

Antenatal counselling for prospective parents whose fetus has a neurological anomaly: part 1, experiences and recommendations for service design

HART, AR <<http://orcid.org/0000-0003-4018-3238>>, VOLLMER, B <<http://orcid.org/0000-0003-4088-5336>>, HOWE, D, BOXALL, S, FOULDS, N, DE LACY, P, VASUDEVAN, C, GRIFFITHS, PD and PIERCY, Hilary <<http://orcid.org/0000-0002-7663-8858>>

Available from Sheffield Hallam University Research Archive (SHURA) at:

<http://shura.shu.ac.uk/29080/>

This document is the author deposited version. You are advised to consult the publisher's version if you wish to cite from it.

Published version

HART, AR, VOLLMER, B, HOWE, D, BOXALL, S, FOULDS, N, DE LACY, P, VASUDEVAN, C, GRIFFITHS, PD and PIERCY, Hilary (2021). Antenatal counselling for prospective parents whose fetus has a neurological anomaly: part 1, experiences and recommendations for service design. *Developmental Medicine and Child Neurology*.

Copyright and re-use policy

See <http://shura.shu.ac.uk/information.html>

Antenatal counselling for families whose fetus has neurological anomalies: Part one – prospective parents’ experiences and recommendations for service design

Anthony R Hart MRCPCH PhD [1]
Brigitte Vollmer MD, PhD, FRCPCH [2]
David Howe DM FRCOG FRCS(Ed) [3]
Sally Boxall BSc, MSc, RGN, RM [3]
Nicola Foulds BSc PhD MBChB FRCP [4]
Patricia de Lacy MBChB FRCS(SN) [5]
Chakra Vasudevan MBBS DCH DNB MRCPCH Med [6]
Paul D Griffiths PhD FMedSci FRCR [7]
Hilary Piercy BSc, MA, PhD, RGN, RM, PGCE, SFHEA [8]

- [1] Department of Perinatal and Paediatric Neurology, Sheffield Children’s NHS Foundation Trust, Ryegate Children’s Centre, Tapton Crescent Road, Sheffield, S10 5DD
- [2] Clinical and Experimental Sciences, Faculty of Medicine, University of Southampton, UK; Paediatric and Neonatal Neurology, Southampton Children’s Hospital, University Hospital Southampton NHS Foundation Trust, Southampton, UK.
- [3] Department of Fetal Maternal Medicine, University Southampton NHS Foundation Trust, E Level, Princess Anne Hospital, Coxford Road, Southampton, SO16 5YA
- [4] Department of Clinical Genetics, University Southampton NHS Foundation Trust, Mailpoint 627, Princess Anne Hospital, Coxford Road, Southampton, SO16 5YA
- [5] Department of Paediatric Neurosurgery, Sheffield Children’s NHS Foundation Trust, Western Bank, Sheffield, S10 2TH
- [6] Department of Neonatology, Bradford Royal Infirmary, Duckworth Lane, Bradford, BD9 6RJ
- [7] Academic Unit of Radiology, University of Sheffield, Royal Hallamshire Hospital, Glossop Road, Sheffield, S10 2JF
- [8] The Centre for Health and Social Care, Sheffield Hallam University, Collegiate Crescent, Sheffield, S10 2BP

CORRESPONDING AUTHOR

Dr Anthony Hart
Department of Paediatric and Neonatal Neurology
Sheffield Children’s Hospital NHS Foundation Trust
Ryegate Children’s Centre
Tapton Crescent Road
Sheffield
South Yorkshire
S10 5DD
United Kingdom

Fax 0114 2678296

Email: A.R.Hart@sheffield.ac.uk

WORD COUNT: 3338

ABSTRACT : 125

TABLES: 1

BOXES: 3

FIGURES: 0

SUPPLEMENTARY TABLES: 0

KEYWORDS / MESH HEADINGS:

Pregnancy – High Risk; Fetus; Infant – Newborn; Child Development; Health Communication

FINANCIAL DISCLOSURE

None

CONFLICTS OF INTEREST

No conflicts of interest are known.

ACKNOWLEDGEMENTS

None

WHAT THIS PAPER ADDS

- Prospective parents report poor experiences of antenatal counselling, which simple changes to service design could improve
- Discussions of risk are frequently poorly understood, and we recommend methods for improving communication of numerical data

ABSTRACT

Prospective parents whose fetus is diagnosed with a neurological abnormality go through a complex range of emotions. Their experiences of antenatal counselling from health care professionals is that the discussion focusses too much on the nature of the abnormality and involves unintelligible medical terminology, when they really want a picture of the best and worse-case scenarios instead. Whilst a figure to estimate the level of risk for their fetus is important, it is not prospective parents' primary concern. When figures for risk are given, it is not certain they are understood as well as health care professionals think. This review discusses the published evidence on antenatal counselling and recommendations for explaining risk. From this data, we make recommendations for the organisation of antenatal counselling services.

INTRODUCTION

Screening for fetal structural and genetic abnormalities is a routine aspect of pregnancy care, and includes ultrasound imaging between 18 and 20 weeks' gestation.¹ When suspected neurological anomalies are found in a fetus, additional investigations, such as a congenital infection screen, genetic testing, and in-utero Magnetic Resonance Imaging (iuMRI), may be requested to confirm the anomaly is present, identify additional abnormalities, and determine aetiology. For example, iuMRI has superior diagnostic accuracy compared to ultrasound²⁻⁵ and provides clinical information that both improves obstetricians' diagnostic confidence and influences their recommendations on the management of the pregnancy.^{2, 3} Improved diagnostic capability may create an expectation of better prognostication for fetal neurological abnormalities; however, there is limited data on how prospective parents view antenatal counselling. The published data comes from qualitative research studies of women, and rarely includes partners' or other family members' experiences; where data from partners is captured, it takes a strictly heteronormative approach. In addition, the data on developmental outcomes for most fetal abnormalities is limited and of poor quality. All of these factors mean that antenatal counselling is a challenging area for health care professionals.

This review discusses the experiences of prospective parents who receive antenatal counselling and the published recommendations on how to communicate risk, from which we make pragmatic recommendations on how services should be organised. Because families come in many forms and not all pregnant individuals identify as women, we use the term "prospective parents" throughout this article, unless the published data refers specifically to women or male partners. Part two of our review discusses the published risks of atypical developmental outcome in a range of common fetal neurological abnormalities.⁶

PROSPECTIVE PARENTS' VIEWS ON SCREENING TESTS IN PREGNANCY AND ANTENATAL COUNSELLING

Views on screening tests in pregnancy and diagnosis of a fetal abnormality

The literature shows pregnant women see ultrasound as a social rite of passage and a bonding opportunity, and it also improves the involvement of their partner in the pregnancy.⁷⁻

¹² However, many women report they are not adequately prepared for an ultrasound scan, with a third not understanding it could detect fetal malformations,^{9; 11; 13; 14} and others assuming that abnormalities are found in other people's babies, not theirs.^{11; 14-17} After prospective parents have been told their baby has an abnormality, they face a complex, confusing and potentially overwhelming range of emotions, including fear, anger, grief, and anxiety, for which they are largely unprepared.^{9; 10; 18; 19} At the same time, prospective parents have to grapple with complex medical information, terminology, confusing statistics, challenges to their ethical, moral or religious beliefs, and social and family expectations.^{16; 19}

Information needs and experience following diagnosis of a fetal abnormality

Almost immediately, prospective parents want information on the best and worst case scenarios for their baby's outcome ^{16; 17; 20} to help them understand what emotional and financial resources will be needed to care for the child after birth.¹⁸ Many report that the information they receive is too focussed on the technical and biological aspects of the anomaly, is given using incomprehensible medical terminology, ^{17; 19; 21} and does not include sufficient information on potential developmental implications. For the health care professional, there may be a number of reasons why they do not give this prognostic information as clearly as prospective parents want: the literature on outcomes of fetuses with abnormalities is limited and of poor quality, the health care professional may not be up to date with the most recent literature, or they have limited experience of following up children with similar abnormalities.^{19; 22} If prospective parents see more than one health care professional during their journey, the information they receive can appear to be conflicting, which leads to uncertainty about whom to believe, a loss of trust, and feelings of insecurity about the right course of action to take.^{16; 17; 20; 21}

Health care professionals should also be aware of how they are phrasing information and their potential non-verbal communication: prospective parents report they can infer from verbal or non-verbal communication what decision the health care professional thinks they should make, which is usually that the pregnancy should be terminated.¹⁹ Studies of women suggest they perceive health care professionals see their health as the most important aspect in the decision and that the baby is 'a problem to be solved'.¹⁹ Prospective parents also report feeling a change in the behaviour of health care professionals if they choose "the wrong decision": clinicians appear to step back and become less involved in their care.¹⁹ As a result of all of these difficulties, prospective parents seek further information from other sources, for example the internet, including support groups for families whose child has the same condition.^{17; 19; 22} Published research, mainly from qualitative studies, have found a number of factors are associated with positive counselling sessions for prospective parents, which are summarised in Box 1.^{16; 17; 19; 21; 23; 24}

Family or societal challenges following diagnosis of a fetal abnormality

Having gathered information, prospective parents may be faced with a choice about termination of pregnancy, which can be particularly difficult in families or communities with strongly polarised ethical views.¹⁰ Societal values influencing these decisions include: the belief that women should love their fetus from conception, that women should not deliver "disabled" children, and the belief that termination of pregnancy represents prejudice against children with disabilities.⁽⁵⁾ Another assumption that can prevent appropriate support being given to prospective parents is the belief they will not experience grief if they choose to have a termination of pregnancy.¹⁰ In the literature, women commonly report pain, grief, and trauma for a long time after termination of pregnancies, irrespective of the circumstances within which conception occurred and how they arrived at the decision to terminate the pregnancy, which leads them to doubt they made the right decision.^{25; 26}

The behaviour of partners

The data on a partner's feelings is lacking in the literature, and there is no data at all on the experiences of same sex partners or extended family members, such as prospective grandparents. The literature suggests that male partners experience similar traumatic emotions to termination of pregnancy as their partner.²⁵⁻²⁹ Men may see their partner's distress as their primary concern and adopt the position that, given she is carrying the baby, the final decision about how to manage the pregnancy is hers and they should support whatever decision she makes.²² This can result in men adopting a passive role during information gathering, in some cases allowing their pregnant partner to function as the conduit through which they receive information. If a couple are not communicating effectively, a male partner may not receive information at all.²² Where partners want different outcomes and a woman elects for a termination of pregnancy, her partner has to cope with feelings of being powerless and despondent, whilst grappling with the belief that the decision should reside with their partner.²² As a result, men can withdraw from discussions about the management of the pregnancy to avoid conflict and reduce trauma, which can be perceived by their partner as an unfeeling or uncaring attitude towards either them or the fetus.¹⁰

The role of extended families and friends

Trauma can also be accentuated when the decision prospective parents make is disapproved of by partners, friends or family. For example, studies have shown women report that other family members may oppose their decision to have a termination of pregnancy on moral grounds, or attempt to over-ride her decision to continue the pregnancy because they perceive she does not have the ability to cope with a potentially disabled child.^{19; 30} The literature shows this conflict leads pregnant women to feel isolated,¹⁹ and health care professionals should not simply assume prospective parents will be supported by their families and friends. There is no published evidence on how a partner feels where disagreement with the wider family occurs, nor the long-term consequences of this conflict.

Recurrence of a fetal abnormality

We are not aware of published data on how prospective parents feel when recurrence of a fetal neurological abnormality occurs. In our experience, this is a devastating diagnosis, made harder if an opportunity to diagnose the abnormality early in the pregnancy was missed; for example, if appropriate genetic testing was not offered in the initial pregnancy or there was an incomplete discussion of recurrence risk. It is our experience that prospective parents may not necessarily make the same decisions in second affected pregnancies than the first. For example, prospective parents who struggled with strong emotions, such as guilt, about a previous termination of pregnancy, may choose to continue with a subsequent affected pregnancy, particularly if their opportunities for future pregnancies are diminishing. Alternatively, if the previous child survived with a poor quality of life and demanding needs that challenged the family's ability to cope, a termination may be considered more readily in subsequent pregnancies. Health care professionals should be careful to listen to prospective parents about their experiences and feelings on previous pregnancies, without making any assumptions on what decision they will make for the current pregnancy.

Pregnancies achieved by in-vitro fertilisation

Prospective parents may experience similar feelings of devastation and disappointment where a pregnancy was achieved by in-vitro fertilisation, as this will often follow many years of attempting to become pregnant, or multiple miscarriages, which could have been an emotionally turbulent experience in its own right. There is no evidence that the emotional journey of prospective parents who achieved pregnancy by IVF is any less or more severe than families who achieved pregnancy naturally; nevertheless, careful discussion about recurrence, liaison with the fertility and genetics team, and psychological support should be available.

COMMUNICATING THE POTENTIAL OF ADVERSE OUTCOME

Health care professionals may see the most important aspect of antenatal counselling as giving prospective parents a numerical figure for the estimation of the risk their child might

have atypical developmental outcome. In fact, prospective parents report that descriptions of the best and worst case scenarios for their baby's outcome are the most useful part of antenatal counselling.^{16; 20} They want health care professionals to "paint a picture" of what expected difficulties will be like with functional descriptions and comments on independence, participation and quality of life. If medical terminology is used, its meaning should be explained in lay-terms. Health care professionals should be aware that some terminology may already be known by prospective parents, but they may not know the full scope or spectrum of the condition. For example, the term cerebral palsy may conjure up pictures of a child with profound motor disorder in a wheelchair, with profound learning disabilities and a range of other comorbidities, without prospective parents understanding the full extent of the spectrum. Prospective parents may have different views on the quality of life experienced by someone with a disability, and health care professionals are also known to underestimate the quality of life of children with disabilities,³¹ so they should be careful of any assumptions they are making during counselling.

At some point, prospective parents are likely to want an estimation of how big the risk is of their child having atypical developmental outcome. Reviewing the evidence on how to communicate the degree of risk in medical practice is beyond the remit of this article, although helpful reviews have been published,³²⁻³⁵ but we outline some important observations below.

Many people struggle to understand numerical data

Lipkus et al found that 40% of individuals over 40, 84.4% of whom had more than high school education, could not solve basic probability puzzles or convert percentages to proportions. For example, 21.8% could not answer correctly which of 1/100, 1/1000 or 1/10 represented the highest risk of getting a disease, and 19.2% could not correctly determine how many people in a population of 100 would develop a disease if the risk was 10%.³⁶

Yamagishi et al found university undergraduates rated cancer as being riskier when it killed

1286 out of 10,000 compared to 24.14 out of 100, presumably because the numerator was larger.³⁷

Understanding numerical data and probability is not the same as understanding personal risk

Having a grasp on numerical data does not mean a person understands their personal risk.³⁵

Research in other fields of medicine shows individuals minimise or exaggerate their personal risk, assuming 'bad things happen to other people'.³² Perception of risk is also affected by how serious the hazard is perceived to be and the person's views of what living with the condition is like.³²

Figures for risk do not always relate to changes in behaviour or decisions

Whatever an individual's understanding of the numerical data, knowing a baby has a high risk of a condition does not necessarily alter prospective parents' behaviour: evidence from genetic antenatal counselling clinics shows factors like maternal age, parity, and religion also play critical roles in decision making.^{24; 38-41}

When communicating a single risk, it probably doesn't matter whether percentages or fractions are used

There is no clear answer as to whether a single risk is best presented as a percentage or a fraction. If several risks are communicated using fractions, the denominator must remain the same to allow for direct comparison.³²⁻³⁵

Verbal descriptions of the level of risk are problematic and not better than numbers

Verbal descriptions of risk, such as "very unlikely", "doubtful" or "almost certain to happen" are more likely to influence outcome than numerical data, and individuals report that they are easier to understand.³² However, these verbal terms use "fuzzy logic" and are not comparable across different specialities, conditions or situations because they do not take

account of the baseline risk in the population. Recommended phrasing for the level of risk (table 1) can bear little relationship to the level of risk one wants to express. For example, in table 1, a risk of 1 in 100 is described as “high”, but the population risk of a child having early developmental impairment is a between 2 and 3 in 100,⁴² so this “high risk” is actually a third of the baseline population risk.

A further problem relates to an individual’s personal assessment of risk and what level of risk they are prepared to accept. Prospective parents may attribute different terms for the degree of risk to the figures than their health care professional. We have experienced how verbal terminology can lead to confusion and frustration, as demonstrated in box two. Experts in medical risk recommend verbal descriptions are avoided or always accompanied by numerical values to put them into context.³³⁻³⁵

Health care professionals should ensure they use positive and negative framing

Behavioural work suggests that numerical information should be framed in different ways to present information with a negative and positive focus.^{33; 34} Negative framing is “25 out of 100 children (or 25%) will have this condition”; positive framing is “75 out of 100 (or 75%) will not have the condition”. Both positive and negative framing should be given when discussing risk.

Pictures and visual aids are good

Visual aids can help patients understand risk. For presentation of the risk of a single event, pictograms or icons should be used, with the population in the background and risk demonstrated by colouring in the number of likely affected patients. The “at risk” figures in the foreground need to be placed together, rather than scattered throughout the background figures, because the reader needs to count them quickly when scanning the diagram. If multiple risks are presented in a document, the icons should be the same shape, size or volume. Numerical data should accompany pictures because some people have a low

graphical literacy. Pie charts are a poor way of explaining and comparing risks. Where the risks between groups are being compared, histograms should be used. Line graphs are recommended to show trends or change over time.³³

PREPARING TO MEET PROSPECTIVE PARENTS

Prior to meeting a family, the health care professional should ensure they are aware of all relevant information, including family history, general health and medication use, the sex of the fetus, fetal growth parameters, whether other abnormalities have been detected in the fetus, and the results of any congenital infection or genetic testing. Where this is not available, they should ask the prospective parents and their families for further information prior to giving prognostic information.

Professionals should be aware of the published figures for risk and providing information only after a quick internet search is not appropriate! In our experience, fetal counselling works best when delivered by a small subgroup of clinicians who have greater knowledge of the data and experience of following up children with similar neurological abnormalities. It is helpful to have a multidisciplinary discussion, which may include Obstetricians, Midwives, Geneticists, Neuroradiologists, Paediatric Neurologists, Neurosurgeons, and Neonatologists, on the consequences of an abnormality before meeting the prospective parents, where this is possible.

When reviewing the published data on outcome, it is also essential to understand how abnormal outcome is defined because this varies between studies, which may inflate or deflate the risk value published compared to other studies.

RECOMMENDATIONS FOR ANTENATAL COUNSELLING SERVICES FOLLOWING THE DIAGNOSIS OF A NEUROLOGICAL ABNORMALITY

Based on these experiences of prospective parents', common-sense recommendations can be made, as summarised in box 3. In short, antenatal counselling teams should include a wide variety of specialities, led by the Feto-Maternal team, including Neonatologists, Paediatric Surgeons, Paediatric Neurosurgeons, Urologists, Geneticists, Radiologists with expertise in fetal imaging, and Paediatricians with expertise in neurological disorders and neurodevelopmental follow-up. The team should meet regularly to build relationships, review cases, discuss what information is given to women whose babies have common anomalies, and review outcome of cases. There should be access to written information, either produced locally or from national bodies and charities. Referral pathways should be agreed and access to other specialities available quickly.

In our experience, a period of time is helpful for health care professionals to review the case and published evidence prior to meeting prospective parents. Where counselling is performed on the same day the diagnosis of the abnormality is made, we find prospective parents are in a state of shock and don't know what to ask. Allowing them a few days to think about the diagnosis and search for data online ensures that counselling sessions are more detailed and fruitful. Where there are specific time frames, such as mandated by the law on termination of pregnancy, then members of the team should collect opinions as fast as possible, and counselling appointments on the same day as diagnosis may be the only option.

It is tempting to assume that antenatal counselling would be ideal for telemedicine, especially with the greater availability of health care technology during the COVID-19 pandemic, alongside the frequent appointments prospective parents must attend, and the large distances they may need to travel to hospital. In our experience, however, prospective parents have preferred face-to-face appointments when offered the choice.

Support should be available following counselling for a pregnant people and their partner, such as from a counsellor or psychologist. Postnatal plans should be clear and long-term follow-up arranged ideally with the person involved in the antenatal counselling. Where a fetal abnormality may have a genetic aetiology and recurrence risk, the case should be reviewed with colleagues in the Genetics department during multi-disciplinary team meetings, or sooner where there are time pressures relating to decisions on managing the pregnancy. Genetic studies like exome sequencing may be appropriate, even if prospective parents decide to have a termination of pregnancy. Appointments with the Clinical Geneticists and / or Genetic Counsellors should be made available to prospective parents where genetic causes and recurrence risks are identified. Health care professionals and families should work together on collecting developmental outcome data to improve future prognostication.

CONCLUSION

Antenatal counselling after diagnosis of fetal neurological abnormalities is a challenge, particularly where the information is used to decide whether a termination of pregnancy is appropriate or not. We provide recommendations on service design and how information should be communicated to women and their families, but ultimately more information from prospective parents and their families on what they need and how to provide it is needed to drive forward improvements in care.

REFERENCES

- 1 Public Health England. (2018) NHS Fetal Anomaly Screening Programme Handbook. London, UK: Public Health England. p 15-20.
- 2 Griffiths PD, Bradburn M, Campbell MJ, et al. Use of MRI in the diagnosis of fetal brain abnormalities in utero (MERIDIAN): a multicentre, prospective cohort study. *Lancet* 2017;389: 538-46.
- 3 Griffiths PD, Bradburn M, Campbell MJ, et al. Change in diagnostic confidence brought about by using in utero MRI for fetal structural brain pathology: analysis of the MERIDIAN cohort. *Clin Radiol* 2017;72: 451-7.
- 4 Turkyilmaz G, Sarac Sivrikoz T, Erturk E, et al. Utilization of neurosonography for evaluation of the corpus callosum malformations in the era of fetal magnetic resonance imaging. *J Obstet Gynaecol Res* 2019;45: 1472-8.
- 5 Di Mascio D, Sileo FG, Khalil A, et al. Role of magnetic resonance imaging in fetuses with mild or moderate ventriculomegaly in the era of fetal neurosonography: systematic review and meta-analysis. *Ultrasound Obstet Gynecol* 2019;54: 164-71.
- 6 Hart AR, Vasudevan C, Griffiths PD, et al. Antenatal counselling families whose fetus has neurological anomalies: Part two –risks of adverse outcome in common abnormalities. *Dev Med Child Neurol* 2021;in press.
- 7 Wertz DC, Fletcher JC. A critique of some feminist challenges to prenatal diagnosis. *J Womens Health* 1993;2: 173-88.
- 8 Sandelowski M. Channel of desire: fetal ultrasonography in two use-contexts. *Qual Health Res* 1994;4: 262.

- 9 Mitchell LM. Women's experiences of unexpected ultrasound findings. *J Midwifery Womens Health* 2004;49: 228-34.
- 10 McCoyd JL. Pregnancy interrupted: loss of a desired pregnancy after diagnosis of fetal anomaly. *J Psychosom Obstet Gynaecol* 2007;28: 37-48.
- 11 Garcia J, Bricker L, Henderson J, et al. Women's views of pregnancy ultrasound: a systematic review. *Birth* 2002;29: 225-50.
- 12 Ekelin M, Crang-Svalenius E, Dykes AK. A qualitative study of mothers' and fathers' experiences of routine ultrasound examination in Sweden. *Midwifery* 2004;20: 335-44.
- 13 Layng J. Screening for nuchal translucency. Counselling should be considered an integral part of screening programmes. *BMJ* 1998;317: 749.
- 14 Liamputtong P, Watson L. The voices and concerns about prenatal testing of Cambodian, Lao and Vietnamese women in Australia. *Midwifery* 2002;18: 304-13.
- 15 Crang-Svalenius E, Dykes AK, Jorgensen C. Organized Routine Ultrasound in the Second Trimester - One Hundred Womens' Experiences. *J Matern Fetal Invest* 1996;6: 219-22.
- 16 Asplin N, Wessel H, Marions L, Georgsson Öhman S. Pregnant women's experiences, needs, and preferences regarding information about malformations detected by ultrasound scan. *Sex Reprod Healthc* 2012;3: 73-8.
- 17 Lalor JG, Devane D, Begley CM. Unexpected diagnosis of fetal abnormality: women's encounters with caregivers. *Birth* 2007;34: 80-8.

- 18 McCoyd JL. "I'm not a saint": burden assessment as an unrecognized factor in prenatal decision making. *Qual Health Res* 2008;18: 1489-500.
- 19 Redlinger-Grosse K, Bernhardt BA, Berg K, Muenke M, Biesecker BB. The decision to continue: the experiences and needs of parents who receive a prenatal diagnosis of holoprosencephaly. *Am J Med Genet* 2002;112: 369-78.
- 20 Walser SA, Werner-Lin A, Russell A, Wapner RJ, Bernhardt BA. "Something Extra on Chromosome 5": Parents' Understanding of Positive Prenatal Chromosomal Microarray Analysis (CMA) Results. *J Genet Couns* 2016;25: 1116-26.
- 21 Larsson AK, Crang-Svalenius E, Dykes AK. Information for better or for worse: interviews with parents when their foetus was found to have choroid plexus cysts at a routine second trimester ultrasound. *J Psychosom Obstet Gynaecol* 2009;30: 48-57.
- 22 Werner-Lin A, Barg FK, Kellom KS, et al. Couple's Narratives of Communion and Isolation Following Abnormal Prenatal Microarray Testing Results. *Qual Health Res* 2016;26: 1975-87.
- 23 Chitty LS, Barnes CA, Berry C. Continuing with pregnancy after a diagnosis of lethal abnormality: experience of five couples and recommendations for management. *BMJ* 1996;313: 478-80.
- 24 Werner-Lin A, McCoyd JL, Bernhardt BA. Balancing Genetics (Science) and Counseling (Art) in Prenatal Chromosomal Microarray Testing. *J Genet Couns* 2016;25: 855-67.
- 25 Korenromp MJ, Page-Christiaens GC, van den Bout J, et al. A prospective study on parental coping 4 months after termination of pregnancy for fetal anomalies. *Prenat Diagn* 2007;27: 709-16.

- 26 Khoramroudi R. The prevalence of posttraumatic stress disorder during pregnancy and postpartum period. *J Family Med Prim Care* 2018;7: 220-3.
- 27 Aite L, Zaccara A, Mirante N, et al. Antenatal diagnosis of congenital anomaly: a really traumatic experience? *J Perinatol* 2011;31: 760-3.
- 28 Sullivan N, de Faoite E. Psychological impact of abortion due to fetal anomaly: a review of published research. *Issues Law Med* 2017;32: 19-30.
- 29 Rychik J, Donaghue DD, Levy S, et al. Maternal psychological stress after prenatal diagnosis of congenital heart disease. *J Pediatr* 2013;162: 302-7.
- 30 Tijnstra TJ, Bosboom GJ, Bouman K. Experiences of women who decided to continue the pregnancy after diagnosis of Down's syndrome. *Ned Tijdschr Geneesk.* 2000;144: 2104-7.
- 31 Dickinson HO, Parkinson KN, Ravens-Sieberer U, et al. Self-reported quality of life of 8-12-year-old children with cerebral palsy: a cross-sectional European study. *Lancet* 2007;369: 2171-8.
- 32 Weinstein ND. What Does It Mean to Understand a Risk? Evaluating Risk Comprehension. *J Natl Cancer Inst Monogr* 1999;25: 15-20.
- 33 Spiegelhalter D. Risk and Uncertainty Communication. *Annu Rev Stat Appl* 2017;4: 31-60.
- 34 Trevena LJ, Zikmund-Fisher BJ, Edwards A, et al. (2013) Presenting quantitative information about decision outcomes: a risk communication primer for patient decision aid developers. *BMC Med Inform Decis Mak* <http://www.biomedcentral.com/1472-6947/13/S2/S7>: BMC. p s7.

- 35 Paling J. Strategies to help patients understand risks. *BMJ* 2003;327: 745-8.
- 36 Lipkus IM, Samsa G, Rimer BK. General Performance on a Numeracy Scale among Highly Educated Samples. *Med Decis Making* 2001;21: 37-44.
- 37 Yamagishi K. When a 12.86% Mortality is More Dangerous than 24.14%: Implications for Risk Communication. *App Cog Psychol* 1997;11: 495-506.
- 38 Bernhardt BA, Soucier D, Hanson K, et al. Women's experiences receiving abnormal prenatal chromosomal microarray testing results. *Genet Med* 2013;15: 139-45.
- 39 Grimes DA, Snively GR. Patients' understanding of medical risks: implications for genetic counseling. *Obstet Gynecol* 1999;93: 910-4.
- 40 Hunt LM, Castañeda H, de Voogd KB. Do notions of risk inform patient choice? Lessons from a study of prenatal genetic counseling. *Med Anthropol* 2006;25: 193-219.
- 41 Jacobs M, Cooper SA, McGowan R, Nelson SM, Pell JP. (2016) Pregnancy Outcome following Prenatal Diagnosis of Chromosomal Anomaly: A Record Linkage Study of 26,261 Pregnancies. *PLoS One*. p e0166909.
- 42 Horridge KA. Assessment and investigation of the child with disordered development. *Archives of disease in childhood. Education and practice edition* 2011;96: 9-20.
- 43 Mohanna K, Chambers R. Risk matters in healthcare: communicating, explaining and managing risk. Abingdon, UK: Radcliffe Medical Press.2001
- 44 EMA (Eur Med Agency). (2014) Benefit-Risk Methodology Project. Update on Work Package 5: Effects Table Pilot (Phase I). London, UK: Eur Med Agency.

Table 1: Examples of different recommended verbal terminology from experts or governmental agencies

Reference	Suggested verbal descriptions of risk		
	Descriptor	Percentage	Fraction
Mohanna and Chambers (2001) ⁴³	High	1	More than 1 in 100
	Moderate	0.1	1 in 100 to 1 in 1000
	Low	0.01	1 in 1000 to 1 in 10,000
	Very Low	0.001	1 in 10,000 to 1 in 100,000
	Minimal	0.0001	1 in 100,000 to 1 in 1,000,000
	Negligible	0.00001	Less than 1 in 1,000,000
European Medicines Agency ⁴⁴ Recommended terminology for the risk of adverse drug events	Very common	≥ 1/10	
	Common	≥ 1/100 to < 1/10	
	Uncommon	≥ 1/1000 to < 1/100	
	Rare	≥ 1/10,000 to < 1/1000	
	Very rare	< 1/100000	
	Frequency not known		
Royal College of Anaesthetists Recommended terminology for the risk of adverse anaesthetic events	Very common – “someone in your family”	10%	1 in 10
	Common – “someone in a street”	1%	1 in 100
	Uncommon – “someone in a village”	0.1%	1 in 1000
	Rare – “someone in a small town”	0.01%	1 in 10,000
	Very rare – “someone in a large town”	0.001%	1 in 100,000
Spiegelhalter et al ³³ Probability intervals and terminology used by the Inter-governmental panel for climate change	Virtually certain	99-100%	
	Extremely likely	95-100%	
	Very likely	90-100%	
	Likely	66-100%	
	More likely than not	50-100%	
	About as likely as not	33-66%	
	Unlikely	0-33%	
	Very unlikely	0-10%	
Exceptionally unlikely	0-1%		

Box 1: Positive features of discussions with health care professionals 16; 17; 19; 21; 23; 24

Parents find discussions with health care professionals are positive when the following occurs:

- the information given is understandable
- the health care professional ensures the information is understood
- active listening, empathy, warmth, respect, and authenticity is employed
- two-way discussions are used to share the work in finding a pathway through uncertainty, rather than 'talking at' prospective parents
- using diagrams
- involving both prospective parents in the discussion
- health care professionals avoid imposing their beliefs or judgements on prospective partners
- the baby is discussed with respect as if they had "meaning", using a name, if the family have one for their baby
- written information is provided
- follow-up appointments or contact details for additional questions are offered
- prospective parents feel supported, whatever choice they make for their pregnancy
- continuity of care occurs after delivery, i.e. where the same health care professional who met the family antenatally provides paediatric follow-up.

Box 2: Paraphrased conversation with prospective parents during antenatal counselling about risk

Health care professional: Now we have discussed what the MRI scan has found, I imagine you would like to talk about what that means for your baby? We know that children with this abnormality can have problems with their learning and development....

Partner: Yes, we have heard that from the previous doctor, but we don't know what that risk is.

Health care professional: The evidence in the literature suggests the risk of some form of learning or development difficulties is around 5%, or to put it another way 5 out of 100 babies with this finding on scan will have some development or learning difficulties.

Partner: That's terrible!

Health care professional: Well, if you look at it the other way, that means that 95% or 95 out of 100 babies with this finding will not – they will have normal development.

Partner: No, no, no...that's too high! That's a really high risk! I don't understand.... The doctor we saw previously said that our baby had a low risk. But 5%? That is huge!

Health care professional: It's important to know what the risk is in children who have a normal MRI. Around 3%, or 3 in 100, children with a normal scan have problems with the learning or development. So, yes, the risk is a bit higher, but the figures suggest it's much more likely that your baby's development will be OK.

Partner: No. No. We were told the risk was low, and now you're saying 5%. That's a high risk. I don't understand why we were told it was low by the other doctor.

Health care professional: Well, it depends on how you view risk. I don't really like using terms like high or low risk – they can be confusing. One person's high risk is another person's low risk. We all see things different. It's true your baby's risk is higher than children with a normal scan. The figures are, though, that 95 out of 100 children with this finding will be normal. Personally, I would say your baby's risk was low.

Partner: But that can't be right. We were told during pregnancy about Down syndrome and the risk was high if it was more than 1 in 150. How can you possibly now say that 5 in 100 is low risk?

Box 3: Recommendations for design of antenatal counselling services

Area of care	Recommendations
Timing of counselling	<ul style="list-style-type: none"> • An initial discussion about the result of a test should occur at the time of diagnosis, such as on ultrasound or in-utero MRI, as parents know from health care professionals' behaviour when a test result is abnormal. • Initial discussions should explain an abnormality has been found, what it is, and what further investigations are needed • Referral pathways for other investigations and advice from professionals should be agreed and timely, taking into account national abortion laws • Advice from other professionals can occur on the day of diagnosis, although staff may not always be available and women may feel overwhelmed. Where it is not possible, follow-up appointments should be timely: it is better to wait a few days and provide high-quality information than see a family quickly and give wrong advice. • Technology, such as video clinics, should be available where families have difficulty attending hospital
Environment	<ul style="list-style-type: none"> • Rooms for counselling should be available in maternity services • Paediatricians should not see families in their own clinics, where families may wait surrounded by children • Rooms should be warm, welcoming, and calm • There should be no time pressures to vacate rooms to ensure there is time to discuss concerns fully. It is not uncommon for an appointment to take over an hour • A computer screen should be available to allow families to see images, such as iuMR, and to review webpages of useful information and relevant organisations • Full information should be available to those providing counselling, including family history, genetic results, general health issues of women and their partner, and neuroimaging
Who performs counselling and is part of the team	<ul style="list-style-type: none"> • There should be a cohesive team who work together, respect expertise, create referral pathways, guidelines and information leaflets, review cases regularly, and provide emotional support to each other • Initial consultations and breaking of news will occur in obstetric services with sonographers, midwives and obstetricians • Women and their families should have access to specialist professionals to explain the abnormality and talk about whether it put the baby at risk of medical or neuro-developmental problems • If complications at birth are possible, a neonatologist should be present to discuss postnatal management • If the child is likely to die after birth, paediatric / perinatal palliative care teams should be involved • Where neurological or developmental morbidity is a possibility, counselling should be provided by an expert who performs antenatal counselling regularly and has experience of managing children with similar problems throughout childhood.
What is communicated and how	<ul style="list-style-type: none"> • A discussion should start with asking the prospective parents what they understand or have found out about the abnormalities, their hopes and expectations, and listening to any thoughts or experiences they have about children with disabilities and quality of life • Medical terminology should be explained in lay terms • Diagrams of normal anatomy and the abnormality should be available • If the family want, they should have the opportunity to see the scan • Discussions should not be overly technical / biological • A clear plan should be made for any other necessary tests, where appropriate • An explanation of the range of possible outcomes should be offered, including best and worse-case scenario

	<ul style="list-style-type: none"> • Outcomes should be functional and not purely medical diagnoses, i.e. “what does it mean in real life?” • Where there is hope, acknowledge it, no matter how small • Where a range of experts are required, a joint meeting will reduce conflicting or contradictory information being given • Discussions should not be judgemental, verbally and non-verbally. • Information leaflets should be given to families, where available, or links to reliable internet resources • Professionals should avoid telling families to avoid internet searches, even if you think they will scare themselves. • A summary letter of what was discussed, in lay terms, should be sent to the family and relevant teams following the appointment • Women and their families should be offered follow-up opportunities to clarify information and ask questions • When a woman has made a decision within the law, the clinician should acknowledge this and validate it as the “right decision for them”, even if this is different from their own personal view
How risk is communicated	<ul style="list-style-type: none"> • Tell the family what the base line risk of developmental problems is to allow for comparison • Use either percentages or fractions. If diagrams are available, use fractions • When using fractions, ensure the denominator is the same. Given low risk probabilities (such as 0.1%) are unlikely to occur when discussing developmental impairment, we recommend using 100 as the denominator • Try to avoid using verbal descriptions such as “very high” or “low”. Where you do, present the statistics alongside them • Use icons / pictograms to demonstrate risk and present the numerical figures alongside them • Frame any fraction or percentage you give to show the positives AND negatives, i.e. “your child has a 10 out of 100 chance of epilepsy, which also means 90 out of 100 won’t develop epilepsy” • Avoid using increases in percentages to explain risk, such as “your child is 20% more likely to develop cerebral palsy…” • Avoid relative risks, such as “your child is 4 times more likely to have difficulties than a child without these findings”
Support for pregnant women and their families post diagnosis	<ul style="list-style-type: none"> • Emotional and psychological support should be easily accessed for pregnant women, either by trained counsellors or psychologists • Referral pathways to mental health services should be clear and timely • Support should be provided to both pregnant women and their partner • Male partners should be encouraged to talk about their feelings and to discuss their grief, sense of loss. Although these cases are seen in maternity services, partners also need support • Where prospective families decide to continue their pregnancy, the paediatric professional providing counselling should endeavour to ensure continuity of care, including writing a plan for postnatal care, and reviewing the baby after birth. • Avoid repeated “offers” of termination of pregnancy where a decision has been made to continue with the pregnancy
Postnatal developmental follow-up	<ul style="list-style-type: none"> • Opportunity for postnatal review of the baby in a neurology / development clinic should be available, if parents wish • Clear referral pathways need to be made to facilitate access to paediatric follow-up • Preferably, developmental or neurological follow-up should be initially with the person providing antenatal counselling to allow for continuity of care and improve prognostication • Developmental follow-up should be available until at least school age, unless the family opt out • The professional providing follow-up should facilitate referrals to other appropriate specialisms when they arise
Improving prognostic data	<ul style="list-style-type: none"> • Review postnatally by the team providing antenatal counselling will improve prognostication in the long-run • Further research is required into the developmental significance of many lesions detected antenatally • There needs to be better liaison and communication between teams around the UK on difficult cases to improve prognostication, and involvement in data collection, service evaluation and research.