approved by the Sheffield Hallam University Research Ethics Committee in October 2015 (Ref: 2015-6/HWB-HSC-5) (Appendix 2).

3.4.2 Participants and recruitment

The study recruited PwMS who currently use fabric orthoses. Recruitment occurred between December 2015 and May 2016, according to the following criteria: (1) a confirmed diagnosis of MS, (2) have used a fabric orthosis for at least two months on a regular basis and (3) remember their initial experiences with the orthosis. Participants were excluded according to the following criteria: (1) not meeting the inclusion criteria, (2) orthosis not used in the month preceding the interview and (3) lived more than three hours travelling distance from Sheffield. The aim was to recruit a relatively small sample of between three to six people, as is usual in phenomenological studies (Finlay, 2011).

Invitations to participate in the study were sent by therapy staff at DM Orthotics. Therapists forwarded the invitation and Participant Information Sheet (Appendix 3) to individuals whom they knew were long-term users of fabric orthoses. In addition, the study was advertised on the webpage of the MS Society UK, including a link to the Participant Information Sheet.

Potential participants contacted the researcher directly by phone or e-mail. There was an initial conversation to confirm that the information about the study was clear and that the potential participant met the eligibility criteria for the study. If the participant was interested in being interviewed, an appointment was made for the researcher and potential participant to meet at a convenient time and location.

In the face-to-face meeting, there was a further opportunity for participants to ask any questions prior to consenting. A written consent form (Appendix 4) was completed immediately prior to the face-to-face interview and verbal consent recorded at the start of the interview.

3.4.3 Data collection

Data were generated using face-to-face interviews. Interviews over telephone or a Voice over Internet Protocol connection were considered, however, these were ruled out because it was anticipated that the participants and the researcher may need gesture and movement to communicate effectively.

Interviews were semi-structured and directed by a topic guide (Appendix 5), designed according to the guidance provided by Bevan (2014). The key tips suggested by Finlay (2011) were used to maximise the quality of the interview. These included the researcher preparing herself and the participant to focus on the purpose of the interview, asking simple open questions, listening carefully and being responsive to what was said.

Interviews were transcribed verbatim from the audio recording. Transcripts included non-verbal features such as laughter, pauses and gesture.

3.4.4 Data analysis

Data were analysed by the researcher according to the guidelines suggested by Smith et al. (2009). Transcripts were read repeatedly and annotated with three different types of exploratory comments. These were (1) descriptive, taking the content at face value; (2) linguistic, exploring the potential significance of the words used and how ideas were communicated and (3) conceptual, the possible meanings revealed by the participants. The individual exploratory comments were typed, printed and cut out and organised into emergent themes for each individual. Once emergent themes had been identified for each individual, they were compared across the four participants, looking for similar themes and polarised ideas (Smith et al., 2009). From this analysis, draft crosscutting themes were identified that expressed commonalties across the participants' stories. The findings from this initial analysis were presented and discussed at two professional conferences (Snowdon et al., 2017a and 2017b).
Later in the doctoral programme, the initial analysis was examined by a peer reviewer (SB), a physiotherapist in paediatric practice who had used IPA in her Masters' dissertation. She read the transcripts, added her own exploratory comments and produced her own list of emergent themes. She audited the principal researcher's analytical trail from annotated transcripts through production of crosscutting themes and the initial report of the findings. She compared the final report to her own interpretation of the data. In addition, the supervision team reviewed the thematic structure in the initial report. These reviews prompted a re-structuring of themes to reduce repetition and expand the focus on the meaning that the orthoses held for these individuals and this iteration was further reviewed by the peer reviewer and

supervision team. Appendix 6 shows an extract of one transcript illustrating the exploratory comments, the initial emergent themes identified for this individual and the final crosscutting themes. Appendix 7 shows how the emergent themes for each participant mapped onto the final crosscutting themes.

3.4.5 Trustworthiness

Rodham, Fox and Doran (2015) suggested that it is particularly important to demonstrate trustworthiness in IPA research because IPA encourages researchers to find their own interpretation and to be "creative" (p60). They stressed the importance of reflexivity to ensure that IPA findings are trustworthy, ensuring that the researcher fully examines the role of self in creating knowledge and shares this reflexivity with their readers to allow others to understand the potential impact of the researcher on the findings. Mason (2018) suggested ensuring trustworthiness through the processes of both data generation and analysis, keeping these true to the research question, being thorough and satisfying yourself, and others, that data are accurately represented.

In this study, trustworthiness was assured through (1) the reflective activities described below, (2) the researcher conducting and transcribing interviews herself, ensuring familiarity with non-verbal elements of communication, minimising the risk of misinterpretation of participants' words, (3) prolonged engagement with the transcripts with thorough annotation and (4) an audit of the process of producing the final report from the transcripts by an independent peer reviewer.

3.4.6 Reflexivity

I engaged in a number of reflective activities to facilitate assessment of my impact on the study and its findings. As recommended by Dowling (2007), I kept a reflective journal during the study, from before the interviews into the write-up. I attempted the phenomenological practice of "bracketing" pre-existing assumptions and ideas before, during interviews, and during the analysis process (Tufford & Newman, 2010). I engaged in a written, in-depth analysis of my first interview transcript, reflecting on each of my questions, my probes and my manner, considering how each might have influenced the participant and the data. An account of these reflective activities is provided in the Discussion.

3.5 Results

3.5.1 Recruitment

DM Orthotics invited four people and three of these contacted the researcher. All were eligible and willing to be interviewed. Three potential participants responded to the MS Society webpage advert but two had inadequate experience of fabric orthoses. Four participants were recruited, three via DM Orthotics and one via the MS Society. Recruitment ended because the response rate had been low and it seemed unlikely that further recruitment was possible.

3.5.2 Participants

Table 3.1 describes participants' characteristics. Two participants used orthoses primarily for control of involuntary movements, whereas two used orthoses primarily to control voluntary movement. Two participants used orthotic sleeves, one used an ankle support and one used shorts. In addition, three participants had been provided with orthoses that they no longer wore. This was fortuitous in that it enabled exploration of factors leading to discontinued use. Three participants were interviewed in their own homes, one in a café.

Table 3.1 Participant characteristics and experiences of orthoses

	Gender	Age	Overview of presentation	Fabric orthosis currently worn	Purpose of orthosis	Fabric orthoses trialled but not regularly worn
Sarah	Female	32	Independent with daily living; walking unsteady but independent; worked full-time. Holmes tremor in upper limbs, left arm more severe.	Sleeve - extended from axilla to tips of fingers on left (non-dominant) arm.	Decreased severity of tremor in left upper limb.	Sleeve that extended from axilla to wrist on right arm, provided for tremor control.
Rebecca	Female	25	Independent with daily living; walked independently and was a keen runner; worked full-time, dystonia in right upper limb, poor proximal control.	Sleeve - extended from axilla to tips of fingers, right (dominant) arm.	Decreased severity of dystonia in right upper limb.	Orthotic shorts and a shoulder support provided for poor control.
Marion	Female	53	Full-time wheelchair user; did not work, independent in transfers; uncoordinated right upper limb, significant weakness in trunk and lower limbs.	Ankle support.	Prevented inversion of ankle during transfers.	Orthotic sleeve that extended from elbow to tips of fingers provided for ataxia.
David	Male	46	Independent with daily living; was seeking full-time employment, walked independent but slow and unsteady with spasticity in both legs.	Commercially available cycling shorts	Stabilised hips and improved control of walking.	Orthotic shorts trialled for hip stability but funding for provision not available.

(All names are pseudonyms)

3.5.3 Themes arising

Two superordinate themes (Table 3.2) explained the meaning of a fabric orthosis. These were giving back control and learning to live with an orthosis. "Giving back control" included subordinate themes of the impact of MS, reclaiming my body and my autonomy and maintaining my self-image. "Learning to live with an orthosis" included subordinate themes describing the stages through which participants adjusted to orthotic use.

Giving back control

The impact of MS

In order to provide context as to why they used their orthoses, participants explained how MS had affected them, both physically and emotionally. Three aspects were especially important in understanding motivations for using the orthoses. These were the feeling of dissociation of one's body from oneself, the sense of distrust of their own body and the feeling of being out of control over what their body did.

The dissociation of body from self was evident in Sarah, Rebecca and Marion's descriptions. Sarah and Rebecca both used language such as "*the left arm*" and *"the toes"* as though they were talking about a separate entity. Rebecca talked about the possible benefits of cutting off her right arm and twice described it as *"useless"*. Marion used the word *"props"* to refer to her legs.

Table 3.2 Superordinate themes and corresponding subordinate themes

Superordinate themes	Subordinate themes
Giving back control	The impact of MS Reclaiming my body Reclaiming autonomy Maintaining self-image
Learning to live with an orthosis	Initial experiences Getting to know my orthosis Compromising and adapting Establishing routines

The sense of distrust of their body came across for all participants. They talked about various ways in which their bodies let them down, including poor awareness of their body's whereabouts and an inability to predict how their body might respond in different activities. David described falling down the stairs because his foot would seem to be securely on a step when it was not. Marion's description was particularly strong, indicating a sense that her body was deliberately trying to deceive her.

"I've just grown to accept that your muscles tell you lies with multiple sclerosis." [Marion]

Sarah and Rebecca experienced involuntary movements that gave both women a sense of being out of control, living with a body that was almost completely unpredictable. Sarah described the jerky tremor in her left arm as *"wild"*, *"random"* and *"uncontrollable"*. She used terms linked with fire to describe the way the tremor *"sparks"* from specific points in her arm and then *"spreads"* elsewhere. At its worse, the tremor would happen in bed at night, preventing her from sleeping. Sarah had problems with her balance and her walking as well but it was the *"craziness"* of the tremor that impacted most upon her feeling of being out of control. Rebecca had dystonia in her right arm, which worsened with movement. Similar to Sarah, she described her movement as *"crazy"* and *"triggered"* in one part of her body before spreading elsewhere.

Reclaiming my body

For all participants, there was a sense that their orthoses gave them back elements of control over their bodies, making their bodies feel more a part of them. Marion felt that her fabric ankle orthosis decreased her clonus, made her ankle more predictable and improved her awareness of her foot position. Rebecca described improved awareness of her muscles with her orthotic shorts. Sarah's sleeve seemed to control the spread of her tremor. It helped her maintain a still posture, with her arm by her side. It slowed down her tremor and reduced its amplitude.

"I will still get a slight tremor in my fingers but that almost feels disconnected to the rest of this, in my arm. If I didn't have my splint on, a tremor in my fingers would set off a tremor in my arm, which would set off a tremor higher up in my arm." [Sarah]

Rebecca's sleeve helped her control her dystonic movements and this made her feel more in control of herself. Similar to Sarah, Rebecca's sleeve decreased the movement amplitude and seemed to contain the involuntary movement to where it had originated, rather than spreading elsewhere.

"(The dystonia) tends to get triggered from my right side, so if that's controlled, the rest of the body seems to be OK." [Rebecca]

David had difficulty walking. He described weakness and tightness in his legs and *"problems with stability"* around his pelvis and hips. He usually walked with a stick and described tripping and falling when his weaker foot caught the ground. He had been assessed, in a physiotherapy session, walking with some orthotic shorts and described the increased control the shorts gave him.

"It was quite remarkable, the difference in my walking. It was a lot more even. I was a lot stronger in the mid-section. … With the orthotic shorts, the limp more or less disappeared. I could feel that I wasn't wavering when I was walking, which was a big improvement, a huge improvement for me." [David]

David had been unable to gain funding for the provision of tailor-made orthotic shorts but started wearing his old cycling shorts instead, which provided some of the same effect. He described the shorts decreasing the cognitive demand of walking.

"What I find particularly draining is that I have two things to think about at once. Number 1, picking my feet up and moving forwards without tripping over myself; but number 2 is the wobbliness as well and taking care of that wobbliness actually is a lot more than it seems because suddenly I don't have to worry about one thing, I can just concentrate on the other one." [David]

Reclaiming autonomy

Sarah, Marion and David related the influence of the orthoses on their bodies to their ability to function independently. David explained that he wore his cycling shorts every day and felt that he needed them every day. He felt they enabled him to walk further and be less reliant on his walking stick.

For Sarah, tremor significantly influenced her function; it was described as tiring, off-putting and embarrassing. Without the orthotic sleeve, she would struggle to drink, because the movement of her left arm would cause her whole body to shake. Without the orthosis, she was unable to drive and unable to use a computer keyboard, both tasks that were crucial to her work. Sarah wore her orthosis every day, all day and expressed a feeling that she was dependent upon it.

"It's very stressful knowing that I have one glove that I am hanging on to and if the zip breaks on it, then that's me out." [Sarah]

Marion directly related the improvement in control of her ankle posture to an improvement in her functional ability. Without support, her ankle inverted and this prevented her from taking weight on her leg, which in turn prevented her from transferring from one chair to another.

"It stops that ankle from turning over and it's so frustrating, when you're there, trying to transfer and your ankle's letting you down." [Marion]

Maintaining self-image

The importance of self-image was evident in a number of ways. Sarah, Rebecca and David talked at length about the importance of how they appeared to others. For David, the appearance of his walking was extremely important. He explained that his shorts were like an invisible walking stick, in that he has less need for a stick when wearing the shorts. He believed the appearance of his walking had an impact on people's perceptions of him, particularly considering that many of his colleagues were unaware of his diagnosis and that David's work role was an important aspect of his identity.

"If I don't wear the shorts, then I find that my walking is absolutely the worst that it can be and, to the outside world it looks like I'm drunk and I can't take the risk at work of not wearing them. Without them on, it would just be horrendous. It definitely has an impact on people's perceptions of you… I know because I can see their reactions. And… that just makes me feel even worse." [David]

Sarah started with MS as a child, aged 14 and, when in public, was constantly striving to prevent others from seeing the wild tremor in her arm. She talked about trying to *"cover"* her tremor maybe indicating a feeling of the orthosis concealing the arm. She described that the orthosis greatly increased her confidence, hinting that the tremor had prevented her seeing a future for herself. Similarly, Rebecca expressed her feeling about the importance of preventing others from seeing her involuntary movement.

"I wanted to just be normal and fit in, and not thinking I can't go out places because of this. But [the orthosis] *gave me a lot of confidence to go out to do things and more drive for the future really."* [Sarah]

"My arm will be in a state that it's not going to be seen." [Rebecca]

For Sarah and Rebecca, their orthotic sleeves would usually be visible to others and they described their feelings about how their orthoses looked to others and how that influenced their self-image. Sarah had adjusted over time to wearing the orthosis, describing that when she was younger she would always choose clothes that concealed it. More recently, she was happier to allow the splint to be visible to others and described it as *"part of me"*. However, when tremor began to be a problem in her right arm, she did not want to use a second orthosis. This was partly because a second orthosis felt to her like confirmation of her deterioration but also because of the fear of how others might view her.

"When it was talked about having a second garment on my right arm, the big thing for me was that I didn't want to be seen with two." [Sarah]

Rebecca expressed a number of issues with the appearance of the orthoses themselves. She did not like the shorts or the shoulder support to show beneath her everyday clothes. She was concerned that wearing extra layers under everyday clothes would cause others to see her perspire. She chose bold, primary colours for her orthotic sleeve so that it would be compatible with her "sporty" self-image but that prevented her from wearing it unless she was exercising. We discussed at some length whether sports compression clothing might have a similar enough effect to her orthotics and her interest in this was possibly prompted by her preference for sports clothing.

Learning to live with an orthosis

A number of aspects of the participants' stories gave the sense of a journey travelled with the orthosis. This second superordinate theme is structured around that journey. As explained in Table 3.1, three of the participants had experiences of orthoses they no longer wore. Unless explained otherwise, the descriptions given below are centred on those orthoses that participants have continued to use.

Initial experiences

All participants described their initial experiences with orthoses as "*experiments*" into solving their movement problems. This feeling of experimentation came across as an important aspect of living with MS, particularly in Marion's account.

"*You know… it's all workarounds and you just do what you can and if you can't do it this way then you might be able to do it that way.*" [Marion]

In Sarah and Rebecca's accounts, use of *"we"* and "*us*" implied collaborative problem solving with healthcare professionals. For Rebecca, language such as *"throw on some shorts"* makes this experimentation seem a little random, as though they were trying anything in an almost desperate attempt to make a difference. For all the orthoses used in the longer-term, the initial responses were positive with at least some benefits evident when the orthoses were first trialled. There is a sense in many of the accounts that the participants' therapists had low expectations of what the orthoses might achieve.

"We were just playing around" (Rebecca)

"It was almost a bit of an experimental thing on me. It was like, let's try it." (Sarah)

"*(My physio) made independent observations of her own and said "wow" these really do seem to make a difference*." (David)

Marion's initial experience of her, now discarded, orthotic sleeve contrasted with the collaborative experimentation described by the other participants. She was given the sleeve as part of a research study and this involved neither collaborative problem solving nor a focus on a problem that was important to her at the time. Although, she had ataxia in her right upper limb, she was able to use her left arm to stabilise her right. She could cope well, was not actively seeking a solution to her ataxia and did not feel any immediate benefit from the sleeve. These different elements probably all contributed to her discontinued use.

Getting to know my orthosis

This theme encompasses the process of becoming familiar with an orthosis. Participants described their early adjustments to wearing an orthosis, how they envisioned their orthoses and their beliefs about how their orthoses worked. Sarah and Rebecca experienced some initial reaction against their orthotic sleeves and both described a period of adjustment.

"I don't really like stuff on the arm […] so I was thinking, you are just going to shove something really tight on my arm and my arm is not going to like it. And, I mean, the first time they put it on… it took a while to settle." (Rebecca)

“It is going to feel like an alien thing on your body but you have to be quite patient with it.” [Sarah]

There were contrasting viewpoints across our participants around whether orthoses should be used in the longer-term. David did not discuss this, seeming more interested in the short-term impact. However, Sarah, Rebecca and Marion all expressed opinions around the impact of long-term use, informed by what they had been told by their physiotherapists. Marion had been told her ankle orthosis might make her worse, by allowing her to be less active. Because of this she deliberately restricted the amount of time that she wore it for, carefully judging her ability and only using it if she felt she needed to. This contrasted sharply with Sarah's account of having been told that her orthosis would induce positive changes over time. Her experiences had strengthened her belief in her orthosis as a treatment. She saw her improvement over time as being, at least in part, related to use of her sleeve.

“*The only negative effect is that if you wear it too much it may stop you from doing things properly yourself. I have had a physio say, well, don’t wear it any more than you have to*.” [Marion]

"*I think that what was sold to me in terms of […] how it was going to help in the longer term as well, […] when I'm not even wearing the splint." (Sarah)*

"The overlay is actually very good now in terms of what I get beyond when I'm not even wearing it." [Sarah]

Both Sarah and Rebecca described their orthoses as replacements for physiotherapy. Sarah had experienced her physiotherapist inhibiting her tremor by holding the point in her arm where the tremor originated. She said her orthosis felt as though "*someone had got hold of my arm*". She had joked with her physiotherapist about wanting to take her everywhere to inhibit her tremor and explained that her orthotic sleeve had enabled that to happen. Rebecca described that following earlier relapses, physiotherapists had used handling to help stabilise her, improving her confidence and providing her with feedback. She explained that orthotic shorts could provide such feedback, enabling people to do more and be less reliant on physiotherapy input.

Rebecca suggested that orthotic shorts could prompt a change in movement patterns, explaining that, once someone's movement pattern had changed, there would be no need for longer-term wear.

All participants described the importance of the compression provided by the orthosis. All had experienced changes in effectiveness due to changes in compression. David felt his shorts to be more useful when they were freshly washed and Sarah explained how her orthoses aged and lost elasticity. Rebecca explained a similar effect. Having lost weight after she was first provided with her orthosis, she found a change in effectiveness as the orthoses became looser.

“*This is an old splint and I can tell the difference. I start to lose certain abilities*.” [Sarah]

“They are not as tight as they used to be so they don't do as much as they should do.” [Rebecca]

Compromising and adapting

Sarah, Rebecca and Marion explained a number of disadvantages to orthotic use both in describing the orthoses that they no longer used and in describing the orthotics that they still use. There was a feeling that they were compromising, weighing up when the advantages of orthotic use outweighed the disadvantages.

Sarah’s orthotic sleeve provided a lot of compression, more than would usually be recommended by her orthotics provider. She felt that this was necessary for the sleeve to be effective but it caused a number of disadvantages. The sleeve caused fatigue in her arm, painful sores in her hand and restriction of circulation, causing pins and needles if she was not active. She stressed her choice to accept these risks and that the fact that she coped with such difficulties indicated how important the orthosis was to her.

“*Even though it is painful, I will still need to put that glove on…. I make that choice to wear it all day*." [Sarah]

Other disadvantages cited were Sarah and Rebecca's concerns about how the orthoses appeared to others, as described earlier. Sarah, Rebecca and Marion expressed difficulty in getting their orthoses on and off but all participants could get their orthoses on and off independently.

Specifically for the participants who used orthotic sleeves, all sleeves had integral gloves covering participants' hands and this caused practical difficulties around hygiene. Sarah described the importance of wearing a rubber glove over the orthosis for activities such as cooking, to keep the orthosis clean and to avoid it taking on food smells. In contrast, Marion explained that being unable to wash her hand easily during the day was one of the reasons she stopped wearing the orthotic sleeve. She related this disadvantage directly to her perception of the effectiveness of the sleeve.

“Every time I went to the loo, I would take it off and I did get to the point where I didn’t put it on. I didn’t put it on and it didn’t really do what it was supposed to do anyway.” [Marion]

In most cases, the participants had decided what they would continue to use and what they would not, however, Rebecca was still actively weighing up whether she should use her orthotic shorts. Rebecca considered her orthotic shorts uncomfortable, “*high-waisted*” and “*not elegant*”. She was initially reticent to explain that she rarely used them. She was certain that they were beneficial for her posture. However, she explained that she preferred to manage without the shorts and likened this to preferring to manage without help and, conversely, to choosing to avoid exercises that she found difficult.

"I walk quite badly without my stick. When I am at my worse, my legs just kind of turn in and I kind of walk a bit weird. The shorts can kind of counteract that[…] I should probably wear them more often, they would probably help […] but I'll just battle on and get through it." [Rebecca]

Alongside the feeling of compromise is the importance of "adapting" the orthosis to get the most out of it whilst minimising the disadvantages. This element came across particularly for Sarah and Marion with Sarah describing a lot of "*toing and froing*" when she was younger, trying to get the fit and the design of her sleeve correct.

Marion was able to adapt her ankle support herself, *"playing around"* to alter the amount of tightness that she applied. In contrast, she described her ideas for adapting her orthotic sleeve, such as taking the sleeve higher up her arm to where she felt particularly unstable but she had been unable to explore such adaptations with the orthotics provider.

Establishing routines

There were two contrasting elements to the theme of "establishing routines". The first captures the routines involved in using an orthosis and the second captures the routine of discontinued use.

Routines of orthotic use included some basic practical strategies such as ensuring that the orthosis was ready for use, including washing and drying and storing the orthosis so that it was available when required. Sarah probably faced the greatest challenges in using her orthosis because it seemed to be a far tighter fit than those used by other participants. In addition, she had many more years of experience of orthotic use. Consequently, she had established an extensive repertoire of coping strategies. These included her strategy for getting her orthosis on, her choice of clothing and her habit of always keeping talcum powder in her holiday bag to enable the splint to be fitted in a hot climate. She conveyed a sense of pride in her own ability, seeing herself as an expert user. She described herself as "*a bit of a pro*" and explained how she gave advice to new orthotists around the fit of her sleeve. The challenges of orthotic use appeared to reflect positively on her self-image.

"*Whenever anybody measures them I am quite good at knowing whether that fits or not […] (*I say*) you are talking to someone with experience and I need it that tight.*"
Sarah

In comparison, the rarely worn orthoses disappeared into the background. As Marion explained, after repeatedly struggling and seeing limited benefit she "*got to the point where I didn't put it on*". The rarely used orthoses were not actually thrown away but might be mislaid or kept in a cupboard where they were mostly forgotten.

3.6 Discussion

3.6.1 Summary of main findings

This study explored the experiences of PwMS using fabric orthoses. These long-term users felt MS had caused them to distrust and feel disassociated from their bodies. Their orthoses restored a feeling of control and ownership. Participants' described that their orthoses decreased involuntary movement, improved awareness of their body and improved stability. Participants felt orthoses altered body posture, to positions that inhibited involuntary activity or to positions that were more stable. They experienced improved autonomy and described the importance of their orthosis with respect to their self-image. A journey of orthotic use was described, starting with an initial experience involving experimentation, searching for anything that might improve their movement. This moved into a process of weighing up the advantages and disadvantages of orthotic use. For most participants, ongoing orthotic use required some persistence in overcoming the disadvantages, by adapting skills and routines or adapting the orthosis itself. Other aspects of the meaning of a fabric orthosis included the importance of a tight fit, the feeling that an orthosis is like a supportive physiotherapist and a controversy around whether fabric orthoses have positive or negative effects with longer-term use.

3.6.2 Discussion of main findings

This discussion will consider the perceived impact of the fabric orthoses on physical and psychosocial factors, the concepts of the "journey of orthotic use" and "weighing up" pros and cons of orthotic use, the strengths and limitations of the FabO IPA study and the implications of the findings for the thesis as a whole.

The perceived physical effects of fabric orthoses

The physical effects of the fabric orthoses of decreased involuntary movement, improved posture and improved stability were similar to those reported in previous fabric orthosis research. Blair, Ballantyne, Horsman and Chauvel (1995) observed an impact of fabric orthoses on involuntary movement in children with cerebral palsy, although movements were simply rated subjectively by blinded assessors. An impact of fabric orthoses on posture is a commonly reported feature in previous studies, though again these are rarely studied using objective measures (for example, Blair et al., 1995;

Flanagan, Krzak, Peer, Johnson & Urban, 2009). The two previous studies that investigated perceived effects with robust qualitative methods were published after the FabO IPA study was first designed (Miller et al., 2016; Stone, 2014). Their accounts of perceived benefits and disadvantages included similar issues to those found in the FabO IPA study. Participants in the Stone (2014) study felt improvements in limb position, stability, muscle tone and pain relief. Miller et al. (2016) found perceived benefits including increased strength and stamina and smoother movement. In terms of disadvantages, participants in the Stone (2014) study had mostly experienced solid thermoplastic splints and compared their fabric orthoses favourably with these. They considered them easy to remove, lightweight and fitting well under clothing. Disadvantages were that they were difficult to get on, either too tight or too slack and uncomfortable in summer. Miller et al. (2016) reported that nine of their 11 participants described difficulty both donning and doffing and the sleeve not staying in place over the upper arm. Other issues included a feeling of impaired circulation, increased pain and irritation. Participants in both studies described a period of adjustment to coping with using the orthoses. Overall, previous research confirms the nature of the perceived effects reported in the FabO IPA study, the challenges of using a fabric orthosis and the importance of getting the fit and the design exactly right for each individual.

Psychosocial impact of fabric orthoses

The psychosocial effects described in the FabO IPA study around improved body awareness, improved autonomy and self-image reflect some of the findings in the Stone (2014) and Miller et al. (2016) studies. For example, participants in the Stone (2014) study explained that they looked and felt more "normal"; they felt an increased awareness of their limbs and explained their limbs felt more "part of me" with their orthoses on. Miller et al. (2016) reported that five of their 30 participants felt more confident and had improved overall well-being with their orthoses. Participants in both studies described stigma associated with looking different when the orthosis was visible to others.

The finding that fabric orthoses might restore a feeling of ownership over an MS-affected body is potentially an important finding. Previous research has found the perception of distrust of your body is common and important in MS (Mozo-Dutton, Simpson & Boot, 2012; Olsson, Lexell & Söderberg, 2005). For some, this is because of the unpredictable nature of the disease process itself but also the lack of bodily awareness and the impact of fatigue on the reliability of one's body.

The FabO IPA study was the first study to evaluate the meaning of fabric orthoses to long-term users. However, there are similarities between the themes described here and themes arising from qualitative research investigating assistive technology. Assistive technology research has considered a wide range of aids such as mobility devices and electronic devices for communication, mobility and environmental control. Participants include stroke survivors, PwMS and elderly people. The meaning of such assistive devices includes the feeling of being in control, for example, the ability to continue with valued roles as a result of device use (Copolillo, Collins, Randall & Cash, 2001; Pape, Kim & Weiner, 2002). Assistive devices can change an individual's feeling of identity or self-image, occasionally enhancing identity but often diminishing it by means of providing a visible and constant reminder of the disability that has led to assistive device use (Gitlin, Luborsky & Schemm, 1998; Pape, Kim & Weiner, 2002; Verza, Carvalho, Battaglia & Uccelli, 2006). Ploughman et al. (2012) investigated self-management in older people with MS and found that assistive devices polarised opinions. Some people considered them *"a godsend"* and others were concerned that assistive devices were markers of disability and caused stigma. They perceived a risk that reliance on such devices might subsequently increase dependence on others. Squires, Williams and Morrison (2019) conducted focus groups with PwMS, their carers and occupational therapists and found that the perceived stigma around using assistive devices was a key factor in discontinued use.

Hocking (1999) reviewed research investigating device use and abandonment and concluded that individual responses to assistive devices are heavily informed by psychosocial factors. She suggested that many healthcare professionals underestimated the importance of psychosocial factors influencing device use, even though theories of motivation and self-directed behaviour suggest that psychosocial factors are extremely important in deciding what people choose to do. For example, Ajzen and Madden (1986) studied college students' behaviours and beliefs and proposed a "theory of reasoned action" in which there are two key determinants of behaviour: personal beliefs about whether behaviour will result in an intended outcome and personal beliefs about what society would think of that behaviour. Deci and Ryan's Self Determination Model (Deci & Ryan, 1985, cited in Siegert & Taylor, 2004) suggested three fundamental human needs that drive our goals and behaviours. These are autonomy (control and independence), competence (a sense of mastery in successfully responding to challenges) and interpersonal relatedness (the sense of emotional connectedness to other people). These are similar to the subordinate themes in the "giving back control" theme in the FabO IPA study. Overall, previous research confirms the importance of considering both positive and negative psychosocial effects with respect to the impact of orthoses.

Orthotic use as a journey

The data suggested a journey of orthotic use. Within this journey, there are key issues that might have particular significance for determining whether an orthosis was used in the longer-term or discarded. These included the degree of collaboration with healthcare professionals when the orthosis was first trialled, the importance to the individual of the problem for which the orthosis was prescribed, the users' perception of benefit, the users' willingness to adapt their routines around orthotic use and the degree of support available for adapting the orthosis in the early phases of use. The contrasts in the FabO IPA stories led to creation of a model describing the stages of the journey and the factors that might lead to continuing and discontinuing use (see Figure 3.1). Implied within this model is the sense that good practice in orthotic provision should be in line with the "journey to continued use".

One important element of the "journey of orthotic use" model is the importance of a short-term, orthotic effect. As described in Section 2.4.3, research into orthoses can investigate direct orthotic effect, training effect and total orthotic effect (Taylor, Humphreys, & Swain, 2013). Traditionally, fabric orthosis research has mainly investigated the training effect and the total orthotic effect with limited attention to the direct orthotic effect. The findings of the FabO IPA study further suggest that direct orthotic effect is important to users and may be one of the aspects determining the acceptability of the orthotic.

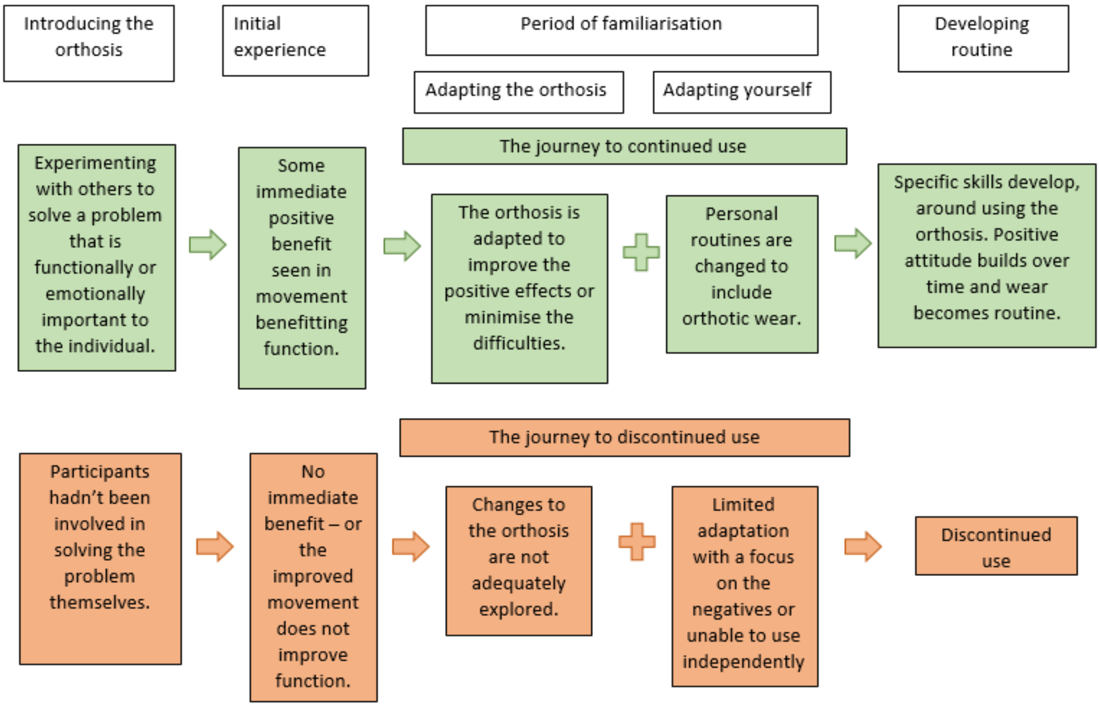


Figure 3.1: Comparing the journey to continued use of an orthosis with that to discontinued use. (The suggested journey to continued use is shown in green and the journey to discontinued use in red.)

When I first constructed the "journey of orthotic use" model, I was unaware of similar work having been done in the field of assistive technology. However, there are considerable similarities between my model and a number of previous models. Lenker and Paquet (2003) reviewed six models of assistive technology use and found similarities across them all. All emphasize the process of initial assessment, all assess person, environment and task with a multi-disciplinary collaborative approach and all aim to match the person with an assistive device for use in a specific environment. Since 2003, one of those six models, Scherer's Matching Person and Technology model, has been developed using assessment tools designed to structure collaborative problem-solving and maximise the match between an individual and an assistive device (Scherer, Sax, Vanbiervliet, Cushman & Scherer, 2005). Other authors have published prescription protocols with a similar purpose, such as the Assistive Device Evaluation and Prescription Protocol (Verza et al., 2006) and the Prescription Process of Assistive Devices (Blomquist & Nicolau, 2003, cited in Hedberg-Kristensson, Ivanoff & Iwarsson, 2006) .

There is some evidence for the effectiveness of assistive device prescription models. For example, Verza et al. (2006) tested their prescription protocol on 54 PwMS, where 67 assistive devices were provided before the protocol was introduced and 84 devices were provided after the protocol was introduced. The rate of abandonment of devices was 37% without the protocol and only 9.5% with the protocol. A one-third abandonment rate is similar to that reported in other assistive technology studies (e.g. Phillips & Zhao, 1993), indicating that such approaches have significant potential to improve wider practice. Despite many years of experience in neurological rehabilitation, I was unaware of such guidelines and have found no previous research applying these models to orthotic practice.

"Weighing up" and the link to acceptability

The concept of weighing up the pros and cons of orthotic use came across strongly in our data. Participants did not minimise the challenges they had faced in using their orthoses. Instead, they described these as tasks they had mastered and challenges they had overcome. In explaining why they felt their orthoses were worth using, they described positive effects and weighed these against the challenges of use. Rebecca was still weighing up whether to wear her orthotic shorts and her considerations included her values; for certain tasks, she preferred to *"battle on"* without the help of an orthosis.

The concept of weighing up different influences in order to inform an overall opinion about a healthcare intervention has been explicated within a model proposed to explain acceptability (Sekhon, Cartwright & Francis, 2017). Sekhon et al. (2017) designed the Theoretical Framework of Acceptability using iterative steps including (1) review of previous systematic reviews of the acceptability of health care interventions; (2) a consensus group exercise with research psychologists on the possible definitions of acceptability derived from the reviews and (3) defining, mapping and re-mapping potential constructs that might make up the multi-dimensional construct of acceptability. Acceptability was defined as *"a multi-faceted construct that reflects the extent to which people delivering or receiving a healthcare intervention consider it to be appropriate, based on anticipated or experienced cognitive and emotional responses to the intervention"* (Sekhon, Cartwright and Francis, 2017, p1). The component constructs of acceptability are shown in Table 3.3. The themes arising in the FabO IPA study clearly relate to burden, perceived effectiveness and self-efficacy. In addition, FabO IPA participants referred to several other constructs such as their feelings towards their orthosis (affective attitude), their interpretations of how their orthoses worked (intervention coherence) and how their orthotic fitted with their values (ethicality). This indicates the relevance of the Theoretical Framework of Acceptability for investigating acceptability of fabric orthoses.

Table 3.3: The component constructs of acceptability proposed by Sekhon et al. (2017)

Component construct	Definition
Affective attitude	How an individual feels about the intervention
Burden	The perceived amount of effort that is required to participate in the intervention
Ethicality	The extent to which the intervention has a good fit with an individual's value system
Intervention coherence	The extent to which the participant understands the intervention and how it works
Opportunity costs	The extent to which benefits, profits or values must be given up to engage in the intervention
Perceived effectiveness	The extent to which the intervention is perceived as likely to achieve its purpose
Self-efficacy	The participant's confidence that they can perform the behaviour(s) required to participate in the intervention

Sekhon, Cartwright and Francis (2018) suggest that the Theoretical Framework of Acceptability be used for research purposes, both to develop the acceptability of interventions in the feasibility stage of the MRC "Developing Complex Interventions" framework or in the evaluation phases (Craig et al., 2008). They proposed that acceptability be assessed at various time points in a research participant's experience. They used the term "anticipated acceptability" to refer to acceptability assessed prior to actually having experience with an intervention. They proposed that the constructs of ethicality, intervention coherence and self-efficacy can be assessed simply from explaining the proposed intervention. They suggest assessing "experienced acceptability" either during an intervention or after an intervention. This approach contrasts with the concept of assessing satisfaction with interventions, which relates only to "*a subjective evaluation of a treatment or therapy upon treatment completion*" (Sidani, Epstein, Fox & Collins, 2018, p573).

3.6.3 Limitations of this study

It is important to remember that the participants in this study represent people with a positive outlook on fabric orthoses. They were invited because they were long-term users. Two were invited by the company who manufactures their orthoses and one (David) was partly motivated to be involved in the study by the desire to be provided with a pair of orthotic shorts as part of a follow-up study. Participants could have been reticent to provide negative feedback. Nevertheless, the participants openly shared stories of the difficulties they experienced in using fabric orthoses.

The sample size for this study was small. Smith et al. (2009) and Finlay (2011) suggest an IPA study should have between three and six participants and the sample should be relatively homogenous in terms of the participants' experiences. Smith et al. (2009) explain that "homogenous" is considered relative to other qualitative traditions such as grounded theory, where researchers deliberately seek variability in the samples in order to test theories around whether people with different characteristics have different beliefs or behaviours. They suggest that a more "homogenous" sample enables a more in-depth examination of people who have relatively similar experiences. The participants in the FabO IPA study group were similar in that they all had MS but they differed markedly in the nature of their symptoms, their functional ability and the types of fabric orthoses that they experienced. It is possible that a larger group or participants who have more similar symptoms would reveal data that are more illuminating.

3.6.4 Reflections on the impact of the researcher on the research

I have influenced this study in two main ways. Firstly, this was the first qualitative study I had designed, my first experience of using IPA and of conducting qualitative interviews. Secondly, my professional background as a physiotherapist influenced my approach and may have influenced the opinions expressed by the participants. As a novice, I struggled with recognising and limiting the impact of my professional skills and identity.

It could be argued that having set out on a doctoral programme investigating fabric orthoses I might have been prejudiced towards seeking and hearing a positive viewpoint. However, because this project took place at a relatively early stage in my doctoral journey, I was not invested in any particular outcome. My approach to the study was one of wanting to learn about fabric orthosis and about qualitative research. Having never used fabric orthoses clinically, nor met a service user who used them, I found it easy to approach the project with an open mind and honest curiosity.

My lack of previous experience and my professional background have influenced the content of the interviews. I reflected in some depth on my interview with Sarah because this was the first interview. I found a number of areas for improvement. Firstly, I had a tendency to revert to the objectives of a subjective patient assessment; prioritising questions about the possible cause and impact of physical problems over the search for meaning. Secondly, there were occasions where I used prompts that were too leading and too specific (such as "body image?" see extract in Appendix 6). I suspect I have used specific prompts in clinical practice as a means of communicating my understanding and building trust in a therapeutic relationship. I tried to limit this but used two leading prompts in the later interviews. Reflecting upon each of these, I found participants corrected me where I had suggested something they disagreed with. Finally, I did not always follow up adequately on topics of importance. I failed to follow-up on a difficult conversation around a second orthosis being an unwelcome marker of Sarah's deterioration and I did not fully explore what she had been told about her orthosis as a means of improving her underlying ability. I considered myself to be a good listener but this highlighted my deficiencies. In the later interviews, I used more open, generic prompts and left fewer relevant avenues unexplored.

Participants were aware of my professional and clinical role, either because they were told when first invited or because they asked, prior to the interview. There were indications in the interviews that participants were influenced by their knowledge that I was a physiotherapist. For example, some used medical terminology to describe their problems and sometimes appeared to assume that my opinions would match those of other physiotherapists they knew.

Key messages in the data were evident from early in the analysis process and were used to shape the later studies in this thesis. However, it was relatively late in the doctoral programme that I engaged the second reviewer and discussed the thematic structure with supervisors experienced in qualitative research. The second reviewer felt that my analysis was true to the method of Smith et al. (2009), that my prompts and probes were effective in exploring important points and that the list of emergent themes was comprehensive. She highlighted a number of areas where the final interpretation could be enriched to draw out the double hermeneutic and where unnecessary description of functions and activities had been included. On reflection, this was a further indication of my tendency to approach each participant primarily from the perspective of a physiotherapist. The support of the peer reviewer and supervision team was essential in broadening my approach.

Overall, I suggest that the findings presented here are a relatively unbiased account of experiences with PwMS around using fabric orthoses but that elements of meaning may be missed or not fully explored due to the project being conducted from a physiotherapist's perspective.

3.7 Implications for the thesis

The FabO IPA study has contributed to the thesis both in making a contribution to knowledge in its own right and in informing the design of the final study presented in this thesis: the Orthotic Shorts Feasibility in Multiple Sclerosis (OSFeaMS) study. In terms of a direct contribution to knowledge, the overriding importance of psychosocial issues with fabric orthoses, the similarities to good practice in assistive device prescription and the theme of "weighing up" the pros and cons of orthotic use have not been recognised previously. In terms of informing the design of the OSFeaMS study, the findings of the FabO IPA study have (1) prompted the OSFeaMS study to focus on the direct orthotic effect as this was suggested to be necessary for longer-term acceptability; (2) highlighted the importance of investigating the various constructs within acceptability that appear to be weighed against each other to determine ongoing effect; (3) prompted the inclusion of an initial interview in the OSFeaMS study to investigate acceptability at the first assessment; (4) supported the choice of focussing on orthotic shorts due to the suggested concept of "an invisible walking stick", (5) prompted the inclusion of a dual task cost measure due to the suggested impact on concentration during walking, (6) suggested specific prompts around potential disadvantages to be included in the final qualitative interview and (7) informed the analytical framework used to analyse the qualitative data.

3.8 Conclusion

The FabO IPA study provided an important exploration of users' perspectives on fabric orthoses. It has highlighted key themes around what an orthosis means to users, how they can give back a sense of control over one's body, one's independence and one's self-image. The study focussed attention on the important process of providing an orthosis and highlighted similarities between users' experiences of fabric orthoses and users' experiences of other assistive devices.

The subsequent chapters of this thesis describe the testing and development of a specific fabric orthosis, orthotic shorts, in the Orthotic Shorts Feasibility in MS (OSFeaMS) study. Through focussing on one specific type of fabric orthosis, key principles around acceptability and the "journey of orthotic use" will be further developed. The next chapter reports a review and pilot study used to inform measurement choices for the OSFeaMS study.

Chapter 4: Outcome measures for walking ability in multiple sclerosis - a critical review and methods pilot

Summary

This chapter provides a review of outcome measures for assessing walking ability in MS and an account of a single-case methods pilot conducted to determine the burden associated with measurement and trouble shoot unforeseen challenges. These evaluations informed the measures used for the final study in this thesis, the OSFeaMS study, reported in Chapters 5, 6, 7 and 8.

4.1 A critical review of outcome measures for assessing walking ability

4.1.1 Introduction

Appropriate choice of outcome measurement is vital in any quantitative investigation and involves choosing the most appropriate things to measure and the most appropriate tools with which to measure them. As explained in Chapter 1, a key objective of this thesis was to investigate the feasibility of using orthotic shorts in PwMS for improving walking ability. The focus on walking ability was prompted by the importance placed on walking by PwMS (LaRocca, 2011; Pike et al., 2012) and its relationship to independence, social participation and employment status (Kierkegaard, Einarsson, Gottberg, Von Koch & Holmqvist, 2012; Pike et al., 2012). In this section, various aspects of walking ability will be discussed, with respect to their importance to PwMS. Then specific measurement tools will be explained and critiqued.

4.1.2 Search strategy and inclusion criteria

To understand the measurement approaches used to study walking in PwMS, a broad review was conducted. An initial search was carried out on MEDLINE, using the search strategy "measure*" AND "multiple sclerosis" AND ("walking" OR "gait"). Studies were included in the review if they were either reviews of measures used in MS, intervention studies relevant to improving walking in MS or studies investigating the validity and reliability of measures. From this review, a list of relevant variables and potential measurement tools was drawn up. A review of the importance of the different variables was conducted, working from the papers already selected, additional resources available from reference lists and additional searches focussed on the tools themselves. This review has been updated during the process of writing up the thesis. The review of variables was followed by a review of potential measurement tools, including their measurement properties and potential feasibility.

4.1.3 Variables relevant to walking ability in MS

Variables considered as components of walking ability include maximal walking speed, maximal walking distance, spatiotemporal gait parameters, kinetics and kinematics of walking, gait variability, energy expenditure, cognitive demand, balance, patient-perceived walking ability, patient perceived balance and fatigue.

Maximal walking speed is considered to be a good indicator of walking function. For example, it is related to activity levels, in that there is a moderate correlation between maximal walking speed and step count measured over a week (Motl et al., 2013). It is highly correlated to employment status and social participation ($r = 0.71$-0.81; Kierkegaard et al., 2012). In addition, maximal walking speed is closely correlated to the maximal distance that a PwMS can walk in six minutes, with an R^2 of 0.78 in mildly affected PwMS and an R^2 of 0.81 in moderately affected PwMS (Gijbels et al., 2012). Maximal walking speed is usually considered as being the fastest that an individual can walk over a relatively short distance, typically 25 feet or 10 metres.

The ability to walk longer distances is relevant to exercise tolerance, endurance, cardiovascular fitness and fatigability, which are important for integration within a community (Andrews et al., 2010). Gijbels et al. (2010) argued that continuous walking for a period of six minutes is more typical of community ambulation than walking a short distance at maximal speed. They found that the distance walked over six minutes in PwMS was a better predictor of step count over a week than maximal speed over 25 feet.

Spatiotemporal gait parameters include velocity, cadence, step length, stride time, proportion of gait cycle in double support and step width, which is the perpendicular distance between one foot and a line visualised between successive footfalls of the opposite limb (see Figure 4.1). All these variables are impacted upon in MS (Givon, Zeilig & Achiron, 2009) and related to fear of falling (Delbaere, Sturnieks, Crombez & Lord, 2009). Givon et al. (2009) suggested that different parameters were relevant in different sub-types of MS: velocity, stride time and % of gait cycle in double support indicated lesions in the pyramidal system and step width indicated cerebellar lesions. Spatiotemporal gait parameters have been extensively investigated in MS, so a lot is known about how such variables are related to function (Givon et al., 2009). It is relatively unusual for spatiotemporal gait parameters to be used as outcome measures in MS trials (Leone et al., 2018) but they have been shown to change with rehabilitation, can be measured with precision and can provide insight into potential mechanisms by which therapy might be effective (Gutierrez et al., 2005; Kalron, Pasitselsky, Greenberg-Abrahami & Achiron, 2015; Newman et al., 2007)

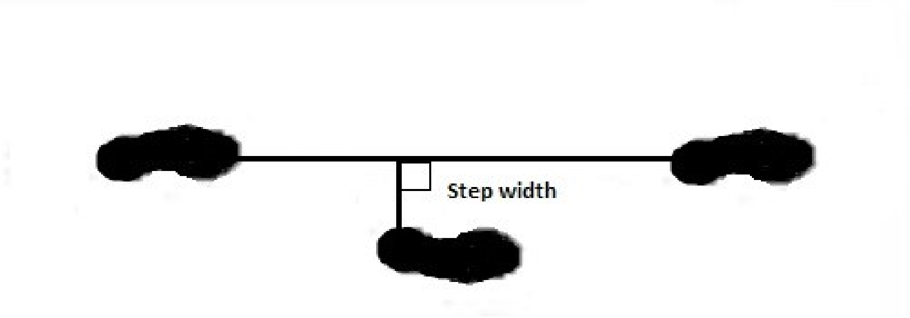


Figure 4.1: An illustration of step width

Kinetics is the study of the actions of forces that produce walking and involves measuring variables such as torque, gravity and ground reaction force; kinematics is the study of motion of body segments. Both are extremely relevant to understanding exactly how MS and orthotics impact upon movement of the lower limbs, head, trunk, pelvis and upper limbs during walking. Measurement of kinematics can be highly precise and capable of exploring potential mechanisms behind changes in walking ability (Cameron & Wagner, 2011). Kinematics is commonly measured in studies investigating lower limb orthotics in MS (Andreopoulou, Mercer & van Der Linden, 2018). Andreopoulou et al. (2018) suggested that the reliability and validity of many kinetic and kinematic variables have not been determined specifically in PwMS.

Although these are known to be highly accurate methods, the relevance of changes seen in PwMS might be unclear.

Trunk and pelvic kinematics are particularly important variables, as these might change either as compensation for weakness in the lower limbs (Psarakis et al., 2018) or as a direct result of neurological damage (Nilsagård, Carling, Davidsson, Franzén & Forsberg, 2017). For example, it has been shown that PwMS sway more in standing and walking than do non-neurologically impaired people and that more disabled PwMS tend to have more trunk sway in standing and walking than those who are less disabled (Corporaal et al., 2013).

Variability of walking has been shown to be a key feature that distinguishes people with MS from non-neurologically impaired people (Flegel, Knox & Nickel, 2012) and is closely related to falls risk (Allali et al., 2016a; Socie et al., 2013a). Gait becomes more variable early on in the progression of MS, often before there are any clinical signs of difficulty walking (Flegel et al., 2012). Variability is, therefore, an important construct to investigate in PwMS who are less severely affected. Measures of variability of gait may be more sensitive to change than other gait parameters (Hausdorff, 2005) and have been recommended for and used as outcome measures in trials investigating older people (Beauchet, Launay, Annweiler & Allali, 2014; Zhang, Low, Gwynn & Clemson, 2019). No previous trials have been found that have used gait variability as an outcome measure in PwMS.

Energy expenditure during walking is higher in PwMS compared to healthy controls and this may contribute towards fatigue in MS (Chung et al., 2016). A lower limb orthosis should improve the efficiency of walking and therefore, measuring energy expenditure is recommended and frequently used in orthotic research (Andreopoulou et al., 2018; Brehm, Bus, Harlaar & Nollet, 2011). Energy expenditure has been shown to be more responsive to orthotic assistance than distance walked (Mcloughlin, Lord, Barr, Crotty & Sturnieks, 2015).

The cognitive demand of walking is higher in PwMS than in non-neurologically impaired people, suggesting that PwMS compensate for their walking difficulties by increasing cognitive control (Etemadi, 2017; Wajda, Motl & Sosnoff, 2013; Nilsagård, Denison, Gunnarsson & Bostrm, 2009). It is a potentially important construct to measure because cognitive demand of walking may increase fatigue in MS (Hamilton et al., 2009) and it is argued that it is a valid representation of daily life, in that it is usual to think whilst walking (Leone, Patti & Feys, 2015). To date, the cognitive impact of motor tasks has only been assessed in trials where the interventions involved practicing simultaneous cognitive and motor tasks (for example, Kramer, Dettmers, & Gruber, 2014; Monjezi et al., 2017).

Balance is commonly affected in PwMS in a wide variety of tasks and across the disability spectrum (Comber, Sosnoff, Galvin, & Coote, 2018). Balance is closely related to walking ability (Brincks, Andersen, Sørensen & Dalgas, 2017); Callesen, Dalgas, Brincks & Cattaneo, 2019) and, because it is so relevant to function and safety, it is frequently measured in intervention trials in PwMS (for example, Gunn, Markevics, Haas, Marsden & Freeman, 2015).

Participant-perceived ability is commonly measured in trials of interventions in multiple sclerosis. Capturing participants' perceptions of their own abilities has the advantage of capturing their experience of everyday life, which should complement data obtained from objective measures. In addition, participant-perceived ability may be influential in determining physical activity and social engagement in PwMS (Kieseier & Pozzilli, 2012; Motl et al., 2013; Motl & Snook, 2008a). Improvements in participant perceived ability have been shown to be only weakly associated with improvements in performance-based measures, stressing the importance of evaluating multiple constructs when evaluating an intervention (Schwartz, Ayandeh & Motl, 2014). Self-report measures for walking ability in MS measure constructs such as perceived walking ability, balance confidence and fatigue.

4.1.4 Variables relevant to efficacy of orthotic shorts for walking

Brehm et al. (2011) proposed that orthotics should be evaluated in a number of different ways, to ensure practice is well informed. They suggested assessment should include (1) the biomechanical impact of an orthosis, (2) the impact on a patient's activities such as walking and stair climbing and (3) the impact on participation and personal factors relevant to the success of an orthosis. In other words, measurement across the ICF is recommended.

To assess the efficacy of orthotic shorts for walking ability, the variables of walking speed, distance, spatiotemporal gait parameters, kinematics of pelvic movement, gait variability, energy expenditure, cognitive demand, patient-perceived walking ability and patient perceived balance were chosen from the review. Kinetics was not considered further as a measurement because it required force plates and a three dimensional (3D) motion analysis system that would not have been readily available. It was decided to not measure fatigue because the likelihood that fatigue might change in the short-term following a two-week home trial of a novel orthotic appeared low. Objectively measuring balance was not considered because it was felt that inference about balance might be drawn from assessing walking alone and that including objective measures of balance might add too much to participant burden. Participant perception of balance was included as this might enable exploration of potential impact upon balance, without contributing too much to participant burden.

4.1.5 Tools to measure walking ability

Walking measures that have been suggested for use in clinical practice in MS are the Timed 25-foot Walk (T25FW), a measure of maximal walking speed, the Two Minute Walk Test (2MWT), a measure of walking distance and the 12-item Multiple Sclerosis Walking Scale (MSWS-12, Appendix 8), a measure of patient-perceived walking ability (Bethoux and Bennett, 2011; Capra et al., 2014; Gijbels et al., 2012). In the section below, objective measures will be discussed first, followed by self-report measures.

The T25FW is one of the constituent measures in the Multiple Sclerosis Functional Composite (Rudick, Cutter & Reingold, 2002) and is commonly used in clinical practice and research. It involves measuring the time taken to walk 25 feet at maximal speed, using a standing start and a stopwatch. The test is performed twice and a mean taken of the two times. A practice effect has been demonstrated showing improvement in T25FW between the first and second trial (Larson, Larson, Baumgartner & White, 2013). However, Learmonth, Dlugonski, Pilutti, Sandroff & Motl (2013a) found an Intraclass Correlation Coefficient (ICC) of 0.991 with 82 PwMS assessed 6 months apart and Nieuwenhuis et al. (2006) found an ICC of 0.96 across a 1 - 2 hour period in 151 PwMS, indicating strong reliability. Construct validity has been demonstrated by moderate significant correlations between T25FW and quality of life (Cohen et al., 2014) and between T25FW and disability in MS (Learmonth, Motl, Sandroff, Pula & Cadavid, 2013b; Sandroff, Motl, Sosnoff & Pula, 2015a). Strict adherence to a protocol for instructions provided is crucial to maintain reliability (Gijbels et al., 2012). Studies utilising both anchor-based and distribution-based methods for determining a value for Minimum Clinically Important Change have concluded that a change of 20% should be considered clinically meaningful (Coleman, Sobieraj & Marinucci, 2012; Hobart, Blight, Goodman, Lynn & Putzki, 2013; Kaufman, Moyer & Norton, 2000).

The 2MWT is a variant of the Six Minute Walk test (6MWT), traditionally used in cardiac and pulmonary disease to assess exercise tolerance. Participants are asked to walk continuously, at a comfortable pace, as far as possible within two minutes and the distance walked is recorded. The nature of the walk differs between studies, either a back and forth route of 15 metres (Bohannon et al., 2014) or 30 metres (Gijbels, Eijnde & Feys, 2011) or by walking around a square (Sandroff et al., 2014). The 6MWT has been shown to be reliable in PwMS, with an ICC of 0.98 for a spectrum of PwMS and 0.95 for less mobile PwMS (Toomey and Coote, 2013). However, the 6MWT can be fatiguing for PwMS (Barr, Mcloughlin, Lord, Crotty & Sturnieks, 2014; Mcloughlin et al., 2016) particularly those with lower levels of walking ability (Feys et al., 2013), whereas the 2MWT has been found to have minimal impact upon gait variables, indicating that the test is not overly fatiguing (Feys et al., 2013). The Two Minute Walk Test (2MWT) is an equally valid measure of exercise tolerance to the 6MWT (Bohannon et al., 2014; Gijbels et al., 2011; Eijnde & Feys, 2011) and so is preferred in MS. Baert et al. (2015) suggested that a difference of seven to ten metres on the 2MWT can be considered to be clinically important.

Spatiotemporal gait parameters can be measured using a range of different methods, for example, accelerometry, 3D optical motion capture, foot switches or instrumented walkways (Muro-de-La-Herran, Garcia-Zapirain, & Mendez-Zorrilla, 2014). An instrumented walkway, a 5.18m long GAITRite 3.8 system, was available for the duration of the doctoral programme. This has been shown to have excellent agreement with the Vicon motion analysis system (Webster, Wittwer & Feller, 2005) with ICCs of between 0.91 and 0.99 for various parameters. Reliability was investigated in PwMS by Sosnoff, Klaren, Pilutti, Dlugonski & Motl (2015) with a 6-month gap between trials. They found that gait speed, step time, step length and cadence all had excellent reliability with ICCs over 0.9. ICCs for step width and percentage of gait cycle in double support were lower at 0.56 and 0.6 respectively. Menz, Latt, Tiedemann, Mun San Kwan and Lord (2004) conducted a reliability study with healthy young participants and older participants and found ICCs for step width of 0.85 and 0.49 respectively (*n* = 61). They suggested that the lower reliability of step width might be related to the resolution of the GAITRite mat or, more likely, the higher intra and inter-individual variability of step width, compared to other gait parameters. A further important issue is that the mean values obtained by Sosnoff et al. (2015) were obtained from only two passes of a 4.9m GAITRite mat and those obtained by Menz et al. (2004) were from three passes of a 4.6m long walkway. Wong et al. (2014) found ICCs of 0.81 - 0.84 for step width reliability in stroke (*n* = 46), based upon 2-4 passes of the mat (18 footfalls). It might be concluded that for spatiotemporal gait parameters that are more inherently variable, more passes of the GAITRite mat may be required in order to gain reliable mean values.

Kinematics of the trunk and pelvis can be assessed using an ordinal scale such as the Dynamic Gait Index (DGI; Forsberg, Andreasson & Nilsagard, 2013). This is a clinical assessment of gait stability that uses observation of stability during eight different walking tasks. This was considered but rejected because a more accurate, instrumented assessment of stability was required in order to have a strong chance of detecting any changes in stability with the shorts and because observation alone is not accurate enough for detecting changes in gait parameters (Williams, Morris, Schache & McCrory, 2009). In addition, assessment of the DGI requires walking in eight different tasks, which would add to participant burden. In the absence of a 3D motion analysis system, other options were considered and a set of Opal sensors (provided by APDM Wearable Technologies) were available for use during the project. These are Inertial Measurement Units (IMUs), a form of body-worn sensor containing an accelerometer, gyroscope and magnetometer. These detect acceleration and angular velocity, both of which can be used to measure stability of the trunk and pelvis. There were a number of options available for extracting data from IMUs and the simplest of these were reported by Corporaal et al. (2013) and Huisinga, Mancini, St. George and Horak (2013). Corporaal et al. (2013) used gyroscopes to directly measure angular velocity in different planes during a range of gait and balance tasks and demonstrated that trunk sway was higher in PwMS than in healthy, non-neurologically impaired people. They found strong correlations between trunk sway and both disability and dizziness severity. Huisinga et al. (2013) used peak-to-peak amplitude, mean and root mean squared of acceleration in the mediolateral plane and successfully identified differences in the variability of trunk and pelvic movement between PwMS and healthy participants. The approach used by Huisanga et al. (2013) was chosen because I felt that acceleration data might provide a more complete picture of unsteadiness in walking.

Gait variability, is relevant to evaluating orthotic shorts both in terms of the variability of trunk and pelvic kinematics described above and variability of spatiotemporal gait parameters. Reliable assessment of gait variability requires multiple walks, with an optimal reliability coefficient obtained at 50 gait cycles in healthy people (König, Singh, Von Beckerath, Janke & Taylor, 2014). Previous research on spatiotemporal gait variability in MS had utilised at least two passes over a 7.9m or 10m GAITRite walkway (Socie et al., 2013a; Flegel et al., 2012) suggesting that four passes over a 5.18m walkway would enable valid and reliable measures of variability. Movement variability is expressed as standard deviation (SD) and coefficient of variation (CV; SD divided by mean, multiplied by 100). To date, the reliability of spatiotemporal gait variability and trunk and pelvic movement have not been determined in PwMS.

Energy expenditure can be assessed using oxygen consumption but is often calculated from heart rate using a Physiological Cost Index. This requires participants to exercise for long enough to reach a steady state before heart rate is measured. An alternative approach, assessed in non-steady state conditions is the Total Heart Beat Index (THBI), which was shown to be reliable in people with spinal cord injury over a 5 - 10 minute walk (ICCs = 0.89-0.97; Hood, Granat, Maxwell & Hasler, 2002). The THBI was considered to be the most practical solution for measuring energy expenditure but has not been tested previously in PwMS and might be inaccurate in MS due the potential impact of autonomic problems on heart rate regulation (Bethoux and Bennett, 2011).

Cognitive demand of walking is assessed using a dual task cost paradigm, in which walking is assessed as a single task and then assessed again whilst the participant undertakes a simultaneous cognitive task. The dual task cost is the percentage change in walking ability when the cognitive task is added. Walking ability can be assessed in many different ways for a dual task cost assessment but Leone et al. (2015) suggest using maximal walking speed, because it requires more cognitive and motor resources than self-selected walking speed. Dual task cost is the percentage change in walking speed (walking speed as a single task minus walking speed as dual task divided by walking speed for single task, multiplied by 100). To discourage participants from prioritising walking over the cognitive task, participants are asked to divide their attention equally between the cognitive and walking task (Wajda, Roeing, Mcauley, Motl & Sosnoff, 2015). The impact of the cognitive task on walking speed and the impact of walking on the cognitive task should both be assessed (Leone et al., 2015). At the time the OSFeaMS study was being designed, reliability of this dual task cost assessment in PwMS had not been established. Unfortunately, recently, dual task cost assessed using the T25FW was shown to have poor reliability in MS (ICC = 0.45; Decavel, Moulin & Sagawa, 2019) and the implications of this will be discussed in later chapters.

The MSWS-12 is the only tool designed specifically to measure participant-perceived walking ability in MS (Kieseier & Pozzilli, 2012, Appendix 8). This measure asks 12 questions, rated 1-5 about the extent to which MS has impacted upon various aspects of mobility over the previous two weeks, such as running, stairs, distance, speed, smoothness of walking and need for support. It is believed to capture quality of movement, not just speed and endurance (Pilutti et al., 2013) and correlates well with overall walking function (Kieseier & Pozzilli, 2012; Motl et al., 2013; Motl & Snook, 2008a). There is evidence of strong psychometric properties, including internal consistency and test-retest reliability (Cronbach's alpha 0.97; ICC 0.94; Hobart et al., 2003). The MSWS-12 was found to be more responsive than the T25FW, the Expanded Disability Status Scale (EDSS) and various earlier self-report measures of walking

(Hobart et al., 2003). Mehta et al. (2015) suggested a difference of 8 points to be clinically important based upon a triangulation between anchor-based and distribution-based methods. In comparison, Baert et al. (2014) conducted a more thorough analysis in which they evaluated important change in groups with different levels of disability and used both patient and therapist perspectives as the anchors. They suggest 11.4 as the largest estimate for the whole MS group but a 14-point change for the more disabled participants.

Balance confidence can be assessed with two commonly used measures: the Falls Efficacy Scale - International (FES-I; Appendix 8) and the Activities-specific Balance Confidence measure (ABC; Appendix 8). As the names suggest, the FES-I asks specifically about fear of falling, whereas the ABC asks more generally about people's perception of their ability to maintain balance and keep steady. Orthotic shorts are designed to steady the pelvis and so, balance confidence, as measured by the ABC seemed more likely to be relevant. The ABC scale asks questions about confidence performing a range of tasks. It has been shown to be a reliable and valid measure of balance confidence having good internal consistency (Cronbach's alpha = 0.95, Nilsagård et al., 2012) and test-retest reliability (ICC = 0.92, Cattaneo, Jonsdottir & Repetti, 2007) in MS. It relates closely to measures of balance function, including frequency of falls (Cattaneo et al., 2007). Parry, Steen, Galloway, Kenny & Bond (2001) reported that language used in the original version, developed in North America, caused some confusion amongst elderly British users. They developed the UK version of the ABC known as the ABC-UK, which has UK English language but is otherwise identical in structure and content. To date, the Minimum Clinically Important Difference for the ABC scale has not been determined in MS but is considered to be 6.8 points for stroke survivors (Botner, Miller & Eng, 2005).

4.1.6 Summary of measurement review

In conclusion, some measures were chosen for use because they appeared to be relevant to walking ability, potentially changeable with the shorts and had strong psychometric properties demonstrated in PwMS. These were the T25FW, 2MWT, MSWS-12, ABC-UK and spatiotemporal gait parameters of step length, cadence and stride time and percentage of gait cycle in double support. Other measures were felt to be potentially changeable with the orthotic shorts but were more exploratory due to the weak or absent evidence around reliability in MS. These were the THBI for energy expenditure, step width, variability of spatiotemporal gait parameters, dual task cost of cognition on walking and the range and variability of mediolateral pelvic sway during walking.

4.2 A single case methods pilot

4.2.1 Introduction

Piloting methods for a research study can enable researchers to discover any problems with their protocol and take corrective action in the design of a planned study (Persuad, 2010). The choice of outcome measures felt particularly challenging for the OSFeaMS study because the measures proposed were potentially fatiguing and burdensome for participants.

During the FabO IPA study (Chapter 3), one participant expressed a desire to trial a pair of orthotic shorts. He had trialled DM Orthotics shorts previously but had not obtained National Health Service (NHS) funding. In the IPA study, he discussed his experiences with using cycling shorts as an orthosis but he believed properly manufactured orthotic shorts would be more effective. David did not fit the eligibility criteria for the OSFeaMS study, precisely because he had previous experience of trialling orthotic shorts. It became clear that a piloting of methods would be mutually beneficial, with David as our only participant.

4.2.2 Study aims

The study aimed to gain participant feedback on the burden associated with proposed objective measures, provide feedback on the relevance of the proposed measures and to provide feedback on the placebo shorts proposed for use in the OSFeaMS study.

4.2.3 Methods

The study protocol for the methods pilot was approved by Sheffield Hallam University Research Ethics Committee in January 2017 (Ref: 2016-17/ HWB-HSC-06; Appendix 9). The participant was a 47-year-old male with MS. He was formally invited to this pilot study in January 2017 by email with a Participant Information Sheet (Appendix 10). In

February 2017, he attended for Visit 1. At Visit 1, the proposed plan for the study was discussed in full and the consent form was signed (Appendix 11). Figure 4.2 shows an overview of the study.

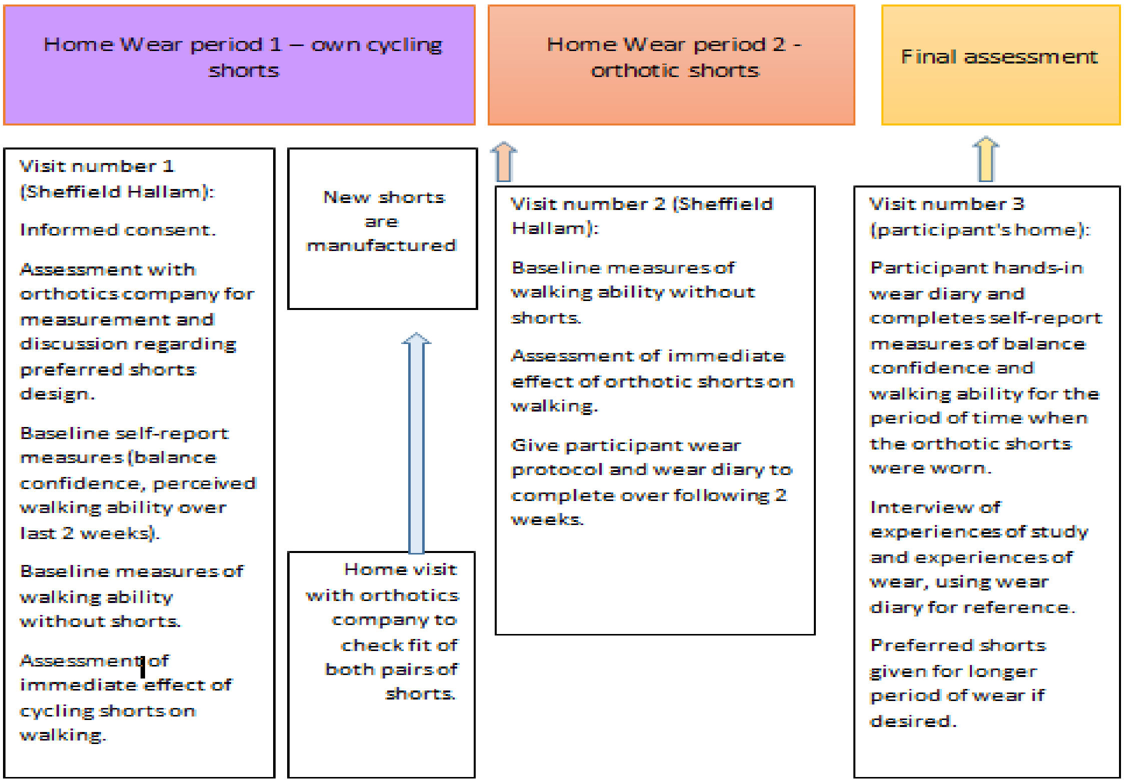


Figure 4.2: An overview of the methods pilot study.

Procedure

At Visit 1, the participant was measured for his orthotic shorts, completed the baseline self-report measures and his walking ability was assessed without, then with, his cycling shorts. Orthotic shorts were manufactured between Visits 1 and 2 and the representative from the orthotics company visited the participant at home to fit the shorts. Two pairs of shorts were made, one was an orthotic pair designed with the intention of improving his walking and the second a "placebo" pair designed to look similar but not be effective. Visit 2 included assessment of walking ability without, then with, the orthotic shorts. Following Visit 2, he trialled both the orthotic and placebo shorts at home and kept a wear diary (Appendix 12). At Visit 3, the researcher visited the participant at home. He handed back the wear diary, completed self-report measures and was interviewed regarding the impact of the orthotic and placebo shorts and the burden associated with the various measures.

Data collection

The measures of walking ability trialled in this pilot have been described and evaluated in Section 4.1. Objective measures were used at Visits 1 and 2 (summarised in Table 4.1). Self-report measures were used at baseline and following the orthotics shorts home trial.

Table 4.1: Objective measures used for assessment of walking ability in the methods pilot

Measures	Methods used
Spatiotemporal gait parameters	Each walking test involved the participant performing four consecutive walks over the GAITRite 3.8 walkway at a self-selected pace. Each walk started two metres before the walkway and finished two metres after the walkway, to assess only the steady state of walking. Data for each walking test was downloaded on mean cadence, gait speed and percentage of gait cycle in double support. Data on individual steps/strides was downloaded for step length, stride time and step width and means and variability calculated and reported. Variability was reported using standard deviation and coefficient of variation.
Trunk and pelvic stability	Opal sensors were attached via straps over clothing, on the sacrum and the sternal angle. Position of the sensors was confirmed by palpation of the spine, the suprasternal notch and the sternal angle. Participant walked at a self-selected pace along a straight corridor, approximately 30 metres in length.
Walking speed/Timed 25 Foot Walk	Start and finish points were marked on the floor using masking tape, 25 feet apart. The time taken to walk the 25 feet from a standing start was recorded. The stopwatch was started when the participant's foot crossed the first line and stopped when their foot crossed the second line. The test was performed twice with a 1-2 minute rest between each trial. The mean of the two trials was calculated and converted from time taken to average velocity in metres per second.
Dual task cost/cognitive demand of walking	Dual Task Cost was assessed following assessment of the Timed 25 Foot Walk by walking the same distance a third and fourth time whilst reciting alternate letters of the alphabet. The participant was asked to divide attention equally between the two tasks and the number of letters correctly given was recorded. Dual Task cost on walking was calculated as the percentage change in T25FW (walking speed as a single task minus walking speed as dual task divided by walking speed for single task multiplied by 100). The impact of walking on the cognitive task was estimated by calculating the number of letters correctly given in each second of walking (Wajda et al., 2015).
Walking distance/Two Minute Walk Test	Distance covered over a two-minute walk was assessed with a stopwatch recording time and the participant walking back and forth along a 10-metre walkway, pivoting at either end.
Energy expenditure	Energy expenditure was assessed at the same time as the Two Minute Walk Test. The participant was fitted with a heart rate monitor prior to the start of the walk test and heart rate was recorded as one-second intervals during the assessment. The total number of heartbeats was calculated from this and the Total Heart Beat Index calculated by dividing the number of heartbeats by the total distance walked.

4.2.4 Key learning from the methods pilot

The participant had a significant relapse between Visit 1 and 2. He chose to go ahead with Visits 2 and 3 and he completed all the objective measures on Visits 1 and 2. However, he deteriorated significantly as Visit 2 progressed, meaning that comparison between the orthotic shorts and his cycling shorts was not valid and neither was the Visit 2 within-day comparison of "shorts off" versus "shorts on". Data for evaluating objective measures was obtained at Visit 1, where he was assessed without and with his cycling shorts.

Aspects confirmed for inclusion in the OSFeaMS study

David felt that the shorts designed as a placebo were comfortable but ineffective in improving his walking and, therefore, would be appropriate in the OSFeaMS study as a placebo. He had not worn them for long during the home trial as he felt he needed the support provided by the orthotic shorts.

The self-report measures (MSWS-12 and ABC-UK) seemed easy to complete and changes in perceived walking ability and balance confidence were apparent and tallied with participant feedback. The wear diary was completed as requested and appeared useful as a means of noting observations on abilities as well as the actual wear times.

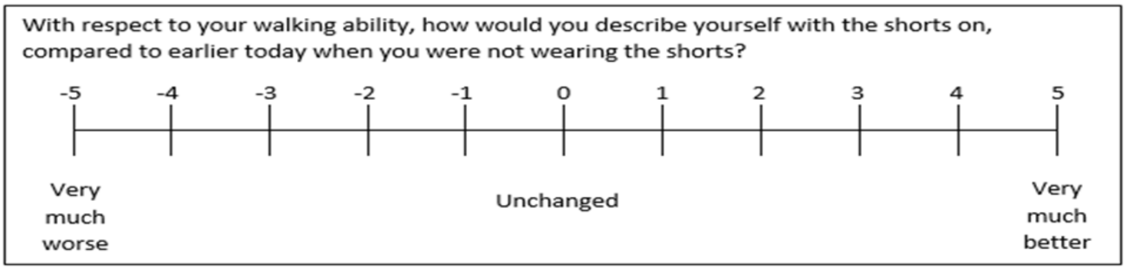
No issues arose with the T25FW, dual task or use of the GAITRite for spatiotemporal gait parameters. David felt that all the objective measures were relevant to the aspects that he believed changed when he walked in shorts. He believed he could walk faster, walk further, be more stable and be better able to think and walk simultaneously with the shorts. The T25FW showed improved gait speed with the shorts. Dual task cost did not change with the shorts but David's ability to manage the simultaneous cognitive task was improved. The spatiotemporal gait parameters obtained from the GAITRite mat changed with the cycling shorts on Visit 1, appearing to indicate an impact of the cycling shorts on step length, step width and gait variability.

Aspects removed from the OSFeaMS protocol

The 2MWT was clearly too fatiguing and was not used in the OSFeaMS study. David felt it was relevant to what the shorts did for him but was aware that fatigue caused by the 2MWT had influenced his performance on the later tests. There was no difference in number of heartbeats when the 2MWT was attempted with "shorts off" compared to "shorts on" but he could not walk as far on "shorts on" tests, largely due to fatigue. Due to lack of space in the facility, the 2MWT was conducted by walking forwards and backwards on a 10m line. It was apparent that he was trying to walk faster with the cycling shorts on and his balance during the 180° turns at either end of the line was poor. There were two near falls during this test on Visit 2. The test was dropped from the OSFeaMS protocol because it seemed unsafe, fatiguing and burdensome.

Aspects added to the OSFeaMS protocol

A participant Global Rating of Change (GRC) was added to the protocol for the OSFeaMS study in order to capture participants' ratings of change in their walking ability at the time the shorts were first trialled. The initial plan had been to discuss participants' first impressions of the shorts only in the final interview but the methods pilot highlighted the potential impact of time as a confounding factor for people remembering their first impressions of the shorts. Participant-rated GRC scores have been found to correlate well with more extensive patient-reported outcome measures (Watson et al., 2005) and measures of patient satisfaction (Fischer et al., 1999). Reliability of such scales is considered strong, with ICC values over 0.84, a minimum detectable change of 0.5 on an 11-point GRC scale and a minimum clinically important change of 2 points (Kamper, Maher & Mackay, 2009). The GRC scale used in the OSFeaMS study was designed according to guidelines suggested by Kamper et al. (2009) and is shown in Figure 4.3.



With respect to your walking ability, how would you describe yourself with the shorts on, compared to earlier today when you were not wearing the shorts?

Figure 4.3: Global Rating of Change Scale

A falls diary (Appendix 13) was added to the OSFeaMS study protocol in response to the methods pilot. David made frequent reference in his final interview to falls he had sustained over just the previous few weeks and felt the shorts impacted upon how likely he was to fall. Falling is not uncommon in PwMS and is a serious issue because it can lead to further injury and increased disability due to physical factors related to a fall or the psychological consequences of falling (Finlayson & Peterson, 2010). I was concerned that the ABC-UK scale in itself would not be sufficient to capture any changes to falls incidence, particularly because David was falling during tasks not included on the ABC-UK, such as mowing the lawn and carrying firewood. Coote, Sosnoff and Gunn (2014) suggested that falls in PwMS are not well predicted by measuring balance confidence and they suggested including actual measurement of falls incidence in studies where there might be a positive or negative impact on falls risk. Coote et al. (2014) recommend a 3-month recording period for falls frequency with bi-weekly reminders by phone and monthly returns for falls intervention studies. Only two-week recording periods were used in the OSFeaMS study, so a valid assessment of fall incidence was not achievable. Nevertheless, a falls diary was included in the OSFeaMS study in order to gain feedback on its relevance for future trials.

Other changes made in response to the methods pilot

The degree of fatigue seen in David prompted counter-balancing of the order in which the different objective tests were performed for different participants. One objective of the OSFeaMS study was to determine which measures might be most suitable for a future RCT and an order effect would have influenced this decision. Counterbalancing minimises the impact of order effects.

The plan for analysing the IMU data changed significantly in response to the methods pilot. The original plan was to analyse acceleration data in the sensor coordinate system, meaning that the mediolateral acceleration of the sensor would be used without any attempt to convert it into a global reference frame, such as anatomical planes. The data obtained in the pilot suggested that trunk and pelvic acceleration changed with the shorts. The range and root mean square of mediolateral acceleration of the sternal sensor were reduced (from 7.4 m/s^2 to 7.2 m/s^2 and from 1.6 m/s^2 to 1.5 m/s^2 respectively), maybe indicating increased thoracic stability in walking with the shorts on. A plot of acceleration over time for the thoracic sensor (Figure 4. 4) indicated changes in the pattern of trunk movement but it was not possible to interpret what these might indicate. Acceleration of the pelvic sensor changed with the shorts but became more variable, with a larger range (25.4m/s^2 without shorts to 31.6m/s^2 with shorts) and root mean square (from 2.1m/s^2 without to 2.6m/s^2 with) and a more complex, "noisier" plot.

Increased variability of mediolateral pelvic acceleration could be due to increased pelvic rotation associated with increased gait speed. The pelvis rotates in the transverse plane during walking. For example, at heel strike, the pelvis is rotated forwards on the side about to make heel contact. With an increase in walking speed, the degree of pelvic rotation increases in order to increase step length (Liang et al., 2014; Whitcombe, Miller & Burns, 2017). When acceleration data were analysed in the sensor coordinate system, as pelvic rotation increased, the sensor's mediolateral axis would have become aligned more closely to the direction in which the participant was walking. What appeared to be an increase in mediolateral acceleration of the pelvis could have been an increase in anteroposterior pelvic acceleration. To overcome this problem, it was necessary to convert the data from the sensor coordinate system to an external coordinate system.

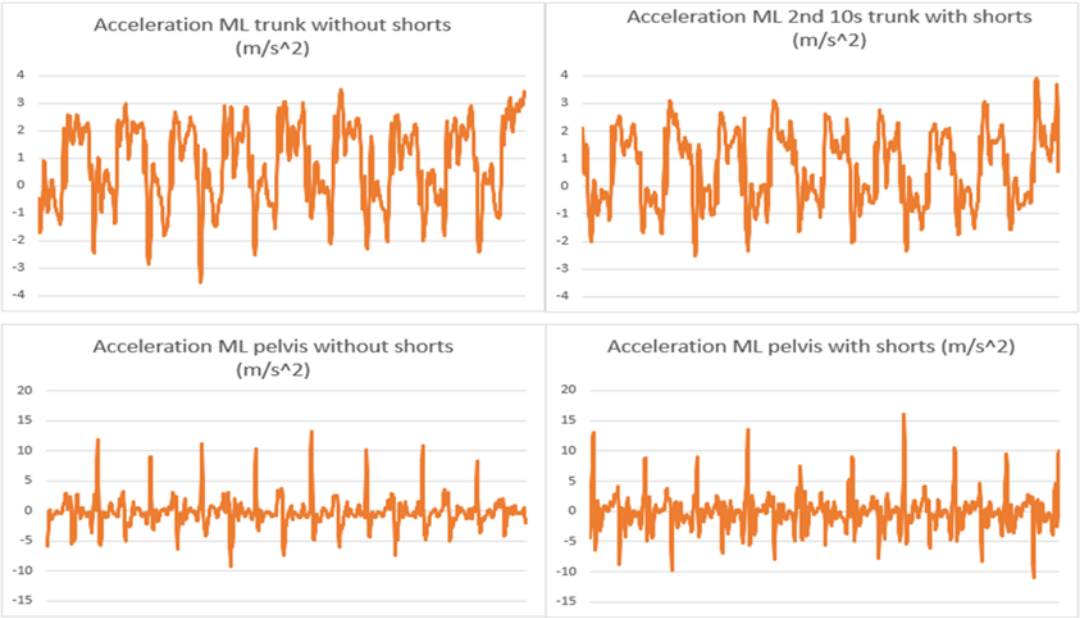


Figure 4.4: Mediolateral acceleration over a 10-second sample of walking with and without cycling shorts at Visit 1 in methods pilot.

There are a number of different techniques available for converting acceleration data into an external coordinate system. The IMUs contain magnetometers that orientate to magnetic North and the accelerometers provide information on the direction of gravity. These data can be used to provide a means of converting data from the sensor coordinate system to a global coordinate system orientated with respect to magnetic North. Unfortunately, data were collected indoors where the metal structures embedded within the building interfered with detection of magnetic North. The manufacturers of the Opal sensors suggested that data could be downloaded without the magnetometers and orientated to an arbitrary reference frame. This was tested by attaching an Opal sensor to a piece of laboratory equipment that rotated slowly in a constant pattern. Unfortunately, data downloaded from this Opal drifted significantly over time, indicating that it was not likely to be valid for assessing a 30-second sample of walking.

A further research review revealed a method devised by Brodie, Psarakis and Hoang (2016), which was superior to the original idea of using acceleration to determine trunk and pelvic stability. Their method enabled the IMUs to be used to determine distance moved in 3D space. They had developed a filter using step frequency to filter out large amplitude changes in acceleration associated with changes in sensor orientation and heel strike. The filter was created by comparing data obtained from the IMUs to data obtained from a 3D optical motion capture system. They then used data on angular velocity obtained from the gyroscope and acceleration from the accelerometer to determine changes in the orientation of the sensors with respect to the direction in which their model was walking (termed the Global Vertical Body Heading, GVBH), which approximately aligns to anatomical planes. This enabled acceleration to be aligned to the GVBH in three axes: the anteroposterior axis, aligned to the direction of heading, the vertical axes, aligned to gravitational vertical and the mediolateral axis, which was perpendicular to the anteroposterior and vertical axes. The actual distance that the pelvis moved in these axes was obtained by double-integrating acceleration. Brodie et al. (2016) validated this technique against a 3D optical motion capture system and used it to demonstrate differences in pelvic sway between PwMS and healthy controls in both the anteroposterior and mediolateral axes. The same group developed a technique to use the IMUs to determine rotation of the trunk and pelvis. This technique was believed to be more accurate than integrating the angular velocity data obtained from the gyroscopes because of the corrections introduced for changes in walking direction. In addition, they calculated sway variability, which was the standard deviation of the trajectories of the trunk or pelvis over multiple strides in all three movement axes (Psarakis et al., 2018).

The technique described above was superior to the original analysis plan because it enabled detailed analysis of trunk and pelvis kinematics. Brodie et al. (2016) suggested that the codes available to analyse IMU data could be shared, so these were requested from the authors. They did not share the code but offered to analyse the IMU data for the OSFeaMS study themselves. In the OSFeaMS study, to ensure that acceleration data could be accurately filtered with reference to step frequency, IMUs were attached to ankles as well as pelvis and thorax.

4.3 Conclusion

This chapter has reviewed and appraised measures for walking ability and balance, ensuring that measurement for the OSFeaMS study was based upon a thorough review of relevant outcomes and measurement tools. The methods pilot provided preliminary indications that the placebo shorts would be an appropriate comparator and that most of the measures proposed would be acceptable and relevant to the impact of the shorts. Measures of energy expenditure and walking distance were removed from the OSFeaMS protocol due to poor tolerance of the 2MWT. By conducting a preliminary analysis of data arising from the methods pilot, it became clear that the initial plan for analysing the IMU data was inadequate and this plan was adapted and improved prior to finalising the protocol for the OSFeaMS study.

The next chapter reports the methodology and methods for the OSFeaMS study.

Chapter 5: The Orthotic Shorts Feasibility in MS study – methodology and methods

Summary

This chapter explains the methodology and methods for the mixed methods Orthotic Shorts Feasibility in MS (OSFeaMS) study. The results of the OSFeaMS study will be reported in later chapters: Chapter 6 is devoted to quantitative data and Chapter 7 to qualitative data. Qualitative and quantitative findings will be integrated in Chapter 8.

5.1 Aims and Objectives

The OSFeaMS study aimed to determine the feasibility of using orthotic shorts for improving walking in PwMS. Feasibility was considered as including acceptability and potential efficacy. In addition, the feasibility of a potential future trial was investigated by evaluating recruitment and retention rates, evaluating which measures might be most relevant and gaining participant feedback on study design.

The study objectives were:

1. To investigate acceptability of orthotic shorts:
    a. Determine how long shorts were worn as a proportion of time recommended
    b. Determine the proportion of people who opted to keep the shorts after the study
    c. Explore participant perceptions around acceptability, including advantages and disadvantages and perception of initial effect

2. To determine the immediate, orthotic effect of shorts on the following objective measures of walking ability:
    a. Spatiotemporal gait parameters and gait variability
    b. Self-selected gait speed and maximal gait speed
    c. Trunk and pelvic stability in walking
    d. Dual task cost of a cognitive task whilst walking

3. To determine the impact of wearing orthotic shorts on:
    a. Participant perceived effect on walking in the movement laboratory
    b. Self-reported balance confidence
    c. Self-reported perception of walking ability

4. To provide specific information to inform a future randomised controlled trial:
    a. Determine the ease of recruitment and retention
    b. Determine the suitability of measurement techniques for assessing the impact of orthotic shorts
    c. Gain participant feedback on research methods.

5.2 Methodological approach

A series of choices were made in deciding upon methods for the OSFeaMS study: firstly, the decision to consider this as a "feasibility study" and to define what that means in this context; secondly, the decision to use mixed methods and, thirdly, the decision to design a crossover study.

5.2.1. What is a feasibility study?

The National Institute for Health Research (2010) stated that feasibility studies are pieces of research done before a main study in order to answer the question "Can this study be done?" They recommended listing aspects of trial design that are uncertain and designing feasibility studies to achieve greater certainty around those factors. Examples might be the willingness of participants to be randomised, recruitment rates and retention rates. Thabane et al. (2010) and Tickle-Degnen (2013) explained that a feasibility study should take place before a Phase III trial and after Phase I and Phase II studies. Phase I and II studies provide reliable information about safety and include "proof of concept" studies (Thabane et al., 2010, p3), investigating the potential efficacy of an intervention. For example, Phase II studies might assess efficacy using surrogate markers or short-term effects, where a short-term effect is required but not sufficient to indicate a longer-term effect. In this definition of feasibility study, feasibility considers mainly the processes involved in a future RCT, the resources and administrative support required to deliver that RCT and scientific issues around safety, dose, estimation of treatment effect and estimation of variance in the treatment effect.

In contrast, other writers consider that a feasibility study can and should include some testing of efficacy (Bowen et al., 2009; Orsmond & Cohn, 2015). Bowen et al. (2009) explained an approach to feasibility studies that focusses entirely on investigating the feasibility of interventions rather than the feasibility of trials. They considered a feasibility study should include acceptability of the intervention, demand for the intervention, how the intervention might be implemented, how practical it is within a specific context, how it might be adapted for or integrated within current systems of support or provision and some limited efficacy testing. Orsmond and Cohn (2015) suggest five objectives for a feasibility study in which the first four are about determining the feasibility of an RCT and the fifth is about "preliminary evaluation of participant responses to the intervention" (p5). A possible reason for the different approaches is that the feasibility approach described by the National Institute for Health Research (2010) is relevant mostly to drug trials in which Phase I and Phase II trials have already established potential efficacy prior to planning an RCT.

The OSFeaMS study aligned most closely to the Bowen et al. (2009) definition of a feasibility study and, therefore, tested the feasibility of orthotic shorts as an intervention. This seemed more in keeping with the guidelines on Developing a Complex Intervention (Craig et al., 2008). Because the mapping review found no previous research on orthotic shorts, it was necessary to include preliminary evaluation of participant responses to the intervention.

5.2.2 Why mixed methods?

A mixed method study includes both qualitative and quantitative data collection and analysis and, crucially, integration between these two types of data collection (Creswell & Plano Clark, 2011).

A mixed methods approach was chosen because it was felt to be the best match to the study aims. For example, whilst it is possible to consider acceptability purely from a quantitative perspective (how many people continued to use the shorts after the trial? how long were the shorts worn for during the home trial?), it is more appropriate to understand acceptability by discussing participants' experiences in depth. For this aim, qualitative data collection was important. In contrast, investigating the short-term effect of the shorts on walking ability necessitated quantitative methods with minimal use of qualitative data. Gaining an overview of the impact of the orthotic shorts required integration of qualitative and quantitative data, to achieve a more in-depth understanding than could be achieved with one method alone. Such an in-depth understanding is important to enable intervention development because it can provide insight into the reasons behind the more quantitative findings, thus directing future research and refining an intervention. To address these aims, the OSFeaMS study required a convergent parallel design, which involved concurrent collection of qualitative and quantitative data (Creswell & Plano Clark, 2011). This design enabled qualitative feedback on the orthotic shorts themselves and the study processes, alongside collection of quantitative data.

Mixed methods were applied from a philosophical stance of critical realism. Critical realism combines the ontological perspective of realism (a real world exists independent of our perception) with the epistemological perspective of constructivism (our understanding of the real world is a construction based upon our own perspectives and points of view) (Maxwell & Mittapali, 2010; Teddlie & Tashakkori, 2010). This stance recognises the difficulties inherent in trying to understand "reality" through research. For example, there needs to be reflection on the researchers' assumptions and perspectives in any research study. In addition, critical realism is extremely relevant to understanding the perceptions of service users because it reminds us that their understanding of reality is based upon their perspectives and points of view. This captures a similar understanding of practice as the biopsychosocial model of practice (Pilgrim, 2015). In other words, whilst an intervention may or may not have biomedical or biomechanical effects, it is an individual's beliefs in those effects and how important they appear that determines perception of effect and acceptability.

5.2.3 Why a crossover study?

A crossover design was chosen for the OSFeaMS study. The orthotic shorts were compared to a "placebo" pair of shorts designed to look similar but provide no compression of the tissues and no changes to movement control.

A crossover design was suited to the objective assessment of walking ability because the study was focussed on determining if there was a short-term, direct orthotic impact. If an intervention has a direct orthotic impact then, by definition, this is detectable immediately, i.e. people walk better with the orthosis than without. It is possible that the experience of wearing an orthosis would alter the way somebody moves after removal, however, by definition, an orthotic effect ought to disappear once an orthosis is removed. Crossover studies are a commonly used and useful research design where the effects of an intervention are short-lived and reversible and the underlying condition is chronic and relatively stable (Mills et al., 2009). One key advantage of a crossover study over a parallel group design is that fewer participants are required; each participant tries out two different interventions, essentially producing two matched groups.

Good practice guidelines for crossover studies were developed by Ding et al. (2015) and include similar guidelines to those suggested for a RCT, plus the following specific quality standards for crossover studies. Firstly, the crossover design should be used when the participants' condition is chronic and stable, the intervention should not provide permanent change but only temporary relief and the effect of the first intervention should not last into the second treatment period. Secondly, the order of interventions needs to be randomised and, thirdly, the researchers must check whether there is a carry-over effect.

5.3 Methods

The study protocol was approved by the National Research Ethics Service (NRES) Committee for Leeds East on the 19th May 2017 (Ref: 17/YH/0140) and received approval from the Health Research Authority on the 11th December 2017 (Appendix 14). Sheffield Teaching Hospitals NHS Foundation Trust confirmed they had capacity and capability to deliver the study on the 7th January 2018.

The study protocol was made publically available on the Clinical Trials.gov webpage (Ref: NCT03164031).

5.3.1 Design and setting

This was a randomised crossover trial investigating the impact of orthotic shorts versus a pair of placebo shorts. The shorts were tested in two main ways: firstly the impact of the shorts on objective walking ability when first worn and, secondly, the impact of the shorts on perceived ability over a two-week home trial period. Each participant was involved with the study for approximately three months, during which they trialled both pairs of shorts. Each participant had seven appointments, four at the university and three in their own homes. Figure 5.1 provides an overview of participant progression through the study.

Each home trial period was followed by a two-week "wash out" period, designed to return the participant to their baseline ability if the shorts had had any longer-term impact on their walking. The length of this washout period was determined in consultation with orthotists and therapists at DM Orthotics, who felt that any carryover effect from a two-week trial was unlikely to last beyond 10 days. Following each washout period, there was an objective assessment of walking ability without any shorts to assess the underlying stability of the participants' condition and to determine if there had been any carry-over effect.

5.3.2 Participants

Sixteen participants were recruited over an eight-month period from January 2018 to August 2018. This sample size was planned, as it was felt that 16 people would be adequate for a sample representative of the different characteristics that might influence shorts use, for example, gender, age and nature of impairment (e.g. ataxia, spasticity, weakness). Participants were recruited according to the eligibility criteria reported in Table 5.1.

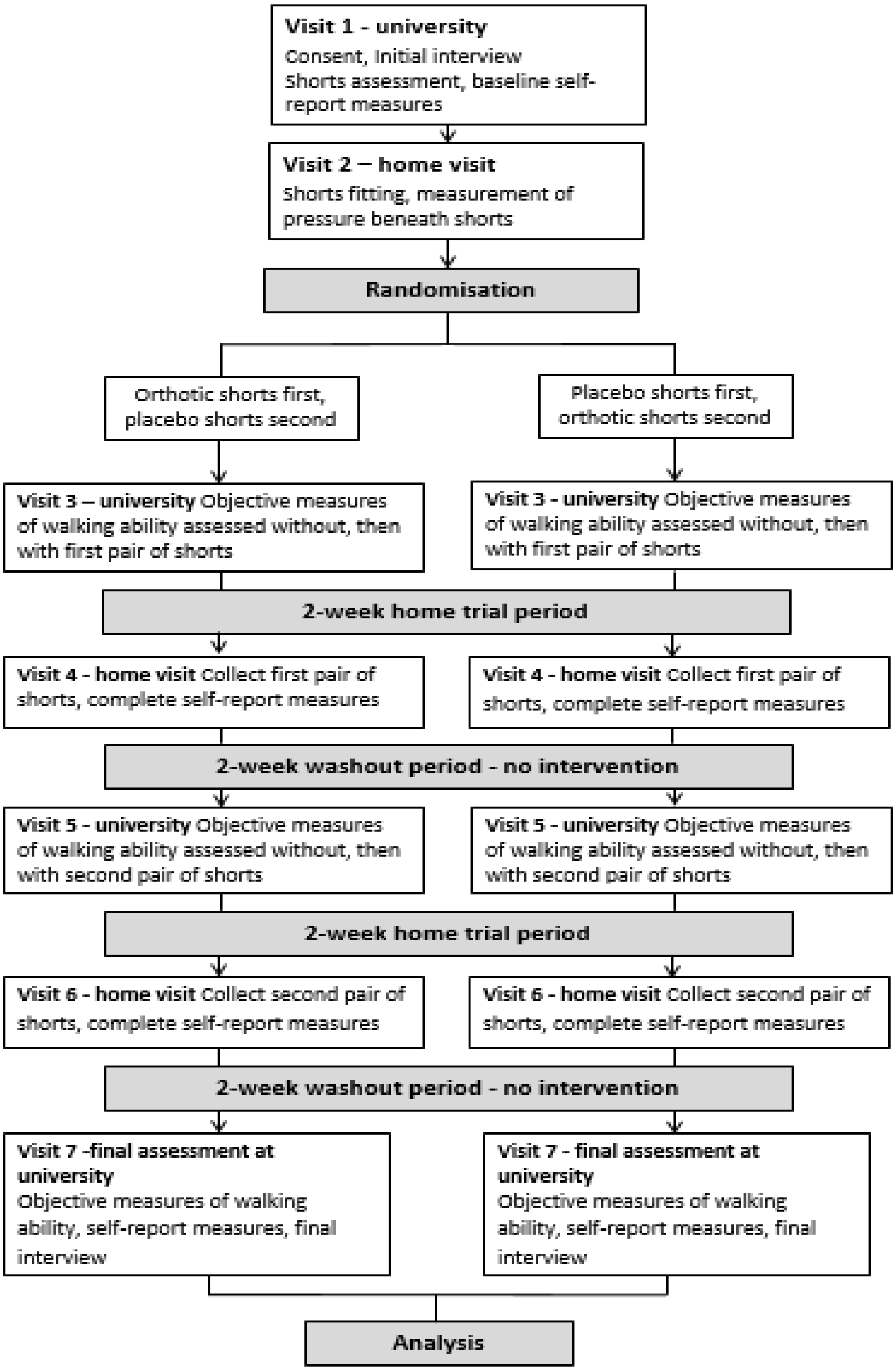


Figure 5.1 Overview of study design

Table 5.1 Eligibility criteria for the Orthotic Shorts Feasibility in MS study

Inclusion criteria	Exclusion criteria
Diagnosed as having multiple sclerosis of any type (relapsing-remitting, primary progressive or secondary progressive). Clinically stable – had not experienced relapse in the last 4 weeks; had not commenced a novel therapy in the last 3 months. Able to travel to Sheffield Hallam University for assessments. Difficulty walking with a subjective feeling of instability around the hips or lower trunk. Able to walk for at least 2 minutes at a time. Able to provide written record of informed consent.	People who did not meet inclusion criteria. Skin conditions that might be exacerbated by tight clothing. Circulatory problems that might be exacerbated by tight clothing, such as varicose veins, previous thrombosis, venous or arterial insufficiency. Cognitive problems including memory disturbance that might influence recall of experience and adherence to guidance. Pregnancy – because of related circulatory problems. Living further than 10 miles from Sheffield if they were unable to provide own transport. Had previously used orthotic shorts.

5.3.3 Recruitment

Participants were recruited from Sheffield Teaching Hospitals NHS Foundation Trust; from the Sheffield MS Therapy Centre and via the protocol published on the Clinical Trials.gov webpage (Clinical Trials, no date).

There were three methods of recruitment within Sheffield Teaching Hospitals. The main strategy was to identify potential participants who had been treated by the specialist physiotherapy service for PwMS. They had created a list of PwMS who had attended physiotherapy between October 2016 and January 2018 and whom the physiotherapists judged to have trunk or pelvic instability. The physiotherapy team contacted potential participants in batches by telephone. Those who expressed initial interest were sent a Participant Information Sheet and recorded in a paper recruitment log, which was given by hand to myself as the chief investigator. Other recruitment strategies were attempted at Sheffield Teaching Hospitals after everyone on the physiotherapists' list had been approached. The MS Support Nurses identified potential participants from their clinic list and the study was advertised via fliers (Appendix 15) given out at the regular Tysabri drug clinic.

The Sheffield MS Therapy Centre distributed paper copies of the flier and forwarded an email invitation to everyone on their membership list. The researcher attended the Centre to discuss the study with anyone who was interested in participating. The study was explained, Participant Information Sheet provided and questions answered. Eligibility was assessed for those who were interested and potential participants provided contact details.

The study was advertised on the Clinical Trials.gov webpage. Some potential participants located this via a search engine and others were forwarded to the site from the webpage for the MS Society UK. Interested individuals contacted the researcher directly by phone or email. In addition, a local private physiotherapy clinic distributed flyers to their clients.

With all of the above recruitment strategies, when contact was first made between the potential participants and the research team, there was an initial informal discussion about the study and the Participant Information Sheet (Appendix 16) was provided as a paper or email copy according to the individuals' preference. Potential participants had a week to read the information and consider their involvement. As chief investigator, I then phoned potential participants and answered any outstanding questions. If people were willing to join the study, we agreed a date for their first appointment at the university. When they attended that first appointment, there was further opportunity for them to ask questions and informed consent was recorded in writing (Appendix 17).

5.3.4 Procedure

As illustrated in Figure 5.1, each participant had seven visits. Visits 1, 3, 5 and 7 were at Sheffield Hallam University.

Visit 1 included informed consent, collection of basic participant details, an initial semi-structured interview around expectations and motivations, completion of the self-report measures on perceived walking ability and balance confidence, assessment and measurement for shorts with a representative from DM Orthotics and provision of a falls diary (Appendix 13) for completion over the following two weeks.

Visits 3 and 5 followed the same structure, with one visit testing one pair of shorts and the other visit the other pair of shorts. Firstly, walking ability without the shorts was measured. Then participants put the shorts on and spent 30 minutes becoming familiar with them before the objective measures were repeated. After the objective assessments, participants were provided with written information on wearing and caring for the shorts, a wear diary (Appendix 12) and another falls diary for them to complete during the home trial.

Visit 7 included a final measure of walking ability without shorts, a completion of the final self-report measures and a semi-structured interview around the participants' experiences of the shorts and the trial.

In between the above visits to Sheffield Hallam, there were three shorter visits. Each participant could choose to attend university or meet in an agreed location. Visit 2 was with the representative from DM Orthotics to check the fit of the shorts and measure the pressure exerted by the shorts. When adjustments were required, the shorts were sent back to the factory and adjustments completed within three days. The shorts were delivered to the researcher. Visits 4 and 6 involved the researcher collecting the shorts, collecting the diaries and completing the self-report measures on walking ability and balance confidence with the participants.

5.3.5 Randomisation and blinding

The crossover study was designed to test differences between the orthotic shorts and a placebo pair of shorts. Therefore, it was planned to randomise and counter-balance the order in which the shorts were tested. This was difficult to achieve because of the lack of funding for an external randomisation service and the fact that, as chief investigator, I was also the assessor and needed to be blinded to the order in which the shorts were tested.

A colleague created a randomisation schedule, according to my instructions, using the Sealed Envelope online system (Sealed Envelope, n.d.) for blocked randomisation. This randomisation schedule was kept in the Site File in an opaque envelope. When a participant needed to be provided with their shorts, in order to maintain blinding, a member of Sheffield Hallam teaching staff was asked to place each pair of shorts into an A4 box file according to the randomisation schedule. The box files were labelled "First pair of shorts" and "Second Pair of shorts" and sealed. The first pair of shorts was given to the participant at Visit 3, the second pair of shorts at Visit 5. I was unable to feel which pair of shorts was in which box. At Visits 3 and 5, participants would go behind a screen and change into their shorts, putting their clothes on top afterwards. They were asked not to make any comments that might reveal whether they were wearing the "looser pair" or the "tighter pair" of shorts.

Unfortunately, counter-balancing was not successful. The randomisation schedule was completed with the words, "Group 1" and "Group 2" instead of "Orthotic shorts are first pair of shorts" and "Placebo shorts are first pair of shorts". This meant that people allocating shorts to boxes needed to cross-reference with the randomisation instructions to allocate appropriately. The confusion resulted in two errors where the orthotic shorts were provided first instead of placebo. Thus, 10 participants received orthotic first and six placebo first.

Blinding of the chief investigator/assessor was successful for most participants who completed the study. For four participants, unblinding occurred because participants commented upon how the tightness of one pair of shorts differed from the other. All participants remembered not to comment on this when the shorts were first provided but this was revealed in conversation after the home trials.

Participants were not told that a placebo pair of shorts was included in the study. The Information Sheet (Appendix 16) explained that they would test two different pairs of shorts, one of which was tighter than the other. Participants were expected to detect the difference between the two pairs of shorts in terms of their tightness. Only one participant could not detect which shorts were tighter. Participants' perceptions around the placebo shorts were explored in the final qualitative interview at Visit 7. Their feedback is reported in detail in Chapter 7. In brief, eight participants had expected one of the pairs to be a placebo and seven of these believed that the looser pair was the placebo pair, indicating that many participants were not successfully blinded.

5.3.6 Interventions - the orthotic and placebo shorts

DM Orthotics provided both the orthotic and placebo shorts, custom-made for each participant. The orthotic shorts (Figure 5.2) were constructed from a base layer that fitted snugly and provided some compression, plus an added layer of panelling. “Standard” shorts included panelling designed to extend the hips. “Abduction assistance” shorts included additional panelling designed to encourage hip abduction in people with a narrow base of support. The placebo shorts consisted of just the base layer fabric with no additional panelling and were a slightly larger fit than the orthotic shorts. To ensure that both pairs of shorts looked similar, both base layer and panelling were the same colour.

Figure 5.2: Illustration of the DM Orthotics orthotic shorts (base layer is red and panelling black)

All shorts extended down to approximately 4 cm above the knee and were waist high or extended upwards to include the lumbar area. At Visit 1, participants' walking patterns were assessed by the chief investigator and the representative from DM Orthotics to determine the optimal height of the shorts and whether to include abduction assistance. Participants were given the following choices for their shorts design, which were all discussed with the chief investigator and DM Orthotics representative: colour, presence or absence of side zips, presence or absence of a "toileting hole".

For both pairs of shorts, participants were instructed to increase the period of time that the shorts were worn for gradually, starting with one hour on the day of testing and doubling the time worn for three subsequent days before wearing up to 14 hours a day for the fifth and consecutive days. This strategy is a standard precaution used by DM Orthotics to guard against unanticipated skin reactions or adverse musculoskeletal effects.

The DM Orthotics representative measured the pressure beneath the shorts at Visit 2 when the fit of the shorts was assessed. A Kikuhime pressure monitor was used, which consists of a small air-filled bladder that is inflated via tubing attached to a pressure monitor. This was inserted from above and placed beneath each pair of shorts over the left gluteus maximus muscle with the participant standing. This location was chosen as potentially being relevant to pelvic stability and because pressure measurement over bony prominences results in greater error (Partsch et al., 2006). This device is commonly used in compression garment studies and is reliable and valid for this purpose (Brophy-Williams et al., 2014b). The error of measurement is reported to be is 1.3 ± 0.9 mmHg (Brophy-Williams et al., 2014b).

5.3.7 Quantitative data collection

Data using quantitative outcome measures were collected at the following time points. Firstly, baseline self-report measures were assessed at the first visit. Secondly, when each pair of shorts was first issued at Visits 3 and 5, participants were assessed on objective measures of walking ability and participant-perceived impact of the shorts. For all objective measures, participants were first assessed without shorts and then with shorts, enabling within-day and between-day comparisons. Thirdly, falls diaries and wear diaries were collected and self-report measures completed at the end of each home trial period (Visits 4 and 6). Finally, participants were assessed at Visit 7, two weeks after the second home trial to determine the stability of objective measures and self-report measures over the course of the study. At the final visit within their qualitative interview, participants were asked whether they wished to keep their shorts.

Decisions regarding the measures used in this study were informed by the methods pilot reported in Chapter 4. In-depth information about the reliability and validity of outcome measures is provided in Section 4.1.5. Below is an overview of the measures used.

Measuring acceptability

Participants recorded in a wear diary (Appendix 12), the times they put their shorts on and took them off each day. Adherence to the recommended wear times was calculated by taking the hours worn each day and expressing this as a percentage of the wear time requested over each trial period, assuming a full day to be 14 hours long. Whether each participant wanted to keep either or both pairs of shorts for future use was recorded during the final interview.

Objective measures of walking ability

Objective measurement of walking ability involved (1) use of a GAITRite mat for gait speed and cadence at a self-selected walking speed and spatiotemporal gait parameters, (2) use of Inertial Measurement Units (IMUs) for assessing trunk and pelvic stability in walking and (3) use of the Timed 25-foot Walk (T25FW) for assessing maximal gait speed and dual task cost. The order in which these three sets of tests were conducted was randomised and counter-balanced to avoid assessment of the utility of the measures being influenced by participant fatigue.

Assessment of self-selected walking speed, cadence and spatiotemporal gait parameters used a GAITRite 3.8 with a 5.18-metre long walkway. Each participant completed four passes of the mat at each test, producing a mean of 24 steps per test (SD 4.3; range 16 - 31). They commenced walking 2 metres before the GAITRite mat and were instructed to walk "straight down the middle of the mat" until 2 metres beyond the end of the mat. They stopped and turned slowly before returning down the mat. Participants were asked to walk at a comfortable but purposeful pace, as though they were trying to get somewhere but without rushing. Mean values for each test were downloaded for speed, cadence and proportion of gait cycle in double support. Values for each individual step or stride were downloaded and used to calculate central tendency and variability for step length, step width and stride time. Variability of step length, step width and stride time were expressed as interquartile range (IQR) for non-normally distributed datasets and coefficient of variation (CV) and standard deviation (SD) for normally distributed datasets. CV allows an estimate of variability that does not alter as mean values alter. SD might change as mean values change but has been recommended for describing variability, to enable easy comparison across gait analysis studies (Lord, Howe, Greenland, Simpson and Rochester, 2011).

Trunk and pelvic stability were assessed using Opal sensors, provided by APDM Wearable Technologies. These were Inertial Measurement Units (IMUs) attached over the sacrum and sternal angle using straps that fitted over clothing. Further sensors were attached at the ankles, in order to detect the timing of each step. Participants walked along a 100-foot corridor. They were asked to walk at a comfortable speed, maximising the stability and smoothness of their walking. The two most severely impaired participants (Frank and Oliver) walked over a smaller distance on one of each of their testing days, due to fatigue. The data for up to 30 seconds of steady state walking was extracted and analysed as described in the Psarakis et al. (2018) paper. A mean of 49 steps was included within the samples (SD 11; range 18-65). The following variables for both trunk and pelvic motion were hypothesized might be impacted upon by the orthotic shorts, therefore, are presented in this thesis: mediolateral 95% sway range, sway variability and range of rotational movement.

Maximal gait speed was assessed with the T25FW. A 25-foot distance was marked on the floor with tape. Participants were asked to walk as fast as they could, whilst remaining safe. They started from a standing start, one step behind the first tape. A stopwatch measured time taken between crossing the first tape and crossing the final tape. The mean of two trials was taken at each test.

Dual task cost was assessed using the T25FW. After assessing maximal walking speed, the participants repeated the T25FW twice more, whilst reciting alternate letters of the alphabet, as described by Sandroff et al. (2015). The number of letters correctly given was recorded on a digital audio device and deleted immediately after letters had been counted and recorded. Mean walking speed and the mean letters correctly provided per second were calculated for the two trials. Participants were asked to divide their attention equally between the cognitive task and the walking task and it was stressed that the number of letters correctly given was being recorded (Wajda et al., 2015). The dual task cost of undertaking a cognitive task whilst walking was calculated as the percentage change in the T25FW (walking speed as a single task minus walking speed as dual task divided by walking speed for single task multiplied by 100).

Self-report measures of walking and balance

A Global Rating of Change (GRC) scale (Figure 4.3 in Section 4.2.4) was used immediately after the objective assessment of walking ability in the shorts, to determine participants' estimates of whether the shorts had changed their walking ability. Self-report measures of balance confidence and perceived walking ability were assessed at baseline, following each home wear trial and at Visit 7.

Balance confidence was assessed with the ABC-UK (Parry, Steen, Galloway, Kenny & Bond, 2001; Appendix 8). Perceived walking ability was assessed using the MSWS-12 (Kieseier & Pozzilli, 2012; Appendix 8). The ABC-UK was totalled by taking a mean of the percentage scores given for each item. The raw MSWS-12 score was converted to a percentage using the equation below:

$$MSWS12\ percentage\ score = \left(\frac{Raw\ MSWS12\ score - 12}{48}\right) X\ 100$$

The ABC-UK prompted a number of questions from the participants as to the context in which they were judging their confidence. For example, they asked whether they would have a walking stick, would have someone with them or would be "allowed" to touch objects in the environment. Where these issues arose, the researcher noted the context described so that the same context would be presented on the next usage of the measure. The GRC, MSWS-12 and ABC-UK were all provided as paper questionnaires, which the researcher completed with the participants.

The participants kept a falls diary during each two-week home trial (Appendix 13). The diary format and the list of possible causes of falls offered to the participants were those suggested by Coote et al. (2014) but to keep the diary as simple as possible, questions around time of day and context of falls were not included.

5.3.8 Quantitative data analysis

Level of measurement

All objective measures of pressure, walking ability, falls incidence and wear times were considered ratio level data. Other self-report measures were considered ordinal (MSWS-12, ABC-UK) or categorical (GRC, to keep or not keep the shorts).

Normality tests and results

Tests of normal distribution used the Shapiro-Wilk test, as this is recommended for sample sizes below 50 (Ghasemi and Zahediasl, 2012).

Ordinal, interval and ratio data were tested for normal distribution. Interval and ratio data were tested for normal distribution to determine whether parametric or non-parametric statistical methods were appropriate. Ordinal data was tested to determine whether datasets should be described using mean and SD or median and IQR.

Datasets were normally distributed with the exception of pressure beneath the shorts, wear times, falls incidence, 40% of the individual datasets for stride time variability and 30% of the datasets for range of trunk and pelvic rotation in the transverse plane (yaw). Therefore, wear times, falls incidence, stride time variability and trunk and pelvic yaw were analysed using non-parametric methods. Other interval and ratio level measures were analysed with parametric statistical methods. ABC-UK and MSWS-12 data were normally distributed, therefore, described using mean and SD.

Analysis of baseline stability

Baseline stability was determined by comparing descriptive data for all measures for those participants who were assessed on every visit. For objective measures of walking ability, the "no shorts" tests on Visits 3, 5 and 7 were compared. For the MSWS-12 and ABC-UK, the data from Visits 1 and Visit 7 were compared.

Calculation of effect sizes

Effect sizes were used to estimate the magnitude of the effect of the shorts on each variable. For all objective measures, effect sizes and mean differences were calculated for (1) within-day comparison of "no shorts" versus "with shorts" for both orthotic and placebo shorts and (2) between-day comparison of walking ability in orthotic versus placebo shorts. For MSWS-12 and ABC-UK data, effect sizes were calculated comparing baseline to after each of the home trials.

For interval and ratio, normally distributed data, a Cohen's d effect size was calculated by subtracting the mean for one shorts condition from the mean for the other shorts condition and dividing by the pooled standard deviation for both shorts conditions. Pooled standard deviations were obtained using the equation provided by Coe (2002). For ordinal and non-normally distributed interval and ratio data, a non-parametric effect size was calculated using the formula suggested by Pallant (2007), in which the Z value from the Wilcoxon Signed Rank test was divided by the square root of the number of observations across both conditions. The terminology suggested by Cohen was adopted for both types of effect size. Values around 0.2 were considered to indicate a small effect, around 0.5 a moderate effect and 0.8 and above a large effect (Cohen, 1988).

Inferential analyses

No inferential analyses for effectiveness were performed, as these would be misleading in a feasibility study that is not powered for such analyses. Correlation between variables was investigated where this seemed helpful in improving understanding of the potential impact of the shorts. For example, relationships were investigated between different variables at baseline, between baseline and changes seen in a variable, between the changes seen on different variables and between the initial participant perceived effect of the shorts (GRC) and whether the shorts were kept at the end of the study. Where this was done, the Fisher's Exact test was used for categorical data, the Spearman Rank for non-parametric analyses and Pearson's for parametric analyses.

5.3.9 Qualitative data collection

Qualitative data collection used individual semi-structured interviews. Participants were interviewed twice during the study: once at Visit 1 and once at Visit 7, approximately 12 weeks later (see Appendix 18 for topic guides).

The first interview explored participants' motivations for joining the study, their current daily activities, their expectations of and anticipated acceptability of the shorts. In addition, participants were asked about other potential solutions they had already explored for their movement problems. The initial interviews lasted an average of 23 minutes (range 10 to 38 minutes).

The final interview aimed to determine the experienced acceptability of the shorts and the participants' views on the study processes. Participants were asked about the perceived advantages and disadvantages of the shorts, the differences between the two pairs of shorts and whether they would use them again in the future. Prompts were informed by the potential advantages and disadvantages of fabric orthoses determined from previous research and the FabO IPA study. The topic guide for the final interview was modified for each participant prior to the interview (see Appendix 18). Questions were added that related directly to the hopes and expectations reported at the initial interview, the content of the wear diaries kept by each participant during their home trials of the shorts and the participants' rating of the initial impact of the shorts on their walking on the day that they had been first issued with the shorts. During preparation for the final interview, the chief investigator/interviewer was unblinded to which pair of shorts was worn first or second. Final interviews lasted an average of 44 minutes (range 29 to 75 minutes).

Three participants were accompanied to the interviews by their husbands, who had provided transport to the study site. The husbands were not formally consented as participants. I clarified that the interview aimed to determine the views of the person with MS and that the interview was being recorded. However, husbands were not actively discouraged from speaking and their comments were transcribed and analysed.

All interviews were audio recorded and transcribed verbatim by the principal researcher. Transcripts noted emphasis, pauses, gestures, laughter and tone of voice.

5.3.10 Qualitative data analysis

Framework Analysis, as described by Ritchie and Spencer (Ritchie et al., 2013; Gale et al., 2013) was used to analyse the data because of the deductive nature of the analysis required. The aim was to determine answers to clearly defined questions around acceptability of the shorts and of the study design; Framework Analysis works well for such a deductive approach (Pope, Ziebland & Mays, 2000). Whereas some qualitative approaches, including IPA, involve interpretation of meaning from the outset of analysis, Framework Analysis typically separates the tasks of managing data and interpreting meaning. The initial stages of analysis are primarily about managing and reducing the size of the dataset whereas the later stages involve interpreting meaning (NatCen Learning, 2012).

Ritchie and Spencer (Pope et al., 2000; Ritchie et al., 2013) described five stages of analysis in the framework approach. These are: (1) familiarisation, (2) identifying a thematic framework, (3) indexing, in which the thematic framework is applied to the data using numerical codes, (4) charting, in which data is arranged into a table where each column is a theme or subtheme and each row is a participant. Each cell attempts to capture a summary of individual participant's views within that theme. The final stage is (5) mapping and interpretation, which involves explaining the themes arising.

In the OSFeaMS study, familiarisation included transcription, which was followed by reading and re-reading the transcripts and producing a list of emergent themes. The thematic framework identified included both inductive and deductive elements. From a deductive perspective, the thematic framework used the Theoretical Framework of Acceptability (Sekhon, Cartwright & Francis, 2018), the themes arising from the FabO IPA study (Section 3.5.3) and aspects of the research process of relevance to future studies. The Theoretical Framework of Acceptability has been introduced in Section 3.6.2 and is summarised in Table 3.3. Sekhon et al. (2018) designed this framework to explicate the constructs suggested to contribute towards an individual’s overall acceptability of a healthcare intervention. No previous research appeared to have used this framework for qualitative data analysis. However, as explained in Section 3.6.2, the framework was closely related to the themes obtained in the FabO IPA study, so was judged likely to work well for analysing acceptability in the OSFeaMS study.

Inductive elements of the thematic framework came from the initial list of emergent ideas created at the familiarisation stage, which could all be located logically within the deductive framework. The thematic framework was applied to the data using indexing and charting on an Excel spreadsheet (Swallow et al., 2003). The framework progressed through a number of iterations, as codes were renamed and refined, removing ideas that were either not clearly supported by the data or were less relevant to the research objectives. This process was supervised and facilitated by an experienced qualitative researcher in the supervision team, who also verified the final thematic framework by reviewing the coding of one of the interview transcripts. The final thematic framework is shown in Appendix 19. An example of an indexed transcript is provided in Appendix 20.

Two elements of the Theoretical Framework of Acceptability were omitted from the final thematic framework. These were the constructs of "opportunity costs" and "self-efficacy". Participants did not appear to lose opportunities through using orthotic shorts and aspects relevant to "self-efficacy" fitted logically within the "burden" construct. The final framework included three subthemes in the "perceived effectiveness" construct derived from the FabO IPA study that effectively communicated the meaning of perceived effectiveness within the OSFeaMS data. These were "reclaiming my body", "reclaiming autonomy" and "managing self-image".

The final stage of analysis included noting the frequency with which different opinions were expressed and interpreting meaning behind the participants' words. Although frequency counts are controversial within qualitative data analysis (Pope et al., 2000), this felt to be a useful method to validate the themes.

Member checking or participant validation is a technique used to check the credibility of findings in qualitative research (Doyle, 2007; Lincoln & Guba, 1985). Member checking was not used because of concerns about the burden on the participants with a study that already involved three months of involvement and seven contacts with the researcher. Instead, credibility was optimised by in-depth interviews, prolonged engagement with the transcripts, careful attention to the extent to which the data supported each theme and verification of the coding by an experienced supervisor.

5.3.11 Reflexivity

Reflexivity was ensured by ongoing reflection before, during and after each interview, which was facilitated by transcribing the interviews myself. In addition, I kept a reflective diary where I recorded my assumptions and feelings before and after each interview and during the data analysis process.

5.3.12 Mixed methods - integration

Within the OSFeaMS study, integration occurred at the stage of interpretation. In other words, the qualitative and quantitative data were analysed separately as described above and key findings were identified from the qualitative and quantitative datasets separately. After this, integration of the key findings took place to enable insights from the qualitative to inform the quantitative and vice versa.

It is important to use a systematic approach to integration to give the process credibility and transparency (O'Cathain, Murphy & Nicholl, 2010). O'Cathain et al. (2010) suggested three approaches to integration: use of a triangulation protocol at the interpretation stage, "following a thread" or comparing findings case by case at the analysis stage. A triangulation protocol was used in the OSFeaMS study because this is designed for use at the interpretation stage and was felt to be a clear and logical approach for a novice mixed methods researcher. The triangulation protocol involved creating a convergence coding matrix to enable comparison of the qualitative and quantitative findings for different elements of the study (Farmer, Robinson, Elliott & Eyles, 2006; O'Cathain et al., 2010). In order to create the convergence coding matrix, research questions were worded in a manner that allowed both qualitative and quantitative perspectives to be relevant. Key qualitative findings on each of these questions were inserted into the matrix first, followed by quantitative findings related to each of the qualitative findings. All of the important quantitative findings could be added to the matrix in this way, that is to say, they all felt to be relevant to qualitative findings.

The next stage of integration involved classifying qualitative and quantitative findings according to whether they were: convergent, meaning that they agreed; divergent, meaning that they disagreed or "silent", meaning that one or other dataset had nothing to say about an important finding. Divergent findings were classified as either complementary, meaning that they highlighted slightly different aspects, or dissonant, meaning that they disagreed, in a manner that seemed incongruous and required re-evaluation of the data, one's initial assumptions or both (Perlesz & Lindsay, 2003).

5.4 Conclusion

This chapter has reported the methodological decisions and methods used in the mixed methods OSFeaMS study. The results of this study are presented and discussed in Chapters 6 and 7, with integration between the qualitative and quantitative data presented and discussed in Chapter 8.

Chapter 6: The Orthotic Shorts Feasibility in MS study - quantitative results and discussion

Summary

This chapter explains the quantitative findings of the Orthotic Shorts Feasibility in MS (OSFeaMS) study. The methodology and methods have been reported in Chapter 5. This current chapter reports the quantitative results and provides a preliminary discussion of the findings. Qualitative methods and results will be reported in Chapter 7 and qualitative and quantitative findings will be integrated in Chapter 8.

6.1 Aims and Objectives

The quantitative aspects of the OSFeaMS study aimed to determine adherence to wear recommendations, intention for future use and the efficacy of the shorts for improving objective measures of walking ability and self-report measures of walking and balance. In addition, the feasibility of a potential future trial was investigated by evaluating recruitment and retention rates and determining the suitability of the research design and measurement techniques used.

Objectives for the quantitative aspects of the OSFeaMS study were:

1. To provide specific information to inform a future randomised controlled trial:
    a. Determine rates of recruitment and retention
    b. Determine the suitability of measurement techniques for assessing the impact of orthotic shorts
2. To gain a quantitative estimate of acceptability of orthotic shorts:
    a. Determine how long shorts are worn as a proportion of time recommended
    b. Determine the proportion of people who opt to keep the shorts after the study
3. To determine the immediate, orthotic effect of shorts on the following objective measures of walking ability:
    a. Spatiotemporal gait parameters and gait variability

    b. Self-selected gait speed and maximal gait speed
    c. Dual task cost of a cognitive task whilst walking
    d. Trunk and pelvic stability in walking
4. To determine the impact of wearing orthotic shorts on:
    a. Participant perceived effect on walking in the movement laboratory
    b. Self-reported balance confidence
    c. Self-reported perception of walking ability.

6.2 Results

6.2.1 Recruitment and retention

Figure 6.1 details the recruitment to the study. Eighteen people were personally invited to join the study, 17 had been identified by the NHS physiotherapy service and one by the MS Nurses service. Fifteen people responded to the various advertising materials: flyers posted at local clinics, an email invitation sent via the MS Therapy Centre and the online proposal. Of these 33 people, two made contact after recruitment was complete and seven either could not be contacted or did not wish to participate. Reasons provided for not participating were that the time or travel involved in the study would be too burdensome.

On screening for eligibility, seven people were not eligible. Reasons for ineligibility were starting a new drug therapy (*n* = 2), did not identify with weakness around the trunk or hips (*n* = 2), varicose veins (*n* = 1), walking ability was too restricted (*n* = 1) and a gynaecological condition that made tight clothing inadvisable (*n* = 1).

Seventeen people attended the initial assessment and all consented. However, one participant was not provided with orthotic shorts because it was felt that she would not cope well with shorts. She had severe loss of dorsiflexion and poor knee control and used compensatory hip and pelvic movement in order to step through and stabilise her more affected knee. It was felt that her hip and pelvic movement might be reduced by the shorts, thus limiting her ability to use her compensatory strategies and worsening her independence and function. She was withdrawn from the study.

The final participant group included four males and 12 females; 10 with relapsing-remitting MS, four with secondary progressive and two with primary progressive; ages ranged from 34 to 57 (mean 48 ± 7). Time since diagnosis ranged from three to 29 years (mean 13 ± 9). Participant characteristics, with pseudonyms are presented in Table 6.1.

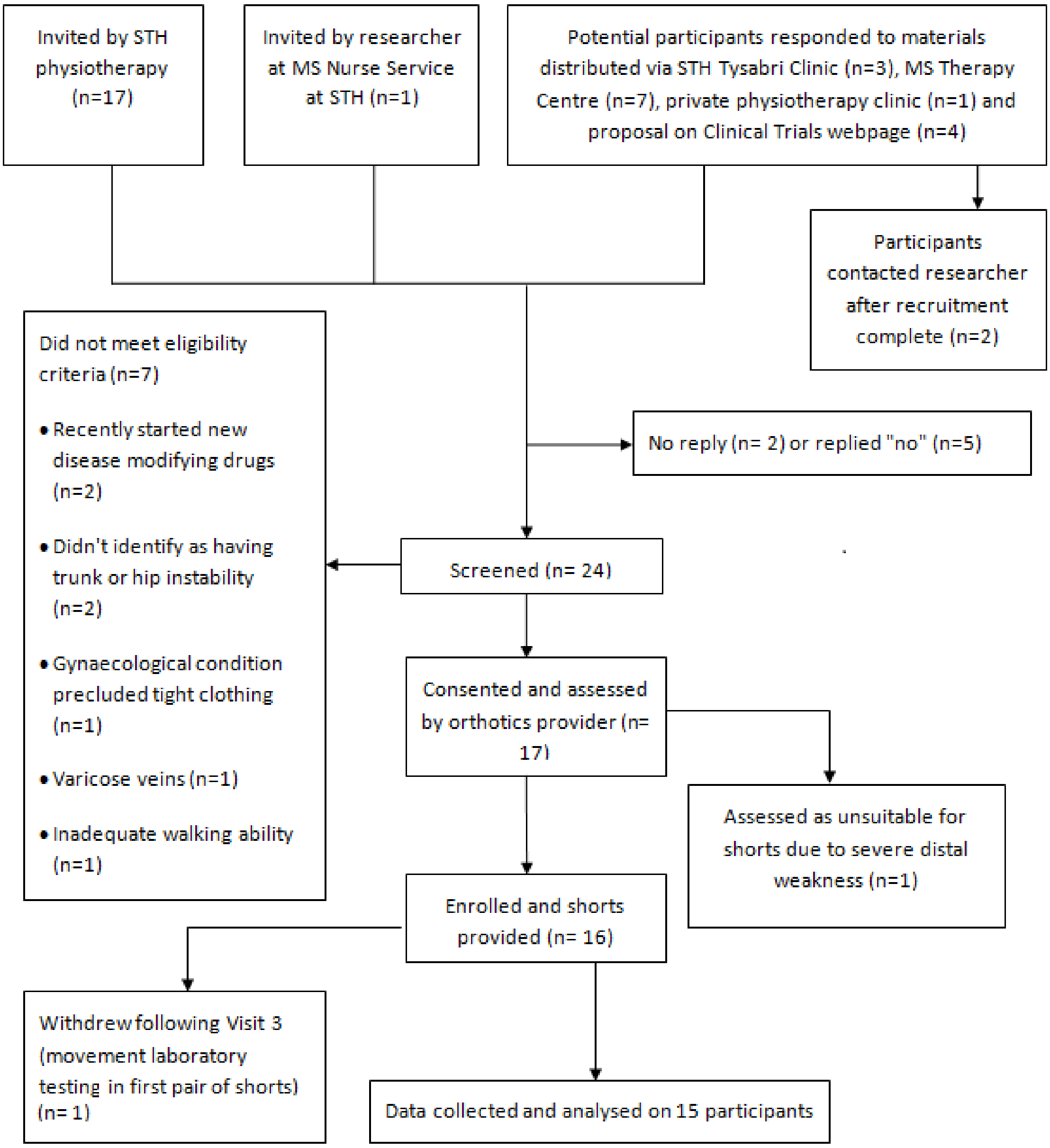


Figure 6.1: Flow chart showing study recruitment and retention

Table 6.1: OSFeaMS participants and shorts provided

Participant pseudonym	Age	Disease type	Time since diagnosis (years)	Walking aids used	Recruited through	Height of shorts	Shorts design	Colour	Side zip provided	Toileting hole included	Pressure orthotic shorts (mmHg)	Pressure placebo shorts (mmHg)
Alison	36	RRMS	18	Two sticks	STH physio	High abdomen	Standard	Beige	On right	No	13	5
Ben	45	RRMS	10	Walking stick	STH physio	High abdomen	Standard	Black	On right	No	13	5
Caroline	42	RRMS	20	Elbow crutch	STH physio	High abdomen	Abd assist	Black	On left	Yes	12	5
Dawn	50	PPMS	4	Walking stick	STH physio	High abdomen	Standard	Lilac	Both sides	No	8	3
Erica	50	RRMS	12	Occasional stick	STH physio	Waist	Standard	Beige	On right	Yes	8	4
Frank	46	RRMS	6	Walking stick	Internet	Waist	Standard	Black	On left	Yes	10	5
Gwen	50	RRMS	29	Occasional stick	STH physio	Waist	Standard	Beige	No	No	11	5
Helen	57	SPMS	20	Occasional stick	STH physio	Waist	Standard	Lilac	On left	Yes	8	3
Ingrid	56	RRMS	5	Occasional rollator	MSTC	High abdomen	Standard	Lilac	No	No	8	2
FSP10	65	SPMS	23	Walking stick	MSTC	No shorts provided due to poor distal function						
Jon	51	PPMS	3	Occasional pole	MSTC	Waist	Standard	Red	No	No	8	4
Kathy	52	RRMS	4	Occasional rollator	MSTC	High abdomen	Standard	Black	On right	No	6	3
Linda	44	SPMS	15	Unable to use	MSTC	Waist high	Standard	Beige	On left	Yes	10	6
Melissa	34	SPMS	8	Elbow crutch	Internet	Waist	Standard	Beige	On left	No	6	3
Natalie	52	RRMS	5	None required	MS Nurse	Waist	Standard	Lilac	No	Yes	9	4
Oliver	51	RRMS	24	Walking stick	Tysabri Clinic	High abdomen	Abd assist	Dark blue	On left	No	10	5
Paula	57	SPMS	26	Walking stick	Tysabri Clinic	Waist	Standard	Beige	On left	No	9	5

(RRMS = relapsing remitting, PPMS = primary progressive, SPMS = secondary progressive, STH = Sheffield Teaching Hospitals NHS Foundation Trust, MSTC = MS Therapy Centre, internet = participants responded to online proposal on clinical trials.gov; Abd assist = shorts designed to increase hip abduction)

Of the 16 participants who were provided with shorts, 15 remained through to the final appointment; one dropped out after her third appointment. She stated that she did not wish to continue because the shorts showed beneath her clothing. She would not wear them and, therefore, did not wish to continue with the study.

6.2.2 Description of shorts provided - pressure applied, colour and design

Table 6.1 shows the shorts provided and the pressure applied by each pair of shorts. There was a difference in the pressure applied beneath the two pairs of shorts with a median of 9 mmHg (range 6 - 13) under the orthotic shorts and a median of 4.5 mmHg beneath the placebo shorts (range 2 - 6). However, there was crossover in that two participants were provided with orthotic shorts providing pressure of 6 mmHg whereas other participants had placebo shorts providing similar compression.

Male participants all chose black or bold primary colours for their shorts, whereas females were more likely to choose beige (six) or pastel (four) colours. The toileting hole was chosen by one male and four female participants. Participants' choices around whether to have a zip or toileting hole were informed by their stated hand function and urgency of bladder function. Four chose not to have a side zip.

6.2.3 Quantitative assessment of acceptability

This section describes wear times and participants' choices as to whether they kept the shorts at the end of the study. A more complete account of acceptability, informed by qualitative methods, is provided in Chapter 7.

Adherence to recommended wear times was good. The orthotic shorts were worn for 86% of the recommended time and the placebo shorts for 88% of the recommended time. Wear times were higher for participants recruited later in the study. Verbal instructions had been adjusted following the first five participants, who believed they had been advised to wear the shorts only when they were active. Erica and Gwen had very low wear times. Gwen experienced an adverse reaction; she developed ankle swelling and was advised to cease wearing the shorts. Erica disliked the shorts and chose not to take them away on holiday.

Twelve participants (75%) chose to keep the orthotic shorts following the study and eight of these kept the placebo shorts as well. Four participants (25%), including the participant who withdrew, chose to keep neither pair. More of the people recruited towards the end of the study chose to keep both pairs of shorts. All participants recruited from outside the NHS chose to keep at least the orthotic shorts, if not both pairs. In contrast, only four of the seven people recruited by the NHS physiotherapist chose to keep their shorts.

6.2.4 Objective assessment of walking ability

Data completeness

A complete dataset was obtained except for the following: data for one participant were excluded from the orthotic shorts testing day due to the impact of fatigue induced by hot weather, one participant was excluded from the dual task cost assessment due to difficulty following the instructions, GAITRite could not be used for the final visit for one participant due to equipment failure and one IMU foot sensor failed on three different trials. The missing foot sensor data did not influence calculation of mediolateral sway or rotational ranges but could have altered accuracy of sway variability, as this required accurate recording of stepping events. Trials with missing foot sensors were not analysed for sway variability. Because the IMU data were analysed externally and data on other measures had demonstrated moderate stability across the study, IMU data for Visit 7 were not analysed. Objective data with and without orthotic shorts, but not placebo shorts, were available for the participant who withdrew.

Walking ability at baseline

The participant group demonstrated a wide range of different abilities, with a mean self-selected gait speed of 1.1 ± 0.37 m/s (range 0.56 m/s to 1.99 m/s). Means for self-selected gait speed, step length (58.4 ± 11 cm), step width (11.6 ± 3.7 cm) and stride time (1.09 ± 0.2s) indicated that the group had a quicker gait, longer step lengths, narrower step width and shorter stride times than previously reported for PwMS. Previously reported means were 0.96 m/s, 50 cm, 13.4 cm and 1.26s respectively (Comber, Galvin & Coote, 2017). Stride time variability had a median CV of 3.5%, which was slightly less than the 4.18% reported by Moon, Sung, An, Hernandez and Sosnoff (2016). Variability of step length and step width was greater than that reported previously (Moon et al., 2016). In the present study, step length variability had a mean CV of 5.8 ± 2.2%, and step width variability a mean CV of 33 ± 19%, compared to previous values of 4.29% and 20.4% respectively (Moon et al., 2016). Six participants required walking aids during the objective assessments whereas nine were assessed without aids.

Baseline stability

Table 6.2 shows the central tendency and variability of measures obtained without the shorts using GAITRite, T25FW and the dual task cost assessment at Visits 3, 5 and 7. Changes over time are noted with a small increase in self-selected gait speed across the three visits, along with a small increase in step length and a small decrease in stride time. Stride time variability and step width variability decrease across the study. Maximal gait speed is slightly increased over the course of the study. There is a notable improvement in performance on the dual task cost assessment during the course of the study, both in a decreased impact of cognition on walking speed and a slightly improved accuracy in the cognitive task.

Table 6.3 shows the central tendency and variability of baseline measures obtained for trunk and pelvic movement using IMUs without the shorts at Visits 3 and 5. Overall, these values appear stable over time.

Table 6.2: Baseline/without shorts values for objective measures at Visits 3, 5 and 7 - GAITRite data, T25FW and dual task cost

Outcome measure	Participants included in data	No shorts conditions at Visit 3	No shorts conditions at Visit 5	No shorts conditions at Visit 7
Gait speed at self-selected walking speed (metres/second)	All participants except Frank (data for Visit 5 not used), Oliver (data unavailable for Visit 7) and Paula (data only available for Visit 3).	1.13 ± 0.37	1.17 ± 0.31	1.2 ± 0.34
Cadence at self-selected walking speed (steps/minute)		114 ± 21	114 ± 16	117 ± 17
Step length (centimetres)		58.4 ± 11	60.7 ± 10	60.8 ± 11
Step width (centimetres)		11.6 ± 3.7	11.7 ± 3.6	12.0 ± 3.0
Stride time (seconds)		1.09 ± 0.2	1.07 ± 0.2	1.04 ± 0.2
% of gait cycle in double support		29 ± 3.7	27 ±3.4	27 ± 3.5
Step length variability SD (centimetres)		3.3 ± 1.0	3.0 ± 1.1	3.8 ± 1.7
Step width variability SD (centimetres)		3.4 ± 1.5	3.2 ± 1.9	3.3 ± 1.8
Stride time variability SD (milliseconds)		Median (IQR) 36 (23)	Median (IQR) 29 (18)	Median (IQR) 23 (28)
Step length variability CV (% of mean)		5.8 ±2.2	5.2 ± 2.2	6.4 ± 3.3
Step width variability CV (% of mean)		33 ± 19	29 ± 14	28 ± 14
Stride time variability CV (% of mean)		Median (IQR) 3.5 (1.6)	Median (IQR) 3.1 (1.4)	Median (IQR) 2.3 (2.1)
Maximal gait speed (T25FW)	All participants except Frank and Paula	1.26 ± 0.31	1.27 ± 0.31	1.30 ± 0.32
Dual task cost (% change to walking speed)	All participants except Ben (unable to follow DTC instructions), Frank (data for Visit 5 not used) and Paula.	18 ± 8	11 ± 8	8 ± 9
Letters correctly provided per second during dual task cost assessment		0.68 ± 0.28	0.76 ± 0.30	0.79 ± 0.30

(Baseline values are expressed as mean ± standard deviation unless otherwise stated; IQR = interquartile range; T25FW = Timed 25-foot walk; SD = standard deviation; CV = coefficient of variation, the standard deviation divided by the mean for each dataset multiplied by 100.)

Table 6.3: Baseline values for objective measures at Visits 3 and 5 obtained from the Inertial Measurement Units

Outcome measure	Participants included in data	No shorts conditions at Visit 3	No shorts conditions at Visit 5
ML pelvic sway (centimetres)	All participants except Frank (data for Visit 5 not used) and Paula (data only available for Visit 3) (n = 14)	5.5 ± 1.8	5.4 ± 1.4
ML trunk sway (centimetres)		5.2 ± 1.6	5.3 ± 1.2
Pelvic rotation in roll (degrees)		11.1 ± 2.7	11.2 ± 3.1
Trunk rotation in roll (degrees)		14.6 ± 7.5	12.6 ± 5.2
Pelvic rotation in pitch (degrees)		10.1 ± 3.1	10.7 ± 2.6
Trunk rotation in pitch (degrees)		10.1 ± 2.2	10.6 ± 2.2
Pelvic rotation in yaw (degrees)		Median (IQR) 15.0 (6.1)	Median (IQR) 16.0 (3.9)
Trunk rotation in yaw (degrees)		Median (IQR) 14.8 (5.4)	Median (IQR) 16.0 (5.0)
Sway variability of pelvis (SD, centimetres)	All participants except Alison and Helen (missing foot sensors), Frank (data for Visit 5 not used) and Paula (data only available for Visit 3) (n = 12)	0.60 ± 0.21	0.55 ± 0.12
Sway variability of trunk (SD, centimetres)		0.64 ± 0.26	0.60 ± 0.21

(Baseline values are expressed as mean ± standard deviation unless otherwise stated; ML = mediolateral; IQR = interquartile range; SD = standard deviation; roll, pitch and yaw are all 95% range; roll is rotation in the frontal plane, which approximates to lateral pelvic tilt and trunk side flexion; pitch is rotation in the sagittal plane, which approximates to anterior-posterior pelvic tilt or trunk flexion/extension; yaw is rotation in the transverse plane which approximates to left/right rotation.)

The impact of orthotic shorts on self-selected gait speed and spatio-temporal gait parameters

There was a small effect size (ES) for the increase in self-selected gait speed with the orthotic shorts (ES = 0.27), with a correspondingly small increase in step length (ES = 0.27) and cadence (ES = 0.21) and a small decrease in stride time (ES = -0.22) (see Table 6.4). Close relationships were found between the change in self-selected gait speed in the orthotic shorts and changes in cadence (r = 0.824, p = 0.002), step length (r = 0.845, p = 0.001) and stride time (r = -0.823, p = 0.002). There were very small or negligible improvements in these variables in the placebo shorts. Self-selected gait speed improved or remained similar in the orthotic shorts for most participants. Only Alison and Caroline were slower in the orthotic shorts. Figure 6.2 shows individual changes in walking speed in the different conditions and compares these to values obtained for “healthy” and MS in other studies, using data obtained from Comber et al. (2017). Differences between the orthotic and placebo shorts for gait speed (ES = 0.18), step length (ES = 0.04), stride time (ES = -0.07) and cadence (ES = 0.27) were small or negligible (Table 6.4).

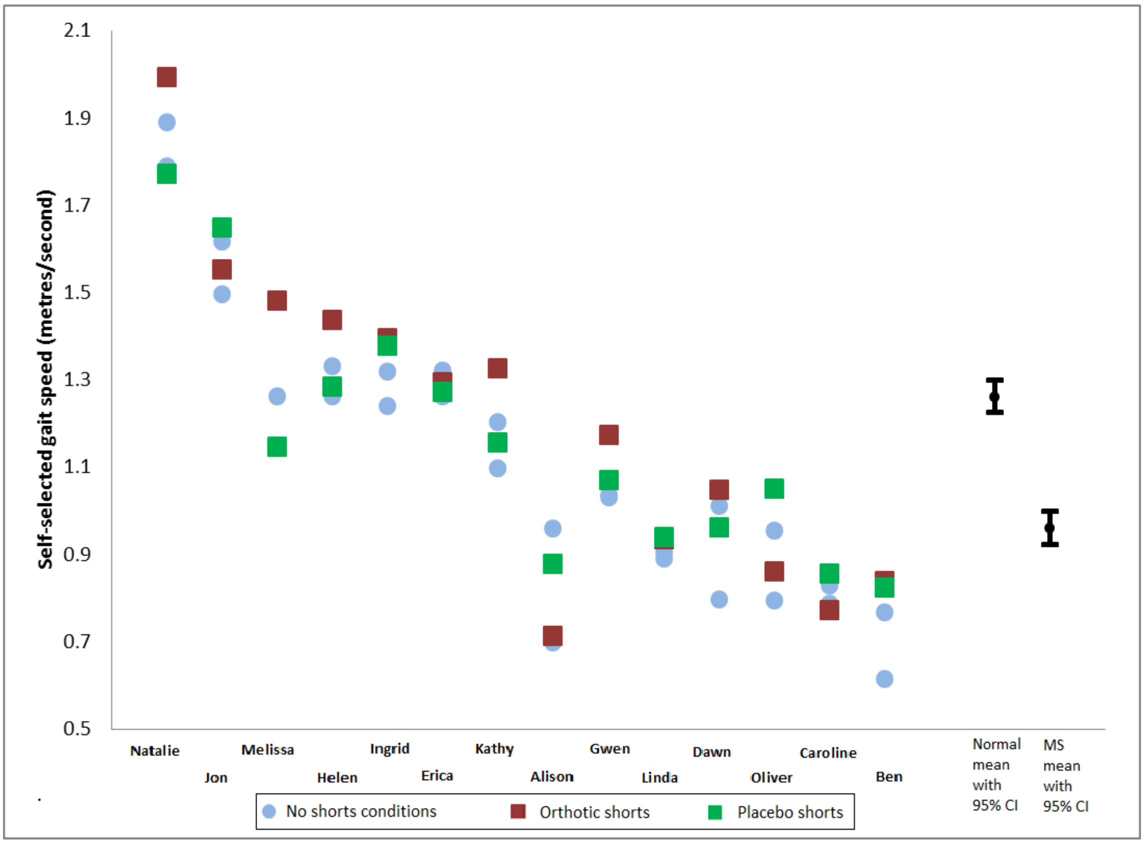


Figure 6.2 Impact of shorts on self-selected gait speed (*Means for normal, non-neurologically impaired people and other people with MS are provided for comparison with 95% confidence intervals (95% CI) for means, using data obtained from Comber et al., 2017*).

There was a moderate decrease in step width (ES = -0.5) with the orthotic shorts but not with the placebo shorts (ES = 0.03). With the orthotic shorts, the group mean for step width narrowed by 1.5 cm. The 95% confidence interval for the mean difference suggested the true mean difference could be as large as 2.6 cm. The difference in step width seen between the two pairs of shorts was small (ES = -0.20), partly because step width was greater at baseline on the day the orthotic shorts were tested. In contrast with the findings for step length and stride time reported in the previous section, there was no relationship between changes in gait speed and changes in step width (r = -0.083, p = 0.808).

Step width did not narrow in the orthotic shorts for all participants. Figure 6.3 shows the different baseline values for each participant, along with the step width in each pair of shorts and comparison values for mean step width in both healthy and MS groups (obtained from Comber et al., 2017). The orthotic shorts appeared to increase step width in people with a narrow base and decrease step width in those with a wider base. Oliver and Caroline had been supplied with shorts specifically designed to increase their step width (“abduction assist”) but step width also widened in Helen and Erica, who had been supplied with “standard” orthotic shorts.

The larger changes in step width were seen in those with the wider base, such as Melissa, Ingrid, Dawn and Ben; however, Alison also had a wide base and did not change with the shorts. There was a strong correlation between mean step width without shorts and the change in step width with the orthotic shorts (*r* = -0.737, *p* = 0.002).

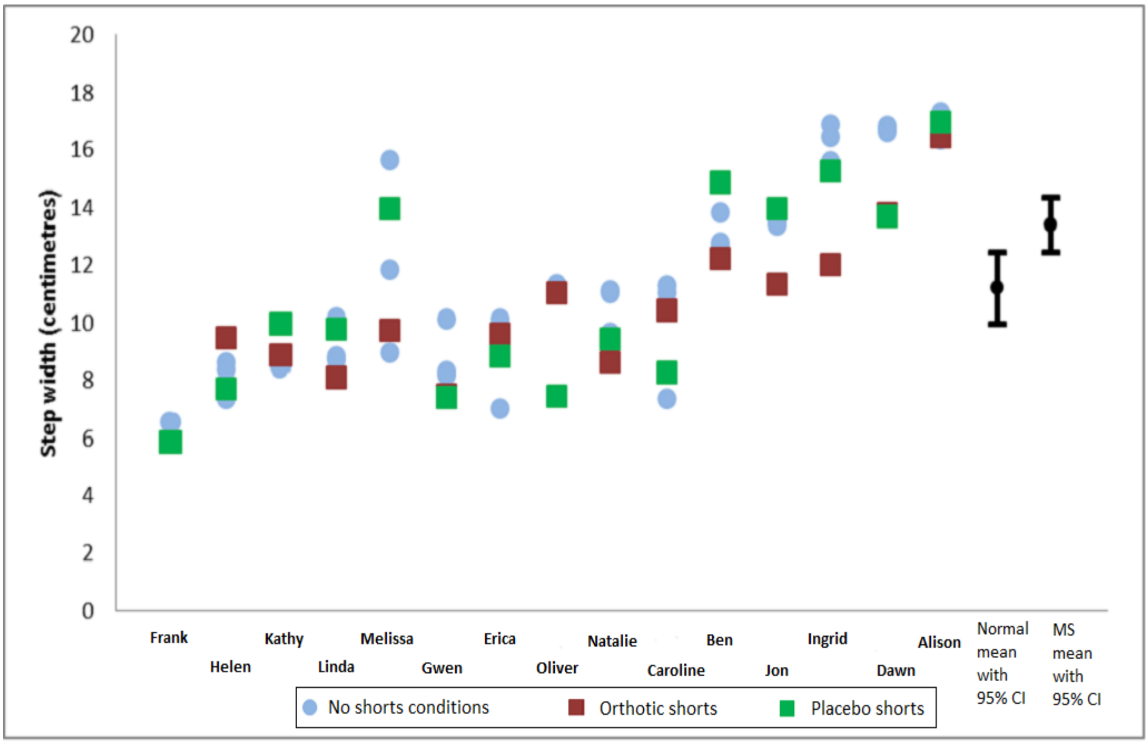


Figure 6.3: Impact of orthotic and placebo shorts on step width (*Means for normal, non-neurologically impaired people and people with MS are provided for comparison, with 95% confidence intervals (95% CI) for means, using data obtained from Comber et al., 2017*).

The percentage of gait cycle in double support did not change in either pair of shorts (Table 6.4).

Table 6.4: Impact of shorts on self-selected gait speed, cadence, step length, step width, stride time and percentage of gait cycle in double support

	Orthotic shorts vs. no shorts					Placebo shorts vs. no shorts					Orthotic shorts vs. placebo shorts				
	n	Orthotic shorts	No shorts	Mean difference (95% CI)	Effect size	n	Placebo shorts	No shorts	Mean difference (95% CI)	Effect size	n	Orthotic shorts	Placebo shorts	Mean difference (95% CI)	Effect size
Self-selected gait speed (m/s)	15	1.2 ± 0.35	1.11 ± 0.33	+0.09* (+0.05 to +1.4)	0.27	15	1.12 ± 0.32	1.09 ± 0.33	+0.03 (-0.01 to +0.06)	0.09	14	1.2 ± 0.35	1.12 ± 0.32	+0.04 (-0.04 to +0.13)	0.13
Cadence (steps per minute)	15	116 ± 19	112 ± 20	+4* (+1.1 to +6.9)	0.21	15	111 ± 17	111 ± 17	+0.7 (-2.1 to +3.4)	0.04	14	116 ± 19	111 ± 17	+2.7 (-1.8 to +7.1)	0.27
Step length (centimetres)	15	61 ± 10	59 ± 10	+2.6* (+1.1 to +4.2)	0.27	15	59 ± 10	58 ± 11	+1.1 (-0.2 to +2.5)	0.11	14	61 ± 10	61 ± 9	+0.3 (-2.1 to +2.8)	0.04
Step width (centimetres)	15	10.7 ± 2.4	12.2 ± 3.4	-1.5* (-2.6 to -0.3)	-0.50	15	10.9 ± 3.5	10.8 ± 3.6	+0.1 (-0.7 to +0.9)	0.03	14	10.7 ± 2.4	11.2 ± 3.4	-0.6 (-1.9 to +0.7)	-0.20
Stride time (seconds)	15	1.06 ± 0.2	1.10 ± 0.2	-0.04* (-0.08 to 0.0)	-0.22	15	1.10 ± 0.2	1.11 ± 0.2	0 (-0.04 to +0.03)	-0.03	14	1.06 ± 0.2	1.07 ± 0.1	-0.01 (-0.05 to +0.03)	-0.07
% gait cycle in double support	15	28 ± 4	29 ± 4	-0.3 (-1.3 to +0.7)	-0.07	15	29 ± 4	29 ± 4	+0.4 (-0.4 to +1.2)	0.10	14	29 ± 4	29 ± 4	+0.1 (-1.1 to +1.3)	0.02

*(Values are means & standard deviations; between-shorts values differ from within-day values because between-day includes only individuals for whom data is available for both pairs of shorts; * indicates confidence interval does not go over zero.)*

The impact of orthotic shorts on variability of spatiotemporal gait parameters

As shown in Figures 6.4 to 6.6 and Table 6.5, variability of spatiotemporal gait parameters decreased in the orthotic and placebo shorts. Step width variability decreased moderately with the orthotic shorts (ES = -0.57; Figure 6.4), with a mean difference in SD of 0.8 cm. Orthotic and placebo shorts had a small impact on stride time variability (see Table 6.5 and Figure 6.5). In comparison, the placebo shorts had a negligible to small impact upon step width variability (ES = -0.12) but a small to moderate impact upon step length variability (ES = -0.32; Figure 6.6). Comparing the two pairs of shorts, stride time variability was similar in both shorts but step length variability was higher in orthotic than placebo (ES = 0.31) and step width variability was lower in orthotic compared to placebo (ES = -0.36). Comparing baseline values to the change seen with the shorts, there was no correlation for most variables, indicating that participants with different baseline abilities responded similarly. The exception was the impact of placebo shorts on step length variability, which was more marked in those with higher variability at baseline (*r* = -0.866, *p* >0.0005).

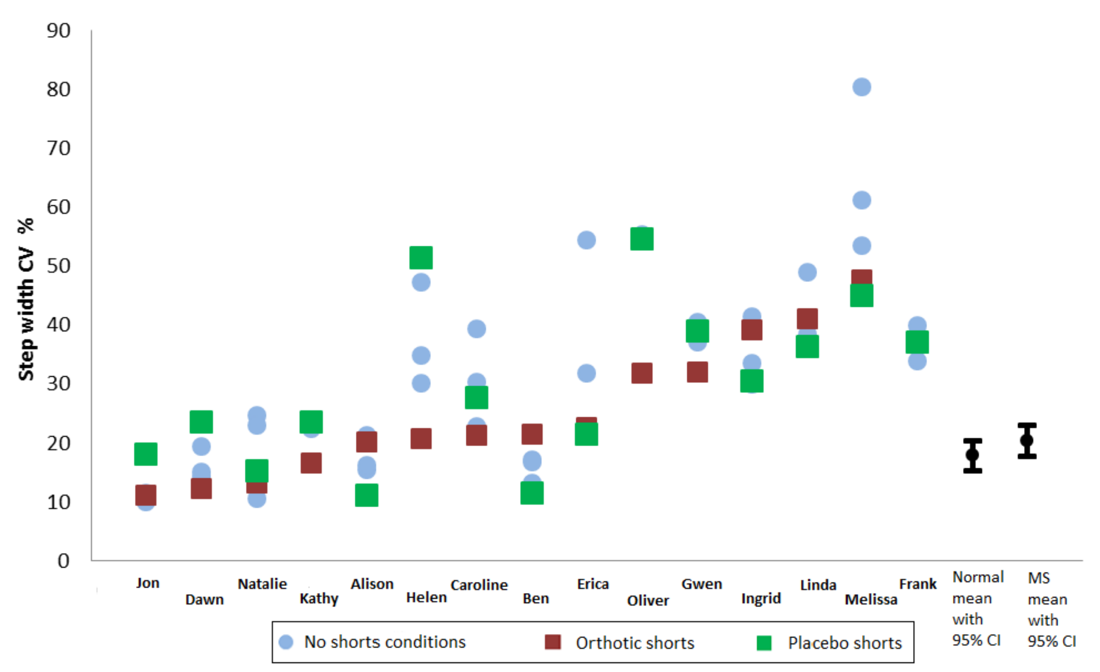


Figure 6.4: Impact of orthotic and placebo shorts on step width variability (*CV =coefficient of variation; mean CVs for normal, non-neurologically impaired people and people with MS are provided for comparison, with 95% confidence intervals (95% CI) for means, using data obtained from Comber et al., 2017*).

Table 6.5: Impact of shorts on variability of spatio-temporal gait parameters

	Orthotic shorts vs. no shorts					Placebo shorts vs. no shorts					Orthotic shorts vs. placebo shorts				
	n	Orthotic shorts	No shorts	Mean difference (95% CI)	Effect size	n	Placebo shorts	No shorts	Mean difference (95% CI)	Effect size	n	Orthotic shorts	Placebo shorts	Mean difference (95% CI)	Effect size
Step length variability (SD, centimetres)	15	3.4 ± 1.0	3.4 ± 0.9	-0.1 (-0.5 to +0.3)	-0.09	15	3.0 ± 0.6	3.3 ± 1.3	-0.3 (-0.9 to +0.2)	-0.32	14	3.3 ± 1.0	3.0 ± 0.7	+0.3 (-0.1 to +0.6)	0.31
Step width variability (SD, centimetres)	15	2.6 ± 1.1	3.4 ± 1.6	-0.8* (-1.3 to -0.3)	-0.57	15	3.0 ± 1.3	3.2 ± 1.6	-0.2 (-0.6 to +0.3)	-0.12	14	2.6 ± 1.2	3.1 ± 1.4	-0.4 (-1.0 to +0.1)	-0.36
Stride time variability (SD, milliseconds)	15	Median (IQR) 32 (28)	Median (IQR) 39 (20)	Median difference -1	-0.22	15	Median (IQR) 28 (14)	Median (IQR) 33 (19)	Median difference -1.5	-0.09	14	Median (IQR) 29.9 (30)	Median (IQR) 28 (14)	Median difference +2.4	0.09
Step length variability (CV)	15	5.65 ± 2.1	6.04 ± 2.1	-0.4 (-1.1 to +0.4)	-0.19	15	5.19 ± 1.4	5.94 ± 2.7	-0.8 (-1.8 to +0.3)	-0.35	14	5.5 ± 2.1	5.04 ± 1.3	+0.5 (-0.2 to +1.1)	0.28
Step width variability (CV)	15	25 ± 11	29 ± 13	-4.2 (-8.3 to +0.3)	-0.36	15	30 ± 14	32 ± 17	-2.4 (-8.9 to +4.0)	-0.16	14	25 ± 11	29 ± 14	-4 (-11 to +2.7)	-0.32
Stride time variability (CV)	15	Median (IQR) 3.3 (2.0)	Median (IQR) 3.6 (1.6)	Median difference -0.04	-0.19	15	Median (IQR) 3.0 (1.0)	Median (IQR) 3.13 (1.5)	Median difference -0.16	-0.20	14	Median (IQR) 2.9 (2.0)	Median (IQR) 2.9 (1.1)	Median difference +0.3	0.08

*(SD = standard deviation, CV = coefficient of variation, the standard deviation divided by the mean for each dataset multiplied by 100. Means and SDs provided for between day/between shorts analysis differ from those for within day analysis because they include only those individuals for whom data is available for both pairs of shorts, * indicates confidence interval does not go over zero.)*

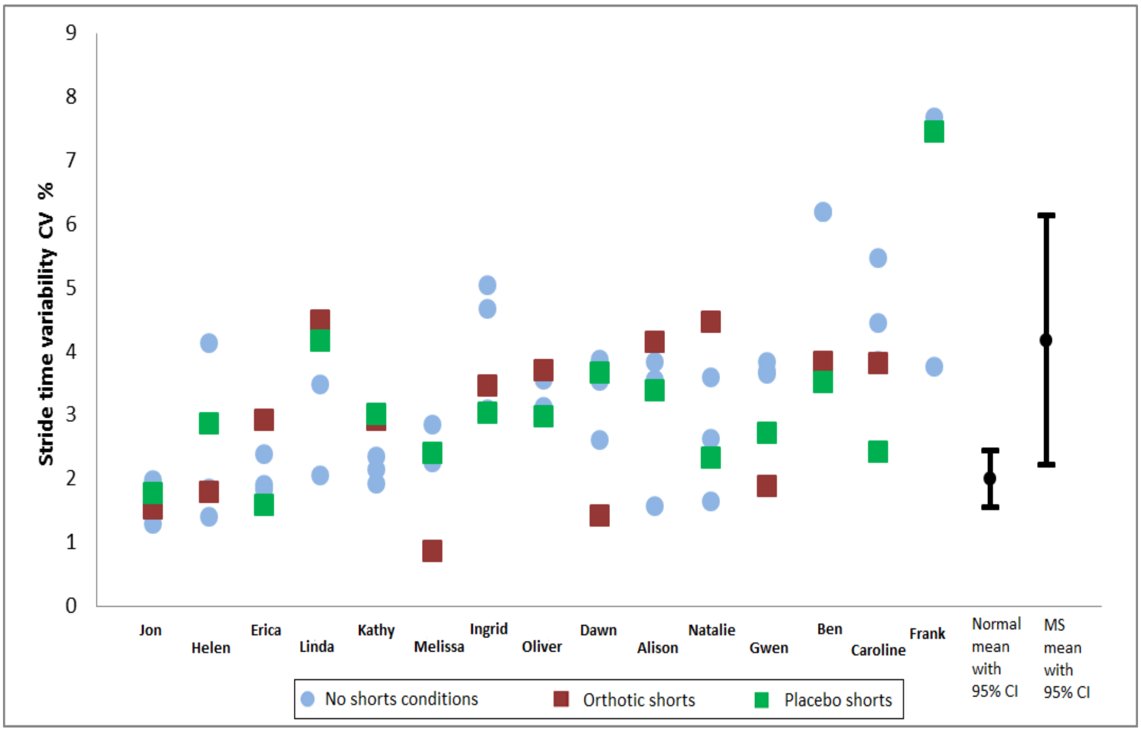


Figure 6.5: Impact of orthotic and placebo shorts on stride time variability (CV = *coefficient of variation; mean CVs for normal, non-neurologically impaired people and people with MS are provided for comparison, with 95% confidence intervals (95% CI) for means, using data obtained from Comber et al., 2017*).

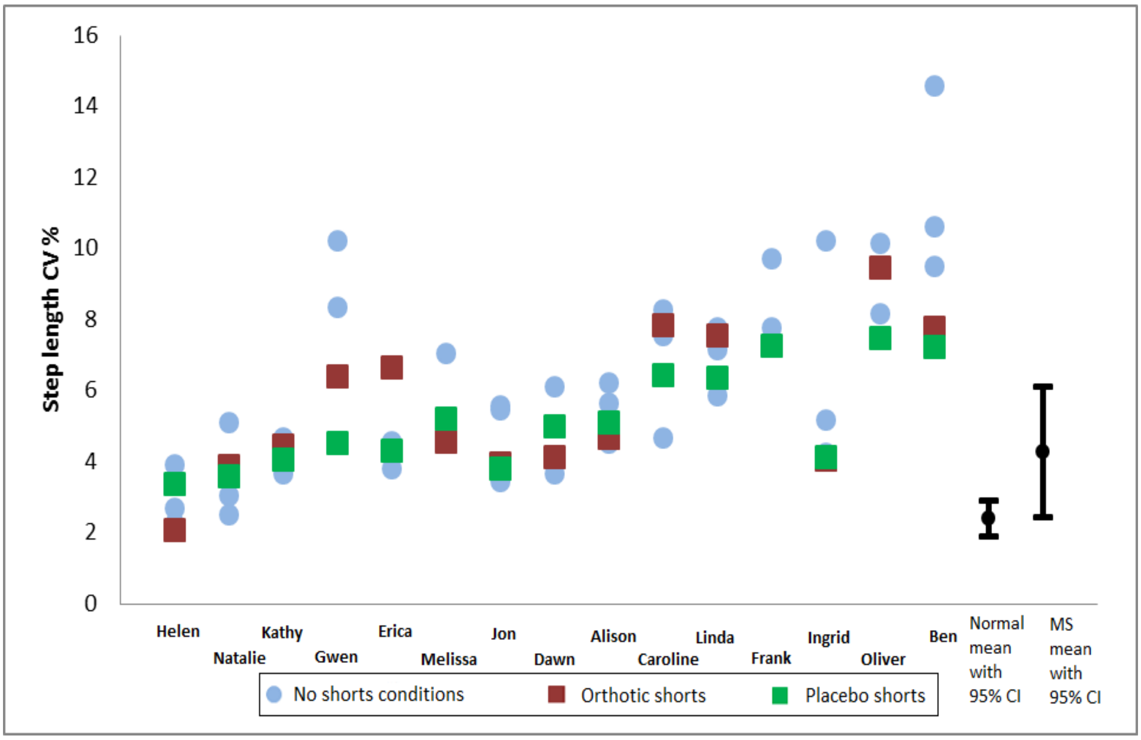


Figure 6.6: Impact of orthotic and placebo shorts on step length variability (CV =*coefficient of variation; mean CVs for normal, non-neurologically impaired people and people with MS are provided for comparison, with 95% confidence intervals (95% CI) for means, using data obtained from Comber et al., 2017*).

Impact of orthotic shorts on trunk and pelvis movement in walking

Even though measures of variability of foot placement and timing showed decreased variability with the orthotic shorts, pelvic sway variability increased (ES = 0.30, Table 6.6). Pelvic sway variability did not increase with the placebo shorts (ES = 0.08). Trunk sway variability increased slightly with the orthotic shorts (ES = 0.10) and decreased with the placebo shorts (ES = -0.20), creating a large difference in trunk sway variability between the two pairs of shorts (ES = 0.71).

As might be expected, mediolateral pelvic sway was moderately correlated with step width at baseline (r = 0.556, p = 0.0031 on orthotic testing day; r = 0.774, p = 0.001 on placebo testing day). However, despite the moderate decrease in step width with the orthotic shorts reported earlier, the orthotic shorts had a negligible impact on mediolateral pelvic sway (ES = -0.05). Inspection of individual data showed that some participants decreased step width and mediolateral pelvic sway in the shorts (Ingrid, Dawn, Jon, Gwen), however, many decreased step width with either no change in mediolateral pelvic sway or an increased pelvic sway (Linda, Helen, Melissa, Caroline, Ben, Natalie). For both orthotic and placebo shorts, the amount by which pelvic sway decreased with the shorts was related to the degree of sway at baseline, i.e. those with more pelvic sway at baseline improved the most with the shorts (r = -0.936, p = 0.011 for orthotic and r = -0.759, p = 0.001 for placebo). There was a small decrease in mediolateral trunk sway in both orthotic and placebo shorts (ES = -0.14 and -0.15 respectively), with a larger trunk sway seen in the orthotic shorts than placebo (ES = 0.29).

Regarding rotational movement of the trunk and pelvis, there was a tendency for movement to increase in range with the orthotic shorts (Table 6.6). There was a small increase in pelvic roll (lateral tilt) but no change in trunk roll. There was a small to moderate increase in pelvic pitch (ES = 0.35; approximates to anteroposterior tilt) and a moderate increase in trunk pitch (ES = 0.65), with an increased pitch range of 1°. There was a moderate increase in pelvic yaw (ES = 0.48; left-right rotation in the transverse plane), with a median increase in rotation of 3.7°. There was a small to moderate increase in trunk yaw (ES = 0.38), with a median increase in rotation of 1.9°.

Table 6.6: Impact of shorts on trunk and pelvic movement

	Orthotic shorts vs. no shorts					Placebo shorts vs. no shorts					Orthotic shorts vs. placebo shorts				
	n	Orthotic shorts	No shorts	Mean difference (95% CI)	Effect size	n	Placebo shorts	No shorts	Mean difference (95% CI)	Effect size	n	Orthotic shorts	Placebo shorts	Mean difference (95% CI)	Effect size
ML pelvic sway (centimetres)	15	5.4 ± 1.4	5.4 ± 1.7	-0.1 (-0.8 to +0.6)	-0.05	15	5.2 ± 1.0	5.3 ± 1.5	-0.1 (-0.5 to +0.3)	-0.06	14	5.4 ± 1.4	5.2 ± 1.0	+0.2 (-0.3 to +0.7)	0.18
ML trunk sway (centimetres)	15	5.2 ± 1.4	5.4 ± 1.5	-0.2 (-0.7 to +0.3)	-0.14	15	5.2 ± 1.8	5.5 ± 2.1	-0.3 (-0.7 to +0.1)	-0.15	14	5.2 ± 1.4	4.8 ± 1.2	+0.4 (-0.3 to +1.1)	0.29
Pelvic roll (degrees)	15	11.7 ± 3.5	11.1 ± 3.0	+0.6 (-0.4 to +1.6)	0.20	15	11.3 ± 2.7	11.4 ± 2.9	-0.1 (-0.7 to +0.5)	-0.03	14	11.6 ± 3.6	11.5 ± 2.7	+0.1 (-0.9 to +1.1)	0.04
Trunk roll (degrees)	15	14.2 ± 6.5	14.3 ± 7.1	-0.1 (-2.0 to +1.8)	-0.01	15	12.9 ±6.3	13.2 ± 5.5	-0.2 (-2.0 to +1.5)	-0.04	14	14.3 ± 6.5	12.9 ± 6.3	+1.5 (-0.7 to +3.7)	0.23
Pelvic pitch (degrees)	15	11.5 ± 4.3	10.5 ± 3.0	+1.0 (-1.1 to +3.2)	0.35	15	10.4 ± 3.2	11.1 ± 3.5	-0.7 (-2.6 to +1.3)	-0.19	14	11.8 ± 4.3	10.1 ± 3.2	+1.8 (-1.3 to +4.9)	0.51
Trunk pitch (degrees)	15	11.4 ± 3.2	10.3 ± 1.7	+1.1 (-0.3 to +2.6)	0.65	15	10.5 ± 2.7	11.2 ± 4.2	-0.7 (-2.2 to +0.8)	-0.17	14	11.3 ± 3.3	10.2 ± 2.5	+1.1 (-0.02 to +2.3)	0.46
Pelvic yaw** (degrees)	15	18.0 (7.4)	13.1 (6.8)	Median difference +3.7	0.48	15	15.9 (8.0)	15.3 (4.2)	Median difference +1.4	0.34	14	18.4 (7.0)	14.9 (8.6)	Median difference +4.3	0.59
Trunk yaw** (degrees)	15	15.8 (5.6)	14.5 (7.3)	Median difference +1.9	0.38	15	16.8 (7.0)	15.2 (2.9)	Median difference +0.2	0.22	14	16.0 (5.9)	16.3 (6.6)	-0.5	0.07
Pelvic sway variability (SD, centimetres)	13	0.65 ± 0.27	0.59 ± 0.21	+0.07*(+0.01 to +0.12)	0.30	13	0.58 ± 0.21	0.60 ± 0.25	-0.02 (-0.07 to +0.03)	0.08	12	0.66 ± 0.28	0.58 ± 0.21	+0.11*(+0.03 to +0.19)	0.33
Trunk sway variability (SD, centimetres)	13	0.67 ± 0.27	0.64 ± 0.25	+0.02 (-0.03 to +0.08)	0.10	13	0.59 ± 0.24	0.65 ± 0.30	-0.06 (-0.13 to +0.01)	-0.20	12	0.67 ± 0.28	0.54 ± 0.18	+0.13*(+0.03 to +0.22)	0.71

*(ML = mediolateral; values are means & standard deviations other than for **trunk & pelvic yaw, which are median (interquartile range); between-shorts values differ from within day values because between-day includes only individuals for whom data is available for both pairs of shorts; * indicates confidence interval does not go over zero; ML sway, roll, pitch and yaw are all 95% range; roll is rotation in the frontal plane, which approximates to lateral pelvic tilt and trunk side flexion; pitch is rotation in the sagittal plane, which approximates to anterior-posterior pelvic tilt or trunk flexion/extension; yaw is rotation in the transverse plane which approximates to left/right rotation.)*

In comparison, with the placebo shorts, trunk and pelvic roll did not change and pelvic and trunk pitch both decreased slightly (ES = -0.19 and -0.17 respectively). Pelvic and trunk rotation in yaw increased with the placebo shorts (ES = 0.34 and 0.22 respectively) but not to the same extent as with the orthotic shorts.

Impact of orthotic shorts on maximal gait speed and dual task cost

There was very little impact on maximal gait speed (T25FW) in either pair of shorts (ES = 0.08 for orthotic and 0.10 for placebo) (Table 6.7) and no difference in maximal gait speed between the two pairs of shorts (ES = 0.04).

There was a small to moderate impact of both pairs of shorts on the dual task cost of cognition on walking speed, indicating that with the shorts on, cognition interfered less with the ability to walk at maximal speed (for orthotic ES = -0.35 and for placebo ES = -0.34). In addition, cognitive ability whilst walking was better with the shorts than without but this effect was more marked in the placebo shorts (ES = 0.45 for placebo and 0.22 for orthotic).

6.2.5 Self-report measures

Self-report measures were participant perceived walking ability (MSWS-12), balance confidence (ABC-UK), participant reported Global Rating of Change (GRC) and falls incidence. The ABC-UK scores were similar at Visit 1 and Visit 7 with mean ± SD of 50 ± 14 at Visit 1 and 52 ± 14 at Visit 7. Conversely, the MSWS-12 scores improved over the course of the study with 73 ± 18 at Visit 1 and 62 ± 22 at Visit 7 (see Table 6.8).

Both shorts appeared to have a moderate positive impact on participant perceived walking ability (see Table 6.8). For the orthotic shorts, most participants improved perceived walking ability during the orthotic shorts trial, with a group mean difference of 21 points (ES = 0.6). The placebo shorts had only slightly less influence, with a mean improvement from baseline of 18 points (ES = 0.52). Confidence intervals indicate 95% confidence that the true sample means reflect improved perceived walking ability. Baert et al. (2014) suggested a change of 11.4 in the MSWS-12 indicated clinically important change. Thus, the group mean improvement was clinically important. Change at an individual level was clinically important for nine participants with the orthotic shorts and eight participants with the placebo shorts.

Table 6.7: Impact of shorts on maximal gait speed and dual task cost

	Orthotic shorts vs. no shorts					Placebo shorts vs. no shorts					Orthotic shorts vs. placebo shorts				
	n	Mean ±SD in orthotic shorts	Mean ±SD in no shorts	Mean difference (95% CI)	Cohen's d effect size	n	Mean ±SD in placebo shorts	Mean ±SD in no shorts	Mean difference (95% CI)	Cohen's d effect size	n	Mean ±SD in orthotic shorts	Mean ±SD in placebo shorts	Mean difference (95% CI)	Cohen's d effect size
Maximal gait speed T25FW (metres/second)	15	1.28 ± 0.4	1.24 ± 0.3	+0.03 (-0.06 to +0.12)	0.10	15	1.25 ± 0.3	1.22 ± 0.3	+0.03 (-0.03 to +0.08)	0.08	14	1.28 ± 0.4	1.29 ± 0.3	-0.01 (-0.1 to +0.07)	-0.04
Dual task cost on walking speed (percentage)	15	10.9 ± 7	13.7 ± 8	-2.7 (-5.7 to +0.3)	-0.35	14	10.6 ± 10	14 ± 10	-3.5 (-5.7 to +0.3)	-0.34	14	11.4 ± 8	10.6 ± 10	+0.8 (-3.8 to +5.3)	0.08
Dual task cost on cognition (letters/second)	15	0.8 ±0.2	0.8 ± 0.2	+0.05 (-0.08 to +0.18)	0.22	14	0.9 ± 0.3	0.7 ± 0.3	+0.12* (+0.02 to +0.3)	0.45	14	0.8 ±0.3	0.9 ± 0.3	-0.04 (-0.19 to +0.11)	-0.17

*(T25FW = Timed 25-foot walk; means and SDs provided for between day/between shorts analysis differ from those for within day analysis because they include only those individuals for whom data is available for both pairs of shorts, * indicates confidence interval does not go over zero.)*

Table 6.8: Mean changes and effect sizes for 12-item Multiple Sclerosis Walking Scale and Activities Balance Confidence Scale.

	Mean score at baseline ± SD	Mean score following orthotic shorts home trial ± SD	Mean score following placebo shorts home trial ± SD	Mean score at final visit ± SD	Comparison	Mean difference (95% confidence interval)	Effect size
MSWS-12 %	73 ± 18	52 ± 24	55 ± 26	62 ± 22	Orthotic vs. baseline	-21* (-12 to -30)	-0.60
					Placebo vs. baseline	-18* (-8 to -27)	-0.52
					Orthotic vs. placebo	+3 (-7 to +14)	0.09
ABC-UK %	50 ± 14	60 ± 19	58 ± 19	52 ± 14	Orthotic vs. baseline	+10* (+0.2 to +21)	0.33
					Placebo vs. baseline	+9 (-0.5 to +18)	0.31
					Orthotic vs. placebo	-1.6 (-9 to +5)	-0.16

*(MSWS-12 = 12-item Multiple Sclerosis Walking Scale; ABC-UK = the UK version of the Activities-Specific Balance Confidence measure; for the MSWS-12, a lower score indicates walking is perceived to be better; for the ABC-UK, a higher score indicates more confidence; * indicates confidence interval does not go over zero)*

Changes to balance confidence were less obvious than those for perceived walking ability (see Table 6.8). There was a small to moderate improvement in balance confidence when both the orthotic (ES = 0.33) and placebo shorts (ES = 0.31) were worn, compared to baseline. The mean change of 10 points is greater than the estimate of clinically important change of 6.8 suggested for stroke survivors (Botner et al., 2005). There is 95% confidence that the true sample mean reflects improvement in balance confidence. Seven participants showed a clinically important improvement with the orthotic shorts, although three (Ben, Gwen and Helen) showed a clinically important deterioration. Eight participants showed a clinically important improvement with the placebo shorts but three (Ben, Frank and Helen) showed a clinically important deterioration.

Participant Global Rating of Change (GRC) was assessed, on a scale of -5 to +5, immediately after the objective assessment of walking ability, to capture participants' assessment of the immediate impact of the shorts on their walking. Ratings ranged from -1 to +5 with a median of 1.5 for the orthotic shorts and 1.0 for the placebo shorts. GRC was measured because of the hypothesis suggested by the FabO IPA study

(Chapter 3) that an initial perception of a beneficial effect might be an important predictor as to whether people find an orthosis acceptable in the long term. However, there was no relationship between the perceived initial benefit of the shorts and whether they were kept at the end of the study (*p* = 0.569 for orthotic and *p* = 0.282 for placebo). Three of the people who kept the orthotic shorts at the end of the trial perceived no benefit at all on first use and a further four perceived only a very small benefit.

Five participants reported no falls during the study (Alison, Erica, Helen, Kathy and Natalie). For the 10 participants who reported falls, these ranged from one to 38 falls in a day. There were differences in the interpretation of a fall, with the individual who reported 38 falls in one day explaining that these were mostly uncontrolled and unexpected descents to a chair or bed. Falls frequency decreased during the shorts home trials compared to baseline. In the baseline period, there was a median of three falls (range 0 - 26), in the orthotic shorts home trial there was a median of one fall, (range 0 to 16, ES = -0.19 baseline to orthotic) and, in the placebo shorts, a median of two falls (range 0 to 38, ES = 0.09 baseline to placebo).

6.3 Discussion

6.3.1 Introduction

This discussion section will consider recruitment and retention, shorts provided, baseline ability, quantitative assessment of acceptability, the changes in baseline measures over the course of the study and the key findings around the impact of the shorts. Discussion will be further developed in Chapter 8, where the findings from this quantitative chapter will be integrated with the qualitative findings with respect to acceptability, the impact of the shorts on walking ability and lessons learnt for future studies.

6.3.2 Recruitment and retention

Recruitment was smooth and successful. The full range of recruitment strategies proposed was used and most contributed towards recruitment. The speed at which recruitment occurred was dictated by the availability of the researcher for data collection rather than the availability of willing participants. It is important to note that the strategy of recruiting via the NHS physiotherapy service was relatively unsuccessful. It took 15 months for a potential list of only 17 people to be identified and there was a relatively high proportion of these 17 who either did not reply (*n* = 1), were unable or unwilling to participate (*n* = 4) or were ineligible (*n* = 5). To some extent, this might have been because people tend to attend NHS physiotherapy when they have a particular challenge and, therefore, their condition was not stable, as required by the eligibility criteria. The relative lack of recruitment via the NHS physiotherapist might have been related to the acceptability of the shorts intervention to the physiotherapists themselves. This important possibility has not been investigated within this programme of research.

The loss of one participant between consent and shorts provision indicated inadequate eligibility criteria. An inclusion criterion of "good or compensated distal function" should be added for future studies. People might still trial the shorts if they have poor distal function but only if they successfully use FES or an AFO to compensate for such difficulties. This would be a difficult judgement for participants to make themselves. Indeed, it is important to note that two of the participants in the OSFeaMS study used FES or a dorsiflexor assist device but did so intermittently. No compensatory pelvic movements were observable in their gait. Nevertheless, highlighting the possibility that orthotic shorts may not be suitable for people with poor distal function would prompt a discussion around this issue with potential participants. This should avoid disappointing and inconveniencing people who are not eligible.

Retention in the study was strong, with only one participant withdrawing. Retention is potentially easier in a single-centre study, conducted by a single-handed researcher, in that participants are dealing with the same person throughout. This might be positive in terms of participants having a clear vision of what each visit would entail and might increase a sense of loyalty to the study. Studies that are multi-centre and involve a larger research team are likely to carry a higher risk of participant withdrawal (Walters et al., 2017).

6.3.3 Shorts provided and pressure applied

A range of shorts was provided in the study, of different colours and styles. Pressure applied by the shorts was greater for the orthotic shorts than for the placebo shorts, as intended. Pressures applied by the shorts were relatively low (6 – 13 mmHg), compared to compression garments used in sport; as reported in Chapter 2, these range from between 10 - 40 mmHg. One recent study measured pressure beneath orthotic garments provided for children with cerebral palsy (Shaari, Osman & Shasmin, 2018). They assessed pressure in a variety of postures and activities and found that, in standing, pressures ranged as low as those exerted by the OSFeaMS orthotic shorts but as high as 26 to 51 mmHg. In sitting, the interface pressures were higher and, for one child, interface pressures of 120 mmHg were recorded at both trunk and pelvis. These findings suggest that pressures exerted by the orthotic shorts in the OSFeaMS study may be much lower than pressures exerted by orthoses used with children, which may be relevant to acceptability.

6.3.4 Participant characteristics – walking ability

OSFeaMS participants had faster self-selected walking speeds and greater step length and step width variability than PwMS in previous studies. This could either be due to the nature of the sample or the data collection methods. There are indications that both factors had an influence. Whilst the OSFeaMS study participants had a faster self-selected gait than found in previous research, their maximal gait speed as assessed with the T25FW was slower than previous studies; the OSFeaMS mean speed for the T25FW was 1.24 ± 0.3 m/s, compared to 1.37 ± 0.6 m/s from the Comber et al., 2017 review. This indicates that the instructions used for preferred gait speed may have resulted in them walking faster than their true self-selected speed. This might explain the finding that gait variability was higher than that found in previous studies as it is known that gait variability is at its lowest at an individual's preferred walking speed and increases at faster or slower speeds (Jordan, Challis & Newel, 2007). Another potential reason for the relatively more variable gait could be related to the moderate disability level in this participant group. PwMS with a moderate disability level have increased gait variability compared to both more severely disabled and less severely disabled PwMS (Kalron, 2017; Socie, Motl, Pula, Sandroff & Sosnoff, 2013b).

The discrepancy in gait variability found between previous research and the OSFeaMS study is particularly noticeable for step width variability. The CV for step width without shorts at Visit 3 was 33 ± 19%. Moon et al. (2016) found a step width CV of 20.4%. This was based on only 49 participants in two studies (Socie, Motl & Sosnoff, 2014; Socie et al., 2013a) but a further study by the same research group found a similar CV of 19.2 ± 11.9% (*n* = 88, Socie et al., 2013b).

The discrepancy in step width variability could relate to walking aid use, disability level or the inclusion criteria of the OSFeaMS study. Socie et al. (2013b) found greater step width variability in their less disabled participants than in those more disabled participants who used walking aids during testing. They suggested that walking aid use might be responsible for decreasing step width variability. The impact of walking aids on step width variability has not been directly investigated but walking aids do improve variability of other spatiotemporal gait parameters in PwMS (Gianfrancesco et al., 2011). Seven OSFeaMS participants either never or only occasionally used a walking aid (Erica, Gwen, Helen, Ingrid, Jon, Kathy and Natalie; see Table 6.1, p109 for details). Two participants habitually used walking aids but chose to walk without aids on the GAITRITE assessment (Melissa and Oliver); potentially increasing their step width variability above what it might be with their aid. A second suggestion is that the high step width variability in the OSFeaMS study may be attributable to the participants' disability level. Kalron (2016) assessed 381 PwMS and found that the 23 people with an EDSS level of 5.0 - 5.5 had similar step width variability to the OSFeaMS participants. Those who were both more disabled and less disabled had the more typical step width variability reported by other studies. EDSS was not measured in the OSFeaMS study so this possible explanation cannot be explored. The final suggestion is that the OSFeaMS study recruited people with particularly high step width variability because the study only included people who self-identified as having instability around the trunk or hips. Step width variability is believed to reflect the control of balance to a far greater extent than step length and stride time variability (Moon et al., 2016), therefore, the particularly high step width variability seen in this study may reflect a group with more pronounced balance instability than a more typical sample of PwMS.

6.3.5 Quantitative assessment of acceptability

Adherence to recommended shorts wear times was strong and 75% of participants chose to keep the shorts after the trial. No pre-determined targets were set in the OSFeaMS study for acceptable wear times or continuing use. Nevertheless, this data can be compared to that obtained in previous studies to obtain an impression of how acceptable the orthotic shorts were compared to other orthoses. Previous research has utilised a range of different methods for estimating acceptability and, therefore, it is difficult to make comparisons. Early research into fabric orthoses reported how many participants said they would continue to use their orthosis after the study was over and this was one reason why this metric was chosen for the OSFeaMS study. The "continued use" percentage of 75% in OSFeaMS study was significantly higher than the low intended continued use found in those early fabric orthosis studies. This was seven out of 15 people in Edmondson et al. (1999), one out of eight in Rennie et al. (2000), one out of 12 in Nicholson et al. (2001) and two out of five in Flanagan et al. (2004). It is possible that the design of fabric orthoses has improved in the intervening years, thus improving acceptability. Serroa et al. (2017) investigated a short Lycra suit in people with ataxia and assessed satisfaction with one single question "Are you satisfied with your suit?" They found 10 out of 11 people were satisfied.

Research into other orthoses indicated strong acceptability. For using FES for foot drop, Taylor (2004, cited in Barrett & Taylor, 2010) reported only a 9% abandonment rate at a two-year follow-up clinic. A questionnaire study for AFOs suggested only a 7% abandonment rate (Holtkamp, Wouters, van Hoof, van Zaalen & Verkerk, 2015). These comparisons suggest that the orthotic shorts may not be as acceptable as other types of orthosis, however, for both FES and AFOs, clinicians and researchers have a wealth of knowledge regarding orthotic design and who is most likely to benefit from the orthoses. For example, when FES is prescribed, this is done using a trial of stimulation at which the impact upon gait is objectively tested (Barrett & Taylor, 2010). Potential users have an opportunity to practice using the stimulators before prescription. If FES does not demonstrate an initial orthotic effect or participants struggle to use the device without support, then FES is not prescribed. As discussed in Section 3.6.2, such collaborative practice and professional support would probably improve acceptability and could not be included within the OSFeaMS study because of the relatively early phase of intervention development.

6.3.6 Baseline stability

There were indications of changes in participants' underlying abilities during the study, with a small improvement in gait speed, a small decrease in variability of step width and stride time and moderate improvements in dual task cost and participant perceived walking ability (MSWS-12). These improvements could be due to chance or may indicate a real change induced by the experience of being on the study. Such changes could result from the experience of wearing the shorts or the repeated measurements. The improvements in dual task cost seem particularly likely to result from the impact of practice, maybe suggesting that this type of measure is not appropriate for a study with so many assessment points. This possible training effect could have been minimised by having a greater number of different cognitive tasks but this in turn would have increased participant burden.

Improvement over time suggests that a crossover study such as this was not an appropriate study design for this intervention and, taken in conjunction with the inability to counter-balance the order in which the shorts were tested, indicates that direct comparison between the orthotic and placebo shorts is not valid.

6.3.7 The impact of orthotic shorts on objective and self-report measures

For this section of the discussion, findings have been grouped to draw out possible implications around underlying mechanisms. Findings are grouped around participant confidence, mediolateral control, gait variability and dual task cost. In each of these sections, findings are summarised and compared to previous research to inform discussion around possible mechanisms of effect and relevance to clinical practice.

Confidence and self-selected gait speed

With the orthotic shorts, small improvements were seen in self-selected gait speed, step length, stride time and cadence. Although these changes were small, there was 95% confidence that the mean differences indicated improvement. No such changes were seen with the placebo shorts. Scores on both the ABC-UK and the MSWS-12 improved in both the orthotic and placebo shorts, indicating improved perceived walking ability and balance confidence. Step length, cadence and stride time would be expected to change to a similar degree to gait speed, as these parameters enable gait speed to increase.

The improvement in speed seen with the orthotic shorts is unlikely to have a direct effect on function. However, self-selected gait speed, cadence, step length and stride time have all been linked to fear of falling in older people (Delbaere et al., 2009), suggesting that people deliberately slow down and take smaller steps if they feel at risk of falling. It could be suggested that the OSFeaMS participants walked more confidently in their orthotic shorts. Maximal gait speed did not improve in the orthotic shorts. This further suggests that the changes seen in self-selected gait speed reflect increased effort or improved confidence, as opposed to any specific biomechanical changes.

If the increase in self-selected gait speed does reflect decreased fear of falling, this could be important because fear of falling is related to physical activity curtailment in MS (Kalron, Aloni, Givon & Menascu, 2018). Gait speed is also associated with risk of falling in PwMS (Giannì, Prosperini, Jonsdottir & Cattaneo, 2014). Giannì et al. (2014) suggested that preferred gait speeds for non-fallers with MS are between 1.1 - 1.31 m/s and gait speed for fallers between 0.91 - 1.1 m/s. Although the group mean for gait speed with the orthotic shorts increased into the non-fallers category, inspection of the individual data (Figure 6.2) indicates that only one participant (Gwen) moved from the fallers to the non-fallers category.

The idea that increased self-selected gait speed might reflect improved confidence is supported by the moderate improvements seen with the self-report measures. Self-selected gait speed and participant perceived walking ability showed improvement over the course of the study, suggesting that the experience of being in the study increased confidence. This may have been in response to the experience of wearing orthotic shorts or due to the wider experience of the study.

Changes seen in MSWS-12 in the OSFeaMS study may be clinically important. The improvement seen following the two-week home trial exceeded the threshold for clinical importance suggested by Baert et al. (2014). In addition, changes were greater than or similar to those found in studies investigating the effectiveness of balance exercise programmes, typically lasting 12 weeks (for example, Carling, Forsberg, Gunnarsson & Nilsagård, 2017; Feys et al., 2019; Forsberg, von Koch & Nilsagård, 2016). Likewise, improvement was greater than that obtained by the drug fampridine, believed to improve walking ability in MS (Hobart et al., 2019).

Overall, the findings on gait speed and self-report measures suggest that orthotic shorts might increase confidence and decrease fear of falling in PwMS. Whilst the changes in objective measures of gait speed are small, the participant perceived effect appears to be important.

Mediolateral control of gait

Moderate improvement was seen with the orthotic shorts in step width and step width variability, with the orthotic shorts appearing to normalise step width and decrease step width variability. At a group level, step width decreased but some participants had narrower step width at baseline and some of these participants increased step width with the orthotic shorts. The placebo shorts had no impact on step width and only a very small effect on step width variability suggesting that the increased structure and support in the orthotic shorts might be required for any effect on step width. Mediolateral pelvic sway did not improve in either pair of shorts, although there was a very small decrease in mediolateral trunk sway in the orthotic shorts. For both pairs of shorts, the degree to which mediolateral pelvic sway decreased was related to the degree of sway at baseline. In other words, those with more pelvic sway at baseline improved the most with the shorts.

To understand the changes described above, it is important to explore the functional relevance of step width and step width variability, consider how mediolateral stability is controlled in normal walking and consider why mediolateral control might be affected in MS.

Step width and step width variability are believed to be particularly reflective of balance ability (Brach et al., 2008; Filli et al., 2018). Step width variability has been linked to falls incidence in healthy elderly people (Brach, Berlin, Vanswearingen, Newman & Studenski, 2005) but appears not to have been studied in MS. Brach et al. (2005) suggested there was an optimal level of step width variability associated with low falls risk and that both higher and lower step width variability were associated with falling. Wide step width is widely believed to be an indicator of poor balance (Givon et al., 2009). Narrow step width is seen in healthy people as they walk faster (Orendurff, Segal, Klute & Berge, 2004) but may also be an indicator of poor balance, in that narrow step width has been associated with inadequate trunk mobility (Arvin, van Dieën & Bruijn, 2016). In addition, narrow step width has been described in elderly people with reduced balance (Nordin, Moe-Nilssen, Ramnemark & Lundin-Olson, 2010), particularly those who fall sideways (Ko et al., 2007).

Mediolateral stability is considered to require far more input from the neurological control system, compared to anteroposterior stability (O'Connor & Kuo, 2009). Anteroposterior control is considered to be dependant upon passive, pendular motion of the legs, whereas mediolateral stability requires active neural control (O'Connor & Kuo, 2009). Mediolateral stability may be controlled by different neural networks to those used for anteroposterior control (Brach et al., 2008). The relatively passive control of anteroposterior movement has been illustrated using computer simulations and robots that can walk down a slope, requiring no active feedback to maintain balance. The increased neural control of mediolateral stability has been suggested as a reason for the greater mediolateral variability during walking, comparing to anteroposterior variability (O'Connor & Kuo, 2009). Wurdeman, Huben and Stergiou (2012) showed that in sideways walking, the opposite is true and variability is lower in an anteroposterior plane, indicating that postural control in the direction of movement is easier to achieve than postural control perpendicular to the direction of movement.

Biomechanically, the control of mediolateral pelvic stability requires complex coordination of trunk, pelvis and the swinging leg. Muscle control around the stance hip is believed to be inadequate to control mediolateral stability during walking; instead, the momentum and direction of movement of the swinging leg (Rankin, Buffo & Dean, 2014) and its placement in the mediolateral plane (Donelan, Shipman, Kram & Kuo, 2004) are believed to be responsible for maintaining mediolateral stability.

Proprioception around the stance hip has been shown to be influential in determining the direction of movement of the swinging leg (Roden-Reynolds, Walker, Wasserman & Dean, 2015). Roden-Reynolds et al. (2015) used muscle vibration to confuse the proprioceptive feedback from gluteus medius on the stance side in healthy people and this caused the swinging leg to respond inappropriately, resulting in loss of mediolateral control. Roden-Reynolds et al. (2015) suggested that proprioceptive feedback from the trunk might also input into a sense of where the swinging leg needs to be placed to maintain stability. The complex coordination required for mediolateral stability could fail at multiple points in the control system in PwMS. It is believed to be particularly sensitive to noise in the sensorimotor system (Roos & Dingwell, 2010).

MS might affect control of stability in a number of ways and the mechanisms are likely to be multi-factorial. MS leads to decreased muscle strength and sensory feedback and both of these can contribute to decreased postural stability (Prosperini & Castelli, 2018). In MS, sensory feedback is slowed by poor spinal and supraspinal conduction, meaning that it can arrive at cortical centres later than required (Cameron et al., 2008a). This is hypothesized to result in responses that are poorly scaled and poorly timed with respect to what might be required, causing hypermetric or delayed movement responses (Cameron et al., 2008a). Delayed or absent sensory feedback may mean that postural sway in standing is increased in MS (Huisinga, St George, Spain, Overs & Horak, 2014). Control of postural sway requires multiple sensory systems, depending upon the specific environmental context and all these sensory systems can be affected by MS (Cattaneo et al., 2016). Poor muscle strength and sensory feedback have been suggested as potential mechanisms for increased postural sway and increased gait variability in MS (Callesen et al., 2019; Moon, Wajda, Motl & Sosnoff, 2015). In addition, cortical and cerebellar networks might be directly affected by MS so that planning, initiation and coordination of motor tasks might be impaired (Moon et al., 2016). Because PwMS have multiple challenges to postural control, their mediolateral stability is likely to be more problematic than control in the anteroposterior direction.

Bearing in mind the potential causes of the increased step width and step width variability seen in MS, there are a number of possibilities as to why orthotic shorts might have a positive influence. With the timescales involved in a direct orthotic effect, there are unlikely to be changes in the central neural networks controlling gait. Step width alters with gait speed, becoming narrower as people walk faster (Orendurff et al., 2004). Because the OSFeaMS participants walked more quickly in their orthotic shorts, this could explain the decrease in step width. However, this is unlikely because there was no relationship between the change in gait speed and the change in step width in the OSFeaMS data. More likely possibilities are direct support and sensory mechanisms. The support provided by the shorts may have restricted hip movement in the frontal plane, decreasing step width in those whose hips were abducted and increasing step width in those with a more adducted posture. Similarly, the support provided by the orthotic shorts may mean that less muscle strength is required to support the pelvis in the stance phase of gait. There is support for a possible sensory mechanism in that, as discussed in Section 2.4.2, joint supports have been shown to improve proprioception. The shorts might have a positive influence on feedback from the trunk or the stance hip. In support of this sensory mechanism, compression garments have been found to improve postural sway and balance in healthy athletes, older people and people with anterior-cruciate ligament deficiencies, all with compression garments that were unlikely to provide any physical support (Kuster et al., 1999; Michael et al., 2014; Woo, Davids, Liukkonen, Chow & Jaakkola, 2018).

The finding that mediolateral pelvic sway does not improve with the orthotic shorts appears, at first, to disagree with the hypotheses suggested above, that orthotic shorts might improve mediolateral stability via mechanisms of support or improved proprioception. On finding the change in step width, I had anticipated finding a decrease in mediolateral pelvic sway as well and, at first, this seemed contradictory. Data on normal values for trunk and pelvic sway in walking might explain this apparent contradiction.

Data from a number of studies suggests that healthy, non-neurologically impaired people walk with mediolateral pelvic sway in the order of 3.5 cm, which is smaller than that seen in the OSFeaMS participants (Galna, Murphy & Morris, 2013; Whittle, 1997). Brodie et al. (2016) found mediolateral pelvic sway in their healthy participants of 3.5 ± 0.6 cm and mediolateral pelvic sway in their MS participants of 6.7 ± 2.2 cm, which suggested that pelvic sway should decrease if gait became more normal. However, mediolateral pelvic sway alters with gait speed and Brodie et al. (2016) did not control for this. Data provided by Orendurff et al. (2004) demonstrated that mediolateral pelvic sway in the order of 5.4 cm is normal for people walking at a gait speed of 1.1 m/s. In other words, at a group level, the pelvic sway amplitude in the OSFeaMS participants at baseline is already in the normal range. It can be proposed that PwMS usually prioritise proximal stability, widening and varying their steps from one step to the next in order to maintain their pelvis and trunk as stable as possible. With the orthotic shorts, they are still able to maintain this relatively normal range of pelvic sway but without needing so much variability and step width distally.

Overall, the changes seen in mediolateral stability indicate that orthotic shorts might improve control and that this could be reflective of decreased falls risk. The mechanism of effect could involve either physical support or improved proprioception, resulting in more precise placement of the swing leg.

Gait variability – distal and proximal

In addition to the improvement in step width variability discussed in the previous section, there were small improvements in variability of step length and stride time with the shorts. Stride time variability improved by a small amount with the orthotic shorts and step length variability improved by a small to moderate amount with the placebo shorts. These changes may be chance occurrences due to natural variability or they may be due to improved control of gait, resulting from increased support and improved proprioception, as hypothesized above. Step length variability and stride time variability are related to falls risk in MS (Allali et al., 2016a; Kalron, 2017). Currently, stride time variability, which was lower in the orthotic shorts, is considered more reflective of falls incidence than step length variability (Allali et al., 2016b). Allali et al. (2016b) reported that fallers with MS have a mean stride time variability (CV) of 3.49 ± 2.83%, compared to non-fallers who had a stride time variability of 1.83 ± 1.14%, similar to that cited for healthy controls (Moon et al., 2016). The OSFeaMS sample were within the "fallers" category, with a stride time median CV of 3.6 (IQR

1.6). This improved with the orthotic shorts to a median of 3.3 (IQR 2.0) but participants would still be classified as at risk of falling.

At first glance, changes to proximal variability appear less positive. In the orthotic shorts, there was a small to moderate increase in sway variability at the pelvis and a very small increase in sway variability at the trunk. Such changes were not seen in the placebo shorts, with no change in pelvic sway variability and a small decrease in trunk sway variability. This is potentially important, particularly because there was a large difference between the orthotic and placebo shorts, with more trunk sway variability in the orthotic pair. In addition, there were marked increases in the range of rotational movement at both the trunk and pelvis.

There are a number of possible explanations for these findings. Firstly, the orthotic shorts might be causing trunk and pelvic movement to become less controlled. This seems at odds with the improvements seen in footfall variability. A second possibility is that the increased movement range and variability seen at the trunk and pelvis might indicate improved movement control. The following sections explain this possibility, discussing the potential role of proximal variability in improving distal function and the potential role of proximal movement in compensating for poor distal function.

In comparison to the values for pelvic sway variability found by Psarakis et al. (2018), the OSFeaMS study participants were less variable. Pelvic sway variability at baseline on Visit 3 in the OSFeaMS study was 0.6 ± 0.21 cm, whereas in the Psarakis et al. (2018) study, pelvic sway variability in the healthy group was 1.25 ± 0.34 cm and for PwMS was 2.24 ± 1.06 cm. This may be because the OSFeaMS participants were walking at their preferred gait speed whereas those in the Psarakis et al. (2018) study were walking at maximal gait speed. Psarakis et al. (2018) showed that sway variability was higher when sway area was higher and that sway area was higher at faster walking speeds. Nevertheless, sway variability was relatively low at baseline in the OSFeaMS participants.

Variability in movement is traditionally seen as error, as an indicator that movement is poorly controlled or less efficient (Lockhart & Stergiou, 2013). The Dynamical Systems Theory and Ecological Dynamics are models of movement control that suggest that movement variability can be a good thing (Cavanaugh, Guskiewicz & Stergiou, 2005). An important distinction is whether variability is considered in relation to a movement goal or the means by which that goal is achieved. Variability in goal achievement is clearly not beneficial but variability in the means by which a goal is achieved may be (Lockhart & Stergiou, 2013). In terms of walking, movement goals could be considered to be foot placement, forward propulsion and maintenance of upright stance. The means by which this goal is achieved includes movement at the hips, trunk and pelvis, referred to as "proximal control" in the language of neurological physiotherapy (Lennon & Ashburn, 2000). Early research that informed the models of dynamic control demonstrated that people who are considered experts at a skill have more variability than novices in the movement patterns and postures used to achieve skilful movement. For example, expert pistol shooters move their pistol in space less than a novice does but experts have more variability at the shoulder, elbow and wrist (Arutyunyan, Gurfinkel & Mirsky, 1969). It is suggested that this proximal variability makes an expert more consistent in goal achievement, partly because they are more adaptable in how they achieve their goal (Vereijken, Emmerik, Whiting & Newell, 1992). Variability is suggested to enable skilled performers to cope with small changes in task and environment (Glazier, Wheat, Pease & Bartlett, 2006). This approach suggests that an increase in trunk and pelvic sway variability in orthotic shorts might indicate more options for achieving the movement goal of stepping, whilst maintaining control over balance.

Movement variability can be measured and analysed in ways that draw out the nature of variability. Such methods highlight how movement changes from moment to moment and step to step, as opposed to simply the total variability over time, as has been measured in this thesis. The former are known as "non-linear" measures of variability and the latter as "linear" measures. Using non-linear techniques, Kaipust et al (2012) demonstrated that PwMS had a more regular stepping pattern than healthy controls. They suggested this indicated a more spinal cord driven walking pattern that was less responsive to subtle environmental changes than that seen in healthy people. Craig, Bruetsch, Lynch and Huisanga (2017a) and Huisanga et al. (2013) used a Lyapunov Exponent (LyE) to analyse acceleration data obtained from sensors on the trunk. Both research groups found that PwMS had higher LyE than healthy people did. LyE quantifies the ability of the motor system to attenuate small perturbations using small adaptations to movement (Mehdizadeh, 2018). If LyE is larger, as in MS, then small perturbations are not controlled causing loss of balance. Non-linear measures were not used in the OSFeaMS study and may require data collection over longer time periods than those used in the OSFeaMS study (Cavanaugh et al., 2005). In future research, such analyses could be used to explore movement variability and the potential influence of orthotic shorts.

Bearing in mind that increased variability of proximal movement might indicate improved adaptability, it is important to consider that PwMS might need more adaptability in their proximal movement to function than healthy, non-neurologically impaired people. PwMS have loss of muscle strength and loss of available joint range in their knees and ankles (Filli et al., 2018). To some extent, such difficulties can be compensated for by using abnormal, exaggerated movements of the trunk and pelvis (Psarakis et al, 2018; Severini et al., 2017). This appears to be the case in the OSFeaMS participants. At baseline, pelvic rotation in the transverse plane was towards the upper end of normal, compared to values cited by Lewis, Laudicina, Khuu and Loverro, (2017), Staszkiewicz, Chwała, Forczek & Laska (2012) and Whittle and Levine (1997) (see Table 6.9). This increased further in the orthotic shorts to a median of 18°, which may simply be in keeping with the increased gait speed and longer step lengths. Alternatively, increased yaw rotation may be a compensatory gait strategy. Rotation in yaw has been found to be higher in MS than in healthy, non-neurologically impaired people and higher in more disabled PwMS compared to less disabled PwMS (Severini et al., 2017). Trunk rotation in the transverse plane increased with the orthotic shorts (see Table 6.10), which would be expected to accompany the increased pelvic rotation in order to maintain trunk counter-rotation.

Pelvic movement in roll (lateral tilt) and pitch (anteroposterior tilt) were greater than normal values at baseline and increased further with the orthotic shorts (Table 6.9). Severini et al. (2017) also found increased range of pelvic pitch and roll in PwMS, with higher ranges of pelvic pitch in their more disabled participants. This suggests that these movements are compensatory mechanisms, for example, for limited hip flexion.

Severini et al. (2017) suggested that compensatory pelvic movements promoted advancement of the lower limb because they noted more pelvic movement during swing phase.

Table 6.9: Comparison of range of pelvic rotation in the OSFeaMS study to values for healthy participants in previous research.

	Whittle and Levine (1997)	Staszkiewicz et al. (2012)	Lewis et al. (2017)	OSFeaMS study – baseline on orthotic shorts testing day	OSFeaMS study – with orthotic shorts
Pelvic yaw	10.4 ± 3.22°	16.8 ± 4.43°	9.5 ± 2.9°	Median 13.1° (IQR 6.8)	Median 18.0° (IQR 7.4)
Pelvic roll	7.72 ± 2.26°	8.8 ± 2.18°	7.4 ± 2.5°	11.1 ± 3.0°	11.7 ± 3.5°
Pelvic pitch	2.79 ± 0.76°	2.2 ± 0.51°	4.3 ± 1.1°	10.5 ± 3.0°	11.5 ± 4.3°

(All values are mean ± standard deviation unless otherwise stated; OSFeaMS = Orthotic Shorts Feasibility in MS; IQR = interquartile range; yaw is rotation in the transverse plane, which approximates to left/right rotation; roll is rotation in the frontal plane, which approximates to lateral pelvic tilt; pitch is rotation in the sagittal plane, which approximates to anterior-posterior pelvic tilt.)

Table 6.10: Comparison of range trunk rotation in the OSFeaMS study to values for healthy participants and people with multiple sclerosis in previous research.

	Spain et al. (2012) – healthy participants	Spain et al. (2012) – multiple sclerosis	OSFeaMS study – baseline on orthotic shorts testing day	OSFeaMS study – with orthotic shorts
Trunk yaw	9.0 ± 0.48°	11.1 ± 0.71°	Median 14.5 (IQR 7.3)	Median 15.8 (IQR 5.6)
Trunk roll	5.9 ± 0.35°	7.5 ± 0.5°	14.3 ± 7.1°	14.2 ± 6.5°
Trunk pitch	5.1 ± 0.3°	5.2 ± 0.3°	10.3 ± 1.7°	11.4 ± 3.2°

(All values are mean ± standard deviation unless otherwise stated; OSFeaMS = Orthotic Shorts Feasibility in MS; IQR = interquartile range; yaw is rotation in the transverse plane, which approximates to left/right rotation; roll is rotation in the frontal plane, which approximates to trunk side flexion; pitch is rotation in the sagittal plane, which approximates to trunk flexion/extension.)

The potential link between increased pelvic movement and increased compensatory movement in MS suggests that compensation became more marked with the shorts. This could be seen as an advantage or a disadvantage. The shorts may have provided the stability and confidence for people to use larger and more effective compensatory strategies, thus improving the efficiency of their swing phase. Alternatively, the shorts may have restricted hip movement, meaning that the participants needed to use additional compensation in order to make up for the loss of hip movement caused by the shorts.

The findings on pelvic movement contrast with those of Serrao et al. (2017) who investigated the impact of a short Lycra suit (upper arm to lower thigh) on adults with cerebellar ataxia (*n* = 10). They found smaller ranges of pelvic rotation at baseline and decreased movement with the orthotic suit. There was a particularly marked reduction in pitch range (anteroposterior pelvic tilt).The discrepancy in findings could be related to Serrao et al. (2017) having supported trunk as well as hips and pelvis with their orthosis or may be due to the different underlying pathology impacting upon walking in very different ways.

Overall, the orthotic shorts could be changing gait variability in a manner suggested to be beneficial to gait, with decreased variability of footfall and increased variability of trunk and pelvic sway. The potential changes to the range of rotational movement at the trunk and pelvis can be interpreted as indicating an increase in compensatory gait strategies but whether this is reflective of increased or decreased function is unclear from the quantitative data alone.

Dual task cost

Small to moderate decreases in dual task cost of cognition on walking speed were seen in both pairs of shorts. The impact of the placebo shorts on cognitive performance whilst walking was moderate, suggesting that walking required less cognitive effort whilst wearing the placebo shorts. There was moderate improvement over the course of the study in dual task cost, suggestive of a training effect with repeated assessment. This means that direct comparison between the placebo and orthotic shorts is likely not to be valid.

Dual task cost is extremely important in MS. It is associated with falls incidence (Etemadi, 2017; Wajda et al. 2013) and is known to impact quality of life (Castelli et al., 2016). It is believed to be challenging in PwMS partly because of the increased cognitive effort that walking requires in the presence of either slow or inaccurate sensory feedback, poor muscle control or both. Gait speed always involves higher-order cognitive systems in its control (Al-Yahya et al., 2011) but walking may be a more cognitively driven task in MS. Specific neurological damage to the cortical centres involved in controlling walking and cognition would also exacerbate problems with dual tasking (Annweiler et al., 2013). However, reliability of dual task cost assessments may be unacceptably low (ICC = 0.45, Decavel et al., 2019) and it is unclear how much change might be required to dual task cost in order to impact falls risk (Leone et al., 2015). Therefore, it is difficult to determine whether the possible changes seen with the shorts might be clinically important.

A recent feasibility study for a 12-week exercise programme designed to improve dual task cost in PwMS showed improvements in walking speed in the dual task condition but no change to cognitive ability whilst walking (Sosnoff et al., 2017). This suggests that if the changes suggested in the OSFeaMS study are valid, then orthotic shorts might contribute something over and above a training programme.

Overall, the OSFeaMS study results suggest that it may be possible to change the dual task cost of cognition on walking and improve cognitive function whilst walking, however, the poor reliability of the assessment means that these findings are particularly uncertain in comparison to the other areas studied in the OSFeaMS study.

6.3.8 Strengths and limitations of the OSFeaMS quantitative study

The counterbalancing in the crossover study was not effective, more participants experienced the orthotic shorts first and there was an improvement in “no shorts” ability over the course of the study. This may have influenced the evaluation of the relative effectiveness of the two pairs of shorts.

Change in the MSWS-12 may have been particularly sensitive to an order-effect because changes in the MSWS-12 with an orthotic intervention might be relatively short-lived, as found in a small study on FES (van der Linden, Hooper, Cowan, Weller & Mercer, 2014). Perceived walking might feel much improved in response to a novel intervention but this response might fade over time, in which case the change in MSWS-12 and ABC-UK would have been more striking for the orthotic shorts. The self-report measures may have been influenced by the context in which they were collected; baseline and the final assessment were assessed in the university but assessments after each home trial were done in the participants' homes. No previous studies have found an impact of context on these specific self-report measures but it is a recognised phenomenon (Lucas, Oishi & Diener, 2016).

The change to gait speed and participant perceived walking ability over the course of the study means that the comparison between the orthotic and placebo shorts may have been impacted upon by baseline changes on these and other measures. For example, step width was larger at baseline on the day the orthotic shorts were assessed, which was the first day for 10 participants. This may have improved prior to the second testing day, meaning that the impact of the second pair of shorts trialled was less obvious. In addition, blinding of the assessors and participants was focussed on the distinction between the orthotic and placebo shorts, yet, most of the findings reported in this chapter relate to the within-day changes for "shorts off" compared to "shorts on". I had chosen to test the "no shorts" condition first every time, so that any impact of the shorts would not be exaggerated as participants fatigued and because Ding et al. (2015) recommended assessing participants at the end of each washout period to determine whether there had been any carryover from the previous treatment phase. However, this meant that there was no attempt to blind the assessor for the within-day comparisons. There may have been a performance bias related to the within-day changes as well. Nevertheless, other than the T25FW and dual task cost assessment, technology provided data with little scope for conscious or subconscious manipulation by the assessor and many of the changes described, such as gait variability, step width and dual task cost, are unlikely to improve purely in response to increased participant effort.

Regarding the findings around the impact of the shorts on spatiotemporal gait parameters and trunk and pelvic movement, there are a number of important limitations. Firstly, data on spatio-temporal gait parameters and trunk and pelvis stability were assessed on different walks with slightly differing instructions to the participants. Thus, the inter-relationship between proximal and distal control cannot be fully explored. The analysis of trunk and pelvic movement has only included so-called “linear analyses”, looking at central tendency and variability, whereas a more detailed approach using “non-linear analyses” may have provided more insight into the step-to-step variability of movement (Craig et al, 2017; Huisanga et al, 2018)). As explained in the “Distal and proximal variability” section of the Discussion, such analyses are important to fully explore whether variability is likely to indicate a poorly controlled, unstable movement system or an efficient and adaptable movement system. Finally, none of the data presented here has investigated symmetry of movement and whether the shorts increase or decrease symmetry. Many PwMS have an asymmetrical presentation (Filli et al. 2018) and exploration of the impact on symmetry may provide useful insight into the issue of whether orthotic shorts increase or decrease compensatory mechanisms in movement. This is suggested as an important area for future investigation.

A key strength of the study was the range of measures chosen both for objective and self-report assessments, enabling insight into outcome measurement choices for a future study. In addition, the participants presented with different abilities and difficulties, meaning that assessment of the impact of the shorts on a range of PwMS was possible.

In retrospect, many of the changes seen with the shorts were related to falls risk or fear of falling and therefore, inclusion of a specific self-report measure such as the Falls Efficacy Scale-International (FES-I, Appendix 8, Jackson & Taylor, 2016) would have been valuable. To determine how relevant falls risk is to the impact of the shorts, it would have been helpful to know how “at risk of falling” each participant was. This may have enabled exploration around the proportions of "fallers" as opposed to "non-fallers" in whom the shorts appeared to be helpful. The most valid means of assessing falls risk in MS has been found to be to ask participants if they have fallen in the last year (Cameron, Thielman, Mazumder & Bourdette, 2013). This would have been a simple addition to the initial assessment.

A final limitation is that the falls diary used is not considered to be a valid measure of falls risk, if used over only a short period (Coote et al., 2014). However, it was included in this feasibility study in order to assess the burden of including such a diary in a later study. The burden associated with the falls diary was low and the finding that other variables linked to falling improved with the shorts confirms that a falls diary would be an important measure in a longer duration trial of the shorts.

6.4 Implications for this thesis

The data presented in this chapter provides a significant contribution to the thesis in that it demonstrates that orthotic shorts are indeed feasible in PwMS. Specifically, the number of participants who chose to keep the shorts after the trial was high. There may be important psychological effects in improving participants' confidence in their own walking ability. In addition, changes seen with the objective measures suggest improved mediolateral stability and decreased variability of footfall, which may be reflective of decreased falls risk. Movement of the trunk and pelvis appears to increase in both range and variability with the orthotic shorts, which may be indicative of improved adaptability in gait, an increased use of compensatory gait strategies or both. The study has informed the choice of outcome measures for future research both in orthotic shorts and more widely in fabric orthosis studies.

6.5 Conclusion

This chapter has presented and discussed findings from the quantitative data of the OSFeaMS study, namely recruitment and retention, quantitative assessment of acceptability, pressure applied by the shorts and the impact of the shorts on objective measures of walking ability and self-report measures.

The quantitative element of the OSFeaMS study has demonstrated potential for orthotic shorts to be effective in improving walking ability in PwMS, specifically around gait speed, mediolateral stability and footfall variability. In comparison, the placebo shorts did not improve gait speed or mediolateral stability but may have had positive effects on dual task cost, cognitive ability whilst walking and step length variability. In the self-report measures, there were large improvements seen in participant perceived walking ability and balance confidence for both orthotic and placebo shorts.

The subsequent chapter will report the qualitative data on acceptability and participant feedback on the OSFeaMS study design. The key themes of how orthotic shorts might affect walking in MS and how research into fabric orthoses needs to be designed will be continued in Chapter 8.

Chapter 7: The Orthotic Shorts Feasibility in MS study - qualitative findings

Summary

This chapter reports the qualitative results of the Orthotic Shorts Feasibility in MS (OSFeaMS) study and a preliminary discussion of the findings around acceptability. The methodology and methods are reported in Chapter 5 and quantitative findings in Chapter 6. Chapter 8 reports integration of the qualitative and quantitative findings from the OSFeaMS study, where these relate to acceptability, perceived effect of the shorts on walking and balance and recommendations for future research.

7.1 Aims and objectives

The qualitative aspect of the OSFeaMS study aimed to determine the acceptability of the orthotic shorts as an intervention for improving walking in PwMS and to gain feedback on study processes in order to inform a future trial.

The objectives for the qualitative aspect of the study were:

1. To investigate the acceptability of orthotic shorts
2. To gain participant feedback on research methods used.

7.2 Results

A description of individual participants and their shorts is provided in Section 5.3.2. Qualitative methods are reported in Sections 5.3.9 and 5.3.10 respectively. In brief, data collection utilised semi-structured interviews at the first and last visit and data was analysed using a Framework Analysis method (see Appendix 19 for the final thematic framework). One participant dropped out during the study, therefore, data from 30 interviews (15 participants) were analysed.

Findings are presented in two themes, which relate directly to the study objectives: “understanding acceptability of orthotic shorts” and “designing a future research study” (Table 7.1). Within the “understanding acceptability” theme, the first five subthemes are constructs defined within the Theoretical Framework of Acceptability (Sekhon et al., 2018), described previously in Section 3.6.2 (Table 3.3).

Table 7.1: an overview of the themes and subthemes for qualitative aspect of the OSFeaMS study

Themes	Sub-themes
Understanding acceptability	Intervention coherence - participants' thoughts about how the shorts might work and how well this fits with their understanding of their own difficulties
	Ethicality - the extent to which the shorts fitted with participants' value systems
	Perceived effectiveness - the extent to which the shorts were perceived to achieve their purpose, defined as reclaiming my body, reclaiming autonomy and managing self-image.
	Burden - the perceived amount of effort required to use the shorts
	Affective attitude - how participants felt about the shorts
	Future intentions for shorts use
Designing a future research study	Feedback on the use of placebo shorts
	Choosing measures for a future trial
	Improving collaborative practice in a research context
	Burden associated with the crossover design

7.2.1 Understanding acceptability of orthotic shorts

The intervention coherence and ethicality subthemes are explained first, since these elements reflect how acceptable participants anticipated the shorts might be, prior to using them. The remaining subthemes reflect participants' experiences with the shorts. These are perceived effectiveness, burden and affective attitude. Finally, I present participants' stated intentions for using the shorts in the future and use the Theoretical Framework of Acceptability to examine individual participant's acceptability of the shorts.

Intervention coherence

This subtheme explored the extent to which participants understood the shorts as an intervention and how they might work. This was explored in the initial interviews by asking participants how they expected they might respond to the shorts. Most participants used terms such as *"stabilise"*, *"balance"*, *"posture"* and *"strength"* to explain how they felt the shorts might help. Some related this to the concept of core stability, which was clearly a familiar concept to all participants. Erica was the only participant who explained that she was not really expecting the shorts would help her.

"Everything's about building your core, there's not much else." (Frank)

"I suppose, if I am brutally honest, I can't imagine what it will change." (Erica)

Prior to hearing about the current study, three participants (Melissa, Natalie and Oliver) had purchased shorts or supportive underwear specifically to see if they helped their movement. They gave only brief explanations for why this had seemed worth trying: "*To see if they make me feel better*" (Natalie) and "*I thought they might just help a little bit*" (Oliver). None of these participants felt any benefit from these previously purchased garments but the fact that they had thought to trial them indicates that they had already considered this to be an intervention that might address their perceived difficulties.

Following the shorts trial, most participants related the impact of the shorts to the same stabilising concept. Some participants mentioned an improvement in feedback with the shorts, which they felt improved their awareness of posture or movement. Four participants expressed some uncertainty around how the shorts might have worked. Kathy and Oliver had been puzzled that the placebo shorts had seemed to work, given they had assumed that the shorts would need to be tight in order to have an effect. In addition, Oliver was puzzled that it took one week of wear before he felt that either pair of shorts was helping him. Both Frank and Natalie expressed uncertainty around whether it was important to remain consciously aware of wearing the shorts in order for them to be effective.

"My legs felt… which sounds daft because I had forgotten I were wearing them… but my legs felt better." (Natalie)

"A couple of days of wearing them all day, you almost forget you are wearing them, which in one sense you think, "oh yeah, that's good". But in another sense you're thinking "oh, I want to know"." (Frank)

Overall, it can be seen that the shorts intervention made sense to the participants who anticipated the shorts would provide a stabilising force around their core, which would improve their control. Once they had experienced the shorts, most still thought of the shorts in this way but some questioned the mechanism of shorts that they were barely aware of.

Ethicality

This subtheme explored the extent to which the shorts intervention fitted with individuals' value systems. Four elements indicate value systems related to acceptability of the shorts. These were: the importance of taking opportunities to help yourself; the perceived value of exercise; the preference for simple, low risk, non-pharmaceutical approaches to managing MS and the desire for assistance that is not visible to others.

The importance of "taking opportunities" came across in some of the initial interviews where participants explained that they take any opportunity to get involved in research and to explore new treatments that might help them. At the final interview, participants were asked what advice they might give other PwMS about whether the shorts might be helpful and all felt that everyone with MS should try anything that might possibly help. Jon, Oliver and Kathy particularly valued interventions they could be responsible for themselves.

"Anything's worth a go. And everybody's different aren't they?" (Dawn)

"I've tried everything else and if you don't do the trial and error you're not going to come to any result." (Ingrid)

"I am open. I will try anything…if maybe it will improve me. I'll try anything." (Oliver)

"I am looking for things that are down to me… on a regular basis to use that I'm responsible for." (Kathy)

All participants explained the value of exercise for managing their MS and most reported being actively engaged in some form of regular exercise, either in their own home, at a therapy centre or in a gym. Ben and Erica were not regularly exercising but both seemed to feel that they ought to be. A number of participants linked the importance of exercise to how they perceived the shorts, indicating that they saw exercise as an important thing and they associated the shorts with exercise.

"That use it or lose it always plays in my head and I think… I don't want to stop moving." (Melissa)

"I used to do a gym … and… (long pause)*… it was alright but then again I got lazy because you just feel like… you know… I don't want to do anything today."* (Ben)

"Well, they're just like exercise shorts, aren't they?" (Oliver)

"The attractive thing about the shorts is that it feels like something you might wear if you were going to the gym." (Erica)

Some participants highlighted a preference for simple, low-risk, non-pharmaceutical approaches to managing MS symptoms, either because they had had negative experiences with disease modifying drugs, did not like the idea of using drugs or wanted something non-pharmaceutical in addition to their drug therapy.

"There's no… err… side-effects… nothing like that. They are just shorts and they do some kind of job." "They make you not stagger as much, not slip quite as much…and all it is… is a <u>pair of shorts</u>" (Frank)

A final element of values is the preference for assistance that is not visible to others, a desire to hide your challenges and to avoid the stigma associated with aids. For Ingrid and Melissa, this was an extremely important aspect of the shorts intervention.

"When I used my rollator, I can walk for further…and I don't have such severe back pain. I am hoping that, in a sense, the shorts are going to do that but they're underneath me skirt and nobody sees them." (Ingrid)

"Nobody's going to say, "Oh, why have you got those funky weird knickers on?" Umm… they are hidden. I like that they are hidden. I love that they are hidden." (Melissa)

Overall, the shorts intervention was in keeping with participants' values of being a low-risk, non-pharmaceutical intervention, something they associated with using exercise to manage their MS and something unobtrusive, hidden from public view. In addition, the principle of trying anything that might be of value seems to be an important influence in prompting shorts use.

Perceived effectiveness

The theme of "perceived effectiveness" included the subthemes of "reclaiming my body", "reclaiming autonomy" and "managing self-image".

Reclaiming my body

The subtheme of "reclaiming my body" explains a sense of regaining trust of and control over one's body. Table 7.2 summarises perceived positive impacts and illustrates that the shorts influenced participants in different ways. Many participants felt improved control over walking ability and some reported improvements in posture, balance, standing ability and strength.

Ben and Jon described their walking having improved because they were better able to move their legs in the direction in which they wanted them to go. Jon related most of the positive benefits of the shorts to this effect. He felt the improved ability to direct his legs meant that he could walk in a straight line without wavering, turn corners more tightly and avoid trip hazards on uneven ground. This feeling of increased ability to direct his legs was reflected in his belief that his could kick a ball more accurately with his orthotic shorts on. Other participants described difficulty controlling their walking direction and found that this improved when they wore the shorts. For example, Caroline and Kathy felt that they had less need to touch walls to help turn or keep steady when walking indoors, Melissa described bumping into furniture less with her orthotic shorts and Natalie felt that the shorts helped her walk in a straight line rather than wavering from side to side.

"There was something there to constantly remind my legs that they should be going in a particular direction whereas in the past when I've turned my head, my legs will just splay out in the direction that I am looking at." (Jon)

"*When I had those shorts on, I wasn't bumping at all. Which is amazing because quite often my hips and especially my arms, my legs, my thighs... I've usually got bruises all over my thighs… and I've not got them when I wear the shorts… at all.*" (Melissa)

"I feel like I can walk in a straight line better… because I was really bad at wobbling and… with them on I feel better." (Natalie)

Table 7.2: Summary of perceived positive impact of shorts.

How I felt with the shorts	Perceived changes to my body	Perceived improvements to function/participation /exercise routine
More confident (A,B,F,G,K,L,M,N,O) More secure (B,C,E,H,I,L) More stable (A,C,F,K,M,N,O) Less pain (A,J,K,N,O) More controlled (J,K,N,O) Less tired (C,H,K,M) Supported (C,F,G,J,K, L) Stronger (C,D,K,N,O) More aware of posture (B,C,D,K) Less wobbly or shaky (C,D,M,N) More aware of leg movement (B.K) Less achy (C,K) More aware of walking ability (D) Firmer (B) Energised (F) Empowered (M) More normal (M) Sturdier (N) Rock hard (N) Better (N)	Kept me upright/improved posture (A,B,C,E,F,K,M) Improved balance (J,K,M,O) Fewer falls or near falls (J,M,O) Reduced need for conscious control (J,K,M,O) Less sway in standing (D,K) Improved bladder control (K) Knee less likely to give way (B) A more controlled core (A)	Able to stand for longer whilst cooking (F,I,K,O), showering (K) and working (O) Squatting lower and more controlled, including getting up from the floor (K,M,O) Housework felt easier (A,K,O) Improved ability to go up and down stairs (K,O) Standing from sitting is easier (F,K) Reaching and turning more easily in standing (A,K) Can exercise for longer (M,O) Improved sitting balance on a gym ball (K) Sitting from standing is more controlled (K) Kicking a ball with more accuracy (J) Able to dance (F) Can run faster (N) Improved control in Tai Chi and Pilates (K)
	Perceived improvements to walking	
	Walking felt better, easier, faster or more fluent (C,F,K,M,O) Could walk further or for longer (F,K,M) Improved walking over uneven/messy ground (J,K,M,O) Catching or dragging feet less when walking (D, F,O) Improved ability to walk in a straight line (J,N) Legs go in the right direction when walking (B,J) Touching the wall less when walking indoors (C,K) Able to walk around corners with more control (J) Longer stride (O) Bumping into furniture less (M)	

(Initials indicate which participants made this comment.)

Other improvements to walking ability included walking further, faster and more smoothly (see Table 7.2). Frank and Oliver explained that they could lift their feet more easily with the shorts on. These men had particularly slow, effortful walking patterns compared to the other participants. Frank linked this improvement directly to an upright posture.

"I feel like I can go further when I've got them on." (Melissa)

"…a smoother walk and a quicker walk…" (Kathy)

"When you've got the shorts on and when you… straighten yourself and get yourself in the right position… because of the shorts, I feel better. That then leads you to not catch your feet as much, not drag as much and then you are able to push a bit further." (Frank)

Jon, Kathy, Melissa and Oliver reported improved ability to walk across messy or uneven surfaces or the ability to walk more confidently at night. They explained that, with the shorts, they felt less need to concentrate and were more stable on unpredictable surfaces.

"I even managed to negotiate rubbish on the floor in the dark at night that… if I had stood on it normally, I would have fell." (Kathy)

"Ground that's a bit pebbly, stony… and gravelly, which usually sort of makes me go "Oh heck" and I didn't feel like that. I didn't feel as… worried or concerned." (Melissa)

Participants who used walking aids were asked to compare the impact of the shorts to the impact of their walking aid. Kathy felt similar benefits with her rollator and her shorts and had started using her shorts as a replacement for the rollator. None of the other participants felt the shorts allowed them to manage without their walking aids. Caroline, Dawn and Frank felt they would be unable to walk without their walking aid or would feel extremely unsafe and did not offer any thoughts about how the shorts compared. Ben and Gwen felt the shorts and their walking sticks had a similar type of effect but that the walking stick was far more effective. Jon, Melissa and Oliver considered their walking aid and their shorts to be complementary. They explained that their shorts actually changed the way they moved, whereas their walking aids did not. Their walking aids had other important functions that the shorts could not achieve.

"Without the shorts… I need my wheels… With the shorts, I can get all the way round. I've tried it without my wheels and I can get all the way round." (Kathy)

"The shorts keep everything moving in the right direction whereas the pole is more for balance." (Jon)

*"They're fantastic because the crutch gives me the… it's the shield… and it's the third leg… (*the shorts*) help me walk taller and it makes me more confident with my crutch as well. Just in how I walk… not shuffling." (Melissa)*

"The walking stick gives me confidence because I know if I fall… if I stumble to one side, I can put that out. The stick doesn't help me legs to work better… it's just there as a safety barrier." (Oliver)

Many participants reported improvements in posture. These included Caroline, Kathy, Frank and Melissa who felt their walking and balance was improved. Alison, Ben and Erica did not perceive much or any change to walking and balance but simply felt *"straighter"* and more *”upright"*.

Kathy felt an improvement in her ability to control movement in standing, specifically in the placebo shorts. Examples of this included that Tai Chi felt easier, she felt she could stand for longer and turn in standing without having to step and without losing her balance. She had worn the placebo shorts in an art gallery and found she could read information posted on the wall, something that she usually found difficult. She had gone to a toilet and removed the shorts to see if this was a change in her or a result of wearing the shorts. Without the shorts, she felt her reading ability worsened. She attributed this to a reduction in sway whilst standing.

A final important aspect of "reclaiming my body" is a perceived improvement in strength. For Natalie, this was simply a feeling that her legs were stronger, sturdier and *"rock hard"*, leading to less wobbliness. Frank, Kathy, Melissa and Oliver reported that their ability to squat, get on and off the floor or move between sitting and standing was noticeably better with the shorts on. Kathy needed to squat as part of her job and believed that she could hold the position longer in her shorts with less reliance on her upper limbs. Melissa and Oliver reported an ability to do more repetitions whilst they exercised, notably in squats and lunges.

"When you've not got the shorts on, you really struggle to get up. When you've got the shorts on, there's a little bit it gives you there… straightening your trunk a little bit and then you think oh, actually, I can do that. Not every time but sometimes it does make a difference. It does help with getting up a little bit from a seated position." (Frank)

"The same exercises weren't… tiring me in the same way. I was thinking oh I could do ten more… easy." (Melissa)

Participants appeared to have taken different approaches to the home trials. Other than Kathy, all participants mentioned things they felt had not improved with the shorts (see Table 7.3). Kathy had tested the shorts in many different tasks and kept written notes on perceived differences. Conversely, at the initial interview, Natalie and Erica appeared to suggest that they were interested in seeing if the shorts improved walking speed (Erica) and balancing on one leg (Natalie). However, neither attempted these tasks during the shorts trial. Helen said that she had not taken much notice of whether she was able to move any differently in the shorts. This indicated a potential relationship between the activities participants engaged with, their curiosity around the impact of the shorts and the perceived benefits of the shorts.

There was a lot of variation in levels of activity during the study. For example, some had relatively sedentary jobs, others more active jobs. Most had regular exercise routines but others did not. During the trial, some participants experimented with activities that they would not usually engage in. For example, Jon tried running for the first time since diagnosis, Kathy tried walking in a shopping centre without her rollator, Frank went out for the evening and stayed on his feet dancing, Melissa tried walking further with her family and Oliver attempted more exercise in the gym than usual and tried walking outside whilst holding rather than leaning on his walking stick. The possible relevance of activity levels is further suggested by feedback from some of the participants. Erica suggested that she might have felt so little impact from the shorts because she was sedentary. Frank expressed his belief that the shorts have most of their effect when you are pushing yourself to do more.

"It didn't help me particularly but I am sat in an office for most of the day, so possibly if you are doing more it might help you." (Erica)

"You see I probably just don't notice. I probably just get on with everyday life." (Helen)

"When you pushed your body a little bit, you could feel the support a little bit more" (Frank)

There are two separate but related implications to the possible relationship between activity and perceptions. Firstly, it is possible that the shorts feel more beneficial when people are "pushing" themselves. Secondly, there is a feeling that some participants were more curious and more engaged in the study than others were and this influenced the perceived effectiveness of the shorts.

Table 7.3: Aspects of reclaiming my body or autonomy that did not change with the shorts

Participants	What did not change with the shorts
Alison	Walking speed; the adducted, flexed posture of her legs in standing
Ben	Balance in walking
Caroline	Walking, the need to concentrate on her walking
Dawn	The ability to carry a drink when walking
Erica	Touching walls when walking indoors, perception of control when walking downhill
Frank	No new activities were possible
Gwen	Hip pain, sitting posture
Helen	Stability, walking distance, balance, walking in a straight line., confidence walking through town, standing up for long periods
Ingrid	Back pain
Jon	Standing up from sitting or squatting
Linda	Independence
Melissa	Confidence to go outside by herself
Natalie	Getting out of a car, balance in single leg stance
Oliver	Tightness in his leg muscles

Overall, around half the participants perceived improvements in their walking with the shorts, with important benefits being improved stability, the ability to control directed leg movement and to lift your feet. Other perceived benefits included improved strength and posture and a number of participants related improvements in their walking ability, balance and strength to experiencing fewer falls or near falls.

Reclaiming my autonomy

This subtheme revolved around the ways in which the shorts helped participants with feelings of independence. With the orthotic shorts in the current study, there were examples where participants felt able to do more or to do their usual activities more easily. Alison, Frank, Kathy and Oliver felt that their ability to stand for longer led to an increased ability to cook, wash-up and work. For Alison and Kathy, other aspects of housework such as cleaning and decorating felt easier due to an improved ability to reach, to squat and to turn in standing.

"The benefits were general walking around, getting about, day-to-day stuff, working… You know, standing in the kitchen, washing and putting the pots away, things like that… umm… and general confidence. You do tend to find yourself doing more and not thinking about it." (Oliver)

An increase in confidence was the most common finding relevant to autonomy. Improved confidence increased participants' sense of freedom of choice and opportunity. Most participants stated that they felt more confident when wearing the shorts and all participants used terms such as "*secure*", "*stable*", "*controlled*", "*supporte*d" or "*strong*" to describe how they felt with the shorts (see Table 7.1). These are all terms than can be applied to both physical ability and emotional status. Alison, Ben and Melissa likened the shorts to a supportive person, saying that they felt *"looked after"* or *"held up"*.

"I don't know if they just…security wise, wearing these shorts, I felt a bit more confident in doing things. That little bit more support." (Linda)

"It felt as though someone was holding me round my waist and it felt so supportive and… reassuring." (Melissa)

Jon and Kathy explained that the change in their confidence had led to at least a partial reversal of the tendency to attempt less in their lives. Both explained that MS can destroy your sense of what you might be capable of. Trialling the shorts helped them reverse that feeling, by prompting them to experiment around what they might be able to do. Many other participants explained that the shorts had prompted them to do more than they might do usually. For some, the increased confidence was related directly to a specific function, such as walking, balance or posture. For others, it seemed to be a more holistic effect, making people "*generally confident*", *"positive"* and *"less ill"*.

"It's a strange thing with MS… you can go outside and because you know you've got it, because you know you might lose your balance… it… it can sort of destroy your confidence a little bit and create a problem. Whereas, if you've got somebody just pushing your legs back into place, then it's a mental stability enhancer." (Jon)

"It's a confidence thing… I've grown… I've thought, oh I can do that now…. so I've done it. When you've not got the shorts on, you just think "I've got MS, I can't get up". "I've got MS, I've got to sit down", "I've got MS, I'm going to struggle to do this"." (Kathy)

"I just… I think having the shorts on made me feel positive and… and more determined." (Dawn)

"I think it's given me the… umph… the… "go on"… The kick up the backside, you know to have another go, let's try a bit more." Alison)

"It gave me more confidence when I had got them on that maybe I can do… more than I would normally do." (Gwen)

Overall, autonomy improved where the shorts improved people's perception of their ability to turn, reach and squat in standing and to stand for long periods. Possibly more importantly, the shorts seemed to give a sense of confidence and empowerment, prompting participants to try activities they might previously have avoided.

Managing self-image

The subtheme of "managing self-image" explored how the shorts influenced self-image, positively and negatively. It encompassed how the appearance of one's movement influences self-image, the dilemma around using a walking aid and the direct impact of the shorts on self-image.

Caroline, Dawn and Ingrid expressed concerns about the appearance of their walking. For Caroline and Dawn, this was a key motivator for participating in the OSFeaMS study. Caroline felt that the shorts did improve the appearance of her walking; Dawn felt the appearance of her walking improved but was not sure whether this was directly because of the shorts or just her increased focus on her walking during the study.

"I mean I have a style of walk that, you know, attracts attention. When I walk into a room, people will see it. Although I know that won't change because of this study, what I would like it to do is… I won't feel as uncomfortable." (Caroline, initial interview)

"Just to walk like a normal person walks. I know I walk awful" (Dawn, initial interview)

"I think my walking looked better." (Caroline, final interview)

"When I first put me shorts on, I did feel that they were… strengthening me… helping me to be conscious of walking better, to be conscious more of being upright and conscious of lifting me legs up and walking properly." (Dawn, final interview)

Melissa felt that not only did her movement impact upon her self-image but also that her self-image influenced her movement. She explained that changes in her posture were not purely physical but associated with her feeling *"smaller*". Somehow, the support of the shorts helped reverse this.

"I don't think my posture's gone lousy because I've just got lousy posture… I think… I think… I have wanted to shrink since being diagnosed and it's made me sort of go smaller. And I think that support helps me… feel taller." (Melissa)

Dawn, Erica, Helen, Ingrid, Kathy and Melissa mentioned their feelings about walking aids and the messages that they convey to others. The impact of an aid on their self-image was a key factor in determining which aid to use and whether to use one at all. For example, Dawn, Ingrid and Kathy used wheeled rollators outdoors but felt very negative about them. Dawn described hers as *"an old granny thing"*, and Ingrid would only use hers where she was unlikely to meet people she knew. Kathy had explained that she *"absolutely hated"* her rollator and, as explained earlier, she decreased use of it as a result of wearing her shorts.

The shorts affected self-image directly due to their appearance and all the female participants in the study discussed the appearance of the shorts. All were concerned around what clothes they might wear with the shorts to avoid them showing to others. Caroline and Melissa questioned whether the shorts could be shorter and still effective, to prevent them from showing beneath a skirt. Similarly, clothing needed to be loose, to prevent the shorts from showing through. Helen felt the ridge where the shorts stopped at the toileting hole was visible beneath her clothing and might attract attention.

The impact of the shorts on self-image appeared to be related to participants' colour choices. Those who had chosen either red, dark blue or black commented very little on the appearance of the shorts. Some of those who had chosen paler colours, either beige or lilac, indicated negative perceptions of the shorts or how the shorts made them feel. This was particularly evident for Erica who explained that she had been drawn to the study partly because of the illustration on the flyer that looked like *"something you might wear if you were going to the gym"*. She chose beige so that they would be less visible under clothing than a darker colour but clearly regretted this choice, describing the colour as *"pretty disgusting"* and wearing them was *"a bit of a downer"*. Paradoxically, the same colour choice was both acceptable and unacceptable with respect to self-image. This depended upon whether appearance was judged with or without a top layer of clothing, both of which seem to be important. Similarly, Melissa felt her beige shorts looked *"clinical"* and explained that a darker colour would have fitted more with the image she was drawn to.

"I think I would probably have felt happier, psychologically, if they looked a bit more like sports shorts because they did make me feel a bit like an old granny." "(On the Information sheet) *they look very much like umm… sports shorts, don't they? Actually maybe in my head that then sends you into a kind of… "Ooh… I can be that physical person", possibly."* (Erica)

"I think I'd prefer them if they looked sporty, which I know is very false advertising but… I know there was a picture on the… umm… on the Information Sheet and they were

black and red and they looked sportier… and they looked less… less…invalid-like…" (Melissa)

Overall, the shorts had some positive effects for self-image around the appearance of walking and the ability to manage without a walking aid. For many participants, the importance of the shorts being invisible to others came across strongly and a direct impact on self-image was closely linked with colour choice.

Burden

This subtheme explored the degree of effort required to use the shorts. Most participants felt the burden of using the shorts was low. The shorts were mostly comfortable, many participants found them easy to get on and off and laundering the shorts was easy. There was burden reported by some participants around difficulty toileting, movement restriction and discomfort. An overview of burden associated with the shorts is presented in Table 7.4.

Many participants described the shorts as *"tight"* but some used more negative terms such as *"restrictive"*, *"constrictive"* and *"uncomfortable"*. Alison, Ben, Erica and Gwen felt a sensation of heaviness with the shorts. The movements felt to be most difficult or uncomfortable in the shorts involved trunk and hip flexion such as climbing stairs, bending over, using an exercise bike and sit-ups. It would appear that hip flexion felt restricted and, for Gwen, the negative impact this had on function was extremely influential on the acceptability of the shorts.

"An awful lot of muscular effort had to go into bending the hip… you know in order to lift the leg properly." (Gwen)

Other elements of burden were that, during the heat of summer, the shorts felt too hot to wear and pressure of the shorts against the stomach felt uncomfortable. Ben and Linda felt uncomfortable when they bent over and Alison, Erica and Melissa felt uncomfortable after a big meal. Alison felt that the shorts pressed upon her bladder and made her more anxious about her continence. Participants felt that the shorts were more of a bother and less relevant when they were less active. Many had simply chosen not to wear them when they were less active. Of those that wore them at rest, some found them irritating.

Table 7.4: Summary of perceived negative impacts

How the shorts felt	Changes required to wear the shorts
Tight (A,B,C,D,E,G,H,J,K) Hot (C,E,H,J,K) Heavy (A,B,E,G) Uncomfortable (E,G,L) Restrictive (E,G,H) Constrictive (A,E) Like sandbags (A) Like dumbbells (B)	Takes longer to dress (B,E,F,G,H,L) Need a belt (trousers drop down over smaller waist and silky material of the shorts) (B,F,K) Takes longer to undress to get to toilet (A,B,E) Need help to dress (B)
Activities that felt more difficult	Negative effects
Prolonged sitting (C,D,I,J,N) Lifting legs to go upstairs (A,B,G,K,L) Eating a big meal (A,E,M) Lifting legs to walk (A,H) Bending down (B,L) Using an exercise bike (A,L) Step-ups (A) Squatting - shorts nipped genitalia (J) Sit-ups - shorts rubbed skin (O)	Fabric above the waist dropped down repeatedly (B,I) Increase in ankle swelling (G) Temporary imprints on skin (A,D,L) Itchiness (L) Pressure on bladder (A) Unable to get to toilet on time (B) Perceived shortening of stride length (J) Chafing - when running with tight clothing over shorts(N) Hook and loop fasteners rubbed against waist (D)

(Initials indicate which participants made this comment.)

"I was sat watching telly in them; I started to think "Oh these are getting on my nerves a little bit"." (Natalie)

"I felt like I am sat in a pair of big knickers most of the day. Whereas if I had had a more active lifestyle maybe I would have… noticed a bit more" (Erica)

Six participants felt that it took them longer to get dressed or undressed (see Table 7.4) with the shorts. Ben required help from his wife to dress, although it was unclear whether this was his usual routine. He had one episode of urinary incontinence due to being unable to get undressed in a timely way. There was a process of familiarisation required around getting dressed and undressed. For example, Frank felt his second pair of shorts were easier to wear despite being tighter because he had learnt how best to manage the zip and the positioning of the shorts during the first home trial. Gwen and Ingrid felt it was difficult to get the shorts into the right place when they first put them on, although neither had a side zip in their shorts. Other participants, including Jon and Melissa, commented on the ease of getting dressed and undressed.

"I needed about three or four minutes at least to get them on…" (Gwen)

"No more difficult than some other clothing that I've got." (Jon)

"There was… a couple of times where it was urgent and I thought, oh no, is it going to be a problem… no, fine… because of the zip." (Melissa)

Of those participants who had chosen to have a toileting hole in their shorts, Caroline, Linda and Frank wore underwear over their shorts and kept the shorts on whilst going to the toilet. Helen and Erica chose a toileting hole but then decided it was unnecessary. Helen and Caroline expressed concerns around the challenges of keeping their shorts clean whilst using the toileting hole. Helen was concerned about getting urine on the shorts. Despite her toileting hole, Caroline removed the shorts *"to sort yourself out"* whilst she was menstruating and expressed that she would not sit on a public toilet in her shorts.

Overall, the burden of using the shorts appeared low for most participants but some found them uncomfortable, restricting to hip and trunk flexion and time consuming in getting dressed and undressed.

Affective attitude

This subtheme explored how people felt about the shorts and was informed by the opinions explained and the language used to describe the shorts. The affective responses to the shorts were varied. Seven participants explained that they liked the shorts and used only positive language to refer to them. For example, Melissa explained that they were *"fantastic"* and *"a pleasure to wear"*. In contrast, Erica and Gwen were consistent in their negative feedback and the language used to express their emotional response to the shorts, with Gwen describing them as *"bloody things… making my life hell"*.

For the other participants, affective attitude was more complex. The positive opinions they expressed were contradicted by negative language. For example, Ben explained, *"they did a lot for me*" but also used words such as *"horrible"* and *"dumbbells"*. Ingrid had provided quite positive feedback about the low burden associated with the shorts but then explained that she would change for swimming in a private area if she was wearing her shorts, so that others would not see them. She used the phrase *"spazzy shorts"*, indicating negative feelings despite her otherwise positive feedback.

Overall, it can be concluded that nearly half the participants liked the shorts, with others expressing ambivalence or dislike of the shorts' appearance or the restrictive feeling of wearing tight shorts.

Future intentions for shorts use

This subtheme described proposed future use of the shorts. All participants who kept the shorts after the study explained that they would use the shorts when they wanted to be most active, for example, walking outside, shopping, going to the gym or doing housework and would not use them when they were more sedentary. This supports the finding, reported in the "perceived effectiveness" theme, that participants felt the shorts were most beneficial and least intrusive when they were more active.

At the final interview, some participants were asked whether they envisaged the shorts as something that would only help them whilst they were wearing them or if they considered the shorts to be a treatment that induced longer lasting effects to their underlying abilities. Alison, Kathy and Natalie expressed that they already felt that they had improved over time because of wearing the shorts. Oliver, Melissa and Natalie explained that because their shorts enabled them to do more, they should get fitter over time.

"They are like… residual build up, I think." (Kathy).

"If your legs feel better, you can do more things. I imagine that's the way it would work." (Natalie)

"In a way, they would help because… they help me exercise… And when I exercise, that's improving health and fitness levels" (Melissa)

Some participants included an element of experimentation with their plans for the shorts. Alison, Jon and Kathy were planning to trial using the shorts for resistance exercises, Oliver intended to test the orthotic shorts again to learn more about how they compared to the placebo shorts and to try swimming in them to see if they improved his swimming ability. Alison and Ben were keen to trial the shorts in the summer months to see if they improved their ease of walking outdoors.

Applying the Theoretical Framework of Acceptability to determine acceptability of orthotic shorts

So far, this Results section has focussed on interpreting the Theoretical Framework of Acceptability with respect to the acceptability of orthotic shorts. Each construct has been explained and used to illustrate the issues arising with respect to the shorts. In addition, I used the framework to analyse individual responses to the shorts, categorising each individual according to whether they seemed to consider the shorts acceptable. In this respect, the construct of "ethicality" did not distinguish between individuals; most mentioned the importance of assistance that is invisible to others, all related the shorts to exercise to some extent and all expressed the importance of "trying anything" as part of living with MS. The construct of "intervention coherence" was helpful in that it highlighted Erica’s uncertainty about how the shorts might work. She was the only person who could not explain at the initial interview why she felt the shorts might work for her. Erica did not keep the shorts after the study, indicating that this construct might be an important aspect of acceptability.

In my analysis of individual's perspectives, the three remaining constructs of affective attitude, burden and perceived effectiveness were weighed up against one another to determine whether individual participants considered the shorts acceptable (see Appendix 21). Unsurprisingly, given that they chose not to keep their shorts, the analysis shows that Erica, Gwen and Helen clearly found the shorts unacceptable. Caroline, Frank, Jon, Kathy, Melissa, Natalie and Oliver clearly found the shorts acceptable, in that perceived effectiveness outweighed burden and they either said how much they liked the shorts or used only positive language in describing them. However, the views of five participants were less certain (Alison, Ben, Dawn, Ingrid and Linda). For Dawn and Linda, it was difficult to gain insight into acceptability of the shorts because they were not very forthcoming in the interviews. For Alison, Ben and Ingrid, there was quite negative language used to describe the shorts and an even weighting of burden and perceived effect indicating that although they kept their shorts after the trial and said they would continue to experiment with them, the actual acceptability may have been low. The analysis of individual's perspectives needed to include a category of "questionable acceptability". Inclusion of such a category may enable a more valid interpretation of how many people found the shorts acceptable, compared to simply counting the number of people who kept the shorts after the trial.

7.2.2 Designing a future research study

This aspect of the OSFeaMS qualitative study aimed to gain insight into the feasibility of a future fully powered trial. Specifically, I had questions about whether a placebo garment should be used in a future trial, what measures should be used in a future trial, how collaborative practice might be improved within the research context and whether the crossover design had involved too much burden for participants. Findings relevant to each of these issues are reported below.

Feedback on use of a placebo garment

In the final interview, it was explained to participants that the placebo shorts were designed as a placebo. The most important finding in relation to this was that many participants stated they already believed this was the case, based upon their general understanding of research trials. They suspected that one pair would be *"the ones"* (Dawn) and the other *"just for show"* (Jon). Gwen had thought the tighter pair might be the placebo but most had correctly identified the looser pair as the placebo pair. For some participants, this had clearly influenced their views of the looser shorts, in that they had disregarded this pair relatively early on in their experience of them. Other participants had taken at face value that they were testing two slightly different versions of the same product. Although, no participants were offended or surprised that one pair had been designed as a placebo, this finding casts doubt upon the effectiveness of a placebo comparator that most participants believed was a placebo.

The inclusion of the placebo garment enabled comparison between two pairs of shorts and this may have enriched the feedback obtained, offering insight into possible mechanisms of effect. Ben and Caroline both believed they were trialling a placebo but felt the contrast between the shorts helped them appreciate what the orthotic shorts did, over and above a close fitting layer of clothing. Jon and Melissa explained the feeling of *"reassurance"* and *"control"* they had with the placebo pair; neither felt these shorts were really changing their movement but explained a psychological benefit that still made them worth wearing.

Many participants perceived a feeling of support in the placebo shorts. For six people (Dawn, Gwen, Helen, Ingrid, Linda and Natalie) there was no difference in perceived effect between the orthotic and placebo shorts; for Oliver, the placebo was more effective. Gwen, Linda and Oliver preferred the placebo shorts because they were more comfortable and allowed more movement. Kathy could feel the difference between the two pairs of shorts and explained the perceived difference in some depth. She described the orthotic shorts as improving her strength and providing support but the placebo shorts as enabling fluidity of movement and decreased sway.

Overall, this feedback suggested that the placebo pair had not fulfilled its proposed role as a placebo but did have some positive effects and helped engage participants in exploring the effects of both pairs of shorts.

Choosing measures for a future trial

Participants gave feedback around their experience of measurement and their ideas for measurement in future trials. Important points that arose were around:

- The falls diary - participants felt "near falls" should be recorded as well as true falls as some believed that the number of near falls they experienced had decreased with the shorts. Participants had difficulty translating the definition of a “fall” provided in the diary into their real life experience of falls and near falls
- The wear diary - this was helpful to prompt reflection on the impact of the shorts but it was suggested that alternative formats should be explored for people who find handwriting difficult

- The focus on walking ability in the objective assessments was felt not to fully capture what they believed they had experienced with the shorts. They suggested that walking assessment should include walking around corners, both because the shorts were felt to have an impact upon this and because it was more reflective of normal life. Additional measures they felt might capture the impact of the shorts were trunk posture, postural sway, balance tests and strength, particularly with regard to standing up, squatting and climbing stairs.
- The dual task cost assessment was highlighted by a number of participants as the most difficult challenge in the measures used. However, some participants considered this a good thing as they recognised their difficulties with dual tasking.

Improving collaborative practice in a research context

Collaborative practice had two elements within the OSFeaMS study. The first related to decision making around the provision of the shorts themselves and the second to engaging participants as co-researchers. In terms of the provision of the shorts, a number of participants suggested that they could have been better supported with choosing the design of their shorts. They felt that photographs or examples of the shorts could have been provided to help them visualise colours and styles. This was particularly important regarding the size and appearance of the toileting hole from both a positive and negative perspective of its use and appearance.

With regard to engaging participants as co-researchers, an important point raised by a small number of participants was that they felt at the end of the trial that they might not have fully explored use of the shorts. Suggestions for improving this included provision of a tick list reminding participants of the things they ought to try with the shorts and provision of a list of the questions they might be asked in the final interview. Participants who learnt most about the shorts had tried activities with the shorts on and compared this to their performance of the same activity without the shorts. Participants, who had done exactly as requested and simply worn the shorts all day, did not learn as much or have the same depth of feedback to share.

Burden associated with the crossover design

Participants were asked whether there was anything they would change for a future trial investigating the orthotic shorts. Two related issues were raised, both of which were related to the period of time participants had been involved in the study. Five participants felt that the period of time that they had been given to trial the shorts needed to have been longer. They had wanted more time to try different activities in the shorts. Two participants specifically commented on the length of time they had been involved in the study compared to the short periods of time for which they had trialled the shorts. They were interested in the reasons for having gaps in the study protocol where they did nothing and both felt this was puzzling and burdensome.

Overall, key points raised with reference to future research design were around: (1) the placebo garment did not work as a placebo but (2) there are advantages of comparing between different shorts designs to engage participants in providing feedback, (3) the impact of the shorts should be objectively measured in a wider variety of tasks, (4) participants should be asked to try activities with and without shorts rather than simply wearing the shorts all day and (5) a longer period of time should be allowed for trialling the shorts. A key point relevant to future research and current clinical practice is to make use of photographs and examples to guide participants' choices around shorts colour and design.

7.3 Discussion

7.3.1 Summary of main findings

This qualitative investigation has shown that orthotic shorts are acceptable to PwMS. In particular, the logic behind the intervention was easily understood and fitted with participants' understanding of how their MS affected their walking and balance. The concept of "core stability" was highlighted frequently. The shorts intervention had a good fit with people's value systems. It was seen as something associated with exercise and physical activity, as a simple, low-risk intervention and as a form of physical assistance not visible to others. Around half the participants believed that the shorts improved their walking ability. Other perceived benefits included improved posture, fewer falls or near falls and improved strength. Participants reported increased confidence related to a comforting, reassuring effect of the shorts. This empowered some participants to increase their activity levels and to feel that this might enable a training effect over time. Shorts had both positive and negative impacts on self-image, with important negative effects related to the "old-lady" appearance of the beige or pastel shorts. The burden of using the shorts appeared low for most participants but a minority found them uncomfortable, restricting to hip and trunk flexion and time consuming to get on and off. There was one incident of urinary incontinence but most people found that, following a process of familiarisation, toileting was easy and the shorts were not intrusive. About half of the participants liked the shorts, but others expressed ambivalence. Where participants did not like the shorts, it appeared mainly due either to the shorts' appearance or because the restriction to hip flexion negatively affected their ability to lift their legs to walk or climb stairs. Participant feedback on research methods provided recommendations for outcome measurement and design of future research studies.

7.3.2 Discussion of main findings

Some of the important findings from this qualitative study will be discussed in Chapter 8, where qualitative findings for the following elements will be integrated with the quantitative findings: (1) perceived effect of the orthotic and placebo shorts, (2) potential training effect of the shorts and (3) feedback on outcome measurement and study design for future research. Discussion in this current chapter will focus on the concept of a "journey" of orthotic use, anticipated and experienced acceptability of the shorts, ideas for improving acceptability, strengths and limitations of the qualitative component of the OSFeaMS study and the implications of these qualitative findings for the thesis as a whole.

The journey of orthotic use

The concept of a "journey of orthotic use" was introduced in Section 3.6.2, where the similarities were highlighted between this and collaborative assessment for assistive devices (Scherer et al., 2005; Verza et al., 2006; Blomquist & Nicolau, 2004). The FabO IPA data suggested that to maximise acceptance of an orthosis, the "journey" should include: (1) collaborative assessment, (2) a positive initial response to the orthosis with an orthotic benefit being obvious to the user and relevant to their function, (3) a supported period of adaptation to users' activities and to the orthosis itself and (4) a period where routines are developed over time until the orthotic becomes a routine part of daily life. The extent to which the OSFeaMS qualitative data is consistent with this model is discussed below.

Because of the limited collaborative assessment possible in a research study, there has been limited opportunity to explore this in the OSFeaMS study. Concerning the importance of a short-term, orthotic benefit, the OSFeaMS study confirmed that this is important. Those who found the shorts acceptable had clear examples of their perceived effect. However, participants felt they required longer to understand what the shorts were doing for them. Participants felt this needed to be determined in real-life activities and contexts; the impact on walking in the research laboratory was not sufficient. This agrees with the finding reported in Section 6.2.5, that there was no relationship between the immediate perceived effect of the orthotic shorts following the laboratory trial and whether people kept the shorts at the end of the study. There was a relationship between whether people felt immediate benefit with the placebo shorts and whether they kept them after the study but our qualitative data suggests this may have been influenced by participants' beliefs that they were testing a placebo. In other words, some may have chosen not to keep the placebo shorts only because they perceived them to be a placebo.

With regard to the need for a period of adaptation and the idea of developing routines of use, there was little sign in the OSFeaMS study that participants experienced this during their two-week home trial. The finding from the FabO IPA study (Section 3.6.1) that fabric orthoses require persistence to learn how to use them, was not present in the OSFeaMS study. This could indicate that the shorts were easier to use than other fabric orthoses. Participants did explore adaptations to the shorts, as there were a number of suggestions made about how the shorts might be altered or alternative choices they wish they had made. Feedback suggested that the home trial was too short to develop routines of use.

A further indication that the home trial period was too short was the provisional nature of some participants' decision-making around longer-term use. One third of the participants were classified by the Theoretical Framework of Acceptability analysis as having "questionable acceptability". The difficulty in determining exactly how acceptable the shorts were might relate to their not feeling able to fully share negative views during the interview. However, they may not have had sufficient experience with the shorts. A number of people referred to ongoing experimentation with the shorts, indicating that they were still in the process of exploring benefits.

Overall, there may be a "journey of orthotic use" being experienced for the OSFeaMS participants and therefore, the findings are consistent with the idea, proposed in Section 3.6.2, that good practice in orthotics is similar to good practice in matching assistive devices to individuals. However, there was limited data to confirm these similarities because of the short home trial used in the OSFeaMS project. Further investigation into the concept of the orthotic "journey" would require a follow-up qualitative investigation or a longer time scale for an initial study, tracking participants' experiences as they adapt to using an orthosis.

Anticipated acceptability

For the shorts intervention, anticipated acceptability appeared strong, particularly in terms of "intervention coherence" and "ethicality". Regarding "intervention coherence", the participants mostly explained the shorts as a stabilising force around the core. The logic of a stabilising force had been introduced in the Participant Information Sheet, which included the phrases *"Support around the hips might make walking smoother and steadier"* and *"These shorts are designed to provide stability around the hips and trunk"*. Nevertheless, the word *"core"* was not used. Fox et al. (2016) suggested that there is a high level of interest in the concept of core stability in the UK in PwMS. They struggled with recruitment to their trial because they targeted PwMS who had no previous experience of either Pilates' classes or core stability exercises. Although many PwMS expressed an interest in their trial, most were ineligible due to having already used these interventions.

Core stability has been defined as *"the ability to control the position and motion of the trunk over the pelvis and leg to allow optimum production, transfer and control of force and motion to the terminal segment in integrated kinetic chain activities"* (Kibler, Press

& Sciascia, 2006, p190). In stroke, there is some evidence that trunk control is correlated with walking ability (Verheyden et al., 2004) and, more importantly, that using exercise to improve trunk control improves standing balance and mobility. However, there is insufficient evidence that the same is true in MS. There are indications that trunk stability may be worse in MS than in healthy controls (Lanzetta, Cattaneo, Pellegatta & Cardini, 2004) and that PwMS use their trunk muscles differently than a healthy sample when walking (Ketelhut, Kindred, Manago, Hebert & Rudroff, 2015). However, there is no evidence that core stability exercise can improve mobility and balance in MS. Some studies found positive effects but included aspects other than trunk training in their rehabilitation programmes (Carling, Forsberg, Gunnarsson & Nilsagård, 2017; Forsberg, von Koch & Nilsagård, 2016). Some studies found positive effects but were preliminary, small-scale trials (Freeman et al., 2010; Normann, Salvesen & Christin Arntzen, 2016) and others were poorly designed trials at high risk of bias (Bulguroglu et al., 2017; Guclu-Gunduz et al., 2014; Keser et al., 2013). Two studies found no effect of trunk training on mobility and balance in MS (Fox, Hough, Creanor, Gear and Freeman, 2016; Sosnoff et al., 2014). The qualitative findings of the OSFeaMS study do agree with qualitative findings around the experience of core stability exercises (Carling, Nilsagård & Forsberg, 2018). For example, Carling et al. (2018) reported an improved bodily confidence and similar functional improvements to those reported by the OSFeaMS participants. These included standing for longer; a steadier, straighter walk; fewer falls and decreased use of walking aids. Despite the lack of evidence of effectiveness, this body of research indicates that PwMS perceive core stability to be important, that interventions addressing core stability are worthy of further investigation and that the effect of the orthotic shorts could be similar to that achieved with exercise.

Regarding the "ethicality" of the orthotic shorts, participants considered the shorts as something associated with exercise and physical activity, as a simple, low-risk intervention and a form of physical assistance not visible to others. The importance of having physical assistance that is not visible to others is unsurprising. It is consistent with the idea that there is a stigma attached to assistive devices, discussed in Section 3.6.2. Similarly, it is unsurprising that people are attracted by a low-risk intervention.

The finding that participants associated the shorts with exercise and physical activity was not anticipated. The perceived link between orthotic shorts and exercise is likely to have a positively influence acceptability of the shorts in the wider population of PwMS because both qualitative and quantitative research indicates that PwMS believe exercise and physical activity are good for them (Ferrier, Dunlop & Blanchard, 2010; Kasser, 2009; Stennett, De Souza & Norris, 2018).

Experienced acceptability - perceived effectiveness

The perceived effect of the shorts on walking ability and balance will be discussed in Chapter 8 in combination with the quantitative findings. This section includes a preliminary discussion around confidence, posture and strength.

Confidence

Improved confidence was the most commonly described effect of the shorts. Increased confidence has been reported in qualitative studies investigating other lower-limb orthotics in both MS (Bulley et al., 2015; Swinnen et al., 2018) and stroke (Wilkie et al., 2012). In these studies, independence and physical activity were believed to improve because of increased confidence. In addition, improved confidence has been reported in other fabric orthosis studies. For example, Stone (2014) reported improved confidence with socks and gloves in relation to better joint stability and improved participation; Miller et al. (2016) reported improved confidence with their orthotic sleeves for PwMS with ataxia. In studies with children using fabric orthoses (Edmundson et al., 1999; Matthews et al., 2009), parents and carers reported improved confidence in their children, though it was unclear whether this was expressed by the children themselves or deduced from observation of mood, activity or movement.

The Oxford English Dictionary defines "confidence" as *"the feeling or belief that one can have faith in or rely on someone or something"* and defines "self-confidence" as *"a feeling of trust in one's abilities, qualities and judgement"* (Mcarthur, Lam-Mcarthur & Fontaine, 2018). Logan et al. (2014) explored the concept of confidence as part of their study into outdoor mobility in stroke survivors. Their exploration included similar elements to those highlighted by the participants of the OSFeaMS study. Logan et al.

(2014) suggested that confidence is multi-faceted. Confidence was damaged by fear (of another stroke, of social interaction and of going outside) and linked to decreased engagement with activities, loss of meaningful roles and the feeling of having become a lesser person. Confidence could be strengthened by regaining meaningful social roles or replacing old roles with new ones. In addition, confidence was strengthened by positive reinforcement from others and mastery of skills. Mastering one task meant people were more likely to attempt the next challenge. They suggested that changes in confidence in one realm were transferred to another. For example, if someone felt more confident in their role within their own family, this spilled over into their confidence outside the home. In the OSFeaMS participants, the fears and skills discussed were different but there are similarities. For example, confidence was not specific to an individual task or function but carried over from one domain into another. As Oliver explained, *"You do tend to find yourself doing more and not thinking about it."* In addition, confidence was related to fear. OSFeaMS participants described fear of uneven ground, of other's negative perceptions of them and of their decreasing abilities to fulfil their work and family roles. With each of these examples, for at least some participants, the shorts decreased these fears and improved confidence.

One constituent aspect of self-confidence is self-efficacy. Whereas self-confidence includes self-esteem and confidence in one's judgement, self-efficacy is specifically concerned with confidence in one's abilities to perform certain actions (Bandura, 1998). Unlike confidence, self-efficacy is specific to particular actions and the most relevant example in this study is exercise self-efficacy. OSFeaMS research participants described feeling empowered to try more and do more than they would usually do and for Melissa, Kathy, Oliver and Natalie, this was explained specifically in relation to exercise. Exercise self-efficacy is important, as higher levels are associated with increased physical activity (Ferrier et al., 2010; Motl & Snook, 2008b). Low exercise self-efficacy in MS is linked to the feelings of distrust of one's MS-affected body and an uncertainty about when or to what extent symptoms might prevent activity (Backus, 2016; Moffat & Paul, 2018). As explained earlier, PwMS believe that exercise is good for them (Ferrier, Dunlop & Blanchard, 2010; Kasser, 2009; Stennett, De Souza &

Norris, 2018) and there is good evidence that it is (Amatya, Khan & Galea, 2019). However, low levels of engagement with exercise and physical activity are common (Motl, 2010). Thus, if orthotic shorts can positively influence exercise self-efficacy for PwMS, then this could have considerable benefit for wider health.

OSFeaMS qualitative data explained that improved confidence was linked with a feeling of security, stability, strength and support. Three participants described that being enveloped by the shorts felt like having a supportive person holding you up, hugging you or looking after you. This was reported by two participants in the FabO IPA study, who both explained that their fabric orthoses felt like their physiotherapist. Such a finding has not been reported in qualitative studies investigating other lower limb orthotics in neurology (Bulley et al., 2015; Swinnen et al., 2018; Wilkie et al., 2012), suggesting that this feeling of comfort, security and emotional support may be specific to fabric orthoses.

An alternative way of interpreting the empowering effect of the shorts is that users feel better and are more likely to try new things, despite their lack of belief in themselves. Their self-confidence may be no different but they attempt more because of confidence in the shorts. In some ways, the distinction is unimportant because in either case, users feel better and do more. However, the concept of faith in an external device rather than oneself has been explicitly investigated by Mothes et al. (2017). They investigated the effect of compression garments on healthy participants' engagement with exercise. They measured exercise self-efficacy in a large group of participants and classified them according to whether they had "low" or "high" exercise self-efficacy. Both these groups were issued with compression garments and were divided into subgroups, each given different information regarding what they could expect from the compression garments. The high self-efficacy group did not change their exercise performance in response to the compression garments, no matter what they were told about their effect. However, those with low self-efficacy who were told that the compression garments would improve their ability did indeed exercise harder and achieve more with the compression garments on. Mothes et al. (2017) concluded that people with low self-efficacy benefit the most from using external devices. It is as though they show faith in the external device that they do not have in themselves. This same psychosocial effect may occur with the orthotic shorts and would be important because as stated earlier, exercise self-efficacy tends to be low in MS.

The potential effect of the orthotic shorts on confidence and activity levels is supported by the finding that participants who found the shorts acceptable reported more activity during their home trials. However, this may have been because participants who were more engaged with the trial and more curious about the shorts put more effort into investigating their effects. Having put this effort in, they had more examples of activities they believed were improved with the shorts and thus greater perceived effectiveness.

Posture

The perception of improved posture was reported in the OSFeaMS study and some participants recommended measurement of posture in future studies. Postural changes were also reported by participants in the FabO IPA study and in previous fabric orthosis studies (Blair et al., 1995; Flanagan et al., 2009), although no studies have measured posture. Postural changes could be due to increased support or improved feedback on body position. As described in Section 2.4.2, studies on joint supports and compression garments indicate improved proprioception, so there is some evidence for this mechanism. Data in the OSFeaMS qualitative study suggests both proprioceptive and support mechanisms, possibly to different extents in different people. Caroline attributed most of the effect of the shorts to improved feedback reminding her to straighten up. Frank felt the shorts supported him, enabling him to achieve a posture he could not achieve without them. Whether shorts might increase support or provide sensory feedback prompting increased activity is potentially important because of the debate around whether supportive orthoses decrease muscle activity and, therefore, strength in the longer-term. Research could distinguish between these by studying long-term effects of orthotics or using electromyography to investigate short-term changes in muscle activity (Geboers, Drost, Spaans, Kuipers & Seelen, 2002). It is possible that increased support does not necessarily decrease muscle activity because, as discussed in Section 2.6.6, Maguire et al. (2010) found that orthotic TheraTogs^{TM} shorts provided support and increased muscle activity.

Strength

The perceived positive effect on strength had two elements to it. Firstly, a number of participants felt improved ability to control hip extension in functions such as sitting down, standing up and squatting. Secondly, one participant described her legs as stronger, sturdier and *"rock hard"*. The feeling of *"rock hard"* legs might reflect the more solid feel of muscles when they are compressed by tight clothing. This can feel similar to muscles that are well toned, providing an impression of strength.

As reported in Section 2.4.2, previous research into the effect of compression garments on strength has been inconclusive, with no improvement in strength or power seen across multiple studies and only a suggestion that they might maintain repeated performances of high powered dynamic actions, which is unlike the effects described by our participants. A more likely mechanism is that the orthotic shorts support hip extension by virtue of their grip on the body and their elasticity. This is in agreement with the finding that hip flexion felt more difficult for some OSFeaMS participants. Similarly, Doan et al. (2003) assessed the impact of compression shorts on a mannequin and found an extension torque in hip flexion and a flexor torque in hip extension.

Experienced acceptability - burden

Overall, burden associated with the shorts was found to be low but there were some disadvantages. The disadvantages of feeling hot in hot weather, movement restriction and worries about the appearance of the orthoses were similar to those found in previous qualitative studies (Miller et al., 2016; Stone, 2014).

Comparing the OSFeaMS findings to user feedback from other studies, there are indications that fabric orthoses vary considerably in fit, design and pressure applied. For example, Nicholson et al. (2001) described problems with fit, discomfort, dressing, restricted circulation and friction sores. Miller et al. (2016) reported problems with both restricted circulation and too loose a fit. Clearly, the individual design and fit of a fabric orthosis has a significant influence on burden.

In the OSFeaMS study, many of the negative comments about tightness and movement restriction came from participants whose shorts exerted a pressure of 10 - 13mmHg (Alison, Ben, Gwen, Linda and Oliver) and many of the participants who found the shorts comfortable had a looser fit (Dawn, Helen, Kathy, Jon, Melissa, Natalie). It should be noted that the error of measurement in the pressure-measuring device is 1.3 ± 0.9mm Hg (Brophy-Williams et al., 2014) so recommendations for the tightness of the shorts cannot be very precise. However, a fit greater than 10 or 11 mmHg may be poorly accepted and unnecessary for achieving a perceived effect. As discussed in Section 6.3.3, these pressures are significantly lower those reported in fabric orthoses for children with cerebral palsy (Shaari et al., 2018).

Acceptability - using the Theoretical Framework of Acceptability to weigh up the pros and cons of orthotic use

When the Theoretical Framework of Acceptability was used to analyse acceptability for each individual, seven participants found the shorts acceptable in every regard, whereas eight participants had both positive and negative views on different elements of acceptability. Of these eight, three found the shorts unacceptable and these participants chose not to keep either pair of shorts after the trial. Five of the eight were classified as having "questionable acceptability" because they reported both positive and negative aspects. All five chose to keep either the orthotic pair or both pairs of shorts after the study. Further validation of this categorisation can be seen by comparing to the wear times recorded in the participants' wear diaries. There was good agreement between the Theoretical Framework of Acceptability informed assessment of acceptability for individuals and their wear times (see Table 7.6). The median wear times were 91.5% and 99.5% for the "acceptable" group, 59% and 61% for the "questionable acceptability" group and 0% and 72% for the "unacceptable" group for orthotic and placebo shorts respectively. This supports the use of the Theoretical Framework of Acceptability as a means of analysing individual responses to a healthcare intervention.

Table 7.6: Comparison between shorts acceptability as determined by wear times and the Theoretical Framework of Acceptability

Participants classified according to the Theoretical Framework of Acceptability		Wear times		Shorts kept after the study
		Orthotic	Placebo	
Participants who found shorts acceptable	Caroline	86%	100%	Kept orthotic
	Frank	100%	100%	Kept orthotic
	Jon	82%	103%	Kept both pairs
	Kathy	91%	82%	Kept both pairs
	Melissa	92%	94%	Kept both pairs
	Natalie	99%	99%	Kept both pairs
	Oliver	70%	63%	Kept both pairs
Participants who found shorts unacceptable	Erica	0%	57%	Kept neither pair
	Gwen	0%	72%	Kept neither pair
	Helen	69%	76%	Kept neither pair
Participants for whom acceptability was questionable	Alison	43%	46%	Kept orthotic
	Ben	59%	61%	Kept orthotic
	Dawn	40%	29%	Kept both pairs
	Ingrid	63%	73%	Kept both pairs
	Linda	79%	95%	Kept both pairs

(Wear times are expressed as the median number of hours worn per day as a percentage of the 14 hours a day recommended.)

Using the Theoretical Framework of Acceptability to interpret individual acceptability involved weighing up the positive aspects of use against the negative aspects, in particular, the perceived effectiveness against the perceived burden. This process felt similar to that described by the participants in the FabO IPA study in the "Compromising and Adapting" theme (Section 3.5.3). Participants in the OSFeaMS study hinted at this "weighing up" process with Alison referring directly to a *"Catch 22"* because her shorts made her trunk felt better but restricted her leg movement. The "weighing up" process could be an essential aspect of determining ongoing use of a medical device and the Theoretical Framework of Acceptability may explicate the process that participants are going through as they consciously or subconsciously decide whether to continue to use their orthotic.

Qualitative findings indicate that using the Theoretical Framework of Acceptability to investigate anticipated acceptability at an initial assessment may predict use of the shorts. Only four participants chose not to keep the shorts so there is limited data to draw upon. However, for Erica, it was clear in the initial interview that her anticipated acceptability was low. This supports use of the Theoretical Framework of Acceptability to assess anticipated acceptability in research and clinical practice.

It is important to note that the number of participants who kept the orthotic shorts (75%) after the trial is significantly greater than the number who found the shorts acceptable according to the Theoretical Framework of Acceptability analysis (47%). As reported in Section 6.3.5, most previous research into acceptability of fabric orthoses judged acceptability by reporting how many kept the orthotic after the initial trial. It is possible that acceptability has been exaggerated in such studies.

Developing orthotic shorts to improve acceptability

Sekhon et al. (2018) have suggested that the Theoretical Framework of Acceptability should be used to develop acceptability of interventions and there are three key areas suggested by this data for developing the shorts. These are the appearance of the shorts and their compatibility with self-image, the size of the toileting hole and the restriction to hip and trunk flexion. In terms of each of these issues, it is important to note that there was no consensus around these issues and what one person expressed as a problem another found no issue with at all. Developing acceptability needs to consider not only potential adaptations to design but also very importantly, the information and advice provided to individuals to inform their choices.

Challenges around the appearance of the shorts related very closely to people's perceptions of the shorts either as underwear or as sports shorts. Participants with paler coloured shorts described them using terms associated with underwear (*"knickers", "sexiest", "corset"*), whereas, they felt they looked like sports shorts when manufactured from more than one colour, bold colours or black. The paler coloured underwear style received comments that were more negative. Therefore, exploring colour combinations that are considered attractive would be an important development. In terms of informing people's choices, it is already usual practice for the orthotics company to discuss which colours are more or less likely to show beneath clothing. This study suggests that it may be useful to discuss whether people envisage sports shorts or supportive underwear as being more in keeping with their self-image. Additional discussion around the visibility of the shorts under different styles of clothing may be important. Under jeans or loose fitting trousers with a belt, participants agreed the shorts were not visible. However, under close fitting trousers, skirts and tights, the shorts were visible. Thus, potential users ought to consider whether they would be willing to alter their clothing choices to use orthotic shorts. Shortening shorts to make them less visible with a skirt might be considered, although this could negatively influence the physical and psychosocial effects reported in the OSFeaMS study.

Although only highlighted by one male participant, the design of the toileting hole could be reviewed. In particular, because urinary urgency and urge incontinence are the most common challenges to continence in MS (Akkoc et al., 2016), the toileting hole could be made smaller for men, so that it allows easy urination but not defecation. This could improve the appearance and the support provided by the shorts.

An important element of burden was the resistance to hip and trunk flexion. This was likely due to the shorts resisting flexion and was probably the same force that improved control in squatting, sitting down and standing up. In other words, the shorts are designed to encourage an extended hip posture, thus they prevented people from collapsing towards flexion as they squatted and sat down but resisted active hip flexion for walking and stairs. Advances in material science have led to the invention of electroactive polymers (Mirfakhrai, Madden & Baughman, 2007), which can change their elasticity and shape in response to electricity and are already being used to develop "smart" socks and smart trousers with application for assistive technology (Green, 2018). Such developments would allow changes in elasticity depending upon the activity the user was engaged in and even provide reciprocal assistance to flexion and extension as people walked. As a development for the future, this idea is attractive. In the meantime, it may be possible to determine which activity a potential user finds more difficult and, for those whose greater challenge is hip flexion, provide orthotic shorts that are less restrictive or that assist flexion, similar to a hip flexion assistance device (Carda et al., 2012; Swinnen et al., 2018). Obviously, such an orthotic might have a negative effect on posture in standing but this option may be worthy of exploration and may result in an orthotic far more acceptable than existing designs of hip flexion assistance device.

7.3.3 Strengths and limitations of the OSFeaMS qualitative study

The most important strength of this study was the use of the Theoretical Framework of Acceptability. This provided an in-depth exploration of acceptability. Inclusion of the initial interview allowed exploration of expectations and hopes, as well as a clear record of participants' activities and challenges.

The study was limited by the fact that the Theoretical Framework of Acceptability was not used to inform the topic guides, meaning that some important aspects of acceptability were not directly queried. For example, although affective attitude was determined from what was said, it would have been helpful to include the simple question "How did you feel about the shorts?" Similarly, Sekhon et al. (2017) recommended assessing anticipated self-efficacy as one aspect of anticipated acceptability. Had I been aware of the Theoretical Framework of Acceptability from the start of the study, the initial interview would have included the question "What, if any, challenges do you think you might face in using these shorts?" Without a framework to guide the interviews, the topic guide included aspects that turned out not to be closely related to acceptability, such as use of previous orthotics and the degree to which participants’ impairments influenced their independence and participation. The initial interview would have been better focussed if informed by the Theoretical Framework of Acceptability.

Further limitations are that, as the researcher leading the study, I conducted the interviews myself. Therefore, social desirability bias may have inhibited participants from provided full and honest responses, particularly around negative perceptions of the shorts. Participant validation was not performed, therefore, participants had no opportunity to check the validity of the findings presented here. Finally, the home trial period did not appear to be long enough for a full exploration of acceptability.

7.3.4 Reflections on the influence of the researcher

My impact is likely to have been considerable in this study, from several perspectives. Participants were all aware that I was the principal researcher. I tried to present the study aims in an objective, balanced manner but there were indications that they thought of the shorts as mine (*"Nicky's Knickers"*). During the seven face-to-face contacts, I built a therapeutic relationship with the participants, which may have made them reticent to give negative feedback. Three of the participants made their positive feelings about the shorts very clear during the study and before the final interview. They expressed such delight with the shorts that my non-verbal behaviour would have mirrored theirs and, thus, encouraged further positive feedback. However, these influences may not have influenced the overall findings of the study because mostly the participants refrained from giving feedback during the study. I approached most of the final interviews with little insight into the participants' perspectives.

During data analysis, I became aware that, although I had started out with a balanced opinion on whether the shorts might be helpful, I developed a bias over the course of the study. I found I felt more positive towards those participants with positive and rich descriptions of their shorts experiences and less positive towards those who found the shorts ineffective, difficult to use or had not engaged in exploring their effect. On a couple of occasions, I recorded findings that I then could not evidence from the data. Therefore, I created Tables 7.2, 7.3 and 7.4 as a means of summarising important words and phrases, providing transparency around which participants said what and how common the different opinions were. Whilst an important finding in qualitative research is not necessarily one that arises frequently (Pope et al., 2000), I felt this improved transparency and credibility, for the sake of my own confidence in the findings as well as other readers.

7.4 Implications for the thesis

The OSFeaMS qualitative study demonstrated that orthotic shorts are acceptable for PwMS and have an important influence on confidence. Findings are consistent with the concept proposed in Section 3.6.3 that there is a "journey of orthotic use" more likely to lead to acceptance of an orthotic and similar to that proposed for other assistive devices. In particular, the importance of a short-term orthotic effect was demonstrated through perceived effectiveness. Ideas for adaptation of the shorts supported the idea that users should receive support from healthcare professionals during early stages of orthotic use, to adapt themselves and their orthotic to best effect. Unfortunately, the short timescale of the home trials in the OSFeaMS study meant that the OSFeaMS study was unable to confirm the importance of the "journey of orthotic use" for longer-term orthotic acceptability. This element could be built into future studies.

Ways of improving acceptability of the shorts have been suggested. These are around (1) assessing anticipated acceptability as part of an initial consultation, (2) clarifying potential disadvantages to inform users' decisions, such as the resistance to trunk and hip flexion and the visibility of the shorts beneath clothing and (3) exploring possible adaptations to the current shorts design such as shorter legs, a smaller toileting hole for men and an alternative design that assists flexion rather than extension.

A number of important qualitative findings have not been explored within this discussion as they will be integrated with relevant quantitative findings and explored in Chapter 8. These are: (1) the measured versus perceived effect of the orthotic and placebo shorts on walking, (2) the potential training effect of the orthotic shorts and (3) the potential choices of outcome measurement and study design for future studies.

7.5 Conclusion

The OSFeaMS qualitative data demonstrated that orthotic shorts are acceptable for PwMS. The perceived links between the shorts and the concepts of core stability and exercise indicate that the anticipated acceptability seen in this study is likely to be transferable to other PwMS. The importance of psychosocial effects of an orthotic were clearly demonstrated, with familiar topics raised around the importance of appearance and self-image. In addition, the shorts appear to have a pronounced effect on confidence, with descriptions of support, stability and control that imply a physical and emotional response. The role of the shorts in empowering PwMS to do more and try more than they might otherwise attempt indicates a possible role in improving engagement with activities that have additional benefits for wellbeing. The next chapter will integrate the qualitative and quantitative findings and develop discussion around the potential mechanisms of effect of the shorts and implications for future research.

Chapter 8: The Orthotic Shorts Feasibility in MS Study - integration of quantitative and qualitative findings

Summary

This penultimate chapter reports integration of the quantitative and qualitative findings of the OSFeaMS study. The discussion section focusses on key findings around mechanism of effect, study methods for future research and the contributions gained from using a mixed methods approach.

8.1 Aims

In order to integrate findings from the qualitative and quantitative data, research questions were worded in such a way as to ensure that both datasets were relevant. Thus, the aims of this integration were to determine answers to the following:

1. Are orthotic shorts acceptable in PwMS?
2. Might orthotic shorts be efficacious in PwMS for improving gait and balance in MS?
3. How should future studies, investigating orthotic shorts be designed?

8.2 Results

Tables 8.1 and 8.2 show the convergence coding matrix, split into two sections. Table 8.1 presents findings around acceptability and the potential efficacy of the orthotic shorts. Table 8.2 presents key findings related to research design. In each of the sections below, I review the ways in which the findings might be considered convergent or divergent. For convergent findings, I attempt to show how qualitative data has enriched the quantitative findings, including some examples not presented earlier in the thesis. Divergent findings are discussed, demonstrating additional insight obtained and, where relevant, questions for future research.

Table 8.1: A convergent coding matrix for the OSFeaMS study - acceptability and potential efficacy

Research question	Qualitative findings	Quantitative findings	Integration
Are orthotic shorts acceptable in PwMS?	Shorts are acceptable = a logical intervention, fitted with values, liked and perceived to be effective; disadvantages = restricted hip flexion, negatively affected self-image, minor issues with dressing and undressing.	Shorts are acceptable = kept by 75% of participants, worn for 86% of time recommended. One adverse reaction regarding development of ankle swelling.	Convergent
Might orthotic shorts be efficacious for improving gait and balance?	↑ confidence in walking	Improved MSWS-12, improved ABC-UK, ↑gait speed, ↑cadence, ↑ step length, ↓ stride time, improvement over time for MSWS-12 & gait speed	Convergent
	Perceived ↑control over leg direction	↓ step width, ↓ step width variability	Convergent
	Perceived ↑control over turning	Not measured	Silence in quantitative
	Walking further and faster	Improved MSWS-12, ↑ gait speed, ↑cadence, ↓ stride time, ↑trunk & pelvic yaw rotation in orthotic shorts	Convergent
		Little change in T25FW	Divergent - dissonant
	Felt more stable, sturdy, better balance, walking smoother and less wobbly, fewer falls and near falls	Improved MSWS-12 and ABC-UK,↓ falls incidence, ↓ step width, ↓ step width variability, ↓ stride time variability, small ↓ in ML trunk sway, ↓ML pelvic sway in those with a larger ML pelvic sway	Convergent
		No change in ML pelvic sway across the group, ↑ sway variability at trunk and pelvis in orthotic shorts, ↑ lateral pelvic tilt	Divergent - complementary
	Lifting legs felt easier, less likely to trip (D,F,O)	Improved MSWS-12 across group, ↓ stride time, ↓stride time variability	Convergent
		↑ trunk and pelvic pitch	Divergent - complementary
	Lifting legs felt harder especially when climbing stairs (A,B,G,H,K,L)	↑ step length, ↓stride time variability	Divergent - complementary
		Hip flexion activities not measured, e.g. steps and stairs	Silence in quantitative
		For orthotic shorts, worse MSWS-12 scores for B,G,L; stairs-specific MSWS-12 question worse rating for G,H and L.	Convergent
	Perceived ↓need to concentrate on walking (J,K,M,O)	↓ dual task cost in orthotic and placebo shorts, ↑cognition in dual task cost assessment - placebo more effective.	Convergent
	Improvement over time due to increased activity (C,M,N,O), shorts had a "residual" effect (K),	Improved baseline values over the course of the study for MSWS-12, gait speed, dual task cost, stride time variability and step width variability	Convergent
Other perceived effects	↑ perceived strength, improved posture, more benefit when more active, decreased sway in standing.	Not measured	Silence in quantitative

(ABC-UK = the UK version of the Activities-Specific Balance Confidence measure; ML = mediolateral; MSWS-12 = 12-item Multiple Sclerosis Walking Scale; PwMS = people with multiple sclerosis; T25FW = Timed 25-foot walk; ↑ = increased; ↓ = decreased, other initials refer to participants' pseudonyms)

8.2.1 Integration of findings related to acceptability

The qualitative and quantitative datasets for acceptability were integrated and discussed in Section 7.3.2 (Table 7.6). Both datasets agreed that orthotic shorts were an acceptable intervention for improving walking in PwMS, with the qualitative component being valuable in explicating what was meant by acceptability and how acceptability might be improved. The quantitative element supported the argument that the qualitative findings were valid. Wear times were good and were higher in those for whom the shorts were most acceptable, lowest in those for whom the shorts were "not acceptable" and at an intermediate level in those classified as having "questionable acceptability". Quantitative data allowed comparison of acceptability with other orthoses, indicating that acceptability might currently be lower for orthotic shorts than for FES and AFOs (Section 6.3.5).

8.2.2 Integration of findings related to potential efficacy

Confidence

There was clear convergence between the qualitative and quantitative data regarding the impact of orthotic shorts on confidence, with no areas of divergence. The impact on confidence came across strongly in the qualitative data. Nine participants described themselves as more confident in the shorts, including some participants who perceived few other benefits. All participants described that in the shorts they felt "*strong*", "*stable*", "*supported*" and "*secure*". All of these adjectives seem to imply a link with confidence. The quantitative data showed improved self-reported walking ability and balance confidence. In addition, the changes to self-selected gait speed, cadence and stride time all indicated a more confident walking pattern. Improvement over time was seen, particularly in variables associated with confidence (MSWS-12 and gait speed).

Qualitative data provided insight into how this improved confidence felt and what the potential mechanisms were for its effect. Participants described doing more, without necessarily noticing at the time. Some described that their increased confidence had influenced areas of their life other than their walking and balance, for example, feeling taller and being less likely to shy away from challenging activities. The concept of the shorts as an enveloping, comforting presence was shared by a small number of participants, suggesting that increased confidence was closely linked to the sensation of increased support.

Some participants questioned the degree to which the effect of the shorts was purely emotional. Gwen felt that confidence was the only change and *"whether it actually gave me truly more stability or not is difficult to tell."* Other participants explained a psychological element that complemented the physical impact of the shorts. Mostly, participants were comfortable believing the shorts had some of their impact via a psychological effect. As Frank explained, "*yes, it was physical but even if part of it was psychosomatic, I don't care, it's done its job.*"

Controlling leg direction and turning

Qualitative and quantitative findings converged regarding the reported improvement in control over leg direction whilst walking, with no areas of divergence. Qualitatively, this was clearly explained by Jon who considered this the most important change for him. He explained that he had little control over leg direction without the shorts, giving examples such as tripping over kerbs, catching his walking pole with his feet and his legs “*splaying out*” if he turned his head to the side. He felt all of these things improved in the shorts. Ben used gesture to describe the sensation of the shorts guiding his legs forwards in a straight line. Melissa described that, without the shorts, she struggled to place her feet accurately. She would tread on her son’s toys on the living room floor, despite trying hard to avoid them. With the shorts, she could direct her feet more precisely. Quantitatively, I suggest that the variables of step width and step width variability reflect this same experience and there was a moderate, measurable improvement in these variables. Theories explained in Section 6.3.5 suggest that PwMS struggle to maintain leg direction when they step because placement of the swinging leg is subconsciously used to maintain a stable trunk and pelvis in the mediolateral plane (Donelan et al., 2004; Rankin et al., 2014). It is normal for the swinging leg to control pelvic stability, however, slow and inaccurate sensory feedback in MS might mean that leg direction is altered late in swing phase, resulting in a hypermetric response and the sensation of uncontrolled, random foot placement. Olsen et al. (2005) provided a similar description of uncontrolled foot placement in their qualitative study exploring fatigue in women with MS. They explained, "The feeling of being fatigued increased because of the extra effort of arranging their footsteps when walking" (p11).

Qualitatively, Jon described being able to turn corners more tightly with his orthotic shorts and he linked this directly to his ability to control his legs better. This seems in keeping with the problems with controlling leg direction; it seems likely that mediolateral stability would require more control whilst turning. Similar suggestions were made by Melissa, Caroline and Kathy who described having more control over their ability to walk inside their homes, touching walls less when turning and bumping into furniture less. Unfortunately, the OSFeaMS study did not measure the ability to turn whilst walking but it was highlighted by a number of participants as a potential gap in the measurement battery.

Smooth, stable and sturdy – not quite as wobbly

Nine participants described changes in trunk control and balance (Alison, Caroline, Dawn, Frank, Jon, Kathy, Melissa, Natalie, Oliver). These problems might be related to difficulty controlling leg direction; however, only three participants reported the leg direction issues described above, indicating that these problems could be unrelated. Alternatively, control over leg direction, balance and control might be very closely related but problems with trunk control and balance may be more evident to the individual than problems with leg control.

Qualitative and quantitative data were largely convergent regarding the impact of orthotic shorts on balance and control. Qualitatively, participants felt more stable and more fluent whilst walking in the shorts and felt they had fewer falls and near falls and improved balance. Expressions of improved strength and stability were common. Quantitatively, this was reflected in the decreased falls incidence and decreased variability of spatiotemporal gait parameters previously linked to falls risk, particularly step length variability and stride time variability. In addition, those participants with a large mediolateral pelvic sway at baseline had decreased pelvic sway whilst wearing the shorts.

The OSFeaMS qualitative data provided insight into how a variable walking pattern feels to the individual with MS. Participants referred to *"wobbling"* and *"wavering*". Many explained that they have difficulty maintaining a consistent direction when they walk, expressed as “*walking in a straight line*”. When they tried to walk straight ahead, they found they wavered about from side to side. Ben described this as *"walking like a type writer"* referring to the lateral movement of a typewriter carriage. Helen described that the first symptom of her MS was that when she walked holding hands with her husband, she would pull him sideways or knock into him. Similarly, she linked her difficulty walking in a straight line to her dislike of crowds. Unless she used a walking stick, she found it difficult to navigate through a crowd because of her tendency to waver from side to side. Helen did not perceive any improvements in these problems with the orthotic shorts but other participants, notably Jon and Natalie, felt that walking in a straight line improved.

Qualitatively, several participants described that improved stability in the shorts had improved their function in standing. Examples included improved ability to stand and turn without moving your feet and an ability to stand for longer, thus improving housework, cooking and coping with work related tasks. Kathy provided a powerful example of improved function in standing. She described being in an art gallery, able to read notices on the wall and examine paintings whilst wearing her shorts but unable to do so without her shorts. She explained that the fatigue she felt whilst standing at the cooker was caused by that same swaying feeling. Postural sway in standing was not measured in the OSFeaMS study but this could be an important future research idea. Although only Kathy specifically mentioned this experience, postural sway and gait variability may have similar underlying causes, such as muscle weakness and slowed sensory feedback (Callesen et al., 2019; Moon et al., 2015).

Quantitatively, the findings that mediolateral pelvic sway in walking did not improve with the shorts, that sway variability increased at the trunk and pelvis and that the pelvis tilted laterally more whilst wearing the shorts could be considered divergent from the qualitative suggestion that stability improved. As discussed in Section 6.3.5, these quantitative findings either could indicate worsening of the control of trunk and pelvic movement in the shorts or improved adaptability with more options available for maintaining control (Glazier et al., 2006). The OSFeaMS qualitative data supports the latter possibility, that the increased variability proximally whilst wearing the shorts is a good thing. No participants reported worsening of their balance and control in walking. The quantitative data on proximal and distal variability allowed exploration of the mechanism by which the sensation of stability might be improved.

Walking further and faster

Qualitatively, five participants felt that they could walk further or faster in the orthotic shorts. Mostly, this finding agrees with the quantitative data in that, the MSWS-12, self-selected walking speed, cadence, step length and stride time all improved. There was an increased range of trunk and pelvic yaw in the orthotic shorts, which Liang et al. (2014) suggested is associated with walking faster and taking longer steps.

However, maximal gait speed, tested in the T25FW, increased by only a very small amount. This may suggest that there is a limit to how much orthotic shorts might improve walking speed. For example, as Jon suggested, they might restrict maximal step length, meaning that at higher speeds, the shorts limited rather than increased speed. Alternatively, the change in self-selected speed but not maximal speed might indicate an effect on confidence but not biomechanics. That is, people chose to walk a little faster due to increased confidence but the actual biomechanics of walking ability were unchanged.

Lifting my legs

The ability to lift one’s legs whilst walking and climbing stairs was discussed by a number of OSFeaMS participants and there was disagreement within the qualitative data regarding whether this was harder or easier in the orthotic shorts. Quantitatively, stairs ability was not measured objectively but there was one question specific to stairs ability in the MSWS-12 and this was rated by three participants (Gwen, Helen and Linda) as being more difficult in the orthotic shorts. Others felt stairs were easier or no different in the shorts. Quantitative data on trunk and pelvic movement may also be relevant to this important issue. Below, the divergence within the qualitative data is discussed first, before comparing qualitative to quantitative findings.

Qualitatively, some participants described improvements in their ability to lift their feet when they walked (Dawn, Frank and Oliver), although Oliver found this effect to be more marked in the placebo shorts. These individuals were amongst the more disabled participants. The tendency to catch their feet when they walked was a cause of them "*stumbling*" and was a key contributing factor limiting how far they could walk. Conversely, others described that lifting their legs felt harder in the shorts. The reported difficulties were mostly related to larger range movements against gravity, such as climbing stairs or lifting legs to step up a kerb. The apparent discrepancy may be because walking on the flat requires less active hip flexion against gravity than stairs. The idea that hip flexion was limited by the orthotic shorts was reflected in difficulty bending at the hips and waist and compression on the stomach in sitting, along with reports of assistance to hip extension in standing up and holding a squatting position. As Linda explained, *"the first pair* (placebo) *seemed better at supporting me; the second* (orthotic) *pair were supporting me a bit too much"*. Overall, the qualitative data strongly supports the idea that hip flexion was restricted by the orthotic shorts so the more important question is why lifting legs whilst walking might have been easier for some individuals.

The quantitative data reflects improved stepping whilst walking with decreased stride time, increased step length and decreased stride time variability at a group level in the orthotic shorts. However, trunk and pelvic pitch (trunk flexion/extension and anteroposterior pelvic tilt) and lateral pelvic tilt were increased in the shorts, despite being significantly larger than normal at baseline. These findings suggest that there may have been an increased need for compensatory trunk and pelvic movement due to restriction of movement at the hips. It also suggests that the shorts helped participants to achieve these compensatory movements. With the shorts fixing the legs more firmly to the trunk and pelvis, compensatory trunk and pelvis movements might be more efficiently transmitted to the legs. This possibility was reflected in Frank's explanation of how the shorts helped him lift his legs by enabling him to "*straighten yourself and get yourself in the right position*" to persuade his legs to move. This may not be the only explanation of these changes but is a credible suggestion that may have either positive or negative effects with longer-term use of the shorts.

Concentrating on walking

The qualitative and quantitative findings converged regarding a reduced need to concentrate whilst walking. Although the reliability of the dual task cost assessment has been found to be poor, there was a small to moderate decrease in the dual task cost of walking and an improvement in performance of the cognitive task whilst walking in both the placebo and the orthotic shorts. Four participants noted a decreased need to concentrate whilst walking, enabling them to think about other things. The convergence between these qualitative and quantitative findings suggests that this could be a real effect and supports the measurement of dual task cost in future studies.

Improvement over time

The qualitative and quantitative findings converged regarding the existence of a possible training effect of the shorts. The quantitative data showed improved values in the “no shorts” conditions across Visits 3, 5 and 7 for a number of variables, namely gait speed, step length, stride time, step width variability, stride time variability and dual task cost. In addition, there was a marked improvement in participant perceived walking ability between Visit 1 and Visit 7. Because of the tendency for previous fabric orthosis studies to focus on training effect rather than a direct orthotic effect, as found in the systematic mapping review (Section 2.4.3), I included a question in the OSFeaMS final interviews around whether participants felt they thought a training effect was conceivable. Kathy was convinced that she had already experienced a training effect over the four weeks that she had worn the shorts, providing many examples of how her underlying ability had improved. Four participants explained that a training effect would happen because they would do more in the shorts, either in terms of repetitions of exercises or in general physical activity. As Natalie explained, “*If your legs feel better you can do more things, I imagine, that's the way it would work*”.

Other perceived effects

A number of perceived effects were highlighted in the qualitative data but not measured in the OSFeaMS study. These were improved strength, particularly in hip extension; improved posture; decreased postural sway; improved standing balance; improved ability to walk around corners and a potential relationship between perceived benefit and the degree to which participants are "pushing themselves" to cope with their MS. These points are noted here in order to highlight the potential importance of further investigation in future qualitative and quantitative studies.

8.2.3 Integration of findings related to future study methods

Integration of the qualitative and quantitative findings relating to future study methods is summarised in Table 8.2. This was broken down into three key areas: study design, outcome measurement and the choice of comparator intervention. The findings are described below; potential implications will be discussed later.

Table 8.2: A convergent coding matrix integrating qualitative and quantitative findings of the OSFeaMS study - planning for future studies

Research question	Qualitative findings	Quantitative findings	Integration
Design of a future study - crossover or parallel group?	Dissatisfaction with washout periods, different shorts allowed participants to compare effect of shorts and appeared to engage some participants to engage with investigating the differential impact of each pair	Improvement over time seen in several variables	Convergent - indicating challenges and advantages of a crossover design
Placebo vs. control vs. active comparator?	Most participants believed the placebo shorts were a placebo; psychosocial elements may be integral to the effect of the shorts; comparison between the two pairs of shorts was interesting and may have prompted exploration of effect	Some beneficial effects of the placebo shorts were difficult to explain as psychosocial effect alone	Convergent – comparison between shorts is helpful but a placebo fabric orthoses may not be realistic
What measures should be used in future studies?	Number of measures used not burdensome; spatiotemporal gait parameters interesting and relevant; dual task cost relevant but challenging	Al least small changes seen in all measures except % of gait cycle in double support and T25FW; gait variability provides important insight into possible mechanisms of effect.	Convergent – most measures were appropriate
	Other constructs measured could include postural sway, posture and strength	Gait variability changed, which may be related to postural sway	Divergent – complementary regarding potential importance of postural sway

(T25FW = Timed 25-foot walk.)

Qualitative and quantitative findings agreed regarding the challenges associated with the crossover design. Most importantly, quantitative data showed improvement over time in several variables, meaning that baseline variables were not similar on the days that the different shorts were tested and the effect of one condition may have carried over into the next. Qualitatively, two participants expressed dissatisfaction that they had been involved in the study for three months but only trialled each pair of shorts for two weeks. They were puzzled by the use of the washout periods and suggested that the long waits between visits had increased the burden of the study. In addition, five participants felt the time allowed for the home trials needed to be longer to enable full exploration of the effect of the shorts.

Qualitative and quantitative findings agreed that there were significant challenges in using a placebo garment. Most participants believed from the outset that one of the pairs tested would be a placebo, which may have been reflected in their relatively low ratings of initial change (GRC). However, many participants felt the placebo shorts were helpful in supporting them and some participants preferred them, partly because they were easier to wear and allowed more movement than the orthotic shorts. The comparison between the two different pairs seemed to help to engage some participants in trying to determine what the shorts were doing for them. From a quantitative perspective, there appeared to be potential effects in the placebo shorts that are hard to explain as a psychosocial effect alone. For example, the improvement in the cognitive task whilst walking was moderate in the placebo shorts, possibly more effective than the orthotic shorts. In addition, there were small to moderate improvements in step length variability in the placebo shorts. These changes may simply be due to chance or result from a training effect. However, they raise the suggestion that the so-called placebo shorts may have an objective, sensorimotor effect. This could due to the sensation of improved support or a change in sensory feedback. Overall, there were advantages to comparing two interventions from a qualitative perspective but the original belief that the placebo shorts would enable a distinction between “real” and psychological effects was flawed.

Qualitative and quantitative findings agreed that most of the measures used in the OSFeaMS study were valuable. All participants felt the choice of measures was appropriate and did not feel unnecessarily burdened by the measurement battery. There were a number of important suggestions for future measures, which have been summarised above, towards the end of Section 8.2.2.

8.3 Discussion

The findings of the integration exercise suggest that orthotic shorts are acceptable and might be efficacious in improving gait and balance in MS. The data most clearly supports that orthotic shorts might improve confidence, improve walking speed for most people, improve control over leg direction in swing phase and improve stability in walking and standing. It has been suggested that those who are more disabled by their MS perceive an improved ability to lift their legs when they walk. However, there is probably restriction of hip flexion in the shorts that might decrease step length at maximal walking speed and increase the effort required to climb stairs. Regarding future study designs, the integration exercise confirms the challenges associated with a crossover design and using a placebo for fabric orthosis studies and suggests ideas for improving measurement.

This discussion section focusses, firstly, on the possible means of effect of the orthotic shorts and, secondly, on future research ideas for orthotic shorts and other fabric orthoses.

8.3.1 A theory explaining mechanism of effect

Figure 8.1 shows a suggested model that summarises various factors identified by the quantitative and qualitative data around what orthotic shorts might be able to offer and how they might achieve a beneficial effect. Improved support is suggested to lead to improved confidence. Confidence, in turn might lead participants to be more active and increase their engagement with exercise. I suggest that confidence and fear of falling are closely interrelated because falling seemed to be a common fear in MS, decreasing confidence. Fear of falling was not directly measured in the OSFeaMS study but the qualitative data, improved self-selected walking speed and balance confidence all support a decreased fear of falling. In turn, fear of falling might decrease due to a decreased falls risk. Decreased falls risk was indicated in the qualitative findings as improved stability, decreased stumbling and decreased tripping and, in the quantitative findings, as improvements in stride time variability, step length variability and falls incidence.

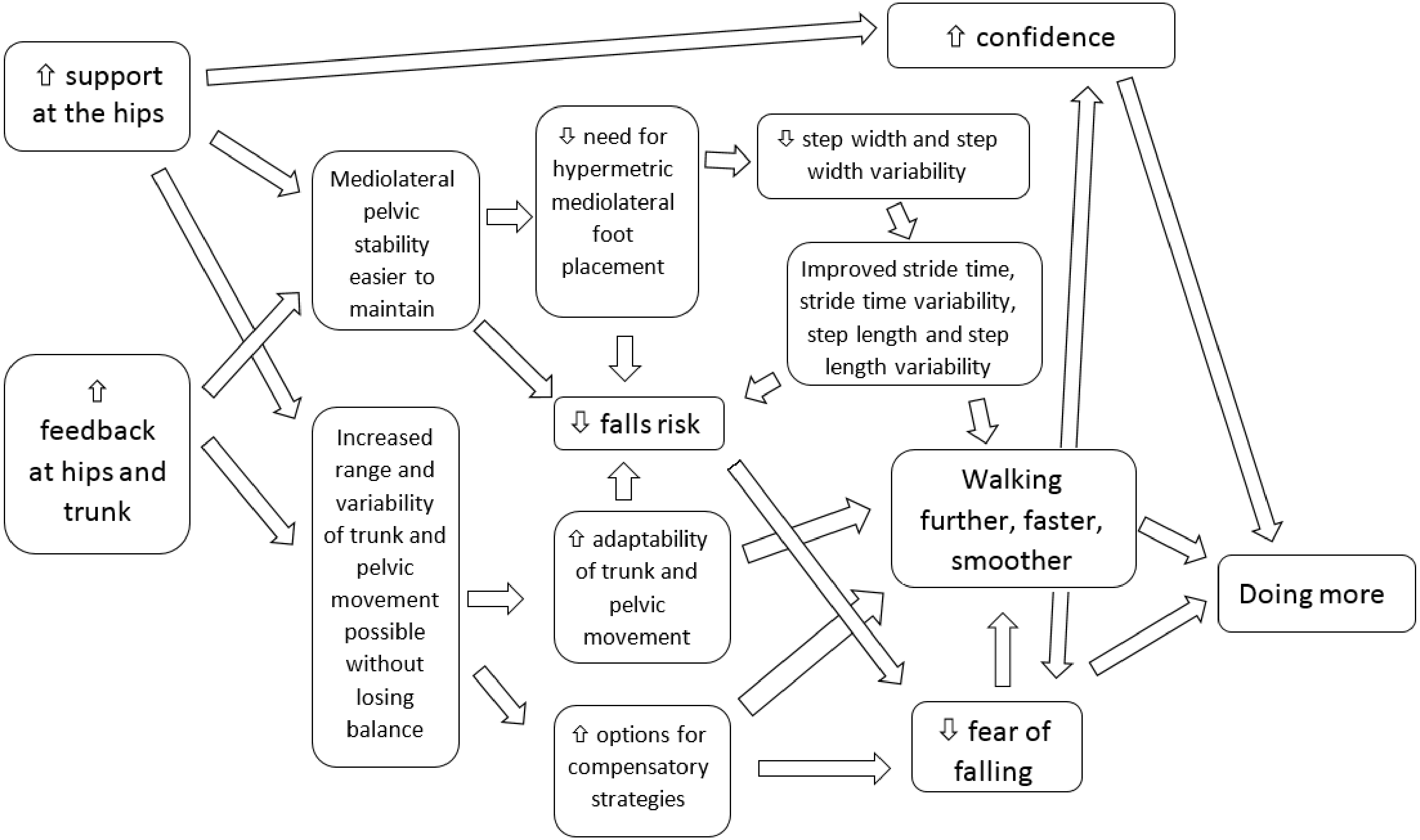


Figure 8.1: An illustration of suggested factors contributing to mechanisms of effect

Other elements of Figure 8.1 draw upon the hypotheses suggested in Section 6.3.5. These hypotheses suggest that the orthotic shorts might work by providing physical support that stabilises the pelvis and improves proprioceptive information from the stance hip and possibly the trunk. The improved stability and feedback could mean that there is (1) improved accuracy in how the swinging leg provides mediolateral stability, as opposed to responding too late and with too large a response and (2) improved options and improved adaptability for compensatory strategies at the trunk and pelvis. The improved compensatory strategies could improve walking speed, efficiency and balance control by improving the ability to lift the feet and stabilise control on the stance leg. It is also possible that improvements in efficiency of mediolateral control could lead to improved anteroposterior control. In other words, because there is less inefficiency in the way in which the swinging leg maintains mediolateral stability, the swinging leg is more able to simply step forwards. Thus, step length becomes longer, stride time shorter and both become less variable. In other words, the improved mediolateral stability could be the source of the improved anteroposterior efficiency. I believe this hypothesis is supported by my finding that mediolateral stability (step width and step width variability) changed more with the shorts than step length and stride time variability. The hypothesis is also supported by previous research suggesting that mediolateral stability requires more input from the neurological system (O'Connor & Kuo, 2009) and, therefore, it is more likely that the mediolateral problems cause the anteroposterior problems than vice versa.

Watson et al. (2007) suggested that fabric orthoses might dampen down error in uncoordinated, involuntary movement, provide a motivational effect via a novel intervention, enhance sensory feedback and provide a flexible "exoskeleton" (p754) for people with weakness. The findings of the OSFeaMS study support their idea of a supportive, external "exoskeleton" and provide qualitative and quantitative data that support that hypothesis. In addition, the OSFeaMS study has demonstrated that variability in voluntary movement might be improved and that fabric orthoses might have a large impact upon confidence, suggesting an important role for emotional support in addition to any physical effects.

8.3.2 Lessons learnt for future studies

Table 8.3 summarises guidelines and recommendations made by a range of research methods experts around how research for a novel intervention should develop over time, assuming that the findings at each stage support ongoing investigation. The OSFeaMS study was a Phase 1 trial aimed at determining whether the intervention might possibly be useful. As reported in earlier sections, the OSFeaMS studies demonstrated that orthotic shorts are acceptable to PwMS, safe and might be efficacious. The findings suggested who might benefit and a model has been proposed as to how the shorts might have their effect. It is, therefore, appropriate to continue research into the orthotic shorts and the next step would be to determine efficacy in an appropriately powered trial. Such a study should not only determine efficacy but also develop understanding of the mechanism of effect (Whyte & Barrett, 2012) and how best to deliver the intervention in a wider practice context (Bleijenberg et al., 2018; Dobkin, 2009). An efficacy study should inform the choice of control intervention and measures for a definitive effectiveness study (Mohr et al. 2009; Whyte & Barrett, 2012).

In truth, design of any research study should be done collaboratively, considering the perspectives of key stakeholders including healthcare practitioners, potential funders, patient and public groups and research staff who may be involved in study delivery (Bagley et al., 2016; Parker & Kingori, 2016). Nevertheless, to draw out the relevance of the OSFeaMS study to informing future research, suggestions discussed here are presented without the benefit of discussion and collaboration with others.

The next section is structured around key reflective points relevant to designing future studies. This discussion covers not only planning for a future efficacy study but also lessons learned from the OSFeaMS study relevant to future feasibility studies.

Table 8.3: A summary of development phases recommended for novel interventions

	Might the intervention work?	Can/does the intervention work at the impairment level?	Does the intervention work for meaningful measures of function or quality of life?
	Feasibility/Phase I trial	Efficacy/proof of concept	Effectiveness
Design features	Might be small sample, within-group design; relatively large number of outcome measures, minimising Type II error is more important than minimising Type I error (Mohr et al., 2009; Whyte & Barrett, 2012).	A "proof of concept" RCT (Eldridge et al., 2016); focusses on efficacy at the level of the immediate treatment target (Rounsaville, Carroll & Onken, 2001; Whyte & Barrett, 2012); must be powered to detect change in the primary outcome measure, which should be sensitive to treatment effects (Hart & Bagiella, 2012); has internal validity re: randomisation and blinding (Whyte & Barrett, 2012); "no treatment" or waiting list control is acceptable, even though these exaggerate treatment effects (Mohr et al., 2009); targets participants most likely to respond (Whyte & Barrett, 2012); may be multi-centre (Whyte and Barrett, 2012).	A multi-centre, fully powered, "effectiveness" RCT; tests intervention against an active control, clearly defined and known to impact upon the same primary outcome measure (Dobkin, 2009; Mohr et al., 2009); determines therapist and site effects (Hart & Bagiella, 2012); multiple outcome measures representing different levels of the ICF (Hart & Bagiella, 2012).
How this stage prepares for the next	Determines whether it is worthwhile continuing, how the intervention can be applied and to whom; informs choice of outcome measures and responses in patient variation (Dobkin, 2009).	Develops and standardises the intervention (Dobkin, 2009; Craig et al., 2008); may develop understanding of mechanism of effect; determines whether further investigation is appropriate; informs choice of measurement, including possible measures for function and quality of life (Whyte & Barrett, 2012); informs choice of control intervention (Mohr et al., 2009).	Evaluates how "transportable" the intervention might be to different practitioners, patients and setting (Rounsaville, Carroll & Onken, 2001); investigates who else might respond positively to the intervention (Whyte & Barrett, 2012).
Other strands of development work	Develop understanding of the main problem being treated, how best to measure it and how it changes in the absence of treatment ((Whyte and Barrett, 2012). Develop in-depth understanding of how the problem of interest is currently managed in practice, to understand how a new intervention might be incorporated (Bleijenberg et al., 2018). Develop the intervention for best effect. Investigate and develop acceptability of the novel intervention to healthcare practitioners (Sekhon et al., 2018).		

(ICF – International Classification of Functioning, Disability and Health; RCT = randomised controlled trial)

The OSFeaMS study was designed as a traditional crossover, in that there were intervention periods and "washout" periods. However, in a traditional crossover design, the effect of each condition is assessed at the end of the intervention period. Other than with the self-report measures, this was not the case in the OSFeaMS study because the aim of the objective assessment of walking ability was to determine if there might be a direct orthotic effect and the primary aim of the home trial was to assess acceptability. Inclusion of the washout periods meant that the assessments of walking ability in the different shorts took place at least 4 weeks apart. This time lapse may have contributed to the differences in baseline “no shorts” ability on the different days. Many participants improved their baseline ability between the two shorts testing days and some deteriorated, the difference being particularly marked for those who had one of the two assessments in the hot summer period.

For future feasibility studies, it would be preferable to separate the two different study aims into different phases of the study. In other words, the question of whether there is a direct orthotic effect and whether the shorts are acceptable should be assessed sequentially. Figure 8.2 illustrates how such a study might be designed. The direct orthotic effect could be assessed first, with different orthoses assessed on different days to allow rest but with the testing days less than a week apart to minimise changes in underlying ability. After the objective assessments, the home trials of the two different orthoses could begin. One home trial could be followed immediately by the next, with no gap between them, thus allowing for a longer trial period within a similar length study. This would enable the benefits of the comparison without the disadvantage of washout periods.

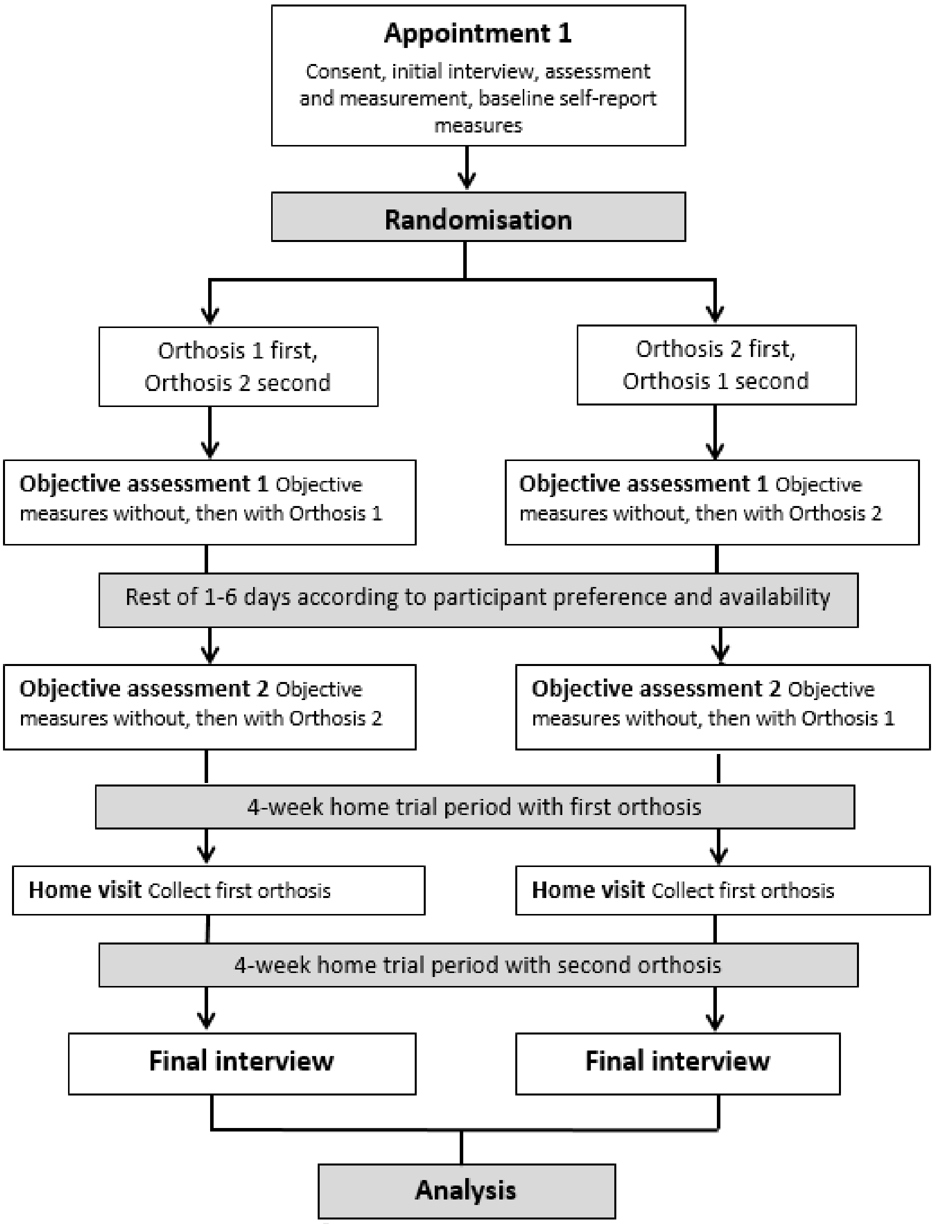


Figure 8.2: Suggested crossover design for future feasibility studies

Reflections on use of a placebo versus an active comparator

A placebo was used in the OSFeaMS study mainly because they appeared helpful compression garment research and because, at the start of the study, the question about whether the shorts had a "real" or purely psychological effect seemed important. Although the so-called placebo shorts enabled insight into the possible effect of close-fitting clothing, I would suggest that similar placebos are not appropriate comparators in orthotic research. Hart and Bagiella (2012) and Peterson and Dieppe (2005) explained that an effective placebo provides effects that are incidental to the treatment but not characteristic of or integral to the treatment. For example, in a pharmacological study, incidental effects might include therapeutic interaction and improved hope for the future. These can be controlled for with a placebo tablet. In rehabilitation and specifically in fabric orthoses, such aspects are integral to the treatment itself. Partly, the difficulty is that rehabilitation and orthotic interventions require perception of effect and an active response to that perceived effect. The logic behind an orthosis and the extent to which it achieves its aim should be clear to the users and should enable them to function better. If an intervention partially achieves that aim, like our placebo shorts, then it is not a true placebo. If users believe a placebo is not achieving anything, this would influence their perception of it and it would not function as a placebo.

In Functional Electrical Stimulation research investigating direct orthotic effect, it is usual to compare movement with and without stimulation (Taylor, Humphreys and Swain, 2013). To investigate training effect, researchers often compare against baseline or against a group who receive no intervention (Barrett & Taylor, 2010; Street, Taylor, & Swain, 2015; van der Linden, Hooper, Cowan, Weller & Mercer, 2014). This has been criticised as an approach and it is recommended that for investigating a training effect of an orthosis, there should be some form of active comparator (Centre for Reviews and Dissemination, 2013). In line with the suggestions summarised in Table 8.1, it would be acceptable to use a "no treatment" or "waiting list control" in an efficacy study but a suitable active control must be identified for a future effectiveness study.

Reflections on outcome measurement choice

Outcome measures in research studies need to have potential to change in response to the intervention, have strong psychometric properties, be relevant to function and not be overly burdening for participants. In the OSFeaMS study, some of the measures that changed most in response to the shorts were those for which psychometric properties were less certain: the spatiotemporal gait parameters, gait variability and the dual task cost assessment. Participants felt the spatiotemporal gait parameters were both interesting and relevant to the impact of the shorts. The dual task cost assessment received both positive and negative feedback. Several people identified having difficulty thinking and walking at the same time and felt that this was important to measure. Others found it challenging and appeared embarrassed with their ability. In addition, since the OSFeaMS study was completed, research was published suggesting that reliability of the dual task cost assessment assessed with the T25FW is poor (ICC = 0.45; Decavel et al., 2019). It may be necessary to use a different means of measuring the impact of cognition on walking. For example, the impact of cognition of spatiotemporal gait parameters has been shown to be reliable in PwMS for all mean gait parameters tested (Monticone et al., 2014).

In efficacy and effectiveness studies, measures defined as most important to the research question should be chosen as primary outcome measures and used to determine sample size calculations (Smith, Morrow and Ross, 2015). They should represent the main reason the trial is being performed. Considering the significant psychosocial impact of the orthotic shorts, it seems important to choose a primary outcome measure for an efficacy study that is unlikely to be sensitive to performance bias. Thus, measures of speed and self-report measures should be avoided. Logical choices for primary outcome measures for assessment of orthotic shorts would be step width or step width variability because these objective measures were most sensitive to the impact of the shorts, the qualitative findings indicate their importance and they are believed to be relevant to balance (Brach et al., 2008; Givon et al, 2009). Step width might change both with a direct orthotic effect and in response to improvement over time (Dixon et al., 2014). A risk associated with choosing step width as a primary outcome measure is that, as explained in Section 6.3.4, there is some uncertainty across different researchers around the mean and variability of this measure in PwMS (Kalron, 2016; Socie et al., 2013a; Socie et al., 2013b; Socie et al., 2014).

In terms of self-report measures, the MSWS-12 has been shown to change in response to the shorts, has strong psychometric properties and is a commonly used and understood metric, allowing easy comparison to effects seen with other interventions.

Measures that might be dropped from a future study would be the GRC, ABC-UK and T25FW. The GRC did not provide useful insights into first impressions, possibly because it was done immediately after the laboratory tests and people needed longer to form an impression of whether their walking had changed. The ABC-UK showed there may be an impact of the shorts on balance confidence but, as discussed in Section 6.3.8, it could be replaced in future studies with the FES-I, a measure of fear of falling. There was only a very small change in the T25FW in response to the shorts in the short term and, therefore, it may not be an important measure. In addition, several participants mentioned their dissatisfaction with being judged according to how quickly they could walk in a straight line, suggesting that control at slower speeds and the ability to remain steady whilst turning were more important. However, the T25FW is very commonly used in MS research, meaning that the effects of different interventions can be directly compared. For example, although the change in T25FW in the OSFeaMS study was very small, the mean improvement seen with the orthotic shorts of 0.03 m/s (95% CI: -0.06 to +0.12 m/s) was only a little smaller than the mean 0.05 m/s improvement found to be achieved using FES (Miller et al., 2017).

Measures that might usefully be added to future studies include assessment of falls incidence, because as explained in Section 6.3.5, many of the changes seen with the shorts have been linked in previous research to either fear of falling or falls risk. Stairs assessment would be important because of the potential disadvantages of the shorts for climbing stairs. For most people, it is a common function and includes active hip extension, which a number of participants felt improved with the shorts. Assessment of stair climbing might help capture whether overall, there is a measurable positive or negative impact and, if people with significant difficulty with hip flexion find the shorts particularly challenging to use, a stairs assessment could be developed into a screening assessment. To date, no studies have been published testing reliability and validity of a stairs measure in MS but a simple timed test could be reliable, such as that suggested by Ni, Brown, Lawler and Bean (2017).

Another commonly used objective measure is the Timed Up and Go Test (TUG). Like stair climbing, this involves large hip extension movements and, like the T25FW it involves fast walking, however, it involves a change in walking direction. Turning is considered more functionally relevant than straight line walking and inclusion of turning in objective assessment enables a closer correlation with participant-reported measures of walking ability (Adusmilli et al., 2018). The TUG has been found to have good psychometric properties (Nilsagard, Lundholm, Gunnarsson & Dcnison, 2007; Sebastião, Sandroff, Learmonth & Motl, 2016) and has been recommended for exercise trials in MS (Paul et al., 2014). It has been found to correlate closely with the T25FW (*r* = 0.9) but is argued to have better face validity (Sebastião et al., 2016). An Instrumented Version of the TUG is available using Opal Sensors, which enables a more accurate total time than that obtained with a stopwatch and provides a breakdown of time taken for the different components of the movement (Craig, Bruetsch, Lynch, Horak & Huisinga, 2017b). Several researchers have used the TUG test for dual task cost assessments (Ciol et al., 2017; Hershkovitz, Malcay, Grinberg, Berkowitz & Kalron, 2019).

Measurement of postural sway was recommended by one of the OSFeaMS participants, whose experiences lead her to believe that she swayed less in standing with her placebo shorts on. Postural sway may be a mechanism associated with gait variability (Callesen et al., 2019; Moon et al., 2015) and, therefore investigation of whether orthotic shorts can change postural sway might enable an improved understanding of mechanism of effect. In addition, because decreased step width was accompanied by increased mediolateral trunk and pelvic sway in some OSFeaMS participants, it is important to understand the impact on mediolateral stability. Postural sway can be measured using instrumented techniques that, although not specifically investigated for reliability in MS have been shown to be responsive to change with balance exercise (Prosperini & Pozzilli, 2013).

In summary, there is potential to develop the outcome measurement battery that might be used in a future study to include measures more specific to everyday function, stability and fear of falling. It is noted that several of the measures considered here do not have clearly demonstrated psychometric properties in MS. It is possible to integrate a reliability assessment into a trial by having an additional baseline assessment, as used by Decavel et al. (2019). By calculating reliability and

Minimum Detectable Change in the same group used for an intervention trial, then any changes from baseline can be more accurately determined.

8.3.3 A suggested "proof of concept" randomised controlled trial

Figure 8.3 illustrates a suggested study design for a proof of concept RCT. This includes two baseline assessments for determining stability and reliability of outcome measures. A 12-week intervention period is suggested, as this is a commonly chosen length of intervention for exercise studies in MS (Carter et al., 2014; Feys et al., 2019) and is the period over which Coote et al. (2014) recommended collecting falls incidence data.

The suggested efficacy trial would assess with and without orthotic shorts at baseline and at post-intervention, enabling assessment of direct orthotic effect, training effect and total orthotic effect, as recommended by Street et al. (2017). As discussed in Section 8.4.2, important candidates for measurement of a direct orthotic effect would be postural sway in standing, mean and variability of spatiotemporal gait parameters, TUG, T25FW and a stairs assessment. Dual task cost could be assessed using either the TUG or spatiotemporal gait parameters. Self-report measures recommended would be falls incidence. MSWS-12 and FES-I. Appendix 22 shows a summary of other learning points from the OSFeaMS study relevant to such a trial. Because the comparator would be different and a number of changes of outcome measurement are proposed, it would be advisable to conduct a pilot RCT prior to a fully powered study. It has been suggested that a sample size of approximately 10% of the planned final study is sufficient to determine the success of a protocol (Hertzog, 2008).

A power calculation was performed from the OSFeaMS data using figures obtained at baseline on the orthotic shorts testing day, assuming that the most important assessment is that of the total training effect at three months and the required inferential analysis would be a t-test. Power was set at 80% and alpha at 0.05. For the step width data, the baseline values were 12.2 ± 3.4 cm and the mean change with the intervention was set at 1.5 cm, the change achieved in the OSFeaMS study. The online calculator provided by Dhand and Khatkar (2014) suggested a sample size of 81 in each group. Using the step width variability data, with baseline values of 3.4 ± 1.6 cm and a mean change with the intervention of 0.8 cm, sample size was suggested to be 63 in each group. The larger sample size would be appropriate to maximise the chances of finding an effect, if the shorts are indeed efficacious.

Dropout rates for studies in PwMS are in the order of 6 - 15% for studies investigating exercise interventions, so it common to plan for a dropout rate of 15% (Carter et al., 2014; Hatton et al., 2016). However, recent studies into orthoses for PwMS have reported a higher dropout rate of 19% at three months (Renfrew et al., 2019) or 19% at nine weeks (Miller et al., 2016). It is wise to plan for this higher dropout rate in the first instance as this decision could be re-evaluated partway through a study. Therefore, the sample required to determine if orthotic shorts are efficacious in improving step width in a between-group study would be 100 participants in each group.

The variability estimates obtained from the OSFeaMS study were from a relatively small sample. They might not be representative of a wider group and this might under or over-estimate the numbers required for a fully powered study (Noordzij et al., 2010). Because of the uncertainty around step width values in previous MS studies discussed earlier, it would be wise to review step width data obtained mid-way through the proposed study and re-run a sample size calculation at this point.

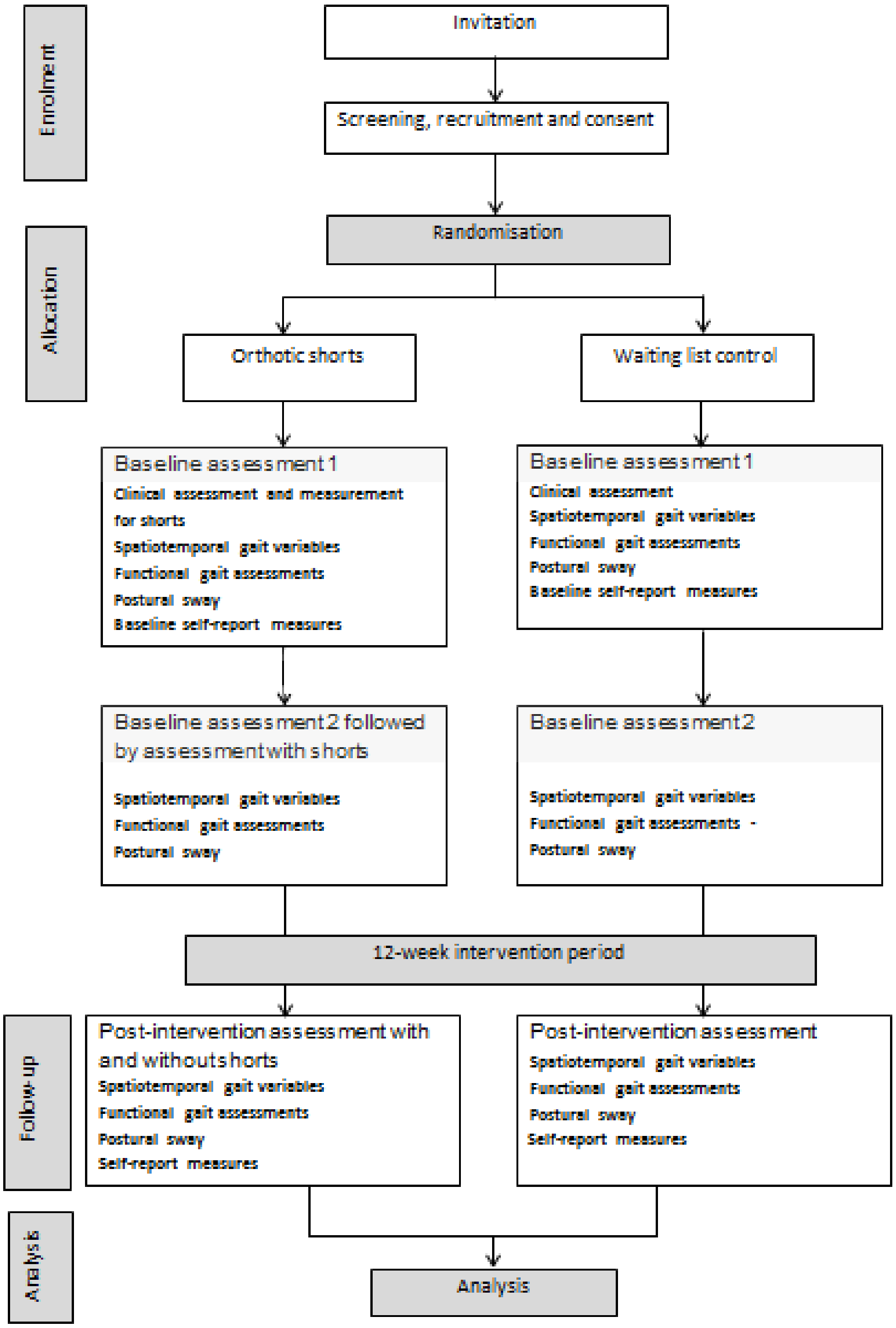


Figure 8.3: A suggested design for a "proof of concept" RCT

8.3.4 The contribution of mixed methods to the OSFeaMS study

The guidelines for Good Reporting of a Mixed Methods Study (GRAMMS; O'Cathain, Murphy & Nicholl, 2008) suggest that it is important to describe clearly what mixed methods have brought to a study, reporting insights gained specifically from integrating methods. It is hoped that many such insights are clear from this chapter. In terms of contribution, the combined qualitative and quantitative investigation of acceptability strengthens the validity of the claim that the shorts are acceptable. The qualitative data contributes to the assessment of acceptability by explicating why the shorts might be acceptable and providing important ideas for improving acceptability, including assessing hip flexor function, improving the colour schemes in which the shorts are made and informing collaborative decision-making.

Integration of qualitative and quantitative data has enabled a far more detailed understanding of the potential effect of the shorts than would have been obtained from quantitative data alone. In particular, qualitative data on perceived stability suggest that the increased variability and range of trunk and pelvic movement are suggestive of improved stability, improved adaptability and an increased range of compensatory strategies, rather than indicating decreased function and stability in the shorts. Qualitative data has demonstrated the day-to-day experience of variables that might otherwise appear irrelevant to human experience. Quantitative data has enabled an in-depth exploration of the possible influence of orthotic shorts on human movement control and has provided a level of objectivity to the study that complements the reports of subjective experience. Objective demonstration of efficacy and effectiveness are necessary to influence the uptake and funding of orthotic interventions and the quantitative element of the OSFeaMS study has laid a foundation for these future studies. Quantitative data has added weight to the idea that the shorts have a physical and possibly sensory effect over and above the important emotional response.

Qualitative data highlighted several areas that have not been investigated quantitatively in the OSFeaMS study, which may be important aspects to measure in future trials, for example, the impact on stair climbing, sit to stand and postural sway.

In summary, the combination of the methods has enabled a more rounded, in-depth analysis than would have been achievable with one method alone.

8.4 Implications for the thesis

The discussion in this chapter has drawn together qualitative and quantitative findings to explicate the ways in which orthotic shorts might affect PwMS. A possible mechanism of effect of the orthotic shorts has been proposed, supported by both qualitative and quantitative findings. Key reflections on research methods have been discussed and a future trial has been suggested to determine efficacy. Integration of methods has enabled key objectives of the thesis to be met and has positioned the OSFeaMS study with reference to a proposed future research programme that may be used to develop the shorts intervention in the future.

8.5 Conclusion

The qualitative and quantitative aspects of the OSFeaMS study have successfully complemented one another to indicate that orthotic shorts are acceptable to PwMS and that there is potential for them to be effective in improving walking. The measured and perceived effectiveness has been explored in this chapter to suggest a theory around how the shorts might have their effect and a future efficacy trial has been suggested that builds upon the findings of this thesis. The next chapter is the final chapter of this thesis. It reflects upon the aims and objectives described in Chapter 1, discusses the suggested contributions to knowledge and discusses further research ideas.

Chapter 9: Summary of findings, clinical implications and recommendations for further research

Summary

This final chapter returns to the aim and objectives outlined in Chapter 1, signposts to thesis content relevant to each objective and considers the extent to which each objective has been met. Original contributions to knowledge arising from the objectives will be summarised and then each contribution discussed in more detail, setting each within the context of previous research. Finally, key clinical implications are discussed and suggestions for future research considered.

9.1 Review of aim and objectives

The aim of this thesis was to investigate the acceptability and feasibility of using fabric orthoses with PwMS. I suggest that this aim has been met and new knowledge generated in relation to understanding fabric orthoses more widely and with respect to the specific application of orthotic shorts in PwMS. The objectives set for the thesis are summarised below.

9.1.1 Objective One

The first objective of the thesis was to identify and evaluate existing evidence into the use of fabric orthoses. This objective was met in Chapter 2. I conducted a comprehensive, systematic mapping review looking across compression garments, joint supports and fabric orthoses and a systematic review evaluating evidence for the effectiveness of fabric orthoses in adults with neurological conditions. The systematic mapping review was the first attempt to describe research across these interrelated fields and was published as Snowdon et al. (2018). The systematic review provided a detailed critical review of the existing knowledge around fabric orthoses for adults with neurological conditions. Together, these provide original insights into the existing evidence base.

9.1.2 Objective Two

The second objective of this thesis was to determine influences on acceptability of fabric orthoses. This objective was met within two qualitative studies, reported in Chapters 3 and 7. Chapter 3 reported the FabO IPA study, which explored the perceptions of long-term users of fabric orthoses. Chapter 7 explored the acceptability of orthotic shorts. These qualitative studies lead to exploration of a number of important and unanticipated areas, resulting in contributions to knowledge around the important psychosocial effects of fabric orthoses, the concept of a journey of orthotic use and use of the recently proposed Theoretical Framework of Acceptability to analyse and interpret findings (Sekhon et al., 2018).

9.1.3 Objective Three

The third objective of the thesis was to determine the feasibility of using orthotic shorts to improve walking in PwMS. Feasibility was considered as potential efficacy and acceptability. This objective was met in that acceptability were shown to be good and the orthotic shorts were shown to be potentially efficacious. Findings relating to this objective were presented and discussed in Chapters 6 and 7. Chapter 8 integrated the qualitative and quantitative findings and demonstrated how the findings from the different approaches largely converged regarding the feasibility of the orthotic shorts. This has been the first research study to investigate the acceptability and potential efficacy of orthotic shorts and, therefore, both are original contributions to knowledge.

9.1.4 Objective Four

Objective four was to develop a theory explaining the possible means of effect of fabric orthoses and, specifically, orthotic shorts. Relevant data were introduced into the thesis across Chapters 2, 3, 6 and 7. Ideas were synthesized and discussed in Chapter 8, where a theory of how the orthotic shorts might have their effect has been proposed.

9.1.5 Objective Five

Objective five was to design a future RCT to investigate the effectiveness of orthotic shorts in MS. Potential further trials into orthotic shorts have been proposed in Chapter 8 and it has been argued that prior to an effectiveness trial, there should be an efficacy trial or so-called "proof of concept" RCT.

9.2 Contributions to knowledge

9.2.1 Identification and evaluation of existing research

The reviews conducted and reported in Chapter 2, revealed a number of important insights. The comparison of fabric orthosis research to compression garment research highlighted the poor quality of the research designs being used in fabric orthosis studies with large numbers of case descriptions and pre-test post-test designs and very few RCTs and crossover studies. The systematic review reinforced this finding by using the Cochrane RoB tool and GRADE guidelines to evaluate fabric orthosis research (Section 2.6.3), concluding that none of the previous adult neurology studies provided more than low quality evidence. Whilst the poor quality of fabric orthosis research had been commented upon previously (Coghill & Simkiss, 2010), this had been based upon reviews of orthotic use for children and had not been identified in the adult literature.

The systematic mapping review highlighted key gaps in the evidence. There were only three qualitative studies, two of which examined fabric orthoses within mixed method feasibility studies (Miller et al., 2016; Stone, 2014). Joint supports had previously been found to improve proprioception but, despite it being a commonly suggested mechanism of effect, it is unknown whether fabric orthoses can improve proprioception and whether improved proprioception improves function. Future systematic reviews were suggested into the effectiveness of compression garments, for variables where there were a number of primary studies investigating a similar group and no previous reviews (Section 2.4.2).

Both reviews revealed that researchers appear to envisage fabric orthoses as a treatment, in that they evaluate use over a number of weeks and rarely investigate whether there is a direct orthotic effect. In addition, researchers described or appeared to apply a range of different theories around how fabric orthoses might work. Most adult neurological studies focussed on spasticity management but there were no topics investigated by more than one study and, even in this one field, theories differed as to how the orthoses might have their effect (Section 2.6.4). Current theoretical understanding around how fabric orthoses might influence movement could be holding back research and development, particularly with respect to the apparent theory that fabric orthoses only work if worn over a number of weeks.

9.2.2 The important psychosocial effects of fabric orthoses

The importance of psychosocial factors in determining the acceptability of orthotic devices and assistive technology has been reported previously; examples have been discussed in Section 3.6.2 and a measure of the psychosocial impact of assistive devices has been validated (Day, Jutai & Campbell, 2002). Previous qualitative studies in fabric orthoses (Miller et al., 2016; Stone, 2014) reported an improved sense of normality, improved body awareness, an improved sense of ownership of your own body, improved confidence and wellbeing, along with an uncomfortable stigma attached to a visible orthosis. The qualitative studies performed in this thesis confirmed and extended these findings. The FabO IPA study highlighted that fabric orthoses can influence identity and self-image in both positive and negative ways and this came across strongly in the OSFeaMS study. Several OSFeaMS participants were motivated to try the orthotic shorts because they were discrete, compared to other assistive devices. People's feelings about the appearance of the shorts were key factors in determining acceptability, with some picturing them as "*sports shorts*" and others as "*granny's knickers*". There were a number of examples in both qualitative studies where participants hinted at or openly described a feeling of emotional support from the orthoses. For example, participants reported that their fabric orthosis felt to them like a supportive person. They felt held or hugged, as though someone had their arms around their waist. They used words such as *"supported", "strong", "stable"* and "*secure*", which can refer to both physical and emotional support. The feeling of being hugged by an orthosis has not been reported in previous literature and may be a sensation peculiar to fabric orthoses. It is potentially important, as it could contribute to the improved confidence reported by the OSFeaMS participants.

9.2.3 The concept of a journey of orthotic use

No previous studies on fabric orthoses have discussed the importance of the way in which an orthosis is introduced to a potential user. The FabO IPA study lead to the proposal that there is a journey of orthotic use, in which certain experiences can make an individual more likely to continue or discontinue using their orthosis (Section 3.6.2). The experiences found to support ongoing use were experiencing a direct orthotic effect, collaborative problem solving during the initial assessment process, support to adapt both one’s behaviours and one’s orthosis, and perceiving that advantages outweigh disadvantages. The OSFeaMS qualitative findings supported the importance of experiencing a direct orthotic effect at first use although participants felt they needed to trial their shorts in a range of different activities to determine what felt easier and what felt more difficult. The suggestion that a direct orthotic effect matters to users supports the importance of investigating short-term changes, rather than longer-term training effects alone in research.

As reviewed in Section 3.6.2, the importance of the prescription process has been researched extensively in reference to assistive technology (Lenker & Paquet, 2003) and protocols guiding healthcare professionals in supporting initial use of assistive technology have been shown to be effective (Verza et al., 2006). Recently, Renfrew et al. (2018) conducted an IPA study with long-term users of FES and found similar themes around the need for persistence, the need to adapt one’s skills and routines and the importance of professional support during the adaptation period. However, these findings have not previously been demonstrated with respect to fabric orthoses. They are an important indication that acceptability is not just about the orthosis itself but also about the way in which it is introduced, explained and adapted to the needs of an individual.

9.2.4 The value of the Theoretical Framework of Acceptability for evaluating healthcare interventions

It is widely accepted that healthcare interventions are only effective if patients engage with them and there is a body of literature investigating facilitatory and inhibitory influences on adherence (Mathes, Jaschinski & Pieper, 2014). Researchers have studied satisfaction with healthcare interventions for many years (Sidani et al., 2018) and the importance of the acceptability of healthcare interventions to users is supported by the Developing Complex Interventions Framework (Craig et al., 2008). However, exactly what is meant by acceptability is sometimes unclear and previous researchers have measured it in a variety of ways (Sekhon et al., 2017). Recently, Sekhon et al. (2017 and 2018) published their Theoretical Framework of Acceptability, in which they proposed that seven different domains make up the construct of acceptability. They suggested that this framework could be used to assess acceptability both before and after a patient has experienced an intervention. No previous research has been published in which the Theoretical Framework of Acceptability is tried and tested as a means of understanding acceptability.

In the OSFeaMS study, the Theoretical Framework of Acceptability was used to analyse the qualitative data on acceptability and was shown to be useful and applicable. There were no findings relevant to acceptability that did not fit within the framework and the framework drew out important elements that might otherwise have gone undetected, such as the fit of the orthotic shorts to an individual's value system. The framework was used to classify participants according to their comments around affective attitude, burden and perceived effectiveness and this process enabled clear distinction between a group of seven participants who clearly found the shorts acceptable and a group of five for whom feedback was mixed and acceptability questionable. I have suggested that this finding could be used as a basis for an assessment of acceptability in future studies. This element of the thesis has wider implications for determining acceptability of other healthcare interventions.

9.2.5 Orthotic shorts are an acceptable intervention for people with multiple sclerosis

No previous research has investigated the acceptability of orthotic shorts as an intervention for improving walking in MS. Orthotic shorts have been used in children with cerebral palsy (Flanagan et al., 2009), with stroke survivors (Maguire et al., 2010) and as a means of supporting athletes with groin pain (Sawle, Freeman & Marsden, 2016) but none of these studies assessed acceptability. Acceptability has been assessed for orthotic socks and sleeves (Miller et al., 2016; Stone, 2014) but the experience of wearing shorts is likely to be very different to that of wearing socks or sleeves.

The OSFeaMS study assessed the acceptability of orthotic shorts using robust qualitative methods and a sample of participants of various ages, genders and abilities. Acceptability was found to be good. In particular, the shorts were a good fit for participants' values; participants associated the shorts with exercise, liked their low-risk nature, felt driven to try anything that might help them and liked the concept of support that was invisible to others. In terms of understanding the shorts as an intervention, participants were mostly familiar with the concept of core stability and identified this as a means through which the shorts might work. Burden was generally low and the shorts were perceived to be effective. However, findings on burden and perceived effect were variable across the participant group and resulted in suggestions for improvements to the shorts. The proportion of participants who found the shorts acceptable was somewhere between 50 and 75%, depending upon whether this was judged according to ongoing use after the trial or by using the Theoretical Framework of Acceptability to classify people into "clearly acceptable" and "questionable acceptability". The acceptability findings suggest that orthotic shorts are worthy of further investigation.

9.2.6 Orthotic shorts could have a direct orthotic effect that improves walking in multiple sclerosis

No previous research has investigated the direct orthotic effect of shorts for PwMS. Previous studies on orthotic shorts have assessed the longer-term effect in children (Flanagan et al., 2009) and the direct orthotic effect on walking in stroke survivors (Maguire et al., 2010). Assessment of direct orthotic effect of fabric orthoses is unusual because, as described in Section 9.2, most previous research has investigated longer-term training effects only.

Methodological limitations to the OSFeaMS study meant that a direct comparison between the orthotic shorts and shorts designed as a placebo could not be trusted. Nevertheless, changes seen with objective measures between the no shorts and orthotic shorts conditions are suggestive of orthotic effects. There were small improvements in self-selected gait speed and the associated variables of step length and stride time, moderate improvements in step width and step width variability, small improvements in step length variability and stride time variability and increased range and variability of trunk and pelvic movement, suggested to indicate improved adaptability and compensatory movement. These findings not only imply that the shorts might be efficacious and worthy of further investigation but are an indication that research should investigate orthotic effects prior to determining whether a training effect exists. The length and burden of these different study designs are markedly different. It can take only a few days to investigate direct orthotic effects, compared to the 2 – 3 months typically used to investigate training effects in orthoses that may not even have been demonstrated to be acceptable.

9.2.7 A theory of change explaining the potential effect of orthotic shorts

Previous researchers have suggested theories to explain the possible effects of fabric orthoses (Gracies et al., 2000; Ibuki et al., 2010a & 2010b; Maguire et al., 2010; Stone, 2014; Watson et al., 2007). Watson et al. (2007) provided the most comprehensive list of potential mechanisms, discussed in Section 8.4.1. Previous research has supported the idea that fabric orthoses provide a flexible exoskeleton that can improve posture, joint range and joint position without removing freedom of movement (Blair et al., 1995; Flanagan et al., 2009; Gracies et al., 2000; Maguire et el., 2010; Stone, 2014). Miller et al. (2016) and Blair et al. (1995) supported the idea that fabric orthoses might dampen down involuntary movement. The relative contribution of psychosocial, sensory and physical support have not been investigated previously. No previous orthotics studies have measured movement variability.

The OSFeaMS study has added to these theoretical perspectives by being the first study to suggest that movement variability might change with fabric orthoses and might be an important mechanism by which walking and balance could improve. The OSFeaMS study found a decrease in footfall variability with the orthotic shorts, no change to mediolateral pelvic sway and an increase in trunk and pelvis sway variability. The different changes associated with the orthotic and placebo shorts suggest that improving step width and step width variability required the increased support found in the orthotic shorts, rather than just any sensory changes that might be associated with close-fitting clothing. A theory has been proposed (Section 8.3.1) linking these different findings, drawing upon the Dynamical Systems Theory (Cavanaugh et al., 2005) and theories explaining control of mediolateral stability in walking (Section 6.3.5). In brief, it has been suggested that the support provided by the shorts and the hypothesized improvement in proprioception make mediolateral pelvic stability easier to control and increase adaptability and compensatory movements possible at the trunk and pelvis. In turn, these decrease footfall variability, decrease falls risk and improve the efficiency of walking. This theory is testable in future studies as many of the variables are measureable. Improved variability might be relevant to other fabric orthoses and I suggest variability would be a relevant, objective measure for fabric orthoses such as shoulder and trunk supports, body suits and orthotic socks.

9.2.8 A proposal for a proof of concept RCT

The final suggested contribution to knowledge is the design of a future RCT that could test the efficacy of orthotic shorts. This proposed trial and the reasoning behind its design was discussed in Section 8.4.3. In brief, the trial would use a waiting list control and be focussed on the impact of orthotic shorts at the impairment level of the ICF. The doctoral programme has provided insight into recruitment and retention for such a study, has informed the choice of outcome measures and provided data to inform sample size calculations. The proposed trial would be considerably different in design to that used in the OSFeaMS study and may need to be multi-centre to facilitate recruitment. Thus, it would be appropriate to test the study design in a pilot RCT. Pragmatic solutions to several challenges faced in the OSFeaMS study have been suggested that would improve a future trial.

9.3 Clinical Implications

This section considers first a summary of potential clinical implications for orthotic shorts in MS and then wider clinical implications of the thesis. Clearly, orthotic shorts should not be recommended for PwMS because all that has been achieved with the OSFeaMS study is evidence of acceptability, support for the idea that there could be an effect, plus a proposed mechanism for such an effect. Nevertheless, PwMS do fund their own assistive devices and will seek advice from healthcare practitioners around their options. Pending a larger trial, I would suggest the following as guidelines for clinical practice:

- Orthotic shorts might benefit PwMS. There could be measurable benefits to movement control, in addition to an important psychosocial effect of improving confidence, so they may be worth trying.
- There is no need to suggest that orthotic shorts should be worn for an extended period in order to feel a benefit. If people want support for particular activities, such as outdoor mobility or standing for long periods, then shorts

could be helpful. They are probably most useful when people are most active and less useful when they are sedentary.

- Those suggested to respond positively are people who feel that they have instability around the trunk and hips, who are challenged by that feeling of instability and, therefore, might be willing to accept the burden associated with wearing an extra layer of clothing in order to ease their difficulties.
- The shorts might be useful for PwMS with a wide range of disability levels, whether they do or do not use a walking aid. However, the burden of the shorts is likely to outweigh the benefits in people who do not walk regularly.
- There is possibly most benefit to people with mediolateral instability, such as increased lateral sway, increased step width and increased step width variability. Such problems may not be measurable without instrumented devices but could be identified, for example, by asking people to walk in a straight line, between two lines and observe if they can do this and how frequently they lose direction or step onto or over the lines.
- People who may be inappropriate for orthotic shorts include those with a previous history of ankle swelling or other circulatory difficulties and those who struggle to lift their legs to climb steps or stairs and need to do so regularly.

Successful prescription of any orthosis relies upon a collaborative assessment process, involving problem solving and discussion of different potential solutions. Assessment of anticipated acceptability is recommended in clinical practice (Sekhon et al., 2018) by asking questions around whether the proposed mechanism of effect makes sense, whether the proposed mechanism fits with their understanding of their own problems and whether they anticipate any challenges in using the proposed intervention. The appearance of orthoses or assistive devices and their potential impact on self-image should be openly discussed. Prescription of fabric orthoses should be informed by photographs, examples and a discussion around how visible the orthosis might be beneath somebody’s clothes. A period of supported orthotic use is recommended, in which adaptations might be facilitated to the individual’s skills and routines and to the orthosis itself. This support should continue until routines of wear are established.

The potential effects suggested for orthotic shorts could be explained by the support provided and, possibly, a sensory mechanism, in other words, the “flexible exoskeleton” concept proposed by Watson et al. (2007, p754). These same mechanisms could explain the increased muscle activity seen with TheraTogs^{TM} shorts in the Maguire et al. (2010) study. Thus, similar uses of fabric orthoses could be effective, where an orthosis provides flexible support for people with weakness. As shown in the OSFeaMS study and Maguire et al.’s study, such orthotic uses could allow improved function with an increase in movement options and may promote, rather than inhibit, muscle activity.

The wider evidence around fabric orthoses suggests that there may be a role in spasticity management; however, it is important to be clear about the mechanism we are hoping to exploit in order to influence spasticity (Section 2.6.4). It is possible that compression decreases reflex excitability. This could be relevant for both spasticity and cerebellar tremor (Section 2.4.2) but is probably only effective with compression over 36 mmHg (Ibuki et al., 2010b). As illustrated by one of the participants in the FabO study, high pressures can cause pressures sores and decreased blood flow (Section 3.5.3), so must only be considered for people who fully understand the risks and for whom alternative treatments have been unsuccessful. Spasticity might be inhibited by fabric orthoses through a biomechanical effect, lengthening shortened muscles or encouraging postures that enable improved function and activity. If this were the potential mechanism being exploited then obviously posture or joint position would need to change when the orthosis was applied.

My final clinical implication is that fabric orthoses deserve a second chance. They appear to have fallen out of favour because of reported problems with acceptability in early research. However, the studies reported in this thesis have demonstrated that a well-designed fabric orthosis need not apply a lot of compression, can be easy to get on and off and has potential to improve movement control. I would suggest that design and fitting have improved over the years and it may be timely to reconsider what these orthoses might be able to achieve.

9.4 Future research priorities

In addition to the proof of concept, efficacy RCT suggested earlier, this thesis has suggested a number of additional lines of inquiry relevant to orthotic shorts and to fabric orthoses more widely. These ideas are discussed here.

As suggested by Andreopoulou et al. (2018), the psychometric properties of measures such as trunk and pelvic kinematics, spatiotemporal gait parameters and gait variability have not been evaluated in PwMS. Although validity has been established by demonstrating relationships between such parameters and several important variables such as falls risk (Allali et al., 2016a; Socie, Sandroff & Pula, 2013) and fear of falling (Delbaere et al., 2009), there is limited information to date on reliability, Minimum Detectable Change and Minimum Clinically Important Change. Such data would all be important to interpret changes in intervention trials. Therefore, reliability studies into these variables are recommended and, as suggested in Section 8.4.2, could be integrated into the design of an efficacy study.

There have been indications in this thesis that it is important to investigate the acceptability of fabric orthoses to healthcare professionals. Sekhon et al. (2018) suggested that this is important to develop and implement an intervention. One FabO IPA study participant reported that her physiotherapist suggested orthotic support should be avoided because it might weaken her muscles. In the OSFeaMS study, recruitment through the NHS physio was relatively unsuccessful and a higher proportion of participants recruited via NHS physio did not find the orthotic shorts acceptable. Whilst there may be other reasons for this finding, it raises the possibility that negative feelings towards the intervention were conveyed by the recruiting physiotherapist to the potential participants. This possibility is extremely important because it might influence recruitment and retention to the proposed efficacy trial, as well as to any future implementation of fabric orthoses into routine clinical practice.

Bowen et al. (2009) suggested that a feasibility study should include assessment of how practical an intervention is, within a specific context. In addition, Bleijenberg et al. (2018) suggested updating the Developing and Evaluating Complex Intervention Guidelines to incorporate in-depth examination of current practice and the context in which the problem of interest is currently being managed. They suggest understanding the organisation of and influences on current services and interventions, prior to introducing a novel approach. Thus, there may be a role for studies investigating what currently happens when someone with MS with a walking difficulty presents to a UK NHS practitioner. Such research might determine what advice and treatment options people currently receive, what influences practitioners in determining whether to recommend an assistive device, what devices are considered, how collaborative that assessment is and how practitioners feel about suggesting orthotics. There may be a need to learn from and influence professional attitudes towards orthoses before or alongside a future trial. Specific study designs might include qualitative investigations, process mapping of current services and a survey of current practice.

Within this thesis, I have made recommendations for future research for other fabric orthoses. I have suggested that direct orthotic effect should be assessed more frequently, that a specific crossover design should be used to investigate potential efficacy and acceptability of different orthotics (see Section 8.4.2, Figure 8.2) and that movement variability be used more widely as an outcome measure. In addition, it would be valuable for future orthotic shorts studies to explore changes to kinematics using 3D optical motion capture. Specifically, this should enable investigation of whether trunk and pelvic posture improve and whether there are changes to the range of hip motion in the sagittal plane. The Opal sensors utilised in this study provided invaluable insight into trunk and pelvic motion because they were able to demonstrate changes in sway and sway variability. The Opal sensors demonstrated changes in the total range of rotation but could not explain exactly how these movements changed. For example, although there was an increase in the range of trunk pitch with the orthotic shorts, the Opal sensors alone could not determine whether this reflected more trunk flexion, more trunk extension or both. This would be possible with optical motion capture. In addition, 3D motion capture could provide insight into the effect of the shorts on lower limb function such as knee joint and ankle kinematics.

Finally, I would like to suggest that the potential changes with the orthotic shorts might be possible in other patient groups. Because many of the variables that changed in the OSFeaMS study have been linked to falls risk or fear of falling, it may be worthwhile exploring whether orthotic shorts have a potential role in other populations at risk of falling.

9.5 Conclusion

This PhD thesis has offered new insight into the potential of fabric orthoses for PwMS. The project was inspired by the enthusiasm of physiotherapists and orthotists at DM Orthotics, which contrasted sharply with a large gap in an evidence base peppered with suggestions of poor acceptability. A series of inter-linked research projects has enabled a systematic and critical approach to investigating the topic. Robust qualitative methods have taken full advantage of the important personal experiences of PwMS. The potential efficacy of orthotic shorts has been explored with a wide range of measures, which integrated with qualitative findings, has enabled insight into potential mechanisms of effect. The work has established the importance of assessing the direct orthotic effect of fabric orthoses and has made specific recommendations around developing and testing acceptability of fabric orthoses and other healthcare interventions. Additionally, factors relevant to future research studies have been explored and a proof of concept RCT has been proposed to test the efficacy of orthotic shorts in MS. Although there is much left to investigate in this field, the work within this thesis has provided a far more solid foundation upon which future studies can be built.

Publication strategy

At the time of thesis completion, the systematic mapping review had been published as Snowdon et al. (2018). Additional publications are planned as reported in the table below.

Proposed title or topic	Approximate date	Co-authors	Target journal	Target audience
The meaning of fabric orthoses to long-term users with multiple sclerosis: an Interpretative Phenomenological Analysis.	Autumn 2019	Supervision team and Susan Booth (peer reviewer/analyser)	Prosthetics and Orthotics International	Orthotists, physiotherapists and occupational therapists
Using the Theoretical Framework of Acceptability to determine acceptability of fabric orthoses to people with multiple sclerosis.	Autumn 2019	Supervision team	Disability and Rehabilitation: Assistive Technology	Healthcare practitioners more widely but with a focus on physiotherapists, occupational therapists and orthotists
Gait variability in multiple sclerosis: insights from a study exploring the efficacy of orthotic shorts.	Spring 2020	Supervision team and Dr Matthew Brodie	Gait and Posture	Movement scientists, bioengineers, physiotherapists
The acceptability and feasibility of orthotic shorts for improving walking in people with multiple sclerosis: a mixed method study.	Spring 2020	Supervision team	Clinical Rehabilitation	Physiotherapists, occupational therapists, rehabilitation specialists.

(All planned outputs are journal articles.)

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Appendix 1
Mapping review presented as published article

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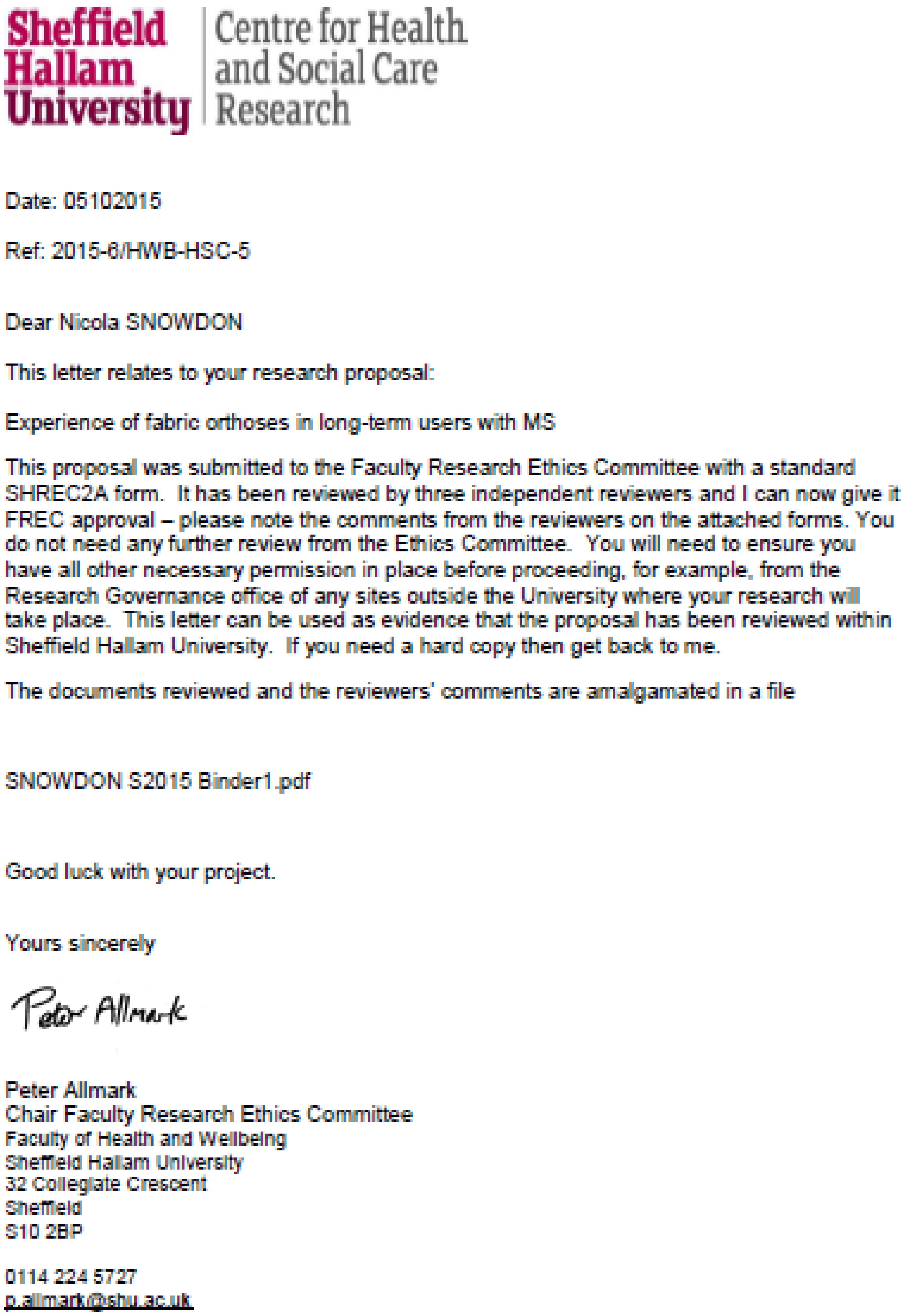
Appendix 2
Approval letter for FabO IPA study



Sheffield Hallam University | Centre for Health and Social Care Research

Date: 05102015

Ref: 2015-6/HWB-HSC-5

Dear Nicola SNOWDON

This letter relates to your research proposal:

Experience of fabric orthoses in long-term users with MS

This proposal was submitted to the Faculty Research Ethics Committee with a standard SHREC2A form. It has been reviewed by three independent reviewers and I can now give it FREC approval – please note the comments from the reviewers on the attached forms. You do not need any further review from the Ethics Committee. You will need to ensure you have all other necessary permission in place before proceeding, for example, from the Research Governance office of any sites outside the University where your research will take place. This letter can be used as evidence that the proposal has been reviewed within Sheffield Hallam University. If you need a hard copy then get back to me.

The documents reviewed and the reviewers' comments are amalgamated in a file

SNOWDON S2015 Binder1.pdf

Good luck with your project.

Yours sincerely

Peter Allmark

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Appendix 3

Invitation letter and Participant Information Sheet for FabO IPA study

Dear…..

We are conducting a piece of research that we hope you will be interested in getting involved with. The research investigates the experiences of people with multiple sclerosis who use fabric orthoses to improve their movement. By fabric orthoses, we mean stretchy, fabric garments such as Lycra shorts, tops, sleeves, socks or whole body suits.

If you:

- Currently use a fabric orthosis at least once a week
- Have used this orthosis for at least a couple of months
- Can remember your experiences when you first received this orthosis
- Have multiple sclerosis and
- Feel that you would like to share your experiences with others, then we would really like you to participate in our study.

The study requires you to give an interview explaining your experiences to the principal researcher. This interview will take place face-to-face and can either be held in your home or somewhere else quiet and private that you can get to easily. The researcher will be travelling from Sheffield for the interview so it is important that we can get to you within about a three hour journey.

If you are interested in being involved, please read the attached information sheet and then get in touch with the principal researcher directly. My contact details are below.

Many thanks

Nicky

Nicky Snowdon, MSc, MCSP.
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Sheffield
Hallam
University

Participant information sheet

A qualitative investigation into the experience of fabric orthoses in long-term users with multiple sclerosis.

We would like to invite you to take part in our research study. Before you decide we would like you to understand why the research is being done and what it would involve for you. Talk to others about the study if you wish. Ask us if there is anything that is not clear.

This research study uses interviews to gain information about the experiences of people with multiple sclerosis of using fabric orthoses such as Lycra garments to improve movement and function. The study is one aspect of the principal researchers' PhD programme.

What is the purpose of this study?

This study aims to explore the experiences of using fabric orthoses. We are interested in people with experience of any orthoses made of a stretchy fabric and worn like a garment, for example, Lycra shorts, shoulder wraps, full body suits, dorsiflexion socks, sleeves. We are hoping to interview between three and six people.

The findings will be used to inform other people with multiple sclerosis about these orthoses to help them decide whether this is an option for them. In addition, the findings will influence the remainder of the PhD programme by shaping the experiments that we conduct into the effect of fabric orthoses on movement.

Why have I been invited?

You have been invited because you are someone with multiple sclerosis who uses a fabric orthosis on a regular basis. You need to have used a fabric orthosis for at least a couple of months and we need you to be able to remember your first experiences of using the orthosis. You do not have to be using the orthosis every single day, so long as you have used it at least every week in the last month or so. It is important that you feel you have experiences that might be useful to others.

Do I have to take part?

Your decision to take part in this study is entirely voluntary. You may refuse to participate or you can withdraw from the study at any time. Your refusal to participate or wish to withdraw would not influence any future services you might receive from the NHS, private practitioners or your orthotics supplier.

What will happen to me if I take part?

If you take part, you will be interviewed about your experiences and this interview is likely to last approximately 45 minutes to one hour. The principal researcher will come to you for the interview. The interview needs to be somewhere quiet and private so can either be in your own home or, if you prefer, could be somewhere that we can agree to meet. The researcher will be accompanied by a postgraduate physiotherapy student and if you are willing, that student will observe the interview. If you prefer, they will wait outside.

The interview will be a conversation about your experiences. Anything that you think is important for the researcher to understand should be discussed in the interview.

What are the possible risks and disadvantages of taking part?

The interview will take up about 1.5 hours of your time including setting up the interview and confirming your consent. There is a risk that discussing your experiences will be upsetting for you. If this is the case, the researcher is an experienced neurological physiotherapist and will be able to discuss any issues arising during the interview and immediately afterwards. However, we would not be able to provide longer term support and if this was required we would help you find someone local to you who could provide support.

Are there any benefits to taking part?

There are no direct benefits to you in being involved in this study, although many people find the process of being interviewed enjoyable and you will be able to help others learn from what you have experienced.

What if there is a problem or I wish to complain?

If you have any queries or questions please contact:
Principal investigator: Nicky Snowdon,
E-mail n.snowdon@shu.ac.uk and phone 0114 225 5751
At Sheffield Hallam University, Faculty of Health and Wellbeing

Alternatively, you can contact my supervisor: Sionnadh McLean
E-mail s.mclean@shu.ac.uk and phone 0114 225

If you would rather contact someone independent of the study, you can contact Peter Allmark, Chair of the Faculty Research Ethics Committee.
E-mail: p.allmark@shu.ac.uk or phone 0114 225 5727

How will you ensure that my taking part in the study will be kept confidential?

The interview will be recorded and then written up word for word. The researcher will check that the recording and the written transcript are the same. The transcript and recording will be kept on a password-protected computer. Identifying details will be taken out of any final report and any publication so people reading these will not be able to identify you.

The documents relating to the administration of this research, such as the consent form you sign to take part, will be kept in a folder called a Site File. This is locked away securely. The folder might be checked by people in authority who want to make sure that researchers are following the correct procedures. These people will not pass on your details to anyone else. The consent forms and transcripts will be destroyed seven years after the end of the study.

What will happen to the results of this study?

The study will continue until February 2016. It will be written up as part of the principal researcher’s doctoral studies. In addition, in 2016, we will seek to report the findings of this study at MS Society events for people with MS and for professionals working in MS and will submit the study for publication in a multi-disciplinary scientific journal.

Who is sponsoring this study?

The sponsor of the study has the duty to ensure that it runs properly and that it is insured. In this study, the sponsor is Sheffield Hallam University.

Who has reviewed this study?

All research based at Sheffield Hallam University is looked at by a group of people called a Research Ethics Committee. This Committee is run by Sheffield Hallam University but its members are not connected to the research they examine. The Research Ethics Committee has reviewed this study and given a favourable opinion.

I would like to participate in this study, what do I do next?

Please contact the researcher directly to arrange the interview, either by e-mail or telephone.

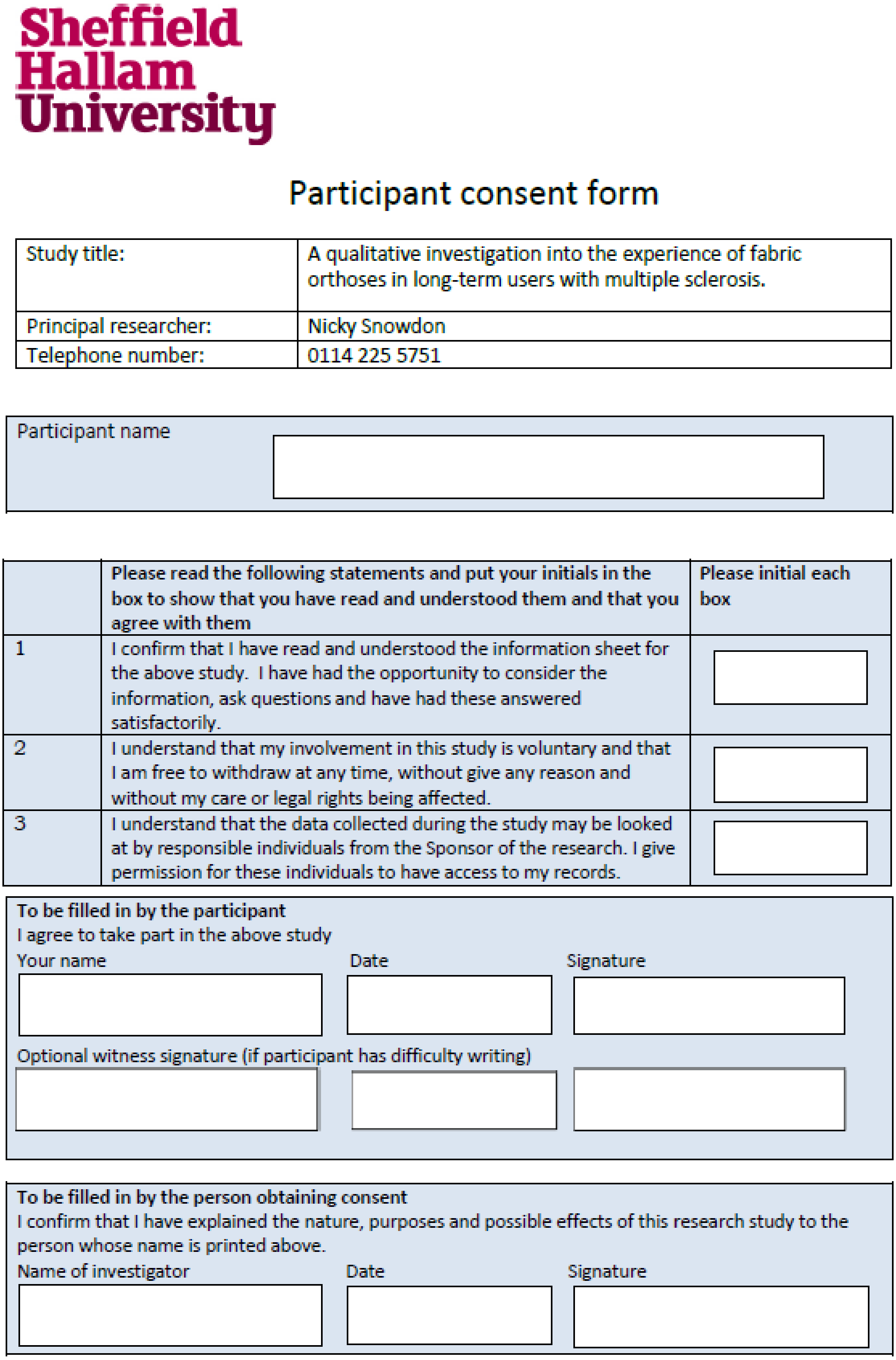
Principal investigator: Nicky Snowdon,

E-mail n.snowdon@shu.ac.uk and phone 0114 225 5751

We will have a telephone conversation to answer any queries that you may have and then we recommend that you have a day or two to think about whether you would like to be interviewed and, if so, where would be convenient for the interview to take place. The researcher will phone you back and if you are still interested then we will make an appointment for a convenient time and location for the interview.

Appendix 4:

Consent form for FabO IPA study



Sheffield Hallam University

Participant consent form

Study title:	A qualitative investigation into the experience of fabric orthoses in long-term users with multiple sclerosis.
Principal researcher:	Nicky Snowdon
Telephone number:	0114 225 5751

Participant name	

	Please read the following statements and put your initials in the box to show that you have read and understood them and that you agree with them	**Please initial each box**
1	I confirm that I have read and understood the information sheet for the above study. I have had the opportunity to consider the information, ask questions and have had these answered satisfactorily.	
2	I understand that my involvement in this study is voluntary and that I am free to withdraw at any time, without give any reason and without my care or legal rights being affected.	
3	I understand that the data collected during the study may be looked at by responsible individuals from the Sponsor of the research. I give permission for these individuals to have access to my records.	

To be filled in by the participant

I agree to take part in the above study

Your name	Date	Signature

Optional witness signature (if participant has difficulty writing)

To be filled in by the person obtaining consent

I confirm that I have explained the nature, purposes and possible effects of this research study to the person whose name is printed above.

Name of investigator	Date	Signature

Appendix 5
Topic Guide for IPA study

In the guide below, the bullet points are examples of optional prompts. The headings in bold should not be obvious to the interviewee but are included to illustrate the proposed structure of the interview.

“Thank you for participating in this interview. As we have discussed, I am hoping to learn all about how you came to use the orthosis, what you use it for and how you feel about it. Before we start, can I check that you are still happy to be interviewed and that you are happy for the interview to be recorded?"

Contextualisation

Could you start by telling about yourself and how you came to be using the orthosis?

- How has the MS affected you?
- How long have you had the orthosis?
- Is it your first orthosis or were you using something different before?
- Who suggested the orthosis and why?
- What made you want to try it?
- What were your hopes for the orthosis?
- How did that first experience feel? How important was it to you?
- Is there anything else you want to tell me about when you first tried the orthosis?

Please tell me more about the orthosis that you wear, for example, how often you wear it.

- What are your usual routines around wearing the garment? - for example, do you always wear it during the day or only for certain activities?
- Do you think other people can see a difference between you wearing the orthosis and not wearing the orthosis?

Apprehending the phenomenon

Please tell me what you can about what it is like to wear a fabric orthosis.

- How does it feel?
- How does it make you feel?
- Please think about the difference between you wearing the orthosis and you not wearing it. How different does this feel?

- What difference does it make to what you can do?
- What difference does it make to your life?
- You have mentioned a lot of positive effects, what to you are the disadvantages?
- What does this orthosis mean for you?
- Has the impact of the orthosis changed since you first started wearing it?
- Are there things that it enables you to do that you wouldn't be able to do otherwise?
- Does it change how you feel about your body?

Clarifying the phenomenon

You have said that the orthosis does….. if it didn't, would it still be worth while wearing it?

You have said… if that were different, for example, ….. would that change what the orthosis means to you?

Imagine that you someone with MS asked you for advice on whether they should try a fabric orthosis. What would you tell them? Would you tell them….? How important is that?

Closing comments

Is there anything more that you want to tell me about your experiences?

Thank you very much for your time today. It has been a really valuable experience for me and my research.

I shall be in touch again in a few months' time with a summary of my findings but in the meantime if you have any queries, do feel free to get in touch with me."

Appendix 6
Example coded transcript

Transcribed interview	Exploratory comments	Emergent themes	Final crosscutting themes
Participant: …even if I wasn't… wasn't trying to do something. So it was an ataxia but actually, I think just with being a 14 year old, and worrying and stressing, I would always try and hold my arm very close to… I would put my hand in my pocket, umm, just try and do whatever I could to cover the fact that I had this tremor.	"IT WAS AN ATAXIA **BUT"** - DOES SHE THINK THE WORRYING AND STRESSING MADE THIS WORSE? OR IS SHE AWARE THAT INTENTION TREMOR SHOULD GO AWAY WITH MOVEMENT? Sense of self/identity. "ATAXIA" - CHOICE OF MEDICAL TERMINOLOGY Hiding/covering the tremor Does the splint cover her embarrassment? It goes over the thing she wants to hide. She tried to hide the thing that made her look different to others. Does the feeling of exposure make the tremor worse or just make her feel exposed?	Tremor impacts your sense of self Desire for control	Impact of MS - loss of control
Researcher: when you say, do you mind, what sort of tremor was it? Was it just like…? Can you…? (laughs, mimes a large amplitude tremor)			
Participant: Well it was with the splint and it was much better than what it would have been like without. You see, at the time, I mean, now I can rest it and keep it still and it isn't an issue at all. So it is really the intention tremor is still there with a splint on but it is reduced. But without the splint, it would literally, go off and do… (gestures a shaking movement) And I wouldn't be able to have my hand by my side. It would be shaking and it would be moving. Not in a gentle shake, not like a Parkinson's shake, but it would be moving in a jerky way, all over the place. So that, obviously, was a huge issue. When I was under paediatrics, at the time, because the recovery had gone so well I was able to move around and walk. Actually the arm, the arm, the use of the left arm was actually the thing that was causing you know tremendous difficulty. So, umm, so what they actually suggested was to go forward to look at the Second Skin garment. Because at that time	"SPLINT" = SOMETHING DESIGNED TO PREVENT MOVEMENT Improvement over time - *she* has more control now. Splint reduces the tremor "GO OFF" = RELEASE, ESCAPE FROM CONTROL, LIKE AN EXPLOSION. Nature of tremor. Tremor was then the main problem she faced. "THE LEFT ARM" = DOESN'T SOUND LIKE PART OF HER Severity of the tremor - causing tremendous difficulty "ALL THAT WAS AVAILABLE" = MAKING DO WITH SOMETHING LESS THAN IDEAL	The nature of tremor Positive effects of the splint What it means to live with tremor	Regaining control - reclaiming my body Impact of MS - loss of control Impact of MS - dissociation of body from self Impact of MS - loss of

that was all that was available… I used to travel up and down to Birmingham and I used that. So that was the start of my journey… in terms of using a Lycra garment, and it did start to have an impact and it did reduce the tremor but I think obviously it is hard to remember that but at the time it had even more impact but if I try and think back to actually before I had it, it was obviously quite significant because it was literally uncontrollable.	Sense of a journey with the splint. "START TO HAVE", "REDUCE THE TREMOR" = DOESN'T SOUND VERY EFFECTIVE AT THE FIRST FEW ATTEMPTS. "LITERALLY UNCONTROLLABLE" = NO ABILITY TO CONTROL HER TREMOR AT ALL. What would it feel like to have a part of your body moving when you want it to be still? IT is part of you but completely outside your control.	The sense of a journey with the splint Getting to know the splint What it means to live with tremor	control Initial experiences - experimentation The sense of a journey with the orthosis Regaining control - reclaiming my body Impact of MS - loss of control
Researcher: Yes, gosh			
Participant: And ah, then obviously the move, it was only when I was in adult services, umm, and I think that was when (my physio) became involved. Obviously DMO were on the scene, because it was actually (a senior orthotist) who came to measure for my very first splint and I think I was pretty much one of the first people to have a Lycra glove.	"VERY FIRST" = SEEMS TO HAVE DISCOUNTED HER EXPERIENCES AS A CHILD Aware this was a bit experimental.	The sense of a journey with the splint Experimentation - might it work?	The sense of a journey with the orthosis Initial experiences - experimentation
Researcher: Oh right, OK. Yes.			
Participant: It was almost a bit of an experimental the thing on me. It was like, let's try it. You've been using… they knew that I had been using the Second Skin glove and obviously this was going to be a better option because it was British. They thought let's have a go with that because the Second Skin garment was a pain because it was from Australia. They had to send it off to be… sort of, if it needed any amendments. Or changes.. or re-measurements or anything. So it was, sort of, a lot of toing and froing. It was difficult but obviously I was younger at the time. I think my parents had just took me where I needed to go.	Definitely experimental! "LET'S HAVE A GO" = EXPERIMENT, LITTLE CONFIDENCE THAT IT WOULD WORK, EVEN FROM THE PROFESSIONALS, "LET'S" = SENSE OF EXPERIMENTING TOGETHER A lot of toing and froing, it was difficult - is this something that happened a lot? Is she implying this happened more with the early splints? Does it need to be more efficient for an adult? Would it be worth it for an adult?	Experimentation - might it work? Adapting the splint	Initial experiences - experimentation Compromising and adapting - adapting the orthosis
Researcher: so can you remember from back then, what your arm felt like without the Lycra garment and then what it felt like with the Lycra garment? Can you remember what that was like?			
Participant: I know that there were a lot of things that… I mean there are still a lot of things that I can't do… but there are a lot of things that I wouldn't even have attempted to do. I think that because obviously, because it was so many years ago… I think the main memories that I have, is it just the fact that it was the things like just walking with my	A possible change in her confidence to try things over the years? Earlier she wouldn't have even tried, she would have assumed she was unable to do things. Arm position is important - not just about keeping it still. BESIDE	Positive effects of the splint How the splint feels	Impact of MS - loss of control

arm beside me. So it was things like that… that was probably more of a significant issue in terms of reducing that tremor. I think, the only way that I can describe it is that by using the glove, straight away by using the glove, was actually feeling that someone had got hold of my arm. Because (my physio) used to be able to locate exactly where the point was.. where the tremor was coming from in my arm. So in actual fact, I always used to laugh to L(my physio) and say if only I could carry you around with me (laughs) and actually that has actually ended up happening. I don't need (my physio) to follow me everywhere, because I have got the Lycra glove…so I think it is just the reduction in the wildness of the intention tremor. So, I can get control of it much more with the glove on it. So, that sort of transition was quite a significant thing because it meant that I felt a lot more confident in myself considering that actually at the time I was much younger so I struggled with a lot of the normal issues that a teenager would have to deal with.	ME = SOUNDS LIKE A RELIABLE FRIEND NOW, STILL NOT QUITE PART OF HER. The glove had an Immediate effect. "GLOVE" = IS THERE ANY SIGNIFICANCE AS TO WHEN SHE SAYS GLOVE OR SPLINT? Her glove feels like a person, like a physiotherapist. "SOMEONE HAD GOT HOLD OF MY ARM" = SOMEONE ELSE TAKING CONTROL, RELIEVING HER, SHARING THE BURDEN OF CONTROL. The glove also gives her independence from needing a real person to help. The tremor has a specific source. "WILDNESS" = RANDOM, UNCONTROLLED, POSSIBLY FRIGHTENING "ICAN GET CONTROL" = THE GLOVE GIVES **HER** BACK HER CONTROL The glove allows her a change in her sense of self, "TRANSITION" = MAYBE NOT IMMEDIATE BUT OVER TIME	The nature of tremor Positive effects of the splint Tremor impacts your sense of self	Regaining control - reclaiming my body Getting to know my orthosis - my orthosis as a person Impact of MS - loss of controlRegaining control - reclaiming my body, reclaiming identity/maintaining self-image - how I see myself
Researcher: body image? [Generally confident?]			
Participant: I wanted to just to be normal and fit in and not thinking I can't go out places because of this. But it gave me a lot of confidence to go out to do things and more drive for the future really. So that was really positive.	Her tremor made her feel different from her peers Confidence to go out and do things = the main impact is on her confidence in herself and how she appears to others. "BE NORMAL" = DESIRE TO CONFORM, FELT ABNORMAL, MAYBE OUTCAST BECAUSE OF THE TREMOR Drive for the future - was the tremor impacting upon her concept of her own future? It wasn't just affecting her there and then but making it difficult to see a future for herself.	Tremor impacts your sense of self Positive effects of the splint	Regaining control - reclaiming identity/maintaining self-image - how orthotic use impacts identity

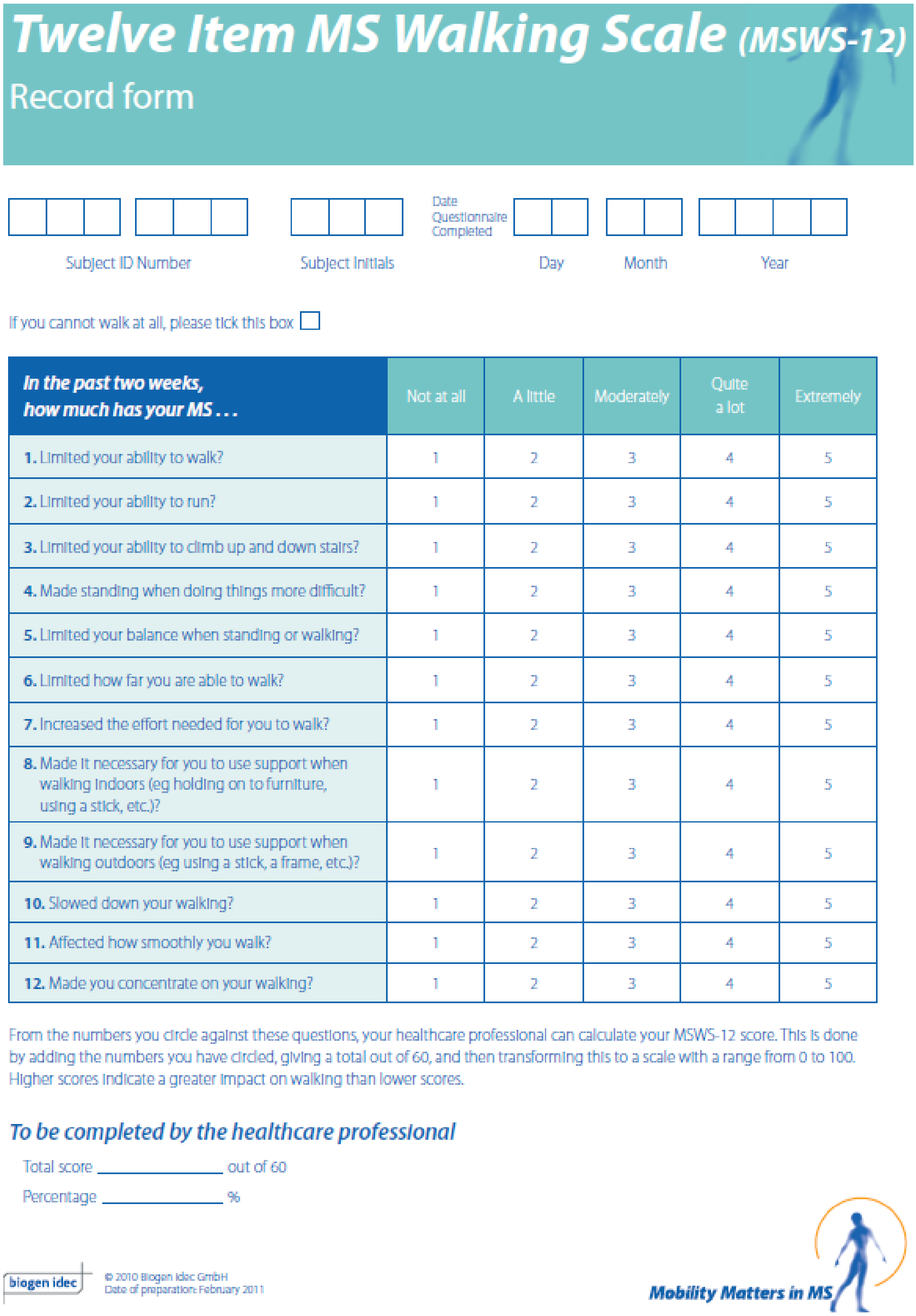
(Coloured highlights relate to the final cross-cutting themes in the right hand column. Normal text *= descriptive comments; BLOCK CAPITALS = linguistic comments;* Underlined *= conceptual/interpretive comments Names of the physiotherapist and orthotist have been removed to maintain confidentiality.)*

Appendix 7

Mapping between emergent themes and cross-cutting themes

	Final cross-cutting themes								Other emergent themes
	Reclaiming control				Learning to live with an orthosis				
	Life with MS	Reclaiming my body	Reclaiming autonomy	Maintaining self-image	Initial experimentation	Getting to know my orthosis	Compromising and adapting	Establishing routines	
Sarah emergent themes	Self-determination Desire for control Living with tremor What changes tremor? The nature of tremor	Importance of arm posture Positive effects of the splint	Practical difficulties with a tremor Positive effects of the splint	Tremor impacts your sense of self The importance of appearance to others Positive effects	Initial experimentation Sense of a journey	How my orthosis feels It's not a cure Orthosis as a treatment How does it work?	Weighing up Practicalities Positive and negative effects of the splint Adapting the splint Adapting yourself	Practicalities Sense of a journey Dependence on the splint	Why did tremor develop? Funding issues
Rebecca emergent themes	Control The nature of dystonia Triggers for dystonia	Control Positive and negative effects		The importance of appearance Identity/self-image Attitudes to coping strategies	Experimentation Alternative means of gaining control Low expectations from the orthosis Sense of a journey	Orthosis as feedback to prompt improvement How does it work? The importance of compression	Weighing up pros and cons Practicalities Positive effects Negative effects	Routines of wear Discontinued use Attitudes to coping strategies Practicalities	
Marion emergent themes	Life with MS An untrustworthy body	Importance of ankle stability Positive effects	A focus on function		Experimentation Problem-solving Unimportance of upper limb stability	How does it work? Features of a good splint Getting to know your splint	Too much support can weaken you Adapting the splint Negative and positive effects Practicalities	Routines of wear	Facing challenges with husband Disappointing outside help
David emergent themes	Life with MS Becoming diagnosed Indicators of deterioration Changeability Physical impact of MS	Core stability Positive effects	Importance of independence Positive effects Coping strategies & compensations The importance of walking	Importance of appearance Self-image and work identity Positive effects	Experimentation Initial expectations Influences of physiotherapists	Essential features of the shorts	Practicalities Positive effects of the shorts Adapting the shorts	Routines of wear	Limiting the impact of stress on MS Funding issues

Appendix 8
Self-report measures considered for or used in OSFeaMS study



Twelve Item MS Walking Scale (MSWS-12)

Record form

☐☐☐ ☐☐☐ Subject ID Number

☐☐☐ Subject Initials

Date Questionnaire Completed ☐☐ Day ☐☐ Month ☐☐☐☐ Year

If you cannot walk at all, please tick this box ☐

In the past two weeks, how much has your MS . . .	Not at all	A little	Moderately	Quite a lot	Extremely
1. Limited your ability to walk?	1	2	3	4	5
2. Limited your ability to run?	1	2	3	4	5
3. Limited your ability to climb up and down stairs?	1	2	3	4	5
4. Made standing when doing things more difficult?	1	2	3	4	5
5. Limited your balance when standing or walking?	1	2	3	4	5
6. Limited how far you are able to walk?	1	2	3	4	5
7. Increased the effort needed for you to walk?	1	2	3	4	5
8. Made it necessary for you to use support when walking indoors (eg holding on to furniture, using a stick, etc.)?	1	2	3	4	5
9. Made it necessary for you to use support when walking outdoors (eg using a stick, a frame, etc.)?	1	2	3	4	5
10. Slowed down your walking?	1	2	3	4	5
11. Affected how smoothly you walk?	1	2	3	4	5
12. Made you concentrate on your walking?	1	2	3	4	5

From the numbers you circle against these questions, your healthcare professional can calculate your MSWS-12 score. This is done by adding the numbers you have circled, giving a total out of 60, and then transforming this to a scale with a range from 0 to 100. Higher scores indicate a greater impact on walking than lower scores.

To be completed by the healthcare professional

Total score ______________ out of 60

Percentage ______________ %

Mobility Matters in MS

Activities-specific Balance Confidence Scale (ABC)

Participant Code Number:

Appointment number:

For each of the following activities, please indicate your level of self-confidence by choosing a corresponding number from the rating scale) 5 to 100%, with 0% meaning you have no confidence and 100% meaning you feel completely confident.

0%	10	20	30	40	50	60	70	80	90	100%
No confidence										**Completely confident**

How confident are you that you can maintain your balance and remain steady when you...

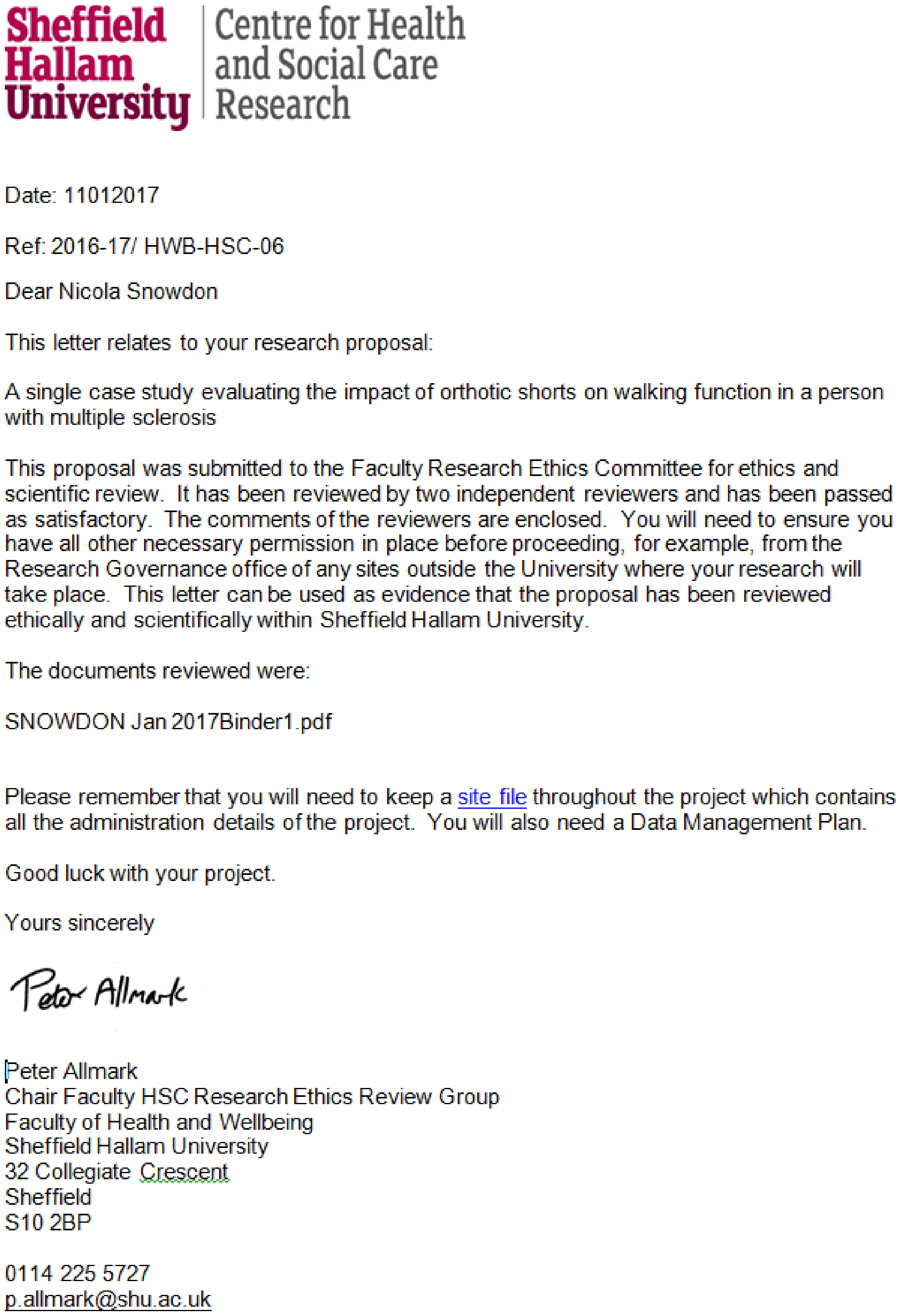
1. ...walk around the house? ____%
2. ...walk up or down stairs? ____%
3. ...bend over and pick up a slipper from the floor at the front of a cupboard? ____%
4. ...reach for a small tin of food from a shelf at eye level? ____%
5. ...stand on your tip toes and reach for something above your head? ____%
6. ...stand on a chair and reach for something? ____%
7. ...sweep the floor? ____%
8. ...walk outside the house to a parked car? ____%
9. ...get into or out of a car? ____%
10. ...walk across a car park to the shops? ____%
11. ...walk up or down a ramp? ____%
12. ...walk in a crowded shopping centre where people walk past you quickly? ____%
13. ...are bumped into by people as you walk through the shopping centre? ____%
14. ...step onto or off an escalator while holding onto the handrail? ____%
15. ...step onto or off an escalator while holding onto parcels, such that you cannot hold onto the handrail? ____%
16. ...walk outside on slippery pavements? ____%

FES-I

Now we would like to ask some questions about how concerned you are about the possibility of falling. Please reply thinking about how you usually do the activity. If you currently don't do the activity (e.g. if someone does your shopping for you), please answer to show whether you think you would be concerned about falling IF you did the activity. For each of the following activities, please tick the box which is closest to your own opinion to show how concerned you are that you might fall if you did this activity.

		Not at all concerned 1	*Somewhat concerned 2*	*Fairly concerned 3*	*Very concerned 4*
1	Cleaning the house (e.g. sweep, vacuum or dust)	*1* ☐	*2* ☐	*3* ☐	*4* ☐
2	Getting dressed or undressed	*1* ☐	*2* ☐	*3* ☐	*4* ☐
3	Preparing simple meals	*1* ☐	*2* ☐	*3* ☐	*4* ☐
4	Taking a bath or shower	*1* ☐	*2* ☐	*3* ☐	*4* ☐
5	Going to the shop	*1* ☐	*2* ☐	*3* ☐	*4* ☐
6	Getting in or out of a chair	*1* ☐	*2* ☐	*3* ☐	*4* ☐
7	Going up or down stairs	*1* ☐	*2* ☐	*3* ☐	*4* ☐
8	Walking around in the neighbourhood	*1* ☐	*2* ☐	*3* ☐	*4* ☐
9	Reaching for something above your head or on the ground	*1* ☐	*2* ☐	*3* ☐	*4* ☐
10	Going to answer the telephone before it stops ringing	*1* ☐	*2* ☐	*3* ☐	*4* ☐
11	Walking on a slippery surface (e.g. wet or icy)	*1* ☐	*2* ☐	*3* ☐	*4* ☐
12	Visiting a friend or relative	*1* ☐	*2* ☐	*3* ☐	*4* ☐
13	Walking in a place with crowds	*1* ☐	*2* ☐	*3* ☐	*4* ☐
14	Walking on an uneven surface (e.g. rocky ground, poorly maintained pavement)	*1* ☐	*2* ☐	*3* ☐	*4* ☐
15	Walking up or down a slope	*1* ☐	*2* ☐	*3* ☐	*4* ☐
16	Going out to a social event (e.g. religious service, family gathering or club meeting)	*1* ☐	*2* ☐	*3* ☐	*4* ☐

Appendix 9
Ethical approval letter for methods pilot



Sheffield Hallam University
Centre for Health and Social Care Research

Date: 11012017

Ref: 2016-17/ HWB-HSC-06

Dear Nicola Snowdon

This letter relates to your research proposal:

A single case study evaluating the impact of orthotic shorts on walking function in a person with multiple sclerosis

This proposal was submitted to the Faculty Research Ethics Committee for ethics and scientific review. It has been reviewed by two independent reviewers and has been passed as satisfactory. The comments of the reviewers are enclosed. You will need to ensure you have all other necessary permission in place before proceeding, for example, from the Research Governance office of any sites outside the University where your research will take place. This letter can be used as evidence that the proposal has been reviewed ethically and scientifically within Sheffield Hallam University.

The documents reviewed were:

SNOWDON Jan 2017Binder1.pdf

Please remember that you will need to keep a site file throughout the project which contains all the administration details of the project. You will also need a Data Management Plan.

Good luck with your project.

Yours sincerely

Peter Allmark

Peter Allmark
Chair Faculty HSC Research Ethics Review Group
Faculty of Health and Wellbeing
Sheffield Hallam University
32 Collegiate Crescent
Sheffield
S10 2BP

0114 225 5727
p.allmark@shu.ac.uk

Appendix 10
Participant Information Sheet for methods pilot

Sheffield Hallam University

Participant information sheet

A single case study evaluating the impact of orthotic shorts on walking function in a person with multiple sclerosis.

We would like to invite you to take part in our research study. Before you decide, we would like you to understand why the research is being done and what it would involve for you. Talk to others about the study if you wish. Ask us if there is anything that is not clear.

This research study will investigate the impact of orthotic shorts. These are close-fitting shorts, made from elastic material, and designed to provide stability around the hips and trunk. This single-case study will evaluate the effect of the shorts on your walking and the researchers will produce a report for you, which can be shared with your healthcare team.

What is the purpose of this study?

This study will trial the methods that we intend to use in a future larger study, in which we will investigate orthotic shorts in a larger number of people with multiple sclerosis. Before we start that study, we would like some feedback on whether we are planning on using too many measures and, if so, which measures seem to best capture the effect of the shorts. We would also like some feedback on the fit and comfort of the shorts.

In addition, this study will determine if the shorts have a measurable effect on your walking.

Why have I been invited?

You have been invited because you are someone with multiple sclerosis who has expressed interest previously in trialling orthotic shorts and determining if they show a measurable effect on your walking. Because you have prior experience of using orthotic shorts, you will not be eligible for our larger research study.

Do I have to take part?

Your decision to take part in this study is entirely voluntary. You may refuse to participate or you can withdraw from the study at any time. Your refusal to participate or wish to withdraw would not influence any future services you might receive from the NHS, private practitioners or your orthotics supplier.

What will happen to me if I take part?

If you take part, you will visit Sheffield Hallam University twice. At the first visit, we will discuss the study and you will be able to change some aspects of the study if you prefer. We will ask you to complete some questionnaires about the severity of your multiple sclerosis, your walking ability and your balance.

At that first visit, you will be assessed for your orthotic shorts by a representative from DM Orthotics. After that, you will be tested using a number of assessments of your walking ability, first wearing just your usual clothing, without your own supportive cycling shorts, and later wearing your supportive cycling shorts.

The measures that we will take involve you walking at different speeds, for different distances and both with and without speaking. We will be recording things like how fast and how far you walk, how you step, your heart rate and how much your trunk moves from side to side as you walk. Some of the assessments require equipment to be attached to your body.

On your second visit to Sheffield Hallam, you will be provided with the orthotic shorts. We will repeat the walking assessments from the first day, first without any shorts and then with the orthotic shorts.

After, this second visit you will be given the shorts to wear at home and we will ask you to record when you wear the shorts and what activities you do in them. You will wear the shorts for only an hour on the first day and gradually increase wear over the first five days.

There will be a small waterproofed monitor in the shorts that will record movement. We want to test that monitor to see if it can tell us when the shorts are being worn. It only measures changes in movement speed and direction; there is no location device within the monitor.

When you have worn the shorts for at least a couple of weeks, we will arrange to see you again. This can be in your own home or anywhere else you prefer, so long as it is reasonably quiet and private. We will interview you about your experiences with the shorts and with the study itself. We will also ask you to complete questionnaires about your walking ability and your balance whilst you were wearing the orthotic shorts.

What are the possible risks and disadvantages of taking part?

The visits to Sheffield Hallam may take up to three hours for each visit. There will be refreshments provided and chance to rest between different tests but it is likely that you will find the testing procedures quite tiring. Because the testing will involve a lot of walking, there is a chance that you will be sore and tired the next day as well.

During the walking tests, there is a small risk of falling. However, you will be supervised by an experienced neurological physiotherapist who will be on hand to steady you if required. If you do not want to complete all the walking tests, then you do not have to.

There may be some disadvantages in trialling the orthotic shorts at home. For example, they can be difficult to get on and take off. However, if you do find them difficult you are not obliged to wear them. We would like to know any feedback on your experiences, whether these are positive or negative.

There is a risk that discussing your experiences will be upsetting for you. If this is the case, the researcher is an experienced neurological physiotherapist and will be able to discuss any issues arising during the study. However, we would not be able to provide longer term support and if this was required we would help you find someone local to you who could provide support.

Are there any benefits to taking part?

We hope that there will be benefits to you in to different ways. If the shorts are helpful to you, then you may keep them for longer-term use after the study. In addition, we can prepare a report on our objective findings that would be given to you and that you may share with your healthcare team. If the shorts are helpful, then this may help you to gain funding for further pairs in the future. You will not be directly identifiable in this report but it can be accompanied by a letter to your healthcare team if required.

What if there is a problem or I wish to complain?

If you have any queries or questions please contact:

Principal investigator: Nicky Snowdon, e-mail n.snowdon@shu.ac.uk and phone 0114 225 5751.

Alternatively, you can contact my supervisor: Sionnadh McLean, e-mail s.mclean@shu.ac.uk and phone 0114 225 2271.

If you would rather contact someone independent of the study, you can contact Dr. Nikki Jordan-Mahy, Chair of the Faculty Research Ethics Committee, e-mail: n.jordan-mahy@shu.ac.uk and phone 0114 225 3120.

How will you ensure that my taking part in the study will be kept confidential?

The testing and the interview will take place in a quiet, private area. All data collected will not have your name attached but just a code identifying you.

The documents relating to the administration of this research, such as the consent form you sign to take part, will be kept in a folder called a Site File. This is locked away securely. The folder might be checked by people in authority who want to make sure that researchers are following the correct procedures. These people will not pass on your details to anyone else.

What will happen to the results of this study?

As described earlier, we are happy to prepare a report for you on the effect of the shorts on your walking ability that you can share with your healthcare team. In addition, we may prepare a report for publication in a research journal of your experiences with the shorts and the effect of the shorts on your walking.

The study will be reported to staff at DM Orthotics, the company who will be providing the shorts for the study, as a verbal presentation and/or a written report.

The data that we collect will be stored safely and anonymously. Your data may be used by other researchers for future projects if this is considered to be of benefit to the wider community and if you explicitly consent to this. Your personal details will not be made available for future projects.

Who is sponsoring this study?

The sponsor of the study has the duty to ensure that it runs properly and that it is insured. In this study, the sponsor is Sheffield Hallam University.

Who has reviewed this study?

All research based at Sheffield Hallam University is looked at by a group of people called a Research Ethics Committee. This Committee is run by Sheffield Hallam University but its members are not connected to the research they examine. The Research Ethics Committee has reviewed this study and given a favourable opinion.

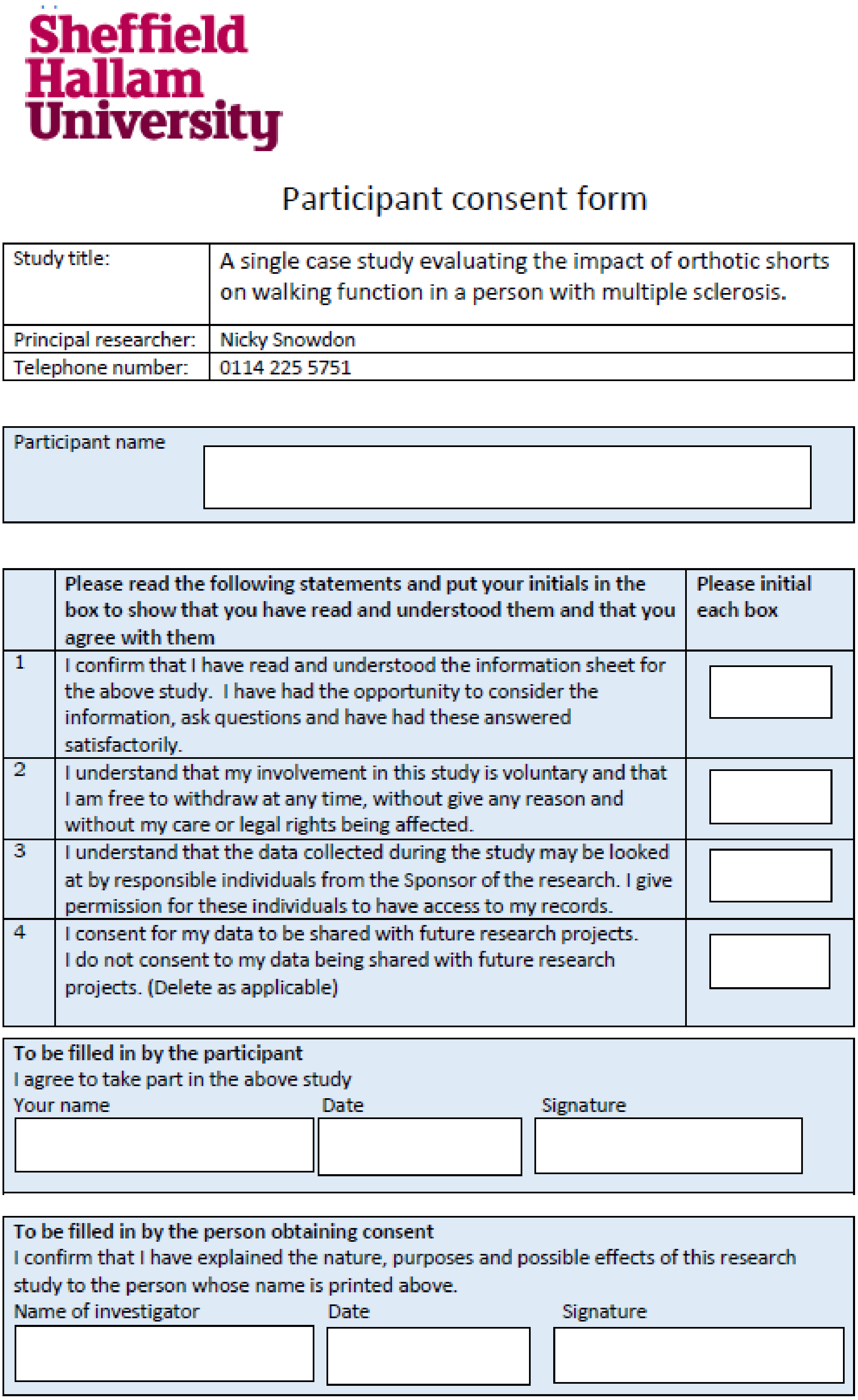
I would like to participate in this study, what do I do next?

Please contact the researcher directly to arrange to start the study, either by e-mail or telephone.

Principal investigator: Nicky Snowdon,
E-mail n.snowdon@shu.ac.uk and phone 0114 225 5751 or 07906 33 4097
We will have a telephone conversation to answer any queries that you may have and to arrange dates for the visits to Sheffield Hallam. The researcher will contact you in writing by e-mail to confirm the times and travel arrangements.

Appendix 11

Consent Form for methods pilot



Sheffield Hallam University

Participant consent form

Study title:	A single case study evaluating the impact of orthotic shorts on walking function in a person with multiple sclerosis.
Principal researcher:	Nicky Snowdon
Telephone number:	0114 225 5751

Participant name

	Please read the following statements and put your initials in the box to show that you have read and understood them and that you agree with them	Please initial each box
1	I confirm that I have read and understood the information sheet for the above study. I have had the opportunity to consider the information, ask questions and have had these answered satisfactorily.	☐
2	I understand that my involvement in this study is voluntary and that I am free to withdraw at any time, without give any reason and without my care or legal rights being affected.	☐
3	I understand that the data collected during the study may be looked at by responsible individuals from the Sponsor of the research. I give permission for these individuals to have access to my records.	☐
4	I consent for my data to be shared with future research projects. I do not consent to my data being shared with future research projects. (Delete as applicable)	☐

To be filled in by the participant

I agree to take part in the above study

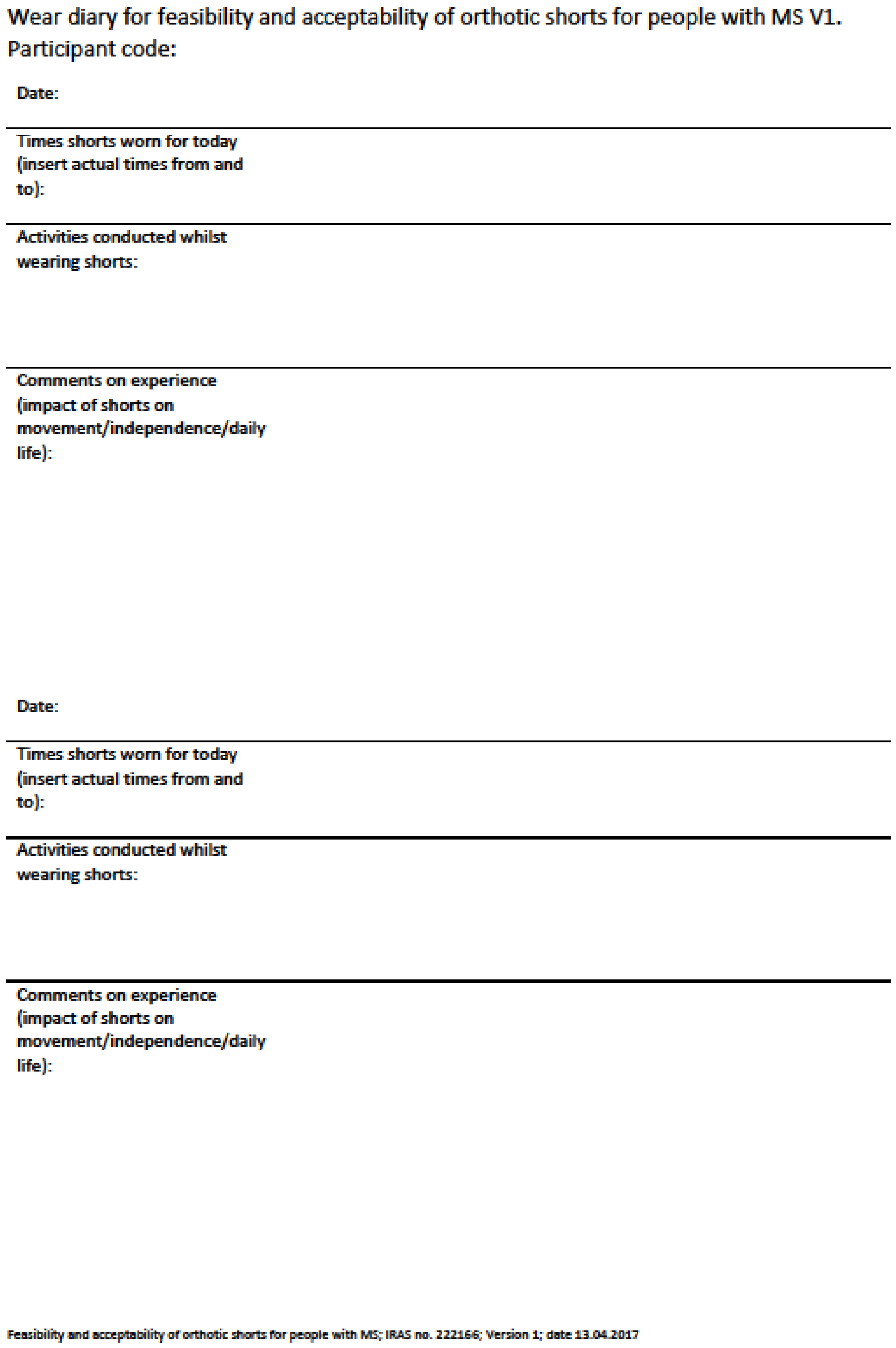
Your name	Date	Signature

To be filled in by the person obtaining consent

I confirm that I have explained the nature, purposes and possible effects of this research study to the person whose name is printed above.

Name of investigator	Date	Signature

Appendix 12
Wear diary



Wear diary for feasibility and acceptability of orthotic shorts for people with MS V1.
Participant code:

Date:

Times shorts worn for today (insert actual times from and to):

Activities conducted whilst wearing shorts:

Comments on experience (impact of shorts on movement/independence/daily life):

Date:

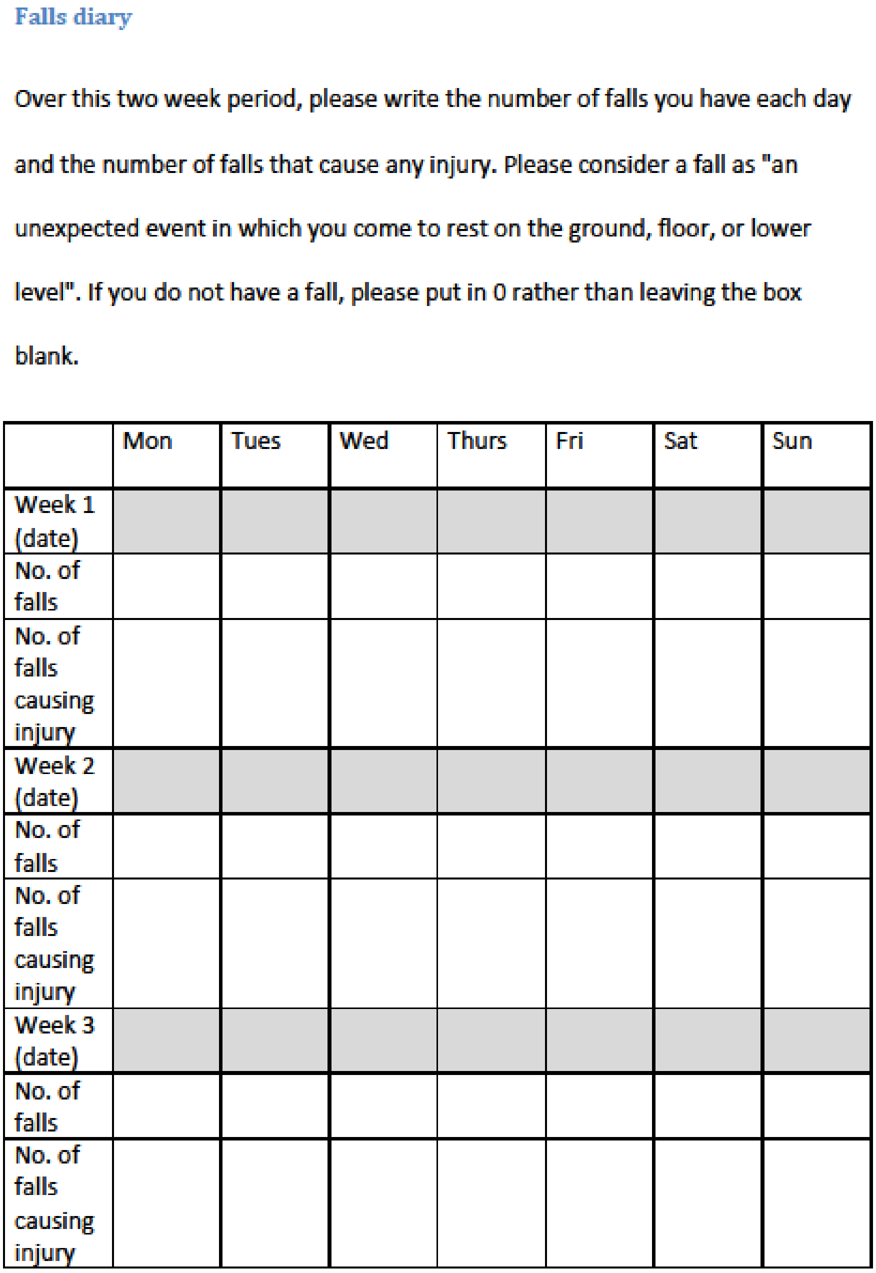
Times shorts worn for today (insert actual times from and to):

Activities conducted whilst wearing shorts:

Comments on experience (impact of shorts on movement/independence/daily life):

Feasibility and acceptability of orthotic shorts for people with MS; IRAS no. 222166; Version 1; date 13.04.2017

Appendix 13
Falls diary

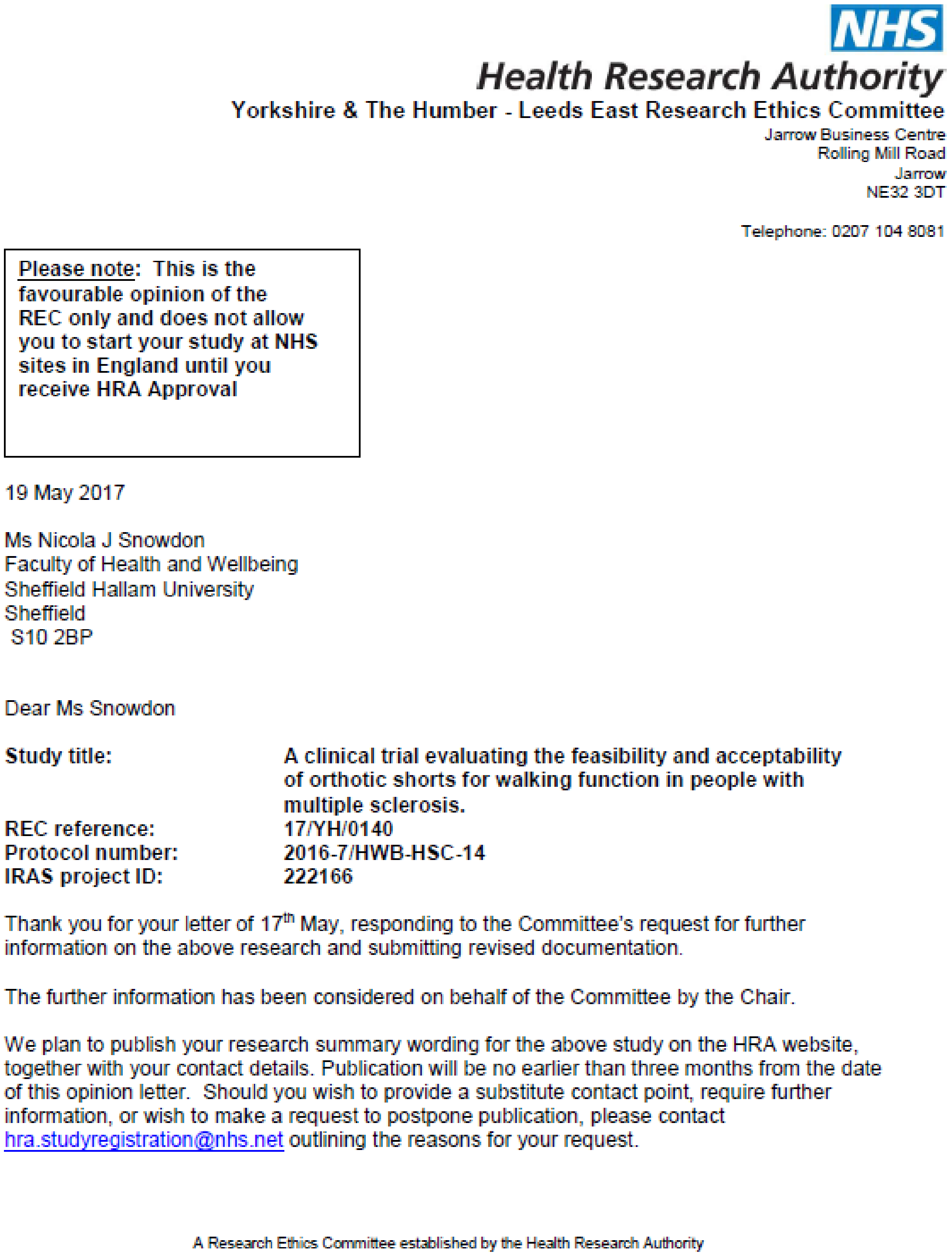


Falls diary

Over this two week period, please write the number of falls you have each day and the number of falls that cause any injury. Please consider a fall as "an unexpected event in which you come to rest on the ground, floor, or lower level". If you do not have a fall, please put in 0 rather than leaving the box blank.

	Mon	Tues	Wed	Thurs	Fri	Sat	Sun
Week 1 (date)							
No. of falls							
No. of falls causing injury							
Week 2 (date)							
No. of falls							
No. of falls causing injury							
Week 3 (date)							
No. of falls							
No. of falls causing injury							

Appendix 14
Approval letters from Research Ethics Committee and Health Research Authority for OSFeaMS study



NHS
Health Research Authority
Yorkshire & The Humber - Leeds East Research Ethics Committee
Jarrow Business Centre
Rolling Mill Road
Jarrow
NE32 3DT

Telephone: 0207 104 8081

<u>Please note</u>: **This is the favourable opinion of the REC only and does not allow you to start your study at NHS sites in England until you receive HRA Approval**

19 May 2017

Ms Nicola J Snowdon
Faculty of Health and Wellbeing
Sheffield Hallam University
Sheffield
S10 2BP

Dear Ms Snowdon

Study title: **A clinical trial evaluating the feasibility and acceptability of orthotic shorts for walking function in people with multiple sclerosis.**
REC reference: **17/YH/0140**
Protocol number: **2016-7/HWB-HSC-14**
IRAS project ID: **222166**

Thank you for your letter of 17th May, responding to the Committee's request for further information on the above research and submitting revised documentation.

The further information has been considered on behalf of the Committee by the Chair.

We plan to publish your research summary wording for the above study on the HRA website, together with your contact details. Publication will be no earlier than three months from the date of this opinion letter. Should you wish to provide a substitute contact point, require further information, or wish to make a request to postpone publication, please contact hra.studyregistration@nhs.net outlining the reasons for your request.

A Research Ethics Committee established by the Health Research Authority

Confirmation of ethical opinion

On behalf of the Committee, I am pleased to confirm a favourable ethical opinion for the above research on the basis described in the application form, protocol and supporting documentation as revised, subject to the conditions specified below.

Conditions of the favourable opinion

The REC favourable opinion is subject to the following conditions being met prior to the start of the study.

Management permission must be obtained from each host organisation prior to the start of the study at the site concerned.

Management permission should be sought from all NHS organisations involved in the study in accordance with NHS research governance arrangements. Each NHS organisation must confirm through the signing of agreements and/or other documents that it has given permission for the research to proceed (except where explicitly specified otherwise).

Guidance on applying for NHS permission for research is available in the Integrated Research Application System, www.hra.nhs.uk or at http://www.rdforum.nhs.uk.

Where a NHS organisation's role in the study is limited to identifying and referring potential participants to research sites ("participant identification centre"), guidance should be sought from the R&D office on the information it requires to give permission for this activity.

For non-NHS sites, site management permission should be obtained in accordance with the procedures of the relevant host organisation.

Sponsors are not required to notify the Committee of management permissions from host organisations

Registration of Clinical Trials

All clinical trials (defined as the first four categories on the IRAS filter page) must be registered on a publically accessible database within 6 weeks of recruitment of the first participant (for medical device studies, within the timeline determined by the current registration and publication trees).

There is no requirement to separately notify the REC but you should do so at the earliest opportunity e.g. when submitting an amendment. We will audit the registration details as part of the annual progress reporting process.

To ensure transparency in research, we strongly recommend that all research is registered but for non-clinical trials this is not currently mandatory.

If a sponsor wishes to request a deferral for study registration within the required timeframe, they should contact hra.studyregistration@nhs.net. The expectation is that all clinical trials will be registered, however, in exceptional circumstances non registration may be permissible with prior agreement from the HRA. Guidance on where to register is provided on the HRA website.

It is the responsibility of the sponsor to ensure that all the conditions are complied with before the start of the study or its initiation at a particular site (as applicable).

Ethical review of research sites

NHS sites

The favourable opinion applies to all NHS sites taking part in the study, subject to management permission being obtained from the NHS/HSC R&D office prior to the start of the study (see "Conditions of the favourable opinion" below).

Non-NHS sites

The Committee has not yet completed any site-specific assessment (SSA) for the non-NHS research site(s) taking part in this study. The favourable opinion does not therefore apply to any non-NHS site at present. We will write to you again as soon as an SSA application(s) has been reviewed. In the meantime no study procedures should be initiated at non-NHS sites.

Approved documents

The final list of documents reviewed and approved by the Committee is as follows:

Document	*Version*	*Date*
Copies of advertisement materials for research participants [Flyer for advertising study]	V1	13 April 2017
Covering letter on headed paper [Covering letter]		
Covering letter on headed paper [Covering letter]	V2	17 May 2017
Evidence of Sponsor insurance or indemnity (non NHS Sponsors only) [Letter regarding professional indemnity]		
Instructions for use of medical device [Wear protocol for participants]	V1	13 April 2017
Interview schedules or topic guides for participants [Topic guides for both interviews]	V1	18 April 2017
IRAS Application Form [IRAS_Form_19042017]		19 April 2017
IRAS Application Form XML file [IRAS_Form_19042017]		19 April 2017
IRAS Checklist XML [Checklist_17052017]		17 May 2017
MHRA Notice of No Objection Letter (Medical Devices) and relevant correspondence [Communication with MHRA]		15 May 2017
Other [Initial Assessment Information for REC]	1	25 April 2017
Participant consent form [Revised consent form]	V2	16 May 2017
Participant consent form [Consent form with marked changes]		16 May 2017
Participant information sheet (PIS) [Revised Information sheet]	V3	13 May 2017
Participant information sheet (PIS) [Information sheet with marked changes]		13 May 2017
Referee's report or other scientific critique report [SHU letter of approval]		28 March 2017
Referee's report or other scientific critique report [SHU scientific		28 March 2017

review]		
Referee's report or other scientific critique report [Protocol at date of SHU review]		28 March 2017
Research protocol or project proposal	V4	13 May 2017
Sample diary card/patient card [Wear diary]	V1	13 April 2017
Sample diary card/patient card [Data collection record sheets]	V2	18 April 2017
Summary CV for Chief Investigator (CI) [CV for NSnowdon]		03 February 2017
Summary CV for supervisor (student research) [CV Sionnadh McLean]		
Summary CV for supervisor (student research) [CV Jon Wheat]		
Summary, synopsis or diagram (flowchart) of protocol in non technical language [Flow chart for each participant]	V1	26 February 2017
Validated questionnaire [Activities-Specific Balance Confidence measure]		
Validated questionnaire [Multiple sclerosis walking scale]		

Statement of compliance

The Committee is constituted in accordance with the Governance Arrangements for Research Ethics Committees and complies fully with the Standard Operating Procedures for Research Ethics Committees in the UK.

After ethical review

Reporting requirements

The attached document *"After ethical review – guidance for researchers"* gives detailed guidance on reporting requirements for studies with a favourable opinion, including:

- Notifying substantial amendments
- Adding new sites and investigators
- Notification of serious breaches of the protocol
- Progress and safety reports
- Notifying the end of the study

The HRA website also provides guidance on these topics, which is updated in the light of changes in reporting requirements or procedures.

User Feedback

The Health Research Authority is continually striving to provide a high quality service to all applicants and sponsors. You are invited to give your view of the service you have received and the application procedure. If you wish to make your views known please use the feedback form available on the HRA website: http://www.hra.nhs.uk/about-the-hra/governance/quality-assurance/

HRA Training

We are pleased to welcome researchers and R&D staff at our training days – see details at http://www.hra.nhs.uk/hra-training/

17/YH/0140 Please quote this number on all correspondence

With the Committee's best wishes for the success of this project.

Yours sincerely

pp

Dr Rhona Bratt
Chair

Email:nrescommittee.yorkandhumber-leedseast@nhs.net

Enclosures: "After ethical review – guidance for researchers" *[SL-AR2]*

Copy to:
Dr Keith Fildes, Sheffield Hallam University
Ms Aimee Card, Sheffield Teaching Hospitals NHS Trust

NHS
Health Research Authority

Ms Nicola J Snowdon
Faculty of Health and Wellbeing
Sheffield Hallam University
Sheffield
S10 2BP

Email: hra.approval@nhs.net

11 December 2017

Dear Ms Snowdon,

Letter of HRA Approval

Study title:	**A clinical trial evaluating the feasibility and acceptability of orthotic shorts for walking function in people with multiple sclerosis.**
IRAS project ID:	**222166**
Protocol number:	**2016-7/HWB-HSC-14**
REC reference:	**17/YH/0140**
Sponsor	**Sheffield Hallam University**

I am pleased to confirm that HRA Approval has been given for the above referenced study, on the basis described in the application form, protocol, supporting documentation and any clarifications noted in this letter.

Participation of NHS Organisations in England

The sponsor should now provide a copy of this letter to all participating NHS organisations in England.

Appendix B provides important information for sponsors and participating NHS organisations in England for arranging and confirming capacity and capability. **Please read *Appendix B* carefully**, in particular the following sections:

- *Participating NHS organisations in England* – this clarifies the types of participating organisations in the study and whether or not all organisations will be undertaking the same activities
- *Confirmation of capacity and capability* - this confirms whether or not each type of participating NHS organisation in England is expected to give formal confirmation of capacity and capability. Where formal confirmation is not expected, the section also provides details on the time limit given to participating organisations to opt out of the study, or request additional time, before their participation is assumed.
- *Allocation of responsibilities and rights are agreed and documented (4.1 of HRA assessment criteria)* - this provides detail on the form of agreement to be used in the study to confirm capacity and capability, where applicable.

Further information on funding, HR processes, and compliance with HRA criteria and standards is also provided.

It is critical that you involve both the research management function (e.g. R&D office) supporting each organisation and the local research team (where there is one) in setting up your study. Contact details and further information about working with the research management function for each organisation can be accessed from www.hra.nhs.uk/hra-approval.

Appendices

The HRA Approval letter contains the following appendices:

- A – List of documents reviewed during HRA assessment
- B – Summary of HRA assessment

After HRA Approval

The document *"After Ethical Review – guidance for sponsors and investigators"*, issued with your REC favourable opinion, gives detailed guidance on reporting expectations for studies, including:

- Registration of research
- Notifying amendments
- Notifying the end of the study

The HRA website also provides guidance on these topics, and is updated in the light of changes in reporting expectations or procedures.

In addition to the guidance in the above, please note the following:

- HRA Approval applies for the duration of your REC favourable opinion, unless otherwise notified in writing by the HRA.
- Substantial amendments should be submitted directly to the Research Ethics Committee, as detailed in the *After Ethical Review* document. Non-substantial amendments should be submitted for review by the HRA using the form provided on the HRA website, and emailed to hra.amendments@nhs.net.
- The HRA will categorise amendments (substantial and non-substantial) and issue confirmation of continued HRA Approval. Further details can be found on the HRA website.

Scope

HRA Approval provides an approval for research involving patients or staff in NHS organisations in England.

If your study involves NHS organisations in other countries in the UK, please contact the relevant national coordinating functions for support and advice. Further information can be found at http://www.hra.nhs.uk/resources/applying-for-reviews/nhs-hsc-rd-review/.

If there are participating non-NHS organisations, local agreement should be obtained in accordance with the procedures of the local participating non-NHS organisation.

User Feedback

The Health Research Authority is continually striving to provide a high quality service to all applicants and sponsors. You are invited to give your view of the service you have received and the application procedure. If you wish to make your views known please use the feedback form available on the HRA website: http://www.hra.nhs.uk/about-the-hra/governance/quality-assurance/.

HRA Training

We are pleased to welcome researchers and research management staff at our training days – see details at http://www.hra.nhs.uk/hra-training/

Your IRAS project ID is **222166**. Please quote this on all correspondence.

Yours sincerely,

Steph Blacklock
Senior Assessor

Email: hra.approval@nhs.net

Copy to: *Dr Keith Fildes*
Ms Aimee Card, Sheffield Teaching Hospitals NHS Trust

Appendix 15
Flier advertising OSFeaMS study

Sheffield
Hallam
University

Participants required for research

Can orthotic shorts help people with multiple sclerosis (MS) to walk?

We are looking for people with MS:

- who have some difficulty walking,
- who feel "unstable" and "wobbly" around their hips or trunk and
- who can trial some shorts and give us feedback.

The study involves having two pairs of shorts made to measure, having your walking tested in a number of ways, wearing the shorts at home for 2 weeks each and being interviewed about your experiences. If the shorts are helpful, you can keep them after the study.

Testing takes place at Sheffield Hallam University at Collegiate Crescent. We can help with transport costs, if you live within 10 miles.

For further information please contact either:

Nicky Snowdon, Lecturer in Physiotherapy, Sheffield Hallam University.
0114 225 5751
n.snowdon@shu.ac.uk

Lee Drake, Clinical Specialist Physiotherapist in Multiple Sclerosis, Sheffield Teaching Hospitals.
0114 271 3090

Feasibility and acceptability of orthotic shorts for people with MS; IRAS no. 222166; Version 1; date 13.04.2017.

Appendix 16
Participant Information Sheet for OSFeaMS study

Sheffield Hallam University

Participant Information Sheet

A clinical trial evaluating the feasibility and acceptability of orthotic shorts for walking function in people with multiple sclerosis.

1. Introduction and purpose of the study

We would like to invite you to take part in our research study. Before you decide, we would like you to understand why the research is being done and what it would involve for you. Talk to others about the study if you wish. Ask us if there is anything that is not clear.

The first part of this information sheet will tell you why we are doing this study. We will then explain what will happen to you if you take part. Then we will give you some supporting information about the study.

This research study will investigate the impact of two types of shorts. These close-fitting, elasticated shorts are designed to provide stability around the hips and trunk. They are usually worn beneath normal clothing.

What is the purpose of the study?

We want to find out whether orthotic shorts improve walking ability in people with multiple sclerosis (MS). Walking can be slow and unsteady in people with MS. Support around the hips might make walking smoother and steadier.

Orthotic shorts are similar to the tight clothing worn to improve performance in athletes. Research suggests that such shorts help people who have had a stroke to walk faster. Some people with MS find that similar garments improve coordination and stability. Currently, some people with MS get funding for garments such as these to be provided on the NHS. However, there is no research investigating whether they work in people with MS.

This research study has two aims. Firstly, we want to know whether the shorts might improve walking. If they do, we want to know how best to measure this. Secondly, we want to know whether the shorts are acceptable to people with MS. In other words, will people choose to wear them or are tight shorts impractical in daily life?

Why have I been invited?

We are looking for people with MS who have difficulty walking and feel unsteady around their hips or lower trunk. You need to be able to walk for at least two minutes in order to do the walking tests. You need to be able to travel to Sheffield Hallam University, where the testing will take place. Your MS needs to be stable. If you are not stable at the moment, you could join the study at a later time.

Do I have to take part?

Your decision to take part in this study is entirely voluntary. You may refuse to participate or you can withdraw from the study at any time. Your refusal to participate or wish to withdraw would not influence any future services you might receive.

2. What the study involves

What will happen to me if I take part?

If you take part, you will try out two different pairs of orthotic shorts that will be made-to-measure for you. The two pairs of shorts differ in the degree of support that they provide. One pair is tighter than the other.

You will test the shorts by wearing them during formal assessments of your walking ability and by wearing them at home. This will involve you visiting Sheffield Hallam University on four occasions and us visiting you three times. When we visit you, this can be at your home or, if you prefer, we can meet in another quiet and private location. Overall, your involvement in the study would last approximately 12 weeks.

The formal assessments of your walking ability capture common difficulties that people experience with MS. They are:

- How fast you can walk over a short distance
- How hard you have to concentrate when you walk (how much you slow down when you have to speak at the same time)
- Exactly how you take steps (for example, step length and how far apart your feet are when you walk)
- How variable your walking is from one step to the next and
- How steady your hips and trunk are when you walk.

At university, one measure will involve attaching two small movement sensors to your body.

We will interview you twice about your experiences. There will be a 20-30 minute interview at the first appointment to explore your current challenges and your expectations of the shorts. After you have tested both pairs of shorts at home, a longer interview will discuss your experiences. We also ask you to complete questionnaires a number of times: one about your walking and one about your balance.

We ask that you trial each pair of shorts at home for two weeks. After each home trial, there will be two weeks where you do not wear the shorts and do not attend any appointments.

As mentioned above, the study will require seven appointments:

- Appointment 1 will be at Sheffield Hallam University. We will discuss the study and you would sign a consent form to confirm your involvement. We will interview you about your current challenges and your expectations. We will ask you to complete questionnaires about your walking and your balance. You will be measured for your shorts, by an employee from the company who makes the shorts. This will be a healthcare professional, either a physiotherapist or occupational therapist.

- Appointment 2: the person who measured you for the shorts will visit you at home to check that both pairs of shorts fit you. We will measure how tight the shorts are and then take them away for any adjustments.

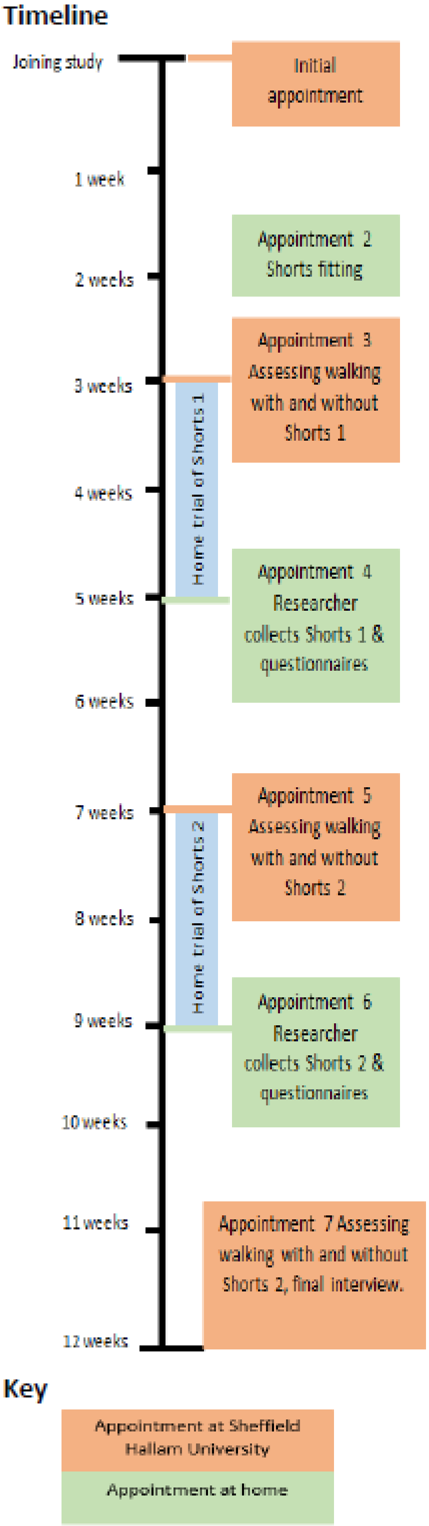
- Appointment 3: you will come to Sheffield Hallam University. We will assess your walking with and without one of the pairs of shorts using the measures described earlier. We will decide which shorts you will test first and which you will test at Appointment 5. After this appointment, you will take the shorts home and wear them at home.

- Appointment 4: about 2 weeks later, a researcher will visit you at home. We will collect the shorts. You will complete questionnaires about your walking and your balance whilst you were wearing the shorts.

- Appointment 5: you will come to Sheffield Hallam University and we will assess your walking in the same way as at Appointment 3. This time you will try out the second pair of shorts. Once again, you will take the shorts away and try them out at home.

- Appointment 6: about 2 weeks later, a researcher will visit you at home. We will collect the shorts and the questionnaires about your walking and balance whilst wearing the shorts.

- Appointment 7: For this last appointment, you will come to Sheffield Hallam University. We will assess your walking one last time, without any shorts. This is to check whether your walking has changed during the study. We will then interview you about your experiences with the shorts and with the study as a whole.



How would I wear the shorts at home?

You should wear the shorts for only one hour on your first day with them at home. You should double how long you wear them each day as follows:

- First day – 1 hour of wear
- 2nd day – 2 hours
- 3rd day – 4 hours
- 4th day – 8 hours
- 5th day and thereafter – all day.

If the shorts become uncomfortable, you must contact the research team for advice. They should not be worn in bed.

We would like you to wear the shorts for a range of activities, particularly when you are walking and standing. The shorts may be more useful in some activities than others. If you find it impractical to wear the shorts all day or every day, you are not obliged to wear them.

We will ask you to keep a diary of when you wear the shorts at home. You can use the diary to record what you did whilst wearing the shorts and how they felt. We would like to know any feedback on your experiences, whether these are positive or negative.

Most people like to wear these shorts next to the skin but over normal underwear. If you sometimes need to use the toilet urgently, we can adapt the shorts to include a "toileting hole". You would wear these adapted shorts underneath your underwear and would not need to remove the shorts to use the toilet.

What are the possible risks and disadvantages of taking part?

The main disadvantage is the time that the study will take up. Each visit to Sheffield Hallam University will last 1 ½ to 2 hours, plus your travel time. You may find the visits tiring. Each batch of walking tests will take about 30 minutes, including a chance to rest every 2-5 minutes. If you are not used to a lot of walking, you may find that you are tired or sore the next day. If you do not want to complete all the walking tests, then you do not have to.

Our visits to your home will last about 30 minutes each.

During the walking tests, there is a small risk of falling. However, you will be supervised by an experienced neurological physiotherapist who will be on hand to steady you if required.

There may be some disadvantages in trialling the shorts at home. You might need to change your usual routines, for example to wash the shorts. There is a risk you will find the shorts uncomfortable or difficult to get on and off. You may find you need help from someone else to get dressed and undressed, even if usually you do not need help. We will ask for feedback on these types of issues as part of the interviews.

Are there any benefits to taking part?

The shorts might help your walking and your balance. If they are helpful, then you may keep them as long as you wish after the study. We would not be able to fund additional pairs of shorts in the future but we can provide you with information about how to purchase them.

By participating in this study, you will be helping us to contribute towards our understanding of walking in MS. If we demonstrate that the shorts are effective, they may become more readily available for others.

3. Supporting information about the study

Can I bring someone with me when I visit the university?

You can bring a friend or family member with you when you come to the university. You may need some help to change into your shorts on Appointments 3 and 5. The research team will be happy to help but you may prefer to have someone you know instead.

How will I get to the university?

We are happy to pay for a return taxi fare for you, if you live within 10 miles. Alternatively, if you are able to drive or be brought in, we have disabled parking immediately outside the building. The testing will take place at the Robert Winston Building at 11-15 Broomhall Road in Sheffield, S10 2BP, which is at the Collegiate Crescent Campus.

How will you ensure that my taking part in the study will be kept confidential?

The testing and the interview will take place in a quiet area in the university. Sometimes there may be other participants present in the testing area at the same time as yourself. There will be screens or toilet facilities where you may change.

All data collected will not have your name attached but just a code identifying you. You will not be identifiable in any reports. The recordings of your interviews will be kept securely, typed out by the Chief Investigator and deleted following the study.

Documents relating to the administration of this research, such as the consent form you sign, will be kept in a folder called a Site File. This is locked away securely. The folder might be checked by people in authority who want to make sure that researchers are following the correct procedures. These people will not pass on your details to anyone else.

How long will this study last?

Your individual involvement in the study will last approximately 12 weeks. We intend to recruit 16 people to the study. The study commenced in January 2018 and we estimate that it will finish in February 2019.

If you join this study, we will keep your contact details for 12 months and may use these to invite you to a follow-up study if you continue to use the shorts. Your contact details will be destroyed 12 months after study completion.

What will happen to the results of this study?

We will prepare a report for publication in a research journal so that people can benefit from what we learn. We will share the findings at professional conferences and at events for people with MS, such as those organised by the MS Society UK. If you would like to be sent a report, please let us know.

The data that we collect will be stored safely and anonymously. At the end of the study, we plan to make some data openly available. It may be used by other researchers and by the company who manufacture these shorts. The interview recordings and transcripts will not be shared, to ensure that you are not identifiable. You will be asked to consent specifically to your data being shared with others; you do not have to consent to this.

Who is organising and funding this study?

The study is organised by the Department of Allied Health Professions at Sheffield Hallam University in conjunction with the Therapy Services Team at Sheffield Teaching Hospitals. The project is part of a programme of doctoral research for the Chief Investigator.

The sponsor of the study has the duty to ensure that it runs properly and that it is insured. In this study, the sponsor is Sheffield Hallam University.

Who has approved this study?

All research in the NHS is reviewed by an independent group of people, called a Research Ethics Committee, which is there to protect your safety, rights, dignity and wellbeing. This project has been reviewed and was given a favourable option by the Leeds East Research Ethics Committee.

Who do I contact if I have any concerns?

If you have any concerns or complaints about anything to do with this study then you can contact the Chief Investigator, Nicky Snowdon:
E-mail n.snowdon@shu.ac.uk and phone 0114 225 5751.

Alternatively, you can contact the main project supervisor: Dr Sionnadh McLean
E-mail s.mclean@shu.ac.uk and phone 0114 225 2271.

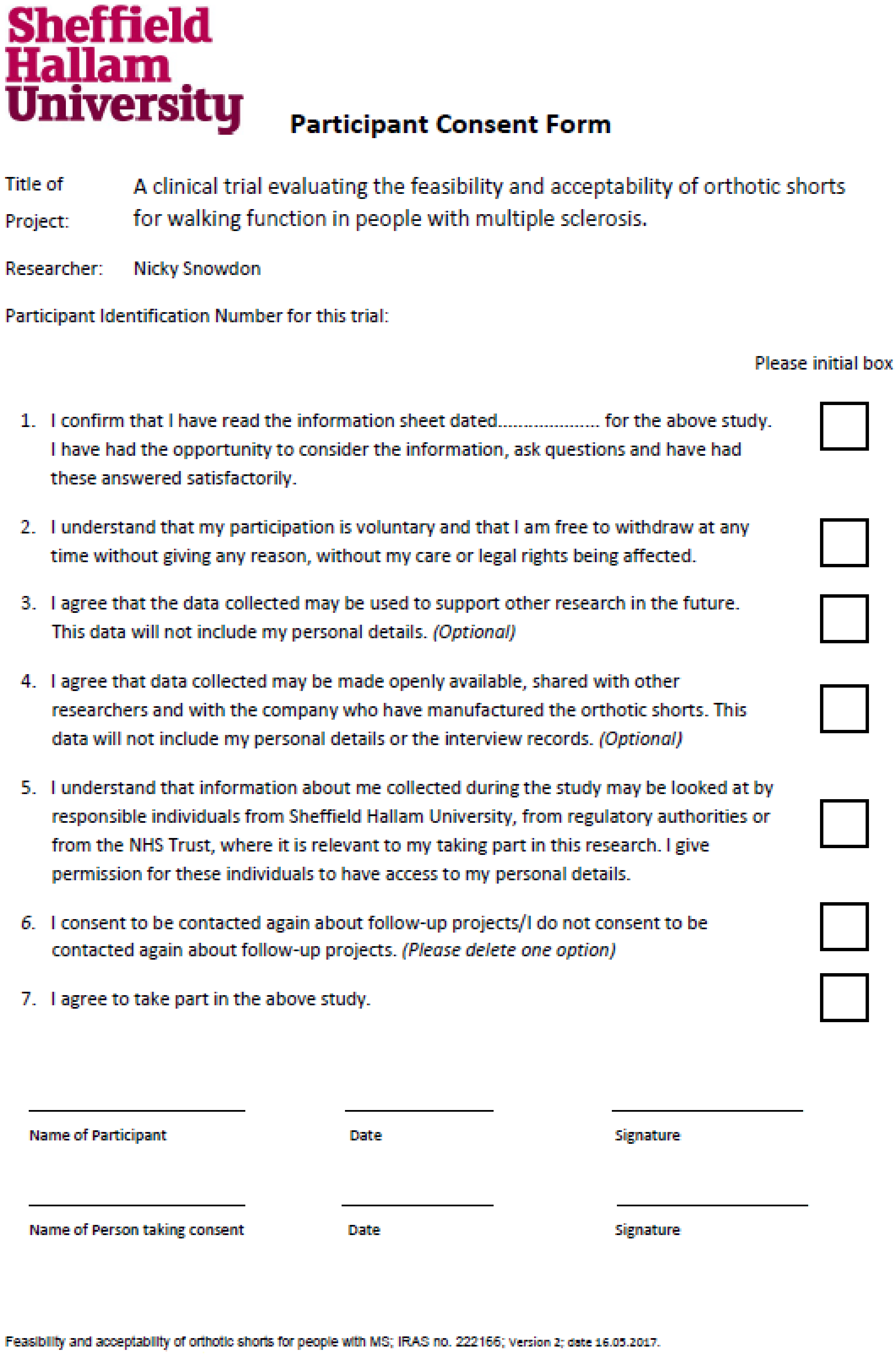
If you would rather contact someone independent of the study, you can contact Dr. Nikki Jordan-Mahy, Chair of the Faculty Research Ethics Committee.
E-mail: n.jordan-mahy@shu.ac.uk and phone 0114 225 3120.

I would like to participate in this study, what do I do next?

Please contact the researcher directly to arrange to start the study, either by e-mail or telephone.
Chief investigator: Nicky Snowdon,
E-mail n.snowdon@shu.ac.uk and phone 0114 225 5751

We will have a telephone conversation to answer your queries. We will check that you meet the study criteria, described above under “Why have I been invited?” We will then arrange provisional dates for the visits. The researcher will contact you in writing by post or e-mail, to confirm the times and travel arrangements.

Appendix 17
Consent form for OSFeaMS study



Sheffield Hallam University

Participant Consent Form

Title of Project: A clinical trial evaluating the feasibility and acceptability of orthotic shorts for walking function in people with multiple sclerosis.

Researcher: Nicky Snowdon

Participant Identification Number for this trial:

Please initial box

1. I confirm that I have read the information sheet dated.................... for the above study. I have had the opportunity to consider the information, ask questions and have had these answered satisfactorily. ☐
2. I understand that my participation is voluntary and that I am free to withdraw at any time without giving any reason, without my care or legal rights being affected. ☐
3. I agree that the data collected may be used to support other research in the future. This data will not include my personal details. *(Optional)* ☐
4. I agree that data collected may be made openly available, shared with other researchers and with the company who have manufactured the orthotic shorts. This data will not include my personal details or the interview records. *(Optional)* ☐
5. I understand that information about me collected during the study may be looked at by responsible individuals from Sheffield Hallam University, from regulatory authorities or from the NHS Trust, where it is relevant to my taking part in this research. I give permission for these individuals to have access to my personal details. ☐
6. I consent to be contacted again about follow-up projects/I do not consent to be contacted again about follow-up projects. *(Please delete one option)* ☐
7. I agree to take part in the above study. ☐

Name of Participant	Date	Signature
Name of Person taking consent	Date	Signature

Feasibility and acceptability of orthotic shorts for people with MS; IRAS no. 222166; Version 2; date 16.05.2017.

Appendix 18

Topic guides for OSFeaMS study: initial interview, final interview and example of a personalised final interview topic guide

Initial interview regarding motivations for trialling shorts

Aim and context of interview

This interview will take place immediately following the informed consent process. The aim of this interview is to explore participants' initial views so that these can be compared to their experiences at the end of the trial, their wear routines and their choice of whether to continue use following the initial 12-week study period. Some of the questions are designed to ascertain the participants' "readiness for change" by asking whether they have already contemplated adaptations to their activities and lifestyles. The data from this interview will enable investigation of whether certain aims, motivations or perceptions impact upon the eventual acceptability of the shorts. In turn, this may enable us to develop advice for potential future users and funders of orthotics regarding factors that predict continued use. This initial interview is likely to last 20 - 40 minutes.

Topic guide

Many thanks for being interviewed. As I have said, we will be talking about your expectations of these shorts and of the wider study. Can I just double check that you are OK that I am recording the interview?

1. So firstly, could you tell me why you are interested in participating in this study?
    - Is there anything about trialling the shorts themselves that appeals to you?
    - Do you think the shorts might help with anything in particular?

2. Have you tried any other possible solutions for managing that problem/those problems prior to joining this study?
    - For example, physiotherapy?
    - Any other aids?
    - Any adaptations to your activities?
    - How did you get on with those other things that you have tried?
    - Are there any other ways of overcoming those problems that you have been considering?

3. Do you have previous experience of using a splint or a support of any kind?
    - For example, an ankle-foot orthosis or “Foot-Up” device? If so, how did you get on with those orthoses?
    - Have those experiences impacted upon your expectations of these shorts?

4. Can you tell me a little about your current day-to-day routines?
    - For example, what is a typical day like for you?
    - What activities might you do in your spare time and at work?
    - Are there any activities that you find particularly difficult at the moment?

5. What are you expecting the shorts might be like?
    - Do you think they might make a difference to what you are able to do?
    - Walking or balance?
    - Independence?
    - Specific participation goals?

Aim and context of final interview

This interview aims to determine the acceptability of the shorts and participants' views on the study processes such as the information and support they received, and the outcome measures used.

The interview will take place at the final visit to Sheffield Hallam (Visit number 4). At this point they will have experienced three assessments of their walking ability; the home trials of using the shorts and the final wash-out period. At this interview, the interviewer can be "unblinded" to when the different shorts were worn. The interviewer will review the data from the initial interview, the wear diaries and the Participant Global Rating of Change scores prior to this interview and will use this to inform the questions that are asked. This will ensure the interviews are focussed on aspects that are important to each participant. This final interview is likely to last 40 - 60 minutes.

Topic Guide

Many thanks for being interviewed. As I have said, we will be talking about your experiences of wearing the shorts, your feelings about whether to wear them in the future and your feedback on being in the study. Can I just double check that you are OK that I am recording the interview?

1. Please tell us a little about the differences between the two pairs of shorts that you have used.
    - How different did they feel?
    - In what way did they differ?
    - Immediate effect and day-to-day effect?

2. When we first talked at the start of this study, you explained that one of the main things that you have difficulty with were …
    - Can you tell me whether either of the shorts had any impact on this?
    - How did the shorts compare to ... (the other things that had been tried previously)?

3. Also, when we first talked, you were thinking that the shorts might help you to achieve…
    - Can you tell me whether either of the shorts had any impact on this?

4. Was there anything that the shorts helped with that you weren't expecting?
    - Movement control?
    - Fatigability?
    - Stability?
    - Any specific functions or activities that you found easier with the shorts?

  - Was there anything you could do with the shorts that you would struggle to do without the shorts?

5. Did the impact of the shorts change over time?
    - For example, as you got used to the shorts did you find it any easier to move in them?
    - Was there any particular point at which you felt the shorts made (the most /a) difference?

6. Overall, did the shorts meet your expectations or were you at all disappointed with their effect?
    - Do you wish we had discussed your expectations more at the start of the study?

7. What were the disadvantages of using the shorts?
    - Getting them on and off?
    - Getting to the toilet?
    - Washing and drying?
    - Restriction of movement?
    - Discomfort?
    - Appearance?
    - Was there anything that you were unable to do with the shorts on that you can usually do without the shorts?

8. Can you tell us more about your experiences of wearing the shorts?
    - When you wore the shorts at home, how did you get on with wearing them for the time periods that we recommended?
    - What do you think about the recommendations we gave you for wearing the shorts?
        - Were they helpful?
        - Were they achievable in your lifestyle?
        - Did you think the gradual increase in wear over the first few days was important?
    - Are there ways in which you have adapted your routines in order to wear the shorts?
        - Getting up earlier?
        - Changing your daily activities?
        - Getting help with dressing or undressing?

9. If someone with MS asked for your opinion on whether orthotic shorts might help them, what would you say?

10. Now that this part of the study is complete, do you want to keep either of the pairs of shorts to wear again in the future?
    - Which pair – or both pairs?

    a. If participants want to continue using shorts:
    - How do you think you will use the shorts in the future?
        - How often might you wear them?
        - Any specific activities – or times of day?

        - Are there particular situations when you think you will wear them – or when you probably wouldn't wear them?
    - Can you explain what the shorts do for you that make you want to continue to use them for these activities?
    - Do you think that when you wear the shorts for longer periods of time, that this might have any impact on your underlying strength and ability?
    - If we were able to adapt these shorts for you, is there anything that you would change?

b. If participants do not want to continue wearing shorts:

- What are the most important factors that have made you choose not to wear the shorts? (use previous comments from earlier in the interview as prompts).
- If we were able to adapt the shorts in any way, would that make you any more likely to want to continue to wear them?

Thank you for explaining your experiences with the shorts. It would be really helpful for us to understand your experiences with the wider research project as well. This will help us to design future studies as well as to help us understand what advice to give people who want to try these shorts in the future.

First, I have some questions about the information we gave you at the start of the study.

11. Is there anything that you think you could have been told at the start of the study that would have helped you better understand what the shorts might be like – or what we were asking you to do?
    - Can you think of anything that you wish you had known at the beginning of the study?
    - Would further information about the shorts themselves have been helpful? If so, what.

Secondly, it is really important for us to understand how you felt about all the measures that we took during the study.

12. Did you feel over-burdened by the number of measures that we used?
    - In the movement laboratory, we measured how fast you could walk, whether your walking was slowed by talking and thinking as you walked and exactly how you were walking.
    - Whilst you trialled the shorts at home, we asked you about the impact of MS on your walking ability, your confidence with your balance and whether you were having any falls.

13. Did any of the measures feel particularly relevant to you?
    - For example, did any of the measures capture the most important ways in which MS changes your ability?
    - Did any of the measures capture the effect of the shorts particularly well?
    - Which, if any of the measures seemed the most important to you?
    - If we were to remove any of these measures in future studies, which would you suggest is least useful?
    - Is there anything you think we should have measured but didn't?

14. That is all extremely helpful. Is there anything else that you would like to tell me that I have not asked you about?

15. If the nature of the placebo shorts has not already come up in the interview, this will be explained at the end. For example:
    - We were not really expecting the looser shorts to make any difference to your walking. They had been designed as a sham or placebo intervention to help us test the effect of the close-fitting shorts. We did this because there was a risk that comparing the shorts to no intervention at all might exaggerate how useful the shorts were. Do you have any thoughts on the use of the loose shorts?

Thank you for your time.

Example of modifications made to the topic guide for the final interview

The earlier part of the topic guide was altered, whereas, the later questions, not reproduced below, remained as per the topic guide provided earlier. In this example, the text in italics was added to the Topic Guide in response to the initial interview, the wear diary and the initial Global Rating of Change recorded when the shorts were first trialled.

Topic Guide

Many thanks for being interviewed. As I have said, we will be talking about your experiences of wearing the shorts, your feelings about whether to wear them in the future and your feedback on being in the study. Can I just double check that you are OK that I am recording the interview?

1. Please tell us a little about the differences between the two pairs of shorts that you have used.
    - How different did they feel?
    - In what way did they differ?
    - Immediate effect and day-to-day effect? *For first 8 days with first pair, felt no difference but after that comments include "better movement", "bit better stability" (at football match), "more stability" (? also at football). Similar with second pair of shorts, no positive comments for first 6 days, then "maybe felt a bit more stable" (work and gym), "felt more stable, slightly easier moving about", "give me a bit more confidence moving about", "more confidence, more stability, easier movement", "felt more stable, more support to legs and waist", "more stability, easier movement round house and gym".*
    - *Maybe compare the difference between the above and his description of strapping his legs to play football - are the shorts not as tight as this? Was he expecting the shorts to be tighter?*

2. When we first talked at the start of this study, you explained that one of the main things that you have difficulty with w*as your balance and your walking. You were hoping the*

shorts might be supportive and might help you to walk more fluently and less slowly and might mean that you fall less.
  - Can you tell me whether either of the shorts had any impact on this?
  - How did the shorts compare to… *the shorts you bought to use in the gym? Using the walking stick? Increasing the Baclofen dose? Being on the Fampyra? Exercising on your wobble board? Strengthening your core at the gym? Seeing the chiropractor?*

3. Was there anything that the shorts helped with that you weren't expecting?
    - *The tightness in your legs.*
    - *The pains in your legs.*
    - Movement control?
    - Fatigability?
    - Any specific functions or activities that you found easier with the shorts?
    - Was there anything you could do with the shorts that you would struggle to do without the shorts?

4. Overall, did the shorts meet your expectations or were you at all disappointed with their effect?
    - Do you wish we had discussed your expectations more at the start of the study?

5. What were the disadvantages of using the shorts?
    - Getting them on and off?
    - Getting to the toilet?
    - Washing and drying?
    - Restriction of movement?
    - Discomfort?
    - Appearance?
    - Was there anything that you were unable to do with the shorts on that you can usually do without the shorts?

6. Can you tell us more about your experiences of wearing the shorts?
    - When you wore the shorts at home, how did you get on with wearing them for the time periods that we recommended? *(He didn't wear them very long - wore first pair for 70% of the time recommended and second pair 63% of the time recommended. Tending to take them off at 4-5pm ish.) Were they uncomfortable for wearing in the evening?*
    - What do you think about the recommendations we gave you for wearing the shorts?
        - Were they helpful?
        - Were they achievable in your lifestyle?
        - Did you think the gradual increase in wear over the first few days was important?
    - Are there ways in which you have adapted your routines in order to wear the shorts?
        - Getting up earlier?
        - Changing your daily activities?
        - Getting help with dressing or undressing?

Appendix 19
Thematic framework from OSFeaMS study

1. Theoretical Framework of Acceptability

- 1.1. Intervention coherence
  - 1.1.1 Core stability and support
  - 1.1.2 Previously trialled shorts
  - 1.1.3 Intervention coherence - other

- 1.2. Ethicality
  - 1.2.1 Try anything
  - 1.2.2 The importance of exercise and links with the shorts
  - 1.2.3 Low-risk, non-pharmaceutical, holistic approach
  - 1.2.4 Assistance that is not visible to others

- 1.3. Perceived effectiveness

  - 1.3.1 Reclaiming my body
    - 1.3.1.1 Walking ability
      - 1.3.1.1.1 Control over leg direction
      - 1.3.1.1.2 Walking further, faster, smoother, easier
      - 1.3.1.1.3 Walking in unpredictable circumstances
      - 1.3.1.1.4 Shorts compared to walking aids
    - 1.3.1.2 Posture
    - 1.3.1.3 Strength, sit to stand to sit, squats and lunges
    - 1.3.1.4 More benefit when you are more active
    - 1.3.1.5 Improved feedback
    - 1.3.1.6 Decreased pain/ache
    - 1.3.1.7 Other advantages
    - 1.3.1.8 What the shorts don't do

  - 1.3.2 Reclaiming autonomy
    - 1.3.2.1 Improved function in standing
    - 1.3.2.2 Improved confidence
    - 1.3.2.3 Secure, stable, strong, supported, looked after
    - 1.3.2.4 Doing more

  - 1.3.3 Managing self-image
    - 1.3.3.1 Appearance of walking
    - 1.3.3.2 How I feel about walking aids
    - 1.3.3.3 Appearance of the shorts and what they represent

- 1.4. Burden
  - 1.4.1 Tightness and comfort/discomfort
  - 1.4.2 Movement restriction
  - 1.4.3 Heat
  - 1.4.4 Dressing and undressing
  - 1.4.5 Toileting and managing the toileting hole
  - 1.4.6 Laundering the shorts
  - 1.4.7 Other aspects of burden

- 1.5. Affective attitude

- 1.6. Future intentions
  - 1.6.1 Shorts as therapy
  - 1.6.2 Future use

2. Feedback on research

- 2.1 Measures
  - 2.1.1 General comments on measurement
  - 2.1.2 What to add
  - 2.1.3 Dual task cost
  - 2.1.4 MSWS-12
  - 2.1.5 ABC-UK
  - 2.1.6 Falls diary
  - 2.1.7 Wear diary

- 2.2 Information
  - 2.2.1 Information provided
  - 2.2.2 The risk of too much information
  - 2.2.3 Gradual increase in wear times

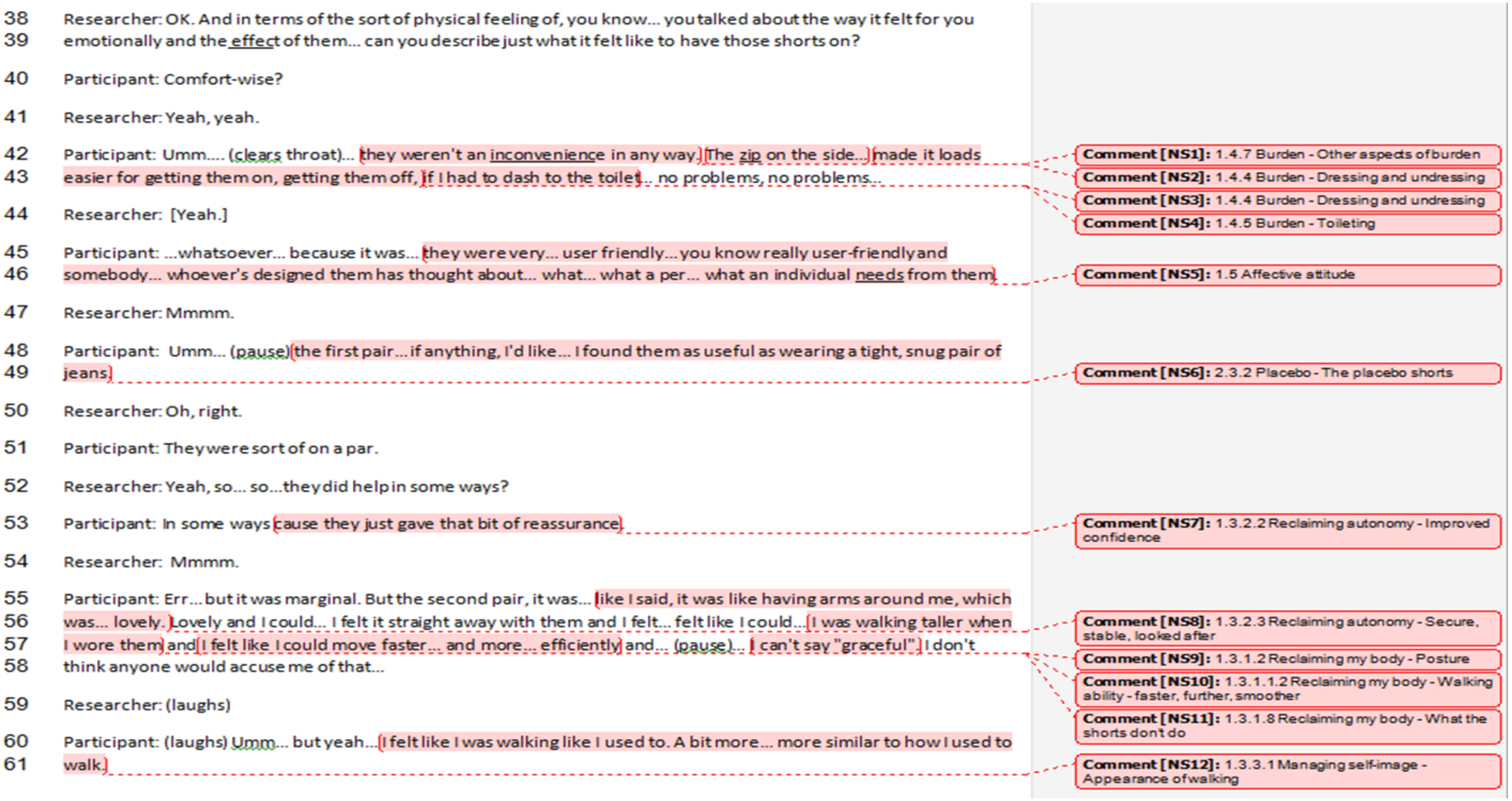
- 2.3 Placebo
  - 2.3.1 The concept of placebo
  - 2.3.2 The placebo shorts

- 2.4 Adapting the shorts

- 2.5 Study design and timeliness
  - 2.5.1 Period of time for trialling the shorts
  - 2.5.2 The crossover design

Appendix 20

Example of an indexed transcript from the OSFeaMS study



Researcher: OK. And in terms of the sort of physical feeling of, you know... you talked about the way it felt for you emotionally and the effect of them... can you describe just what it felt like to have those shorts on?

Participant: Comfort-wise?

Researcher: Yeah, yeah.

Participant: Umm.... (clears throat)... they weren't an inconvenience in any way. The zip on the side... made it loads easier for getting them on, getting them off, if I had to dash to the toilet... no problems, no problems...

Researcher: [Yeah.]

Participant: ...whatsoever... because it was... they were very... user friendly... you know really user-friendly and somebody... whoever's designed them has thought about... what... what a per... what an individual needs from them.

Researcher: Mmmm.

Participant: Umm... (pause) the first pair... if anything, I'd like... I found them as useful as wearing a tight, snug pair of jeans.

Researcher: Oh, right.

Participant: They were sort of on a par.

Researcher: Yeah, so... so...they did help in some ways?

Participant: In some ways cause they just gave that bit of reassurance.

Researcher: Mmmm.

Participant: Err... but it was marginal. But the second pair, it was... like I said, it was like having arms around me, which was... lovely. Lovely and I could... I felt it straight away with them and I felt... felt like I could... I was walking taller when I wore them and I felt like I could move faster... and more... efficiently and... (pause)... I can't say "graceful". I don't think anyone would accuse me of that...

Researcher: (laughs)

Participant: (laughs) Umm... but yeah... I felt like I was walking like I used to. A bit more... more similar to how I used to walk.

Comment [NS1]: 1.4.7 Burden - Other aspects of burden

Comment [NS2]: 1.4.4 Burden - Dressing and undressing

Comment [NS3]: 1.4.4 Burden - Dressing and undressing

Comment [NS4]: 1.4.5 Burden - Toileting

Comment [NS5]: 1.5 Affective attitude

Comment [NS6]: 2.3.2 Placebo - The placebo shorts

Comment [NS7]: 1.3.2.2 Reclaiming autonomy - Improved confidence

Comment [NS8]: 1.3.2.3 Reclaiming autonomy - Secure, stable, looked after

Comment [NS9]: 1.3.1.2 Reclaiming my body - Posture

Comment [NS10]: 1.3.1.1.2 Reclaiming my body - Walking ability - faster, further, smoother

Comment [NS11]: 1.3.1.8 Reclaiming my body - What the shorts don't do

Comment [NS12]: 1.3.3.1 Managing self-image - Appearance of walking

Appendix 21

Theoretical Framework of Acceptability applied to individual participants' perceptions

	Affective attitude	Burden	Perceived effectiveness	Future intentions	Summary of acceptability
Caroline	Liked the shorts.	*"Fitted in"*, comfortable, dressing and undressing easy.	Improved posture and support, walking looked better and more controlled. Less fatigued.	Walking and going to the gym.	Acceptable
Frank	Liked simplicity and ease of use.	Dressing took practice but otherwise no burden perceived.	Improved posture, stability, standing up, lifting feet better, stood longer, walked further, *"energised"*	All day, every day.	Acceptable
Jon	Liked the shorts: *"brilliant", "fantastic", "spot on"*.	Stride shortened, difficulties with heat, uncomfortable squatting. No problems with stairs, dressing or undressing.	Improved control over leg direction and walking; especially uneven ground, in dark and turning corners	Walking and going to the gym.	Acceptable
Kathy	Liked the shorts a lot.	Difficulties with heat, limited walking speed and lifting legs going up stairs. No problems dressing, undressing, toileting.	Convinced shorts were effective, numerous examples of activities easier with the shorts, including walking without rollator.	Going to work (everyday) and going to the gym.	Acceptable
Melissa	Liked the shorts a lot.	No perceived discomfort, restriction or inconvenience. Only disadvantage was appearance of shorts.	Convinced shorts were effective, walking further, posture improved, more stable, fewer near falls	All day, every day.	Acceptable
Natalie	Liked the shorts, *"Oh, I'm glad these are back on"*	Shorts comfortable when active, dressing and undressing not difficult, irritating when sitting still.	Convinced shorts helped, perceived legs stronger, able to run faster in the shorts.	All day, every day.	Acceptable
Oliver	Liked the shorts.	Comfortable, no problems dressing, undressing, toileting. Restricted flexion with sit-ups.	*"Definitely helped me".* Numerous examples of activities that felt easier with the shorts.	All day, every day.	Acceptable
Erica	Clearly disliked shorts: *"pretty disgusting", "big knickers".*	Uncomfortable; didn't restrict movement; dressing, undressing and toileting took longer.	Did not find them effective.	Did not keep shorts after the study.	Not acceptable.
Gwen	Clearly disliked shorts: *"bloody things".*	Restrictive and uncomfortable, increased ankle swelling, difficult to get dressed and to climb stairs.	Felt more supported and confident: *"very, very marginal".*	Did not keep shorts after the study.	Not acceptable.
Helen	Appeared ambivalent: *"disappointed".*	Easy to wear, except some restriction lifting legs, felt hot, felt dressing, undressing, and toileting more effortful.	Only noticed benefit once with feeling of fatigue in prolonged standing. No other benefits noted.	Did not keep shorts after the study.	Not acceptable.
Alison	Appeared ambivalent.	Lifting legs felt restricted, pressed on bladder, dressing easy but toileting harder.	Felt improved posture and housework easier but limited impact on her legs, which were more important.	Housework, exercise routine at home.	Questionable acceptability
Ben	Language appeared negative: " *heavy"* ,"*dumbbells*"	Shorts made lifting legs harder, difficulty bending, shorts fell down from above waist, difficulty dressing, undressing and toileting, one episode of incontinence.	Felt improved confidence, balance and posture. Felt knee gave way less often in shorts. *"They did a lot for me."*	Longer walks, trips to shopping centre.	Questionable acceptability
Dawn	Felt *"positive"* about the shorts.	Feedback contradictory around whether they were restrictive or not, no problems toileting, some difficulty dressing, left imprints on skin.	Unsure about effectiveness. Possibly reticent to share opinions due to uncertainty about the placebo shorts.	Would continue to experiment.	Questionable acceptability
Ingrid	Negative language: *"spazzy shorts" " big underwear"*	No problems with movement restriction, dressing and undressing. Shorts fell down from above waist, uncomfortable when resting.	Unsure about effectiveness. Felt better in the shorts but no impact on pain or fatigue. "*Is mine in me head? That it helped me or not?"*	Housework and days out.	Questionable acceptability
Linda	Appeared ambivalent.	Little burden with placebo shorts but burden with orthotic shorts: lifting legs difficult, going up stairs and bending down harder, dressing and undressing slower.	Felt benefit with the placebo shorts but unable to give examples. *"That extra thing. I've got nothing else"*	Exercising, going for a walk, shopping.	Questionable acceptability

Appendix 22

A summary of additional learning points from the OSFeaMS study informing future research

Method	Learning point	Justification
Eligibility criteria	Exclude people with history of ankle swelling	Adverse reaction to shorts in one participant who had a prior history of ankle swelling
	Exclude people with severe, uncompensated distal weakness.	Shorts might prevent proximal compensations for loss of distal function, thus decreasing independence.
Collaborative assessment process	Discuss whether participants think of the shorts as "sports shorts" or supportive underwear.	Might inform choices of colour and style and improve acceptability.
	Use photographs and examples to illustrate the appearance of the shorts with the different design choices.	May improve acceptability, avoiding people from making choices they are later uncomfortable with.
	Discuss the impact of shorts on clothing choices at the collaborative assessment and possibly in the Participant Information Sheet.	May improve acceptability, avoiding people from making choices they are later uncomfortable with.
	Discus the pros and cons of the high-waist shorts and make this decision collaboratively.	Some participants felt this was important for effectiveness bt others found this to be a considerable disadvantage, impacting upon acceptability.
	Assess weakness of hip flexors, maybe with a stairs assessment and use this to determine the "give" in the shorts.	Restriction to hip flexion was a commonly reported source of burden and may not be necessary for some of the benefits of the shorts.
Intervention	Support orthotic use with therapy contact designed to problem-solve any difficulties and maximise the carry-over from orthotic effect into lifestyle and activity levels.	As suggested in Chapter 3, support is important in the initial stages of orthotic use to maximise the likelihood of longer-term acceptability.
Home trial/longer-term use	Ask participants to trial activities with and without shorts to explore what they might help with. Create a tick list collaboratively of important activities to trial.	To better engage participants in exploring the impact of the shorts.
	Online iPad with audio recording for activity list, wear diary and falls diary.	To overcome challenges with a paper wear diary
	Don't request all day wear but rather wear during activity to avoid discomfort/irritability in shorts at rest.	Shorts felt to be more bother and less relevant at rest.
Measures	Ask at baseline if participants have fallen in the last year.	Subgroup analysis of fallers vs. non-fallers may be important and this question is the most reliable indicator of falls risk.
	Assess near falls	Recommended as important by participants. Suggested that shorts decrease risk of near falls.
	Include self-report EDSS	Subgroup analysis may be important for degree of disability, with some levels responding differently to others.