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Transforming nursing care for children with serious long-term conditions: A mixed methods exploration of the impact of Roald Dahl Specialist Nurses in the United Kingdom

Julie Nightingale *, Nancy Ali, Robin Lewis, Rachel Ibbotson, Helen Monks, Tanya Urquhart-Kelly, Lesley Saunders

Centre for Applied Health and Social Care Research (CARe), Sheffield Hallam University, United Kingdom

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ABSTRACT

Purpose: A new model of paediatric nursing, funded initially by a charitable organisation working in partnership with UK healthcare providers, was implemented to support children living with serious long-term conditions. This study explored, from the perspective of multiple stakeholders, the impact of services provided by 21 'Roald Dahl Specialist Nurses' (RDSN) within 14 NHS Trust hospitals.

Design and methods: A Mixed Methods Exploratory design commenced with interviews with RDSNs ($n = 21$) and their managers ($n = 15$), alongside a medical clinician questionnaire ($n = 17$). Initial themes (constructivist grounded theory) were validated through four RDSN focus groups, and informed development of an online survey of parents ($n = 159$) and children ($n = 32$). Findings related to impact were integrated using a six-step triangulation protocol.

Results: Zones of significant impact included: Improving quality and experience of care; Improved efficiencies and cost-effectiveness; Provision of holistic family-centred care; and Impactful leadership and innovation. The RDSNs forged networks across inter-agency boundaries to safeguard the child and enhance the family experience of care. RDSNs delivered improvements across a range of metrics, and were valued for their emotional support, care navigation and advocacy.

Conclusions: Children living with serious long-term conditions have complex needs. Regardless of the specialty, location, organisation or service focus, this new model of care crosses organisational and inter-agency boundaries to ensure that the healthcare delivered has maximum impact. It has a profoundly positive impact on families.

Practice implications: This integrated and family-centred model of care is strongly recommended for children with complex needs crossing organisational divides.

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Introduction

Roald Dahl was one of the world's most imaginative storytellers; his legacy was the Roald Dahl's Marvellous Children's Charity (RDMCC), founded in the United Kingdom (UK) in 1991. The Charity supports children and young people affected by complex, long-term illnesses that are under-funded, under-resourced and under-recognised. The RDMCC currently funds 82 'Roald Dahl Specialist Nurses' (RDSN) to care for over 24,000 seriously ill children across the UK (Roald Dahl's Marvellous Children's Charity, 2022). The RDMCC funding covers the first two years of the RDSN's salary, and the National Health Service commits to funding the post thereafter.

Advances in obstetric and paediatric medicine have resulted in increasing numbers of children surviving previously non-survivable or significantly life-limiting conditions. The way that health services are organised is crucial to ensuring a good quality of life for these children, however an influential report highlighted an urgent need for improved coordination of children's services in the UK (Kennedy, 2010). The Department of Health (2013) recommended the implementation of family-centred care supported by Clinical Nurse Specialist (CNS) roles. Family-centred care is the delivery of care that supports individuals within their family unit (Institute for Patient and Family-Centered Care, 2023), however two systematic reviews highlight a lack of evidence in terms of measurable outcomes to support its practice (Shields et al., 2012; Watts et al., 2014). Family-centred care brings many challenges, since health professionals and families may have widely varying expectations, experiences, expertise, and motivation to

* Corresponding author.
E-mail address: J.Nightingale@shu.ac.uk (J. Nightingale).

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work collaboratively (Moore et al., 2017; Swallow et al., 2013). With complex conditions, care is often disjointed and fragmented, involving a variety of health professionals in different contexts. The CNS role may add value by improved coordination of services (Vidall et al., 2011), the provision of information and emotional and psychological support, as well as providing expert clinical and technical input. Numerous systematic reviews (Caird et al., 2010; Campbell et al., 2019; Kilpatrick et al., 2014; Randall et al., 2017; Watts et al., 2014) conclude that the CNS role provides high levels of patient satisfaction, quality care, and a reduction in length of hospital stay. The CNS role can lead to greater efficiency and better outcomes (Royal College of Nursing, 2010), however they come at a significant cost, and there is a limited evidence base for their cost-effectiveness (Kilpatrick et al., 2014; Lopatina et al., 2017).

The role and scope of practice undertaken by the Clinical Nurse Specialist varies worldwide. The International Council of Nurses (2020) define a CNS in terms of 'an Advanced Practice Nurse who provides expert clinical advice and care based on established diagnoses in specialist clinical fields of practice' (International Council of Nurses, 2020, p.6). Although the CNS role is firmly embedded in healthcare delivery in Europe, Asia, Canada and the United States of America (USA), there are significant variations in the preparation of the CNS role (Fulton et al., 2016).

In the USA for example, the nature of the CNS role is clearly defined. The role comes under the Advanced Practice Registered Nurse (APRN) 'umbrella'. In the USA, APRNs are Registered Nurses with advanced education and training, typically at master's or doctoral level. In the US, CNSs can assess, diagnose, and treat patients, and their scope of practice is defined and regulated by state-based practice regulations (National Council of State Boards of Nursing [NCSBN], 2015).

In the UK however, the CNS role has evolved 'organically' over the past 20 years and therefore lacks any formal educational framework or structure (Balsdon & Wilkinson, 2014). The ad hoc development of the role in the UK means that although some CNSs are educated to master's or doctoral level, it is not a legal or regulatory requirement to use the CNS title. The required expertise is often gained purely through clinical experience and a professional interest in the particular specialism. This is true of the RDSN, in that they do not undertake a prescribed programme of education such as that required for the APRN qualification. They are referred to as CNSs, which is the cause of some confusion internationally, however for the purposes of this paper the terms RDSN and CNS are used interchangeably.

The care provided by the typical CNS is complex; roles may vary significantly in different contexts, which makes it difficult to objectively measure outcomes and impact or make comparisons between roles in different organisations, even within the UK. While the CNS role has decades of history in supporting patient care, nursing practice, and system improvement (Valdivia, 2022), most evidence is restricted to a single CNS service operating within a single centre. In the current challenging economic climate following the Covid-19 pandemic, there is a real danger that care providers will reduce 'higher end' staffing costs by freezing CNS vacancies to achieve short-term savings. The charity sector may continue to play a vital part in pump-priming public services that would otherwise be considered 'desirable but not essential' by hospital management (King's Fund, 2017), yet they are not immune either to the effects of an economic downturn. Establishing a CNS role is one practical way in which health charities can support publicly funded healthcare services, but it is essential that the impact of these roles is evaluated at scale. This mixed methods study aimed to explore, through the perspectives of multiple stakeholder groups, the impact of a new model of care delivered by Roald Dahl Specialist Nurses (RDSN).

Methods

Services were eligible for inclusion if their RDSN post was established between 01.04.16 and 31.03.19; 21 services were included, 19 based in England, one in Wales and one in Northern Ireland. On site data collection commenced when each RDSN had been in post for a minimum of 18 months. The project was approved by 1) Sheffield Hallam University Research Ethics Committee [ER10500163]; 2) a regional ethics committee specialising in research involving children [North East REC 18/NE/0383]; and 3) the Health Research Authority which governs research within NHS sites in England and Wales. Additional applications were made for the research to proceed in Northern Ireland, and for governance approval to proceed in each participating organisation. All participants provided informed consent.

This study followed a three-phase mixed methods exploratory model (Fig. 1) (Tashakkori & Creswell, 2007). Qualitative data collection was initiated with healthcare staff at each hospital site, followed by a survey of service users (parents and children in the caseload of the RDSNs). Each set of data was analysed separately and then integrated using a triangulation protocol method (O'Cathain et al., 2010), based on the six-step approach developed by Farmer et al. (2006).

Design of phase 1

This phase adopted a constructivist grounded theory methodology, founded upon the co-construction of knowledge between the researchers and participants (Charmaz, 2006). RDSNs and their Nurse Managers were invited separately to a semi-structured interview, undertaken by registered children's nurses [HM, TUK, LS] to capitalise on their professional insight. Interviews were conducted at the participant's place of work though several later interviews were conducted via online technology due to Covid-19 restrictions. A flexible topic guide (Fig. 2) enabled each participant's ideas to inform subsequent questions; interviews were recorded and professionally transcribed. Analysis was led by a qualitative researcher [NA], incorporating two-step coding, memo-writing and the constant comparative method. Rigour was assured by comparing codes across transcripts, and to subcategories, categories and new data (Charmaz, 2008). Discussion within the research team ensured that the generated interpretations were supported by participants' narratives.

Initial themes requiring further exploration were explored via four 'in person' RDSN focus groups ($n = 21$), led by a lead facilitator [JN, RL] with a second researcher taking notes and 'prompting' where necessary. Focus groups were recorded and transcribed, then analysed thematically following a method outlined by Braun and Clarke (2006).

Informed by analysis of the interviews and focus groups, an online questionnaire hosted on the Qualtrics survey platform v2021 (Provo, UT, USA) was distributed to the lead clinicians responsible for the RSDN service. Closed and open-ended questions sought to understand how the roles are regarded by the wider clinical team. Sub-sections included: demographic details; the RDSN business case; the perceived challenges; the RDSN's caseload; and the impact of the nurse on service users. Analysis was via descriptive statistics and thematic analysis of open text comments (Braun & Clarke, 2006).

Design of phase 2

The impact of disease on the families of patients is often unrecognised and underestimated (Golics et al., 2013). An online parental survey aimed to understand the nature of the child's condition and its impact on the family. Placing the RDSN as an 'intervention' at the heart of the child's care, the survey explored the nature and impact of the care received. Parents were also invited to assist their child to complete a brief children's survey, exploring what children liked to do with their RDSN, using a range of 'smiley to sad faces' to ascertain whether children agreed or disagreed with statements. Questions were informed by review of published literature and from the interview themes. Survey sections included: Participant demographics; The participant's household; The child under the care of the RDSN; The impact on the family of managing the child's condition; Interactions with the RDSN. Question

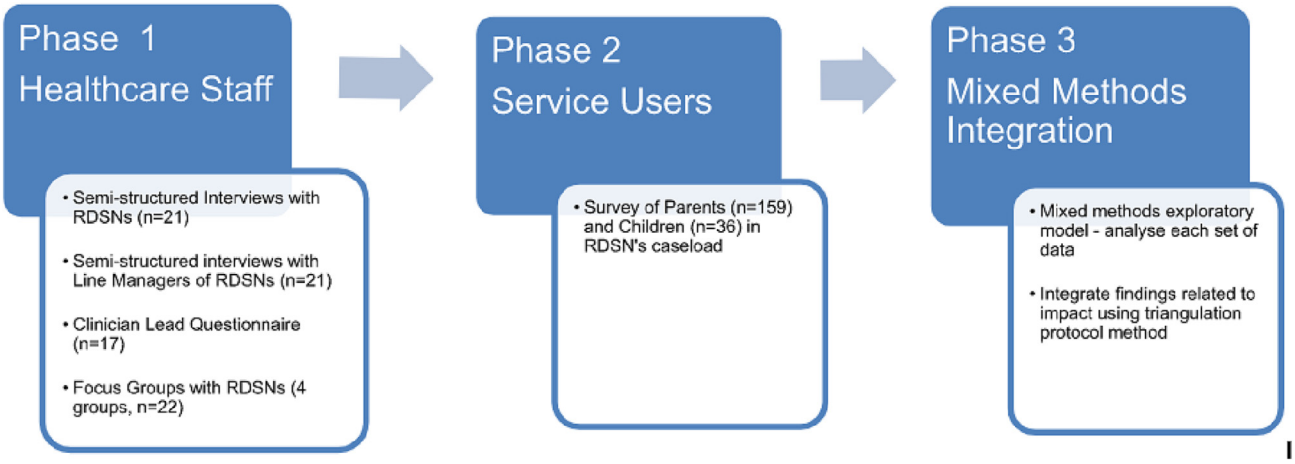

Fig. 1. Mixed methods exploratory research design. Key: RDSN = Roald Dahl Specialist Nurse.

types included Likert scales, sliding scales, forced responses and open text responses. A shortened version of the Impact-on-Family Scale (IOFS-11) (Stein & Jessop, 2003), originally developed by (Stein and Riessman, 1980) to measure the impact of non-specific childhood chronic illness on the family, was incorporated into this survey with the permission of the original authors (REK Stein, personal communication, 25.09.19). The IOFS-11 has been tested successfully in clinical settings including epilepsy (Dehn et al., 2014).

The surveys were tested within the research team, steering group and with representatives of the RDMCC Charity, and were 'field tested' with three parents. The survey was hosted on the Qualtrics online platform v2021 (Provo, UT, USA)(Qualtrics Software, Version, 2021); an email including a survey link and a participant information sheet was distributed by clinical teams to potential parent participants within the RDSN's caseload. Distribution by this method ensured that the research team did not have access to any sensitive data, and gave parents assurance of confidentiality and anonymity as any comments went directly to the research team rather than to their clinical service. Data was analysed via descriptive statistics to enable easily accessible visual displays to be created.

Results and discussion

The clinical services provided by 21 RDSNs were explored, and detailed analysis from the first two phases can be accessed via Supplementary Materials (SM): SM1 nurse and manager interviews; SM2 nurse focus groups; SM3 clinician questionnaire; SM4 service user survey. The key topics identified within each of the stakeholder groups were compared and contrasted following a mixed methods synthesis approach by Farmer et al. (2006); this highlighted the degree of convergence or divergence within the findings as shown in Appendix 1. Each stakeholder group corroborated evidence from other groups and added new information for analysis; there were no significant divergent findings. Topics where a high degree of convergence related to impact was seen across the phases are now presented for further exploration alongside participant quotations and relevant literature.

Impact of Roald Dahl specialist nurses

Role duties were unclear at initial appointment, but as RDSNs transitioned further into the role (Fig. 3), they recognised that they were starting to have a positive impact on their service. However, the impact of complex nursing roles is difficult to quantify; the unique nature of each specialism increases the variability of the roles, making multicentre comparisons challenging. Even in the same specialty there are large inter-centre variations including focus of care (acute/community), geography, levels of deprivation, age ranges and caseload numbers. Measuring the impact of specialist nurses is therefore difficult at scale, and these roles lend themselves to assessment of indirect (e.g. stakeholder perspectives) rather than direct patient outcomes (Campbell et al., 2019; Royal College of Nursing, 2009). It is clear from the parental survey that the RDSNs have a marked effect on the patients and families that they support; in turn managers and clinicians also identify a positive influence on the clinical service. The evidence of impact arising from these stakeholder interactions was categorised into four 'impact

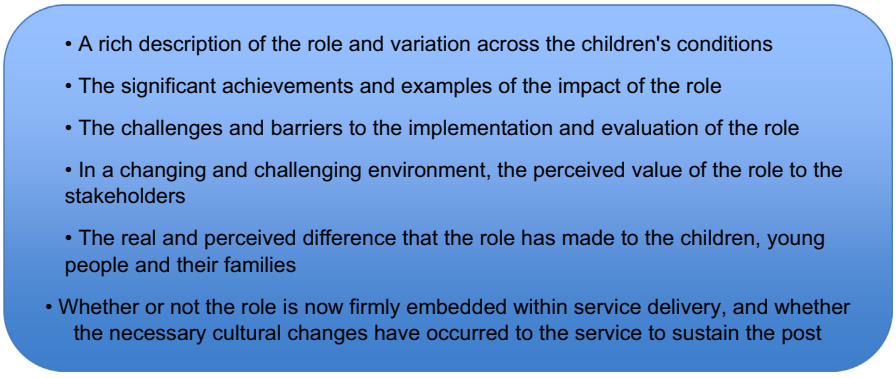

Fig. 2. Guiding research questions and topics.

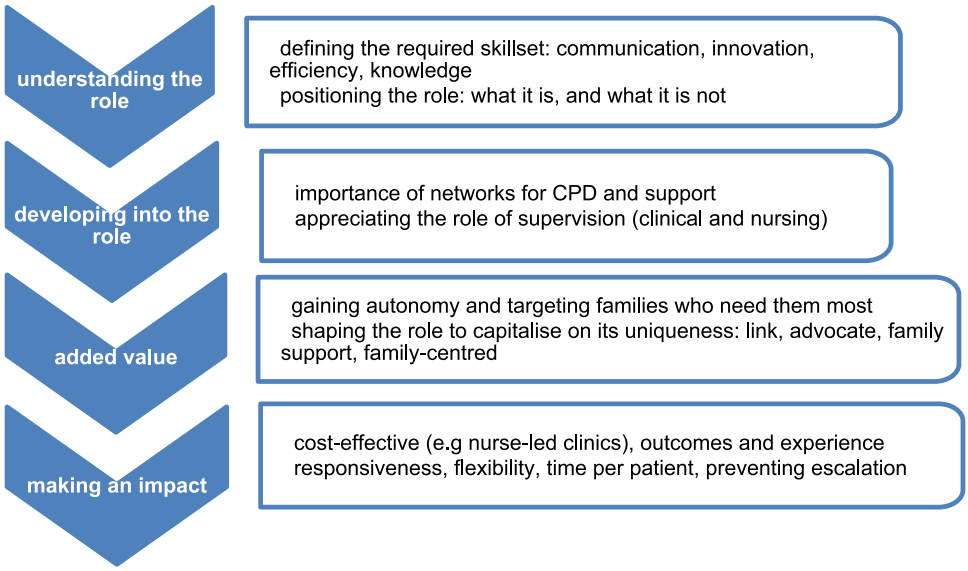

Fig. 3. How Roald Dahl Specialist Nurses transition from being novice practitioners into established roles.

zones' (Fig. 4) adapted from a model previously used by the Macmillan (2023) cancer charity.

Impact 1 - The quality and experience of care

The 21 RDSN services are based within 12 NHS Trusts in England, one in Wales and one Health Board in Northern Ireland. Seven nurses were located at specialist children's hospitals, with the remainder working in mixed adult and child healthcare providers. The nurses worked across eight clinical specialties [epilepsy ($n = 5$); non-malignant haematology ($n = 6$); rare and genetic diseases ($n = 3$); neurology (n = 3); gastroenterology ($n = 1$); child to adult transition services ($n = 2$)]. While the specialties are diverse, the common thread is that the children and young people looked after within these services are all living with complex lifelong conditions. These conditions include progressive disease without curative treatment options, as well as

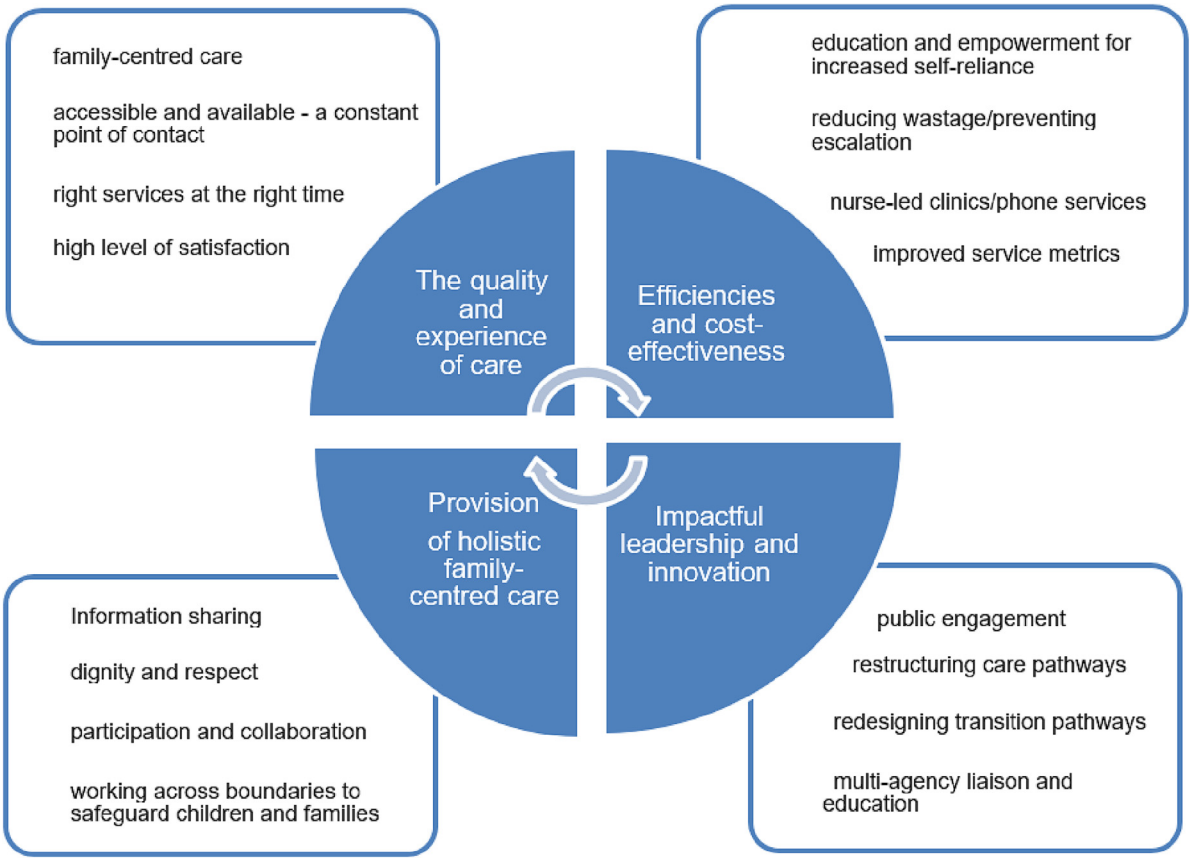

Fig. 4. Four zones of impact embraced by Roald Dahl Specialist Nurses.

irreversible but non-progressive conditions causing severe disability. While some of these children live fairly normal lives for most of the time, others require higher levels of care including support for activities of daily living. In this study, 159 parents completed the service user survey; 41 parents (27.9%) indicated their children were unstable and required round-the-clock care, with 17 of the children (11.6%) requiring at least five emergency (A&E) visits in the last year. The consequences of caring for these children had an impact on the wider family who experienced stress, anxiety and in some cases poor mental health. Completing the Impact-on-Family Scale (IOFS-11), many parents in this study strongly identified with their lives being on a 'roller coaster', needing to give up normal family things or change plans at the last minute due to changes in their child's condition (Fig. 5). This uncertainty and worry was exacerbated when liaising with healthcare services, particularly in emergency situations, with parents recognising that their RDSN had *"a willingness to go the extra mile"* and was *"a credit to her profession"*. The RDSNs provided much-needed support for the family, as outlined by Smith, Cheater, and Bekker (2015) building trust, listening to parent concerns and valuing parents' knowledge of their child; in the parent survey *'providing emotional support for myself or my family'* was one of the most highly valued services.

> *"Our [RDSN] has been a godsend to us at some of the lowest points in my life. I honestly don't think I would mentally be where I am now without her" [Parent].*

> *"She provides us with emotional and practical support when no one else is available. It's really a lifeline sometimes when you feel lost and confused and sad" [Parent].*

> *"Our amazing nurse is a compassionate, knowledgeable voice, who is pretty much always available; this has massively reduced family anxiety, bridged a huge gap and reduced lengthy waiting times trying to contact consultants, and been a regular (sometimes weekly) proactive guide through medication and condition concerns and changes. She has made a dramatic impact on our lives, and we really don't know how we would get through so many days without her support and dedication" [Parent].*

Children and young people (CYP) who completed the survey also recognised how the RDSNs focus not only on their physical health, but also on their emotional wellbeing:

> *"Talk a lot about my meds and how I feel and how things are at school and home" [CYP].*

> *"Answers any questions I have. Always asks me how I have been or if I am worried about anything" [CYP].*

> *"Talks about how I am feeling and how I am coping" [CYP].*

All children in this study were in some form of education, with one third (29.9%, $n = 44/147$) attending a school catering for special educational needs. Many children had missed at least 10 school days in the last year due to their condition (44.5%, $n = 65/146$). These factors all put added strain on parents who had multiple responsibilities. While most parents in this study were married or in steady relationships, 21 (13.2%) were single parents and many were juggling work commitments as well as looking after other children. Sixteen parents (10.7%)

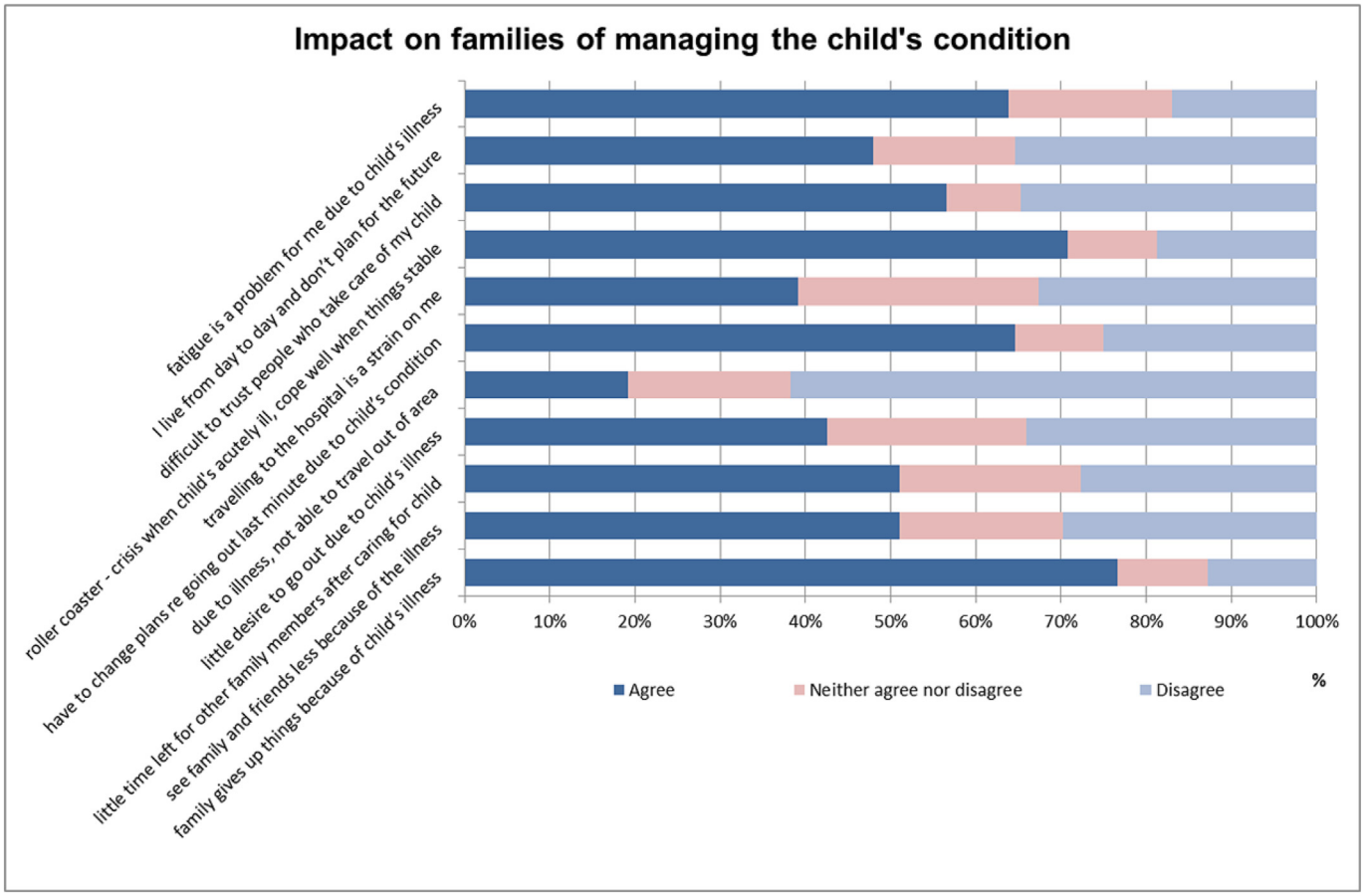

Fig. 5. Visual representation of parental responses to eleven statements in the Impact-on-Family Scale (IOFS-11). The darker colours indicate a stronger agreement.

indicated that they were suffering a great deal of financial hardship, with one third of parents (36.9%, $n = 55/149$) occasionally experiencing hardship. Forty percent of parents (40.1%, $n = 61/152$) stated that their child's illness has had a significant impact on their employment or financial status. A large body of existing evidence has explored the lived experiences of parents whose children have chronic health problems (George et al., 2007; Madrigal & Walter, 2019; Shudy et al., 2006). Unfortunately, these experiences are often dominated by social isolation and significant struggles in coping with work and parental responsibilities on top of looking after a sick child (Wright-Sexton et al., 2020; Yagiela & Meert, 2020). Feelings of despair and helplessness are heightened in young, deprived or ethnic minority families as they often do not know how to navigate the system to get medical, financial and emotional support (Cabizuca et al., 2009; McClellan & Cohen, 2007); Suurmond et al., 2020). Parents in our survey expressed concerns regarding navigation of the different systems within and beyond healthcare; the nursing support most highly valued is *'being a point of contact and coordinating my child's care across hospital services'*. Many of the children had very complex conditions which required input from several different medical specialties, often based in different healthcare organisations. The co-ordination of their child's healthcare was highly challenging and often exhausting for the parents; the RDSNs are a vital link between services:

> *"She [RDSN] has been an amazing support to my family and our little boy, he is extremely complex so therefore has a lot of doctors involved in his care. She has been great at helping them to all work and communicate together…" [Parent].*

> *"With so many medical professionals involved in our daughter's care, it has been great to have a person whom we can turn towards to liaise between them when we are not sure who to contact." [Parent].*

> *"…before Roald Dahl Nurses I spent hours upon hours, days upon days chasing appointments, medication, explaining my daughter's condition, waiting for a call back. Roald Dahl Nurses have relieved the stress of some of my daughter's care … they are absolutely invaluable and worth their weight in gold!" [Parent].*

The majority of parents (72.9%; $n = 102/140$) had required urgent unscheduled contact with the RDSN at least three times in the last year. Parents and children described how this accessibility and availability of the nurses often provided quick answers to their worries, relieving their anxiety and reducing the need for accessing emergency services:

> *"Our Roald Dahl Nurse is always available at the end of a phone call to answer any questions" [Parent].*

> *"She is amazing and always there if we need her. She's always one phone call away" [CYP].*

> *"I am very impressed at how promptly the [Roald Dahl] nurse replied to text / phone calls … I cannot praise them enough for their advice, care and support" [Parent].*

Families indicated that overall satisfaction with the care received was extremely high (mean of 9.47/10). Children and young people who completed the survey ($n = 36$) were also extremely positive about engaging with their RDSN (Table 1), with 88% ($n = 22/25$) stating that their nurse is friendly and 67% ($n = 18/27$) 'agreeing a lot' that they like visiting their nurse. Local service audits also highlighted overwhelmingly positive service user feedback, though RSDNs and managers noted that obtaining quantitative feedback from already overwhelmed and busy families was a challenging process:

> *"It is so subjective what we do, it's difficult to measure … we're asking a lot of families, so it's been very slow to get feedback" [Nurse].*

Nurses highlighted that they provided families with a service which was more responsive, more flexible and provided more time per patient than a consultant could offer. Their work in coordinating services and rationalising appointments, as well as advocating on the families' behalf, had a high impact not only on family experience of care, but also on care outcomes where better care was provided more quickly. Working across professional boundaries in order to mobilise resources to get *"the right services involved at the right time"* appears to be the essence of the RDSNs' work, attributes of specialist nurses previously identified as being highly valued by patients (Corner et al., 2003). In a scoping review of the role of epilepsy nurse specialists, Campbell et al. (2019) cautions that there are elements of the specialist role (being a point of contact and liaising with and linking services) that are poorly recognised and inadequately evaluated in the existing evidence. Our study certainly showcases these two aspects of the role as being pivotal to the smooth running of the service and to patient experience.

> *"… the addition of a Roald Dahl Specialist Nurse has greatly improved the quality of service we offer to our patients and their families, and feedback from these families has all been extremely positive." [Clinician].*

Impact 2 - Efficiencies and cost-effectiveness

Demonstrating the financial impact of the RDSN role is difficult where patients with complex needs interact with a range of services and health professionals. Clinicians recognised that evidence for cost-effectiveness is often limited by the absence of data on costs before and after initiation of the service, as identified by (Eftekhari & Faizel, 2015). They noted that cost savings take time to be demonstrated and costs may increase due to increasing demand. While they cautioned about the challenges of proving the 'cause and effect' relationship as the nurse does not work in isolation, several clinicians outlined how introducing and then evaluating the cost-effectiveness of a service improvement initiative may be a useful indicator of the cost-effectiveness of the wider role. Indeed several RDSNs presented themselves as 'active change agents', introducing a suite of interventions leading to reductions in waiting times, A&E visits, hospital admissions and duration of stay:

> *"I actually get to do service improvement, but also not only at our [service] level, at Trust level and national level..." [RDSN and Manager].*

Table 1
Levels of children and young people's agreement with Roald Dahl nursing activities. Shaded areas = most common responses.

Question	I agree a lot	I agree a bit	I can't decide	I disagree a bit	I disagree a lot	Total
I like going to see my Roald Dahl Specialist Nurse	66.67% (18)	18.52% (5)	11.11% (3)	3.70% (1)	0.00% (0)	27
My Roald Dahl Specialist Nurse is friendly	88.00% (22)	0.00% (0)	12.00% (3)	0.00% (0)	0.00% (0)	25
My Roald Dahl Specialist Nurse talks to me about my condition	77.78% (21)	7.41% (2)	11.11% (3)	3.70% (1)	0.00% (0)	27
My Roald Dahl Specialist Nurse explains things to me so I can understand	80.77% (21)	7.69% (2)	7.69% (2)	3.85% (1)	0.00% (0)	26
My Roald Dahl Specialist Nurse includes me in decisions about me	76.00% (19)	8.00% (2)	16.00% (4)	0.00% (0)	0.00% (0)	25

"I think our team manager and the multidisciplinary team, they can see the value…What would happen if I wasn't doing that? They [family] would just constantly call or attend A&E, so to the wider Trust we are reducing hospital admissions ..." [Nurse].

"Since the [RDSN] started…our regular attenders are not as regular as they used to be, because they get captured before they come into A&E, the correct advice is given, or if they do go to A&E… it reduces bed stays" [Manager].

Many nurses had set up a telephone contact service to improve accessibility, not only for parents but also for the children and young people. The RDSNs typically believe that the more phone calls, cases and teaching a nurse has been able to manage, the more productive they are. Conversely managers recognised the complexity and time-consuming nature of many of their duties and acknowledged that the quality of advice and support that families received were more accurate measures. RDSNs saw the role of the telephone service as 'preventing escalation' which included providing medicines advice out-of-hours to help stabilise the child, and re-arranging appointments so that a child of concern could be seen quickly by a consultant. Nurses who had monitored their caseload over time were able to show reductions in the number of missed appointments, number of A&E visits, number of hospital admissions and reduced duration of hospital stay. John et al. (2019) also advocated the benefits of specialist nursing telephone services as examples of 'open access' models of delivery which reduced demand for emergency services in periods of austerity. These unscheduled phone contacts with the RDSNs were a lifeline for parents, with 40% ($n = 54/135$) stating that this contact had prevented urgent GP (primary care) appointments on more than one occasion in the last year, and 52.7% ($n = 68/129$) indicating that consultant appointments had also been averted on at least one occasion. More significant is the finding that 42.3% ($n = 55/130$) of parents acknowledged that timely contact with the RDSN had averted visits to A&E, with at least one emergency admission to hospital prevented for 37.5% ($n = 48/128$) of parents (Fig. 6). Not only is this a positive impact in terms of reduced costs and time for the NHS, but also in terms of family anxiety and inconvenience. Nevertheless two thirds of clinicians believed that the RDSN role had either no effect or a fairly positive effect on reducing readmission rates.

The introduction of nurse-led clinics to replace some consultant-led clinics for less complex cases is a common RDSN intervention which may be easily equated with cost-effectiveness:

"We earn our keep …we cover the consultants' workload, we are much cheaper than a consultant..." [RDSN and Manager].

However this is a simplified cost analysis, as the clinics are often introduced in addition to the consultant services, rather than as a replacement for them. A systematic review of the impact of nurse-led clinics has shown positive impacts on self-reported patient outcomes, patient satisfaction and access to care, but only mixed results on cost-effectiveness (Randall et al., 2017). In our study 29% of clinicians ($n = 5/17$) noted the challenges in identifying any positive productivity and efficiency impacts in some aspects of the care provided by RDSNs, additionally noting that some RDSN roles required employment of staff at higher grades which could cost more in the short term (Erens et al., 2015). However the RDSNs drew attention to their role in educating and empowering children, young people and their families to self-manage their conditions, thus becoming less dependent upon NHS services. Many nurses gave individual accounts of this gradual handover of care responsibility, often utilised for young people going through the transition to adult services. The ultimate positive impact is when patients and families are supported and empowered to no longer need the RDSN's support.

Impact 3 - Provision of holistic family-centred care

Nursing in adult services is traditionally centred on the person (person-centred care) utilising a medical model. Family-centred care presents a shift of traditional approaches, supporting individuals within their family unit, particularly relevant where children are primarily cared for at home, with parents often taking significant responsibility for care decisions and delivering treatments (Kish et al., 2018; Smith, Swallow, & Coyne, 2015). Family-centred care is linked to four precepts: dignity and respect; information sharing; participation; collaboration (Johnson et al., 2008; Institute for Patient and Family-Centered Care, 2023). Family-centred care is linked to better health outcomes but requires additional encounters to build trust and partnership (Kuo et al., 2011). Evidence presented in all phases of this study suggest that RDSN behaviour and attitudes align with these four precepts, including

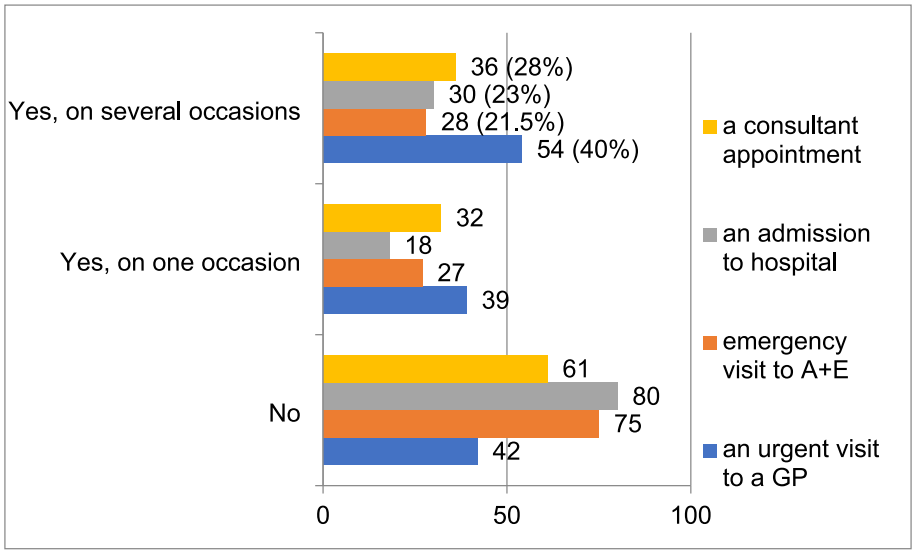

Fig. 6. Parent's views of whether unscheduled contact with the Roald Dahl Specialist Nurse has averted the need to access emergency and urgent services for their child in the previous year.

the provision of independent and unbiased education and counselling [information sharing] leading to empowerment of families and meaningful participation in healthcare decisions [participation], advocacy for patient and family perspectives and values [dignity and respect], and safeguarding and non-healthcare interventions [collaboration] taken on behalf of the child and their family. Examples of family-centred care 'in action' were recounted in the survey by many parents who indicated that they highly valued *'advocating (speaking on my family's behalf) to other authorities or individuals'*, an area of high stress for parents in other studies (Boshoff et al., 2016). The family-centred care provided was appreciated with parents stating that their nurses were: *"amazing", appreciated", "a godsend", "invaluable", "incredible", and "worth their weight in gold"*.

The RDSNs work across healthcare, social services, education and housing boundaries to mobilise resources, striving to secure *"the right services involved at the right time"*. They saw this role as not only the *"missing link"* between families and the medical staff, but also a *"link between families and the rest of the world"*. The term 'care navigation' perhaps better describes the complexity of the role seen from the clinicians' perspectives:

> *"A key role across the entire pathway, helping to ensure continuity between settings, educating service providers and users. Networking with other health professionals and across healthcare boundaries" [Clinician].*

> *"There was a significant need to bridge community, outpatient, and inpatient services…" [Clinician].*

> *"A perceived need to support and educate other clinical staff including community children's nurses, school nurses, teachers, learning disability nurses and medical colleagues" [Clinician].*

The RDSNs' remit appears to be to actively create connections and facilitate care across organisational boundaries (Panton, 2014). Networking across service boundaries is outlined by While et al. (2006) within a five level spectrum of collaboration in nursing; many of the RDSNs were working routinely at *inter-professional* and *intra-organisational* levels, however a major component of their work was situated at the *inter-agency* level (Fig. 7). This inter-agency working is often ongoing, rather than a time-limited process as in many adult nursing 'care navigator' roles (Gilbert, 2016), and is identified as 'process-heavy' (Erens et al., 2015; Gilbert, 2016; Kendall-Raynor, 2012). This cross-boundary working is characterised by negotiation with stakeholders on a daily basis, often demanding post-holders to develop new systems and, importantly, networks (Kessler & Bach, 2007; McEvoy et al., 2011). The staff stakeholders all articulated challenges of working inter-agency, particularly at the start of their role when systems and networks had to be constructed, often from scratch.

> *"She [RDSN] has forged links with other organisations and services to develop 'alert' systems for our patients across the region to develop evidence-based emergency care plans… She is balancing many competing needs… the safeguarding of an individual child against the importance of dealing with a teenager having a 'crisis' against a planned clinic and this requires levels of competence, leadership and initiative that can be daunting" [Clinician].*

The clinician above highlights the importance of the safeguarding aspects of the role. Many RDSNs, however, had been surprised by the volume of safeguarding activity required, exacerbated by workforce shortages in other disciplines such as social work and social care.

> *"I suppose all the services aren't there that you would want out in the community …you've children in here who have been here for weeks and they need discharging, and …it's [care packages] not there. That is all very frustrating…" [Nurse].*

> *"We've got families who have very difficult social circumstances, where we've had to go and help and write letters to the Council…to try and get them a cleaner house, better housing and safer housing for the children" [Manager].*

Safeguarding as an aspect of inter-agency working was often an emergency and a priority over the child's healthcare (HM Government, 2018), and while parents recognised and appreciated this aspect of care, managers noted that it reduced the RDSNs' capacity to deliver healthcare for families. Safeguarding children is challenging and emotionally draining, and benefits from clinical supervision, yet while supervision has been shown to improve patient safety and effectiveness of care (Snowdon et al., 2017) there is little evidence that it can mitigate against burnout (Buckley et al., 2020). Nevertheless, it can be used as a fail-safe mechanism for nurses who need support, as well as offering valuable learning opportunities.

Impact 4 - Impactful leadership and innovation

The nurses stated that clinical skills were a prerequisite for the role, and expressed their core attributes as patient advocacy, being passionate, empathetic and motivational. Managers and clinicians identified RDSN qualities as professional excellence, empathy, proactivity, enthusiasm, resilience and team spirit. Many of these attributes are exhibited by professionals in high responsibility posts, rather than those

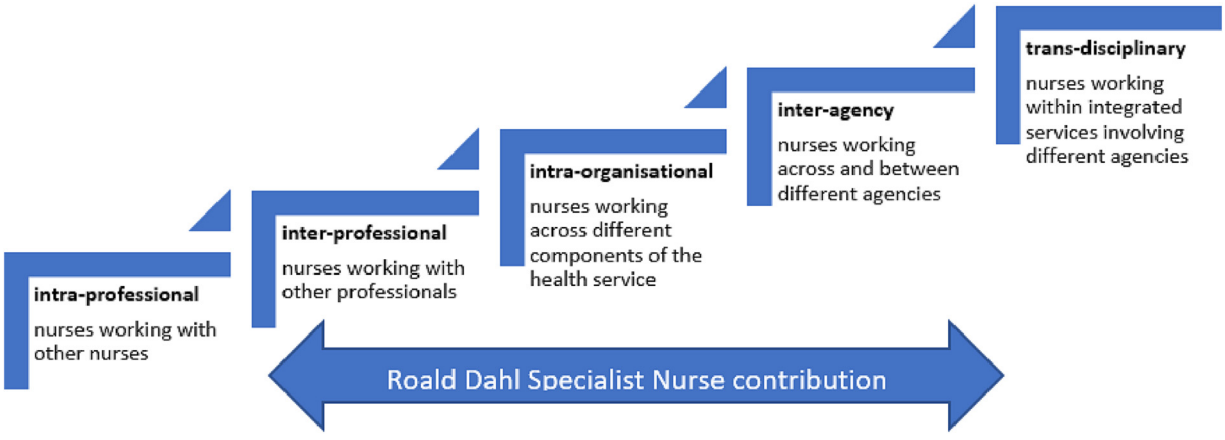

Fig. 7. The Roald Dahl Specialist Nursing contribution across service boundaries, assessed against the five level spectrum of collaboration in nursing (While et al., 2006).

previously reported for specialist nurses (Gibson, 2001 cited in Royal College of Nursing, 2009); examples given aligned clearly with the leadership and innovation elements of advanced clinical practice.

Service innovation represented a significant aspect of the RDSN role. Managers and clinicians gave examples of where RDSN service innovations had a positive impact, including better multidisciplinary team working, improved access to services, better coordination of care and support navigating the healthcare system. Impacts were most profound where nurses had designed or reshaped pathways of care to improve patient experience and care outcomes. Several nurses had designed and implemented nurse-led clinics and services which could offer more frequent and longer appointments. To encourage self-management and transition, some services offered appointments and telephone calls for young people to access without their parents if they wished. Indeed the creation and reshaping of transition pathways appeared to be important for all RDSNs, not just the transition specialists.

> *"The [RDSN] roles are more about influencing and encouraging and engaging our clinical staff to think differently about how they support young people... it's about planting that seed to say actually thirteen is an ideal time to start having those conversations" [Nurse].*

> *"Transition is everybody's business, so what are people, what are they doing about it? And when they're talking about transition, are they actually talking about … transfer of care?" [Nurse].*

The UK Care Quality Commission Report on Transition highlighted significant care gaps in many specialties and centres (Care Quality Commission, 2014). Successful transition is now recognised as an important facet of care in both paediatric and adult medicine; it is not simply transfer of care from children's services to an adult provider, but should be a gradual process taking into account physical and psychological maturity as well as the availability and structure of local resources (Urquhart-Kelly & Wales, 2019). Nearly all of the RDSNs had invested significant time in replacing the outdated 'transfer of care' model with the more apt 'transition pathways' approach. However transition is noted by the RDSN nurses as a 'rocky road', fraught with challenges that could lead to a neglect in care and see some young people slip through the system. Clinicians agreed that the transition from child to adult services was emerging as an important issue requiring specialist support, with some currently exploring transition posts to work alongside their existing RDSNs:

> *"Transition is a huge step for any young adult with a rare disease that has been under paediatric services and there is often so much to coordinate and even create to allow successful transition to adult services, hence the business case for this post" [Clinician].*

Establishing innovations within pathways that spanned across services meant that RDSNs needed to expend significant time and energy in creating and fostering new inter-agency links and networks. Managers noted that this work was often hidden from view, yet it promoted positive changes in organisational culture and openness. However, a more transparent and powerful impact beyond their immediate workplace has been orchestrated by the nurses who educated the wider healthcare team and non-healthcare staff in other agencies and organisations about their specialty. Teaching innovations aimed to not only share knowledge, but to change culture:

> *"They've [RDSNs] spent such a lot of time teaching and training others and the feedback often I get is about, oh it's been marvellous, they've come, we never knew that, we never even thought of it, now we've put this in place. And actually a lot of that stuff is not at a cost, it's only about behavioural differences" [Manager].*

> *"She has 'upskilled' a number of community and school nurses and involved them actively in patient care" [Clinician].*

Education did not stop at the boundaries of care; many RDSNs engaged in media activities to increase public awareness about serious long-term conditions, informing societal perceptions and raising the profile of different health problems. The impact of such work is hard to measure, but will undoubtedly support the better integration of children (and adults) living with long-term conditions into society.

The leadership and innovation activities of the RDSNs appears to be synonymous with practice at an advanced level. The complex conditions and high levels of clinical uncertainty mean that the nurses are not always able to follow standard pathways of care; they regularly have to use their ingenuity and innovation to question the status quo and drive forward change. Their work appears to reflect that of an 'Advanced Clinical Practitioner' (ACP) (Health Education England, 2017; Royal College of Nursing, 2012), yet the majority of the RDSNs self-identify with the role descriptor 'Clinical Nurse Specialist' (CNS). Inconsistency in job titles is not unusual in charity-established roles, with a Macmillan (2023) identifying almost 50 different job titles in use for nurse specialists working in the field of urological cancers. While there is some ambiguity on the ACP role (Leary et al., 2017; East, Knowles, Pettman and Fisher, 2015), the CNS role is expected to have more direct clinical contact than an ACP whose focus is often wider (incorporating leadership, clinical practice, education and research). In this study, however, the RDSNs clinical practice element appeared to be a less significant component, displaying effective mastery of leadership, management and education expertise more aligned to ACP role descriptions:

> *"It's all about coordinating care, being able to support families, encouraging research, to be empathetic and compassionate, ... you've got to be multifaceted to do this role, really, it's unique" [Nurse].*

Numerous examples of autonomous practice, high-level clinical reasoning, decision making and critical analysis provided by the nurses and managers are commensurate with master's level academic criteria. While both the ACP role and the CNS APRN role within the USA are expected to be underpinned by master's level education (Health Education England, 2017), there is no expectation for this level of education to underpin CNS roles in the UK (Royal College of Nursing, 2014). Few of the nurses within this study had undertaken master's level education, which arguably devalues the role and reduces credibility. Lack of investment in academic development to master's (or doctoral level) can affect the potential for impact of specialist nurses (Redwood et al., 2007) as well as harm their career progression. Arguably the role title is not of importance, however the preparation of the nurses for this highly challenging role may be better served with educational underpinning at master's level, instilling the core values of leadership, management and education, as well as providing expertise in clinical reasoning, critical appraisal and decision making.

Practice implications

This study has highlighted that clinical nurse specialists who are facilitating family-centre care, often across organisational boundaries (inter-agency), require knowledge, skills and attributes at an advanced level. Nurses who are aspiring to work with children with serious long-term conditions in a family-centred care environment are recommended to seek opportunities for continuing professional education at Master's level.

Nurses who are providing services for children with long-term conditions should ensure that evidence-based protocols are developed for specific pathways of care such as the gradual transition of children

and young people to adult services. Clinical Nurse Specialists are also advised to capture service-related performance metrics before and after any service interventions to enable the impact of their work to be effectively evaluated and showcased.

Limitations

This study takes a holistic view of the Roald Dahl Specialist Nursing role from a much wider range of stakeholders than is routinely accessed in CNS studies, and at multiple sites (21 RDSN services) rather than the single-centre studies often presented. However the inclusion of different specialties increases variation and heterogeneity of the services studied, though the attempt to capture data at a similar time post-appointment adds greater standardisation to the project. Additionally, the mixed methods approach used in this study has enabled multiple approaches to strengthen the study and yield meaningful data compared to using one method alone.

Standardised data was not collated across the different services prior to the RDSNs commencing in post, so it was not possible to evidence impact 'pre and post intervention' via direct measures. Similarly we did not seek to ascertain the cost-effectiveness of the role. The method of distribution of the patient survey resulted in a relatively low response and difficulties ascertaining a response rate. This study, however, has been able to compromise on these ideals by gathering indirect yet comprehensive (stakeholder) feedback on the role and impact of the RDSN.

Conclusions

The aim of this project was to explore, from the perspectives of multiple stakeholder groups, the role and impact of the Roald Dahl Specialist Nurses (RDSNs) delivering services across the UK. The clinical services provided by 21 nurses were included, representing one quarter of the current RDSN workforce. These nurses worked across eight different clinical specialties, caring for children living with complex long-term conditions. The complexity of many of these conditions mean that healthcare cannot be delivered in isolation; it is inextricably linked to additional needs, for example in education, social services and housing. The RDSNs have forged networks across these interagency boundaries to safeguard the child and family and ensure they are supported in these other aspects of their lives; ensuring that the healthcare that is delivered has the maximum impact. These RDSNs operate at an advanced level; underpinning education at master's level is therefore strongly recommended for those aspiring to the role.

The RDSN role is challenging. The RDSNs require a particular skill set beyond their clinical expertise to equip them for this role, including the core attributes of passion, empathy, motivation, professional excellence, patient advocacy, proactivity, enthusiasm, resilience and team spirit. Innovation is a key driver for the role, yielding new ways of working to improve patient experience and outcomes. It is evident that the RDSNs, regardless of their specialty, location, organisation or service focus, have a profoundly positive impact on the stakeholders around them. The RDSN service model follows the philosophy of family-centred holistic care; parents highly-value the educational and emotional support provided, as well as the care navigation and advocacy for the child and family.

The partnership between the charitable organisation (Roald Dahl Marvellous Children's Charity) and the healthcare providers is effective in pump-priming these innovative posts. This study supports the future establishment of similar posts across the UK and beyond, where the service need includes: 1) *serious long-term conditions that affect both child and the wider family; 2) family-centred care focus; 3) management of own caseload; 4) long-term relationship building and 5) the need for regular multi-agency working*. However the current financial model for pump-priming the RDSN role is inevitably precarious; the charitable funding is not infinite, and the charity sector in the UK is experiencing significant challenges during the current economic climate. This study has showcased the vital services delivered by Roald Dahl Specialist Nurses, yet they are not available to all children living with serious long-term conditions. Addressing the current inequity of access to services should not be the responsibility of the charity alone; public health services must be willing to embrace this innovative model of care to ensure that the healthcare that is delivered has the maximum impact on these children and their families. This research provides independent evidence to underpin future charitable and public sector partnerships, providing a platform from which to input into commissioning negotiations within the UK National Health Service and beyond.

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CRediT authorship contribution statement

Julie Nightingale: Conceptualization, Funding acquisition, Methodology, Formal analysis, Writing – original draft. **Nancy Ali:** Methodology, Formal analysis, Writing – review & editing. **Robin Lewis:** Conceptualization, Methodology, Formal analysis, Writing – review & editing, Funding acquisition. **Rachel Ibbotson:** Formal analysis, Visualization, Data curation. **Helen Monks:** Investigation, Formal analysis. **Tanya Urquhart-Kelly:** Investigation, Formal analysis. **Lesley Saunders:** Investigation, Formal analysis.

Declaration of Competing Interest

None.

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Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.pedn.2023.02.004.

Appendix 1

Mixed methods synthesis of findings.

Topics	Nurse and manager interviews	Nurse focus groups	Lead clinician survey	Parent and child survey	Synthesis
Transition to post and early service development	Stressful when setting up services from scratch; fluid role boundaries. Resistance to change; Unrealistic expectations of some clinicians	Transition to new post stressful. Good training and support an exception rather than the norm. Difficult to move away from 'old role'.	Not always clear in the business case what they wanted RDSN to do. Few had considered providing cover for the nurse's work during absence.	Not highlighted	Weak convergence
Crossing boundaries of care	Emphasis on liaison rather than direct clinical care: *"the right services involved at the right time'*. Cross professional boundaries to mobilise resources: healthcare, social care, education and housing	*'Link between families and the rest of the world'* and the *'missing link'* between families and the medical staff. Multiple contacts, sign-posting and networking, liaison	Key role across entire pathway, ensure continuity between settings, educating service providers and users. Networking across healthcare boundaries. A single point of access and contact for the service	*"Being a point of contact and coordinating my child's care across hospital services."* Providing advice. Liaison with schools and other non-healthcare services highly valued.	Strong convergence
Philosophical approach to role	Clinical philosophies were primarily family-centred care, encompassing holistic management, evidence-based practice and empowerment.	High volumes of safeguarding and non-healthcare interventions. Advocacy and empowerment of selves and others: *"holistic family-centred care that our consultant colleagues do not have the capacity to deliver"*	Recognised a child and family centred approach to care and treatment	Huge difference to own, child's and family's lives. Support for whole family widely recognised: RDSNs are: amazing, appreciated, *'god send'*, invaluable, *'worth their weight in gold'*. Allowed parent to *'be a mum'*.	Strong convergence
Core values and skillset	Patient advocacy, being passionate, empathetic and motivational. Professional excellence, highly proactive, enthusiasm, resilience, team spirit	High degree of insight into their role beyond clinical care. Communication, innovation, efficiency and knowledge, many examples aligned clearly to leadership in advanced practice.	Individual skills highlighted including advocacy, empathy, enthusiasm and being proactive.	Willingness to *'go the extra mile'*. Friendly and impartial, providing emotional support. Advocating for parent and Children and young people. Helpful, caring, a good listener, *'A credit to her profession'*.	Strong convergence
Digital Technology and physical resources (access and bureaucracy)	Multiple systems housing patient data. Poor access to technology, poor physical spaces (offices and consulting rooms)	All re-iterated by many nurses, particularly frustrations with multiple systems requiring many different log-ins etc.	Bureaucratic issues: IT support and the lack of existing infrastructure were cited as challenges in setting up the role	Not highlighted	Weak convergence
Workloads on non-clinically related tasks	Admin takes away from frontline clinical care. Safeguarding role huge. Need for support workers, mental health input, administrators, translators	Not cost-effective for a nurse to be doing work suited to a lower band. Family support workers or administrative support essential. Safeguarding.	Excessive input to caseload administration. Need for additional RDSN, or other healthcare input to support them as caseloads expand.	Not highlighted	Strong convergence
Sustainable caseloads - size and complexity	Lack of clear boundaries, caseload expansion is inevitable: *'exponential caseloads'*. Difficult to transition, yet new ones added every week. Look beyond numbers - many patients highly complex.	Articulated how and why caseloads grow - must gain control quickly with tighter criteria. Emotional blackmail to take more patients, so need support from clinicians and managers from start with clear inclusion / exclusion criteria.	Size of the caseload is less important than the complexity. Caseloads are often too high (1/4 not sustainable), with 65% growing since inception of service. A 'recipe for burnout' if not addressed.	Some children had complex conditions with multiple organ system disorders. 28% children unstable and requiring round the clock care; 14% needed multiple A&E visits in last year.	Strong convergence
Challenges of Evaluating Impact of Nurse rather than service	Complexity of patients means RDSN is one of multiple interventions. No definitive 'before and after' evaluations: collect efficiency, productivity and patient satisfaction data. High subjectivity, difficult to quantify and compare collective impact.	All noted how difficult it is to categorically prove their impact (such as preventing admissions) and recognised the need to gather qualitative data such as parent feedback.	Cannot confidently state cause and effect; nurse does not work in isolation. Takes time for impacts to be apparent. Challenging to identify positive impacts on saving money/ reducing demand. Clinicians divided on impact on readmissions.	Not highlighted	Strong convergence
Stakeholder perceptions of Impact	Interventions reduce A&E visits, waiting times, admissions, duration of stay. Increasing efficiency, reduced wastage, improved patient experience. 'Active change agents'.	All stated impacts in: Cost effectiveness; Responsiveness; Flexibility; Time per patient; Preventing escalation.	Positive service impacts e.g. psychological and practical support; MDT working; improving access to services; co-ordination of care/navigating healthcare system. Positive feedback from families.	For ¼ of parents unscheduled RDSN contact avoided urgent GP/consultant appts. Reassurance prevents escalation. Securing resources; coordinating appointments	Strong convergence
Impact of the child's condition on the family	Recognise stresses on the wider family: provide a support network for parents, always strive to be contactable.	Not highlighted	Not highlighted	Wider impact of illness on the family: on a *'roller coaster'* and need to give things up or change plans at last minute. Stressful.	Weak convergence
The growing importance of Transition	*Re*-shaping of transition pathways important for all RDSNs, not just transition specialists	Not highlighted, other than a reason for caseload expansion	Transition from child to adult services emerging in importance. Some centres exploring transition posts to work alongside RDSNs.	Not highlighted	Weak convergence
Role of Roald Dahl's Marvellous Children's Charity	Enthusiasm for working with Roald Dahl's Marvellous Children's Charity, excellent support early in role, good recruitment feedback, ongoing CPD support	Not highlighted	Importance of charity pump priming for these 'Cinderella services'. Assistance in recruitment and on-going support for nurses CPD highly valued.	Not highlighted, other than affectionate terms such as 'crocodile nurses'.	Weak convergence

Key: Strong Convergence = More than two sets of data agree on key topics; Weak Convergence = Two sets of data agree on key topics; Divergence = Strong findings present in only one set of data, or findings disagree across data sets.

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