Family communication between children and their parents about inherited genetic conditions: a meta-synthesis of the research

Metcalfe, A. , Coad, J. , Plumridge, G.M. , Gill, P. and Farndon, P.

Original citation & hyperlink:
Metcalfe, A. , Coad, J. , Plumridge, G.M. , Gill, P. and Farndon, P. (2008) Family communication between children and their parents about inherited genetic conditions: a meta-synthesis of the research. European Journal of Human Genetics , volume 16 : 1193–1200.
http://dx.doi.org/10.1038/ejhg.2008.84

Please note Jane Coad was working at the University of the West of England at the time of publication.

Family communication between children and their parents about genetic conditions: A meta-synthesis of the research

Authors:
Dr Alison Metcalfe (Corresponding Author)
Senior Research Fellow,
School of Health Sciences,
University of Birmingham,
52 Pritchatts Road,
Edgbaston,
Birmingham
B15 2TT
Tel +44 (0)121 414 2666
Fax +44 (0)121 415 8087
Email: a.m.metcalfe@bham.ac.uk

Dr Jane Coad,
Senior Research Fellow
The University of the West of England,
Centre for Child and Adolescent Health,
Hampton House,
Cotham Hill
Bristol.
BS6 6JS.
Tel – 0117 331 0754
Fax – 0117 331 0893
Email: **Jane.coad@uwe.ac.uk**

Honorary Senior Research Fellow
School of Health Sciences,
University of Birmingham,
Email: **jane.coad@bham.ac.uk**

Gill Plumridge,
Research Associate
School of Health Sciences,
University of Birmingham,
52 Pritchatts Road,
Edgbaston,
Birmingham
B15 2TT
Email: g.plumridge@bham.ac.uk

Dr Paramjit Gill
Senior Lecturer
Department of Primary Care
University of Birmingham,
Edgbaston,

Birmingham
B15 2TT
Email: p.s.gill@bham.ac.uk

Prof Peter Farndon
Consultant Clinical Geneticist / Professor of Medical Genetics
West Midlands Regional Genetics Unit
Birmingham Women Healthcare NHS Trust
Metchley Park Road
Edgbaston
Birmingham
B15 2DP
Email: p.a.farndon@bham.ac.uk

Meta-synthesis of qualitative research on family communication of genetic risk information

Abstract

Background: Family communication with regard to inherited genetic conditions is highly complex. There are communication needs around the illness, its management and morbidity. But there is also a need for intergenerational communication about inherited risk and the implications this has for children, and their future health and reproductive decisions. We aimed to systematically explore and analyse the qualitative and quantitative research to explore the issues surrounding family communication about genetic conditions and genetic risks.

Method and Findings: A systematic review of all major heath and medical research databases was undertaken using current guidelines. In total 9698 abstracts were identified of which 158 research papers were selected and reviewed as potentially relevant. A final 17 papers were identified which met our predefined inclusion and exclusion criteria. The findings from these papers were subjected to a meta-synthesis. Using a meta-ethnographic approach, the studies' findings were analysed as primary data sources by three researchers independently identifying the key concepts to emerge. A high level of congruence emerged between the three researchers and concepts agreed were used to examine similarities and differences between the papers. The findings were used to inform a narrative framework exploring the issues surrounding the communication of genetic risk information between parents and their children. This narrative explored parents' explanations of inherited genetic risk to their children, the reasons for sharing information, children's understanding of parents' explanations, the emotions evoked for all family members, and the support and guidance received from health professionals.

Conclusion: From the narrative we were able to identify key components of successful communication to support children's coping with genetic risk information. However further empirical research is required into developing suitable strategies and materials to support parents' and children's information sharing, through the transitions of readjustment to the impact of the genetic condition at different stages of maturity and role change within the family.

Keywords: Family communication, genetics, meta-synthesis, meta-ethnography, coping, systematic review

Introduction

Family communication with regard to inherited genetic conditions is a highly complex process. There are the communication needs around the illness, its management and morbidity, all of which can be stressful. But there is also a concomitant stressor because there is a need for intergenerational communication about risks of inherited conditions and the implications this has for children†, and their future health and reproductive decisions.

Parents face the dilemma of when, how and what to tell their children about the genetic condition, its morbidity and associated inherited risks, and its implications for their own future children; whilst simultaneously trying to foster a robust self concept and self esteem in their child[1] and limit their anxiety. Conversely, if parents choose to protect the child from the reality of the condition, they have the difficulty of maintaining precarious secrets that others may unwittingly disclose. Revealing the information to the child later in adulthood requires them to rethink their self identity, which may affect their life expectations and aspirations; whereas a younger child has the opportunity to incorporate the genetic information into their self identity[2]. Parents delaying discussion of the genetic condition and its implications, risk their child's resentment and anger which can seriously damage the family's relationships and consequently undermine its support structures[3,4].

In the last 25 years, open communication about family illness has been encouraged by health professionals, based on assumptions that open communication with children allows them to express hidden feelings and discuss and correct distorted notions about the illness in their family[5]. Whilst also expressing parental trust in the child's ability to cope, and assists in preparing them realistically for any role changes[5]. The benefits of open communication observed in families affected by cancer for example, shows children's anxiety levels are lower, all round communication is improved and fewer behavioural problems are noted, compared with families who choose non-disclosure[6,7,8,9].

Footnote: †Children refers to children and young people <18years of age. For the purpose of this review where we refer specifically to young people, these are 13-17 year olds.

The study of family communication about genetic conditions and genetic risk information in the main focuses on the reasons for and against genetic testing of children whilst minors. However a more important focus, which to date has received less attention is the process of parents' and carers' communication with children about genetic conditions affecting their family, and the consequent outcomes for the child in coping and living with this information.

Children's experiences of family communication lay the foundation for their communication skills throughout their lifespan. Successful communication helps families as individuals and as a collective unit, to be responsive to change and to cope with and adapt to internal and external pressures[10]. Experiences of finding out or not knowing about a genetic condition and understanding the implications for self and other family members are therefore likely to have profound reverberations for the functioning of the family unit, and be influential on the individuals' psychological wellbeing and decision-making. This is supported by several studies which highlight that individuals' experiences of finding out about a genetic condition affected their subsequent disclosure decisions[11], choices about genetic counselling and testing[12,13] and the cohesiveness and support of the family unit once faced with dealing with the effects of an inherited disease[3,4].

The aim of this study was to systematically explore and analyse qualitative and quantitative literature to produce a meta-synthesis in a narrative form exploring the issues surrounding family communication about genetic conditions and genetic risks between parents and their children. To achieve this, we had to agree on definitions of 'family' and 'family communication' before commencing the work.

Family

The term 'family' can reflect a different discourse depending on the context in which it used[14,15]. In everyday kinship terminology 'family' is a psychosocial definition

describing a group of people who live together caring for children and each other[16]. All of whom are likely to affect and support the management, understanding and coping of children affected by or at risk from inherited genes causing disease. Therefore our focus is principally on the psychosocial definition of family (table 1), which is inclusive of genetically and non-genetically related individuals and partnerships with a responsibility for raising and supporting children, and communicating information about any genetic condition that may affect them.

Family communication

Family communication is a frequently used term but difficult to define[17]. There are no specific theories *per se*[16,18] but several different models of family interaction have been applied to try to explain it. These models are not exclusive; they tend to focus on a particular aspect that is important in sustaining or understanding the communication process in families. Some models describe regulation of family functioning (family systems theory), others examine the symbols and language used in interactions between family members (symbolic interaction theory[10] and finally, behavioural theories are used to investigate the psychological outcomes of family communication for example in coping, adaptation or cohesiveness (eg. Social Learning Theory[19]).

Having defined family and family communication so that we could agree the types of study that were of relevance, we set out the objective of our meta-synthesis which was to explore the evidence about children and parents' (including carers) communication about genetic conditions to answer the following questions:

How, what and when were genetic conditions and genetic risk information discussed in the family between parents (carers) and children?
What factors affected family communication? (For example: ethnicity, age, level of cognitive development, sex and genetic condition)
What effect did sharing this information have on the children and the parents?
What theoretical frameworks were used to explore family communication?

How did our findings compare with family communication in relation to other childhood illnesses?

Methods

Conventional systematic review methodology is ill suited to examining a range of diverse studies[20] produced on family communication about genetic risk information between children and their parents. There is no single accepted method of synthesizing the evidence from qualitative and mixed methods studies which might also include a quantitative component[21]. However there is a growing move to increase the transparency with which evidence from qualitative and mixed method studies are used to inform and develop a trustworthy consensus of the overall findings within a particular field. This allows research users to use the evidence with a degree of reliability knowing that undue emphasis has not been placed on one finding above another.

There are several examples of qualitative meta-synthesis[20,22,23,24], and we applied a narrative synthesis approach described by Popay et al.,[21] which is based on Noblit and Hare's guidelines for meta-ethnography[25]. Not all of our included studies were ethnographic, but there is agreement that the same guidelines can be applied for synthesizing other qualitative and quantitative data[21,22,24].

Search Strategy

Using Centre for Reviews & Dissemination (CRD)[26] guidelines, eligible papers published between 1980 and 2007 were identified using electronic databases, personal contacts and hand searches. Papers were included if they were directed at family communication relating to genetic conditions, chronic illnesses or cancer. Searches were conducted between May and August 2006 and have been updated by all available alerts in the intervening interval to May 2007. (Figure 1 for a flow diagram of the study identification and selection process).

Of the 158 papers identified as potentially relevant, each was examined to ascertain whether it met the inclusion criteria by two of three researchers (AM, JC or GMP).

Inclusion and exclusion criteria were agreed by the research team prior to commencement of the review in order to focus upon the research questions and to allow for comparison of family communication in other situations ie where a family member is affected by chronic disease or cancer. A total of 30 papers were identified. These included papers on family communication and chronic conditions or cancer. Nineteen papers were focusing on family communication and genetic conditions. These nineteen papers were critically appraised using Mays and Pope qualitative appraisal guidelines[27] by two of the researchers to assess the quality of the paper. All papers were included except for two where the results were so ambiguously written, none of the researchers could interpret them.

From seventeen papers identified on family communication about genetic conditions (table 2), the three researchers independently took the findings of original studies and treated these as primary data to identify first level concepts[21,24]. Three researchers were used to carry out the analysis to increase the reliability of judgements about the findings and reduce personal bias. These first level concepts were analysed to produce a secondary level of conceptualisation, identified as emergent theme concepts (table 2). By comparing and analysing these different concepts across the papers, similarities and contradictions could be observed and explored to produce tertiary level concepts, which informed our conclusions and guided the theory development of our discussion[20,23]. The concepts and interpretations of each researcher were aggregated and examined for similarity and consistency. The consistently derived concepts were synthesized into a narrative to provide a description of the findings from across the studies, to identify the different factors involved and to explore the relationship between them[21]. This narrative was structured around a framework agreed by the three researchers. To test our concepts and conclusions further, we compared our findings with other studies on communication in families affected by cancer or other chronic disease that were uncovered by the literature search.

Findings

A large degree of congruence emerged between the three researchers who had interrogated the data independently. Four over-arching components to the framework were identified, which allowed the incorporation of the derived concepts into a narrative. (Table 3 – summary of papers and the components they informed). Predominantly qualitative studies were identified but several papers also included quantitative results.

Under each component of the framework, a narrative of our triangulated and agreed secondary and tertiary conceptualisation of the data is provided; occasionally divided into sub-components using italicised headings, for clarity.

A large number of the studies identified (N=158) focused the communication of genetic risk information to children by parents, where 'the children' are the adult offspring rather than minors. These studies were not included in the meta-synthesis unless they also included a specific section on communicating with children less than 18 years old.

Narrative framework

1. Parents' explanations of genetic conditions and the risks to their children.

Decision to share genetic risk information

Parents often struggled with what and when to tell their children recognising they had different concerns and questions depending on their ages. An essential aspect of helping parents cope and overcome feelings of panic, fear and anxiety when a child was affected by a genetic condition was access to information. However parents often did not recognise that their affected child and unaffected siblings might have similar feelings and information might help them too.

Over the 27 year time span of the publications included, parents appear to have become increasingly open and honest with their children about genetic conditions affecting their family**.** In more recent studies up to half of parents reported openly communicating with their children about all aspects of a genetic condition affecting their family, including mortality risks where this applied. A further large proportion of parents reported open

communication but did not discuss mortality where this was an issue. Only a small minority stated they did not discuss the genetic condition but this was usually when the children were under 8 years of age.

Strategies used

The process and detail of parents' discussion and explanations about genetic conditions with their children have not been extensively explored. Where they have been, most parents described carefully considering when to share information, what their child needed to know and how much they felt the child could handle at that time. Explanations focused on the management of the condition and promoted positive attitudes sometimes using reframing strategies. For example, making comparisons with other childhood illnesses where symptoms or management could be viewed as more problematic than the condition affecting their own child.

Parents often waited for the child to ask questions before they gave any information or explanation although some said they started to introduce the idea of inheritance from preschool. Such young children were told they were born with a condition and parents elaborated on this by explaining to early school age children that the condition is passed on from one or both parents. Studies from the 1980's found that parents gave one – off explanations and did not check their children's understanding but later work suggests parents viewed information sharing as a continuum, a process that evolved through childhood and adolescence, gradually increasing the child's knowledge with parents wanting to be open and honest relevant to the child's capability of understanding**.**

Who should share information about genetic conditions with children?

The consensus from several studies and the overall expressed view of parent and child participants was that parents should primarily be responsible for discussing genetic conditions and genetic risk information with their children. Parents often wanted to tell their children about genetic inheritance before others told them or information was 'leaked' from other sources such as extended family, teachers or peers. It was believed that children needed information before specific life events such as developing their first

sexual relationship. There was some indication although not fully explored in the literature that parents needed time to make sense of the genetic risk information before they could discuss it with their children.

Mothers were often viewed as the best sources of information and support by children and young people; and in many studies that described research with 'parents', it was predominantly women who participated. This predominant role of female parents is also noticeable because when their communication was inhibited by guilt and grief if a child had a serious maternal X-linked recessive condition, unaffected siblings of the child reported poor family communication about the illness or its implications.

2. Reasons for discussing and sharing information

Some parents who emphasized open communication felt a strong sense of responsibility to discuss the information about inherited risks because it prevented a child from worrying, and promoted trust and open communication. Parents were often motivated to keep their children informed as a reaction to their own experiences as children when information had been withheld from them, leaving them growing up feeling puzzled and confused by what was happening.

Parents reported that they, and their children, found discussion of the condition difficult and that openness did not lessen the psychological and emotional pain of living with the condition and knowledge of your own possible risk. Openly discussing the condition and its effect empowered the family and enabled individuals to discuss matters and concerns as they arose; and increased their support and care for each other. Outcomes of openly communicating genetic risk information to children were not largely considered in the research but where they were, mothers openly discussing hereditary breast and ovarian cancer found it did not affect their children's general behaviour or well-being.

By contrast, in families where the communication was more closed, children often felt upset and frustrated with the family secrecy. Adolescent children maintained the secrecy even though they were unhappy with it. Even when the illness was finally discussed,

some still felt a prevailing atmosphere of secrecy and were anxious that there may be other secrets that were not being disclosed. Whilst limited communication protected the individuals initially, the inability to openly discuss problems and issues as they arose resulted in tense relationships between family members.

Even where parents managed to successfully hide information about a genetic condition in the family from their children until they were adults, these adult children were usually resentful and felt they should have been told. Adult offspring, regardless of whether they personally did or did not have knowledge as children about a genetic condition affecting their family, thought retrospectively that to have such knowledge was important. This would empower the child in making their life and reproductive choices and decisions with time to adjust to the information and avoid family secrets.

3. Children and Young People's Understanding

Parents reported trying to give children information appropriate to their stage of development but there were no comprehensive descriptions of this process provided. Further, none of the studies explored children's understanding based upon the parental reports of the information that had been discussed. However, in the small number of studies involving children, they were often more cognisant than their parents anticipated. For example, adolescent girls placed more emphasis than their parents of the potential psychological risks of carrier testing if undertaken at a young age.

Where more open communication existed, young people as they matured into adulthood were cautious about their reproductive decisions and understood the possibility of genetic testing and its consequent affect on their choices and psychological health. Where the condition affected another family member and may have risks for them in the future, young people emphasized the value of knowing because they were able to offer support to the affected individual and each other and would try not to worry too much for themselves. In contrast, poor communication led to reproductive choices based on inaccurate information and emotionally driven decision-making, which adult children felt with hindsight, more information during childhood would have prevented.

Where parents attempted to protect children by not discussing the genetic condition or the transgenerational risks, children picked up snatches of information but were very often confused by what was happening. Adult offspring recalling their childhood found out information about their condition or that of a family member from a variety of sources including television, other children with the same condition, school and mailings. This often resulted in misconceptions and misunderstanding. Children were unable to clarify their thoughts or interpretations due to the secrecy and felt obligated to protect their parents from having to answer difficult and emotionally taxing questions. Some children thought health professionals were likely to be a good source of information or support, particularly unaffected siblings but few had opportunity to access health professionals.

4. Emotions and feelings evoked for parents and children

Many studies explored communication in terms of what information was shared and by whom. Few however explored the feelings and emotions involved in discussing genetic risk information, either of the parents, the child living with a genetic condition or their unaffected siblings, individually or as a family.

Parents' emotions

Parents' emotions were not overtly explored but feelings of anxiety, worry and concern emerge with many using their own experiences of a genetic condition in the family to inform how they handle information giving to their own children. The majority of parents in all studies report a complete lack of support or advice from health professionals about discussing genetic conditions with their children. Where health professionals did broach the subject, it was usually focused on disease management.

Parents sometimes reported feeling afraid to discuss their child's emotions of worry, depression, frustration or embarrassment. Even if they observed deterioration in their children's behaviour through expressions of anger and aggression, they were afraid of making the child feel worse if too much attention was focused on the problem. By contrast, those parents that discussed feelings said their child could be helped to feel

better because they could provide reassurance that their feelings of anger, upset and frustration were normal and they could discuss ways of coping with the emotions. Parents who openly communicated with their children never expressed regret about discussing the genetic condition with their families. Whereas adult offspring who had the truth hidden from them by their parents expressed resentment and continued distrust and did not appreciate the 'protection' their parents had tried to provide.

Children affected by or at risk of a genetic condition

Children and young people growing up knowing the possible outcomes including their own risk found the information difficult to deal with initially but valued the honesty and openness because it allowed them to discuss, share experiences and learn to cope with the condition. In families where there was more open communication, children were reported to be more emotionally and psychologically resilient. They were often pragmatic in response to genetic risks for themselves.

Siblings

Guilt, fear, resentment and jealousy emerged as key features of studies which included the retrospective perspectives of now adult siblings. Often these feelings had not been discussed with parents. Several different types of guilt were expressed based on their feelings and behaviour toward their sibling at the time of their illness. But also guilt about feeling relieved firstly that they were not affected, and secondly that they could leave the family home upon reaching adulthood.

Some siblings reported intense relationships with an affected sibling and others remoteness. Resentment and jealousy were often described too; the well siblings resented their affected sibling if the parents were heavily reliant on them for helping with the family chores or care provision. Some siblings felt their own developmental needs were often overlooked within the family and some simply felt jealous of the time and attention their ill sibling received, which led to feelings of isolation.

Siblings of a child affected by a genetic condition often expressed feelings of embarrassment and discomfort. They tried to choose emotionally adept friends but often felt their own peers had insufficient knowledge or experiences to have insight into their feelings and feared being stigmatised if less sensitive individuals found out.

Adult children felt that the lack of communication about a genetic condition which resulted in the death of a sibling caused difficulties for the families' mourning and often protracted it. These experiences sometimes affected the siblings' future reproductive choices; girls particularly did not assess their risk objectively of carrying X-linked conditions but relied on family experiences and gut feelings. Many guessed at whether they were carriers and reported basing their life transition decisions on these suspicions rather than requesting genetic counselling.

Discussion

The findings from the narrative suggest that the components of successful communication between parents and children about genetic condition are; provision of information, checking understanding and encouraging discussion, and explaining and managing the emotional feelings that are manifested. These components as the basis of family communication about a genetic condition provide the foundations to support the family members' coping and adaptability and build trust, support and cohesiveness across the family unit.

Our findings advocate open communication with children about a genetic condition and associated risk of inherited disease, appropriate to their level of developmental maturity, which is likely to be more beneficial than trying to protect them by keeping the information secret. Studies that included children's views, suggested that they found the information upsetting and 'difficult to deal with' initially but valued being able to talk openly about the genetic condition in their family, which gave them a strong sense of mutual support. Open communication prevented unnecessary worry, and promoted trust and the discussion of children's feelings. This openness appears to improve children's coping and adjustment to risk information through increased understanding about the

illness, which will effect the gradual realisation of implications for self and future children as proposed by Etchegary[44].

In families where there was less open communication, siblings reported their major concern about the possibility of having a child affected by a genetic condition was for their unaffected children's wellbeing. They were worried that family separation caused by long periods of hospitalisation would be detrimental to the unaffected siblings' health and contentment, perhaps projecting their own feelings in relation to their personal experience where they often described isolation, loneliness and frustration due to poor communication.

Despite the limited number of studies available, this meta-synthesis demonstrates the complexity of family communication with regard to genetic conditions and inherited risk information. Parents have a doubly difficult task. Firstly they have to understand the genetic condition and its management, and cope with and manage their own feelings following their or their child's genetic diagnosis. But secondly, parents also have to explain to their children about the genetic condition and its risk implications which many parents found difficult and struggled to know what and when to tell their children. The difficulties faced in communication across families suggest there is a significant need for increased support. Therefore advice and assistance is likely to be very important in helping parents cope and manage their own feelings and those of their children which can assist the family's functioning. However, little support was available for parents from health professionals about talking to their children.

Parents' reliance on their own experiences to inform how they handle information giving to their children probably underlines that many require support in talking to their children about genetic conditions. Information can be both empowering and threatening depending on the context in which it is used, how it is relayed and delivered, and the level of support in promoting understanding but also managing the feelings evoked. Help is needed for parents and children through the transitions of readjustment to the impact of the genetic condition or the risk to self and other family members, at different stages of maturity and

role change within the family. It is essential that information is given to children appropriate to their developmental stage using suitable strategies and materials for discussing genetic conditions and their implications, which in the longer term will assist children's coping and adaptation to the effects of the genetic condition and risk information.

Young people were concerned about discussing a genetic condition with their peers, which may have consequences for their maturation into adulthood. As children develop into young adults they increasingly turn to their friends and peers for support gradually becoming more independent from their parents. However the development of trustworthy and supportive peer relationships may be inhibited if children fear stigmatisation. Limited family discussion about a genetic condition may reinforce fears about stigmatisation, consequently affecting children's self esteem and identity, and inhibiting communication with their own future families. Further work is required but reducing the stigmatisation the young person fears is most likely overcome by open communication within the family, who will support the young person and assist them in coping with their friends' reactions.

In many families, we observed that it was predominantly women (mothers) who took part in the research studies on family communication. Reflecting previous findings[30,45] showing the responsibility for communicating genetic risk information is either assumed or allocated to women in the family. This needs to be carefully considered by health professionals for families affected by serious or maternal X-linked conditions, where communication may be more problematic due to the mother's feelings of grief and guilt[13,43] and additional support may be beneficial.

Few empirical studies use any family communication theory to underpin their investigation. Most have not explored the effect of family communication across all its members; parents, affected child and siblings. Therefore many of our conclusions are based on a wide variation of studies, often only covering a particular type of family members' (e.g. parent or sibling) perspective. Future work needs to be developed that

takes into account the different facets of family communication theory and apply it to family communication about genetic conditions and genetic risk. This includes an examination of the language and symbols used to convey genetic risk information between different family members and how well they understand it, the effect it has on the family system and the psychological outcomes for individuals' and the families' coping and adjustment. All of which may vary according to which family member is affected by the condition, the morbidity of the disease, the stage of child development and how these change as children mature. Such work will provide insight into how information about genetic conditions and associated risks can be most effectively communicated to children, for the benefit of both the parent and child.

With limited work available on family communication about genetic conditions we examined the literature on family communication and chronic disease, which turned out to be even more restricted. Comparable studies were found in family communication and cancer, and many of the difficulties families affected by genetic conditions described could be juxtaposed with those faced by parents and children affected by cancer [7,46,47,48,49,50].

Limitations

There are several limitations that need consideration in relation to our findings for this study and the methodology used.

Insufficient data do not allow conclusions to be drawn about variations in how and what information parents share with their children depending on the morbidity and mortality effects of the genetic condition, the timeline of disease development or the inheritance pattern; autosomal dominant, autosomal recessive, X-linked or later onset and partial penetrance. Nor do any of the studies examine children's understanding and interpretation of the information they are given.

The majority of studies do not include the child's perspective but rely on parental reports or on retrospective accounts. Other studies only briefly examined the communication of

parents with their children as a subsidiary component. None of the studies explored differences in communications between families of different ethnic backgrounds, in fact ethnicity of participants was rarely described, or variations based on alternative family structures for example one parent families.

The reliability of the meta-synthesis approach for qualitative research is sometimes questioned as it is unclear how reproducible the findings are likely to be between different research teams[20,23]. The benefits and limitation of the methodologies generally have been explored in detail by others (examples[20,21,23]). However we tried to overcome lack of reproducibility by triangulating the findings of 3 researchers who had independently analysed the papers and produced first, second and third order concepts, which were recorded and findings reported where they had been identified by at least two of the three researchers. Whilst we cannot demonstrate reproducibility between different research groups, similar to others developing this methodology[20,21] we would argue that this type meta-synthesis approach merely reflects the inductive approach of qualitative research where variations between researchers on primary data is equally likely. The value of the meta-synthesis is that it is more in depth than a traditional literature review as it draws on all the available research findings; treats them objectively and charts the analysis process in detail to demonstrate the transparency and robustness of the outcomes.

Many of the studies we included could not always be described as high quality methodologically; several for example had only a small sample size. The papers were included however, because their findings added to the breadth of evidence from the other studies. This is increasingly gaining acceptance in meta-synthesis of qualitative and mixed methods studies[20,21,22,24] where the quality levels of papers might be difficult to demonstrate for a variety of reasons but often not directly related to the quality of the research conducted[21].

Conclusion

Based on the limitations uncovered by this meta-synthesis of the empirical research to date, further research is required to explore family communication across a range of

genetic conditions based on a theoretical framework. Examining communication between parents, affected children and their siblings with the aim of assisting parents and health professionals in choosing appropriate strategies to promote children's understanding, help them cope with the knowledge and manage the emotions evoked. With many parents reporting insufficient support from health professionals in advising them how to discuss genetic risk information, insights into children's living and dealing with genetic conditions is essential. This will provide evidence for health professionals to enable them to support families and ensure children are receiving sufficient information to promote emotionally and psychologically adept individuals who can care and help each other. Bearing in mind that what children learn from their families' communication about a genetic condition and associated risk will affect how they cope and adapt to situations in their own future relationships and families.

Acknowledgements
To be inserted

References

[1] McConkie-Rosell A and Spiridigliozzi G A (2004). Family matters: a conceptual framework for genetic testing in children. *J Genetic Counseling*, vol.13; 1: 9-29.

[2] Malpas, P J (2006). Why tell asymptomatic children of the risk of an adult-onset disease in the family but not test them for it? [Review]. *Journal of Medical Ethics*, vol. 32, no. 11, pp. 639-642.

[3] Sobel S & Cowan C B (2000) Impact of genetic testing for Huntingtons Disease on the family system. *American Journal of Medical Genetics* 90:49-59.

[4] Sobel S & Cowan C B (2003) Ambigious loss and disenfranchised grief: the impact of DNA predictive testing on the family as a system. *Family Process* 42; 1: 47-57.

[5] Rosenheim E & Reicher, R (1985). Informing children about a parent's terminal illness. *Journal of Child Psychology & Psychiatry & Allied Disciplines*, vol. 26; 6:995-998.

[6] Barnes J, Kroll L, Burke O, Lee J, Jones A, & Stein A (2000). Qualitative interview study of communication between parents and children about maternal breast cancer. *BMJ*, vol. 321;7259:479-482.

[7] Barnes J, Kroll L, Lee J, Burke O, Jones A & Stein A (2002) Factors predicting communication about the diagnosis of maternal breast cancer to children. *Journal of Psychosomatic Research* pp. 209-214.

[8] Friesen P, Pepler C & Hunter P (2002) Interactive family learning following a cancer diagnosis. *Oncology Nursing Forum* 29;6:981-987

[9] Osborn, T. (2007). The psychosocial impact of parental cancer on children and adolescents: a systematic review. *Psycho-Oncology*, vol. 16; 2:101-126.

[10] Segrin C & For a J (2005) Family Communication. New Jersey, USA. Lawrence Erlbaum Associates.

[11] Wilson B J, Forrest K, van Teijlingen, E R, McKee L, Haites, N, Matthews E, & Simpson S A (2004). Family communication about genetic risk: the little that is known. [Review] *Community Genetics*, vol. 7; 1:15-24

[12] Koehly L M, Peterson S K, Watts B G, Kempf K K G, Vernon S W & Gritz R (2003). A social network analysis of communication about hereditary nonpolyposis colorectal cancer genetic testing and family functioning. *Cancer Epidemiology Biomarkers & Prevention*, vol. 12; 4:304-313.

[13] Fanos J H & Puck J M (2001). Family pictures: Growing up with a brother with X-linked severe combined immunodeficiency, *American Journal of Medical Genetics*, vol. 98; 1:57-63.

[14] Atkinson P, Parsons E & Featherstone K (2001) Professional constructions of family and kinship in medical genetics. *New Genetics and Society* vol. 20;1:5-24.

[15] Finkler K, Skrzynia C., & Evans,J P (2003). The new genetics and its consequences for family, kinship, medicine and medical genetics. *Social Science & Medicine*, vol. 57, no. 3, pp. 403-412.

[16] Koerner A F & Fitzpatrick M A (2002). Towards a theory of family communication. *Communication Theory* 12;1:70-91.

[17] DeGenova M K & Rice F P (2002) *Intimate relationships, marriages and families* (5th Edition) Boston: McGraw Hill.

[18] Kalbfleisch, P J (2002). Communication-based theory development: Building theories for communication research. *Communication Theory*, vol. 12; 1: 5-7.

[19] Bandura A (1977) Social Learning Theory, Upper Saddle River NJ: Prentice Hall.

[20] Dixon-Woods M, Cavers D, Agarwal S, Annandale E, Arthur A, Harvey J, Hsu R, Katbamna S, Olsen R, Smith L, Riley R, Sutton AJ (2006) Conducting a critical interpretative synthesis of the literature on acces to healthcare by vulnerable groups. *BMC Medical Research Methodology* 2006, **6**:35

[21] Popay J, Roberts H, Sowden, Petticrew M, Arai L, Rodgers M, Britten N with Roen K & Duffy S (2006) *Guidance on the conduct of narrative synthesis in systematic reviews. A product from the ESRC methods programme.* Report Version 1 Institute of Health Research, Lancaster University.

[22] Lucas PJ, Baird J, Arai L, Law C, Roberts HM (2007) Conducting a critical interpretive synthesis of the literature on access to healthcare by vulnerable groups. *BMC Medical Research Methodology*, 7:4 (15 January 2007)

[23] Mays N, Pope, C & Popay J (2005). Systematically reviewing qualitative and quantitative evidence to inform management and policy-making in the health field. *Journal of Health Services Research & Policy* Vol 10;S1:6-20.

[24] Britten N, Campbell R, Pope C, Donovan J, Morgan M, Pill R (2002) Using meta ethnography to synthesize qualitative work: a worked example. *Journal Health Services Research & Policy* Vol 7 No 4:209-215.

[25] Noblit GW & Hare RD (1988) *Meta-ethnography: synthesizing qualitative studies*. Newbury Park. Sage.

[26] CRD (2001) NHS Centre for Reviews and Dissemination: *Undertaking systematic reviews of research on effectiveness: CRD's guidance for those carrying out or commissioning reviews.* York: NHS CRD; 2001 *http://www.york.ac.uk/inst/crd/report4.htm Last accessed 24th May 2007*

[27] Mays N & Pope C (1995) Rigour and qualitative research. *BMJ* 311:109-112.

[28] Gallo A M, Angst D, Knafl K A, Hadley E, & Smith C (2005) Parents sharing information with their children about genetic conditions. *Journal of Pediatric Health Care*, vol.19; 5: 267-275.

[29] Kenen R, Arden-Jones A, & Eeles R. (2004). We are talking, but are they listening? Communication patterns in families with a history of breast/ovarian cancer (HBOC), *Psycho-Oncology*, vol. 13; 5: 335-345.

[30] Forrest K, Simpson S A, Wilson B J, van Teijlingen E R, McKee L, Haites, N & Matthews E (2003) To tell or not to tell: barriers and facilitators in family communication about genetic risk. *Clinical Genetics*, vol. 64; 4: 317-326.

[31] Miesfeldt S, Cohn W F, Jones S M, Ropka M E, & Weinstein J C (2003) Breast cancer survivors' attitudes about communication of breast cancer risk to their children. *American Journal of Medical Genetics Part C-Seminars in Medical Genetics*, vol. 119C;1:45-50.

[32] Tercyak K P, Peshkin B N, DeMarco T A, Brogan B M, & Lerman C (2002) Parent-child factors and their effect on communicating BRCA1/2 test results to children. *Patient Education and Counseling*, vol. 47;2: 145-153.

[33] Tercyak K P, Hughes C, Main D, Snyder C, Lynch J F, Lynch H T, & Lerman C (2001). Parental communication of BRCA1/2 genetic test results to children. *Patient Education and Counseling*, vol. 42; 3:213-224.

[34] Canam C (1987). Coping with feelings: chronically ill children and their families. *Nursing Papers: Perspectives En Nursing*, vol. 1987;19, 3:9-21.

[35] Canam C (1986). Talking about cystic fibrosis within the family - what parents need to know. *Issues in Comprehensive Pediatric Nursing*, vol. 1986; 9; 3: 167-178.

[36] Holt K (2006). What Do We Tell the Children? Contrasting the Disclosure Choices of Two HD Families Regarding Risk Status and Predictive Genetic Testing. *Journal of Genetic Counseling*, vol. 15; 4;253-265.

[37] James C A, Holtzman N A, & Hadley D W (2003). Perceptions of reproductive risk and carrier testing among adolescent sisters of males with chronic granulomatous disease. *American Journal of Medical Genetics*, vol. Part 1, 60-69.

[38] Tercyak K P, Peshkin B N, Streisand R, & Lerman C (2001). Psychological issues among children of hereditary breast cancer gene (BRCA1/2) testing participants, *Psycho-Oncology*, vol. 10; 4: 336-346.

[39] Fanos J H (1999). "My crooked vision": the well sib views ataxia-telangiectasia, *American Journal of Medical Genetics*, vol. 87; 5:420-425.

[40] Bluebondlangner M (1991). Living with Cystic-Fibrosis - the Well Sibling Perspective. *Medical Anthropology Quarterly*, vol. 5: 2: 133-152.

[41] Tyler A & Harper P S (1983) Attitudes of subjects at risk and their relatives towards genetic counselling in Huntington's chorea. *Journal of Medical Genetics* 20, 179-188.

[42] Hern M J, Beery T A, & Barry D G (2006). Experiences of College-Age Youths in Families with a Recessive Genetic Condition. *Journal of Family Nursing*, vol. 12; 2:119-142.

[43] Fanos J H, Davis J, & Jennifer M P (2001). Sib understanding of genetics and attitudes toward carrier testing for X-linked severe combined immunodeficiency. *American Journal of Medical Genetics Part A* , vol. 98:46-56.

[44] Etchegary H (2006). Discovering the family history of Huntington disease (HD). *Journal of Genetic Counseling*, vol. 15; 2:105-117.

[45] d'Agincourt-Canning L (2001). Experiences of genetic risk: disclosure and the gendering of responsibility. *Bioethics*, vol. 15; 3: 231-247.

[46] Chesler M A, Paris J & Barbarin O A (1986). Telling the child with cancer: Parental choices to share information with ill children. *Journal of Pediatric Psychology* 11[4], 497-516.

[47] Claflin C J & Barbarin O A (1991). Does "telling" less protect more? Relationships among age, information disclosure, and what children with cancer see and feel. *Journal of Pediatric Psychology*, vol. 16; 2: 169-191.

[48] Shands M E, Lewis F M, & Zahlis E H (2000). Mother and child interactions about the mother's breast cancer: an interview study. *Oncology Nursing Forum*, vol. 2000; 27; 1:77-85.

[49] Sloper P (2000) Experiences and support needs of siblings of children with c ancer. *Health and Social Care in the Community* 8(5): 298-306.

[50] Forrest G, Plumb C , Ziebland S, & Stein A (2006). Breast cancer in the family--children's perceptions of their mother's cancer and its initial treatment: qualitative study *BMJ*, vol. 332;7548:998-1003.

Table 1 Our psychosocial definition of family

Building on suggested definitions of Degenova & Rice (2002) [17] p2, and Koerner & Fitzpatrick (2002) [16], we define family as '*any group of individuals united by the legal ties of marriage or partnership, blood or adoption in which the people are committed to one another in an intimate interpersonal relationship where the members see their individual identities as importantly attached to the group they call 'family' which has an identity in its own right through a shared history and shared future, and the adult(s) cooperate emotionally and financially to support dependent individuals (and each other)'*. This definition is inclusive of genetically and non-genetically related individuals and partnerships responsible for the raising and support of children and young people.

Table 2 – Attached as a separate document

Table 3: Narrative framework components occurring in reviewed papers

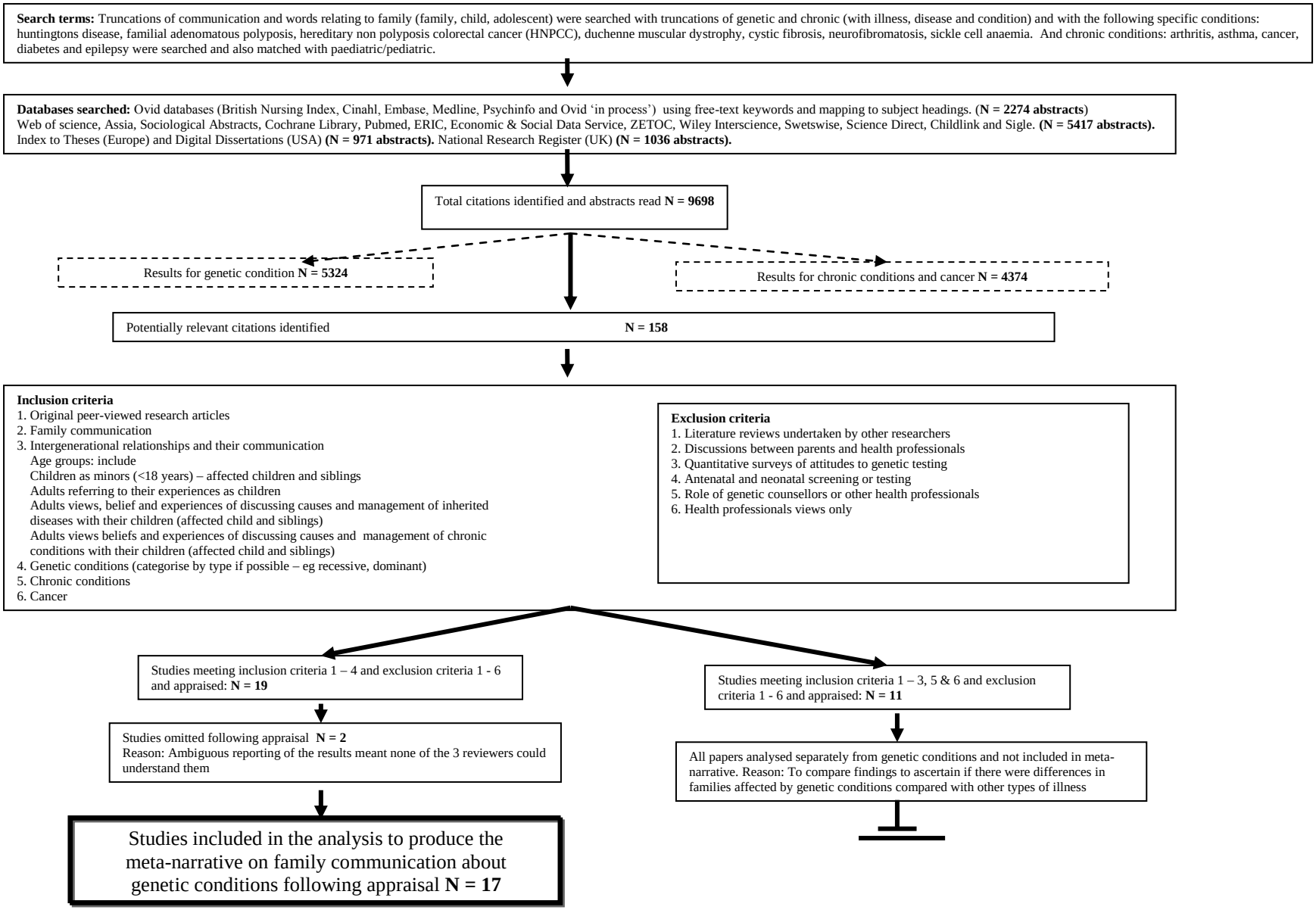
AUTHORS	DATE	THEMES			
		1	2	3	4
Studies primarily involving parents					
Gallo et al [28]	2005	✓	✓	✓*	
Kenen, Arden-Jones and Eeles [29]	2004	✓	✓		
Forrest K et al [30]	2003	✓	✓	✓*	
Miesfeldt, et al [31]	2003	✓			
Tercyak et al [32]	2002	✓	✓	✓	
Tercyak et al [33]	2001a	✓	✓		✓
Canam [34]	1987	✓			✓
Canam [35]	1986	✓		✓*	✓
Studies involving parents and children					
Holt K [36]	2006	✓	✓	✓	✓
James, Holtzman and Hadley [37]	2003	✓	✓	✓	✓
Tercyak et al [38]	2001b	✓			✓
Fanos [39]	1999	✓		✓	✓
Bluebond-Langner [40]	1991	✓	✓	✓	✓
Tyler & Harper [41]	1983		✓		✓
Studies primarily involving unaffected siblings					
Hern, Beery and Barry [42]	2006	✓		✓	✓
Fanos, Davis and Puck [43]	2001a	✓	✓	✓	✓
Fanos and Puck [13]	2001b			✓	✓

* = Children and Young People's understanding from parents point of view

Narrative framework components: Aide Memoir

1. Parents' explanations of genetic conditions and the risks to their children
2. Reasons for discussing and sharing information (including reasons for not doing so)
3. Children and Young People's Understanding
4. Emotions and feelings evoked for parents and children

Figure 1: Flow diagram of study selection process (see p25 or attached Powerpoint slide for clearer version)


Search terms: Truncations of communication and words relating to family (family, child, adolescent) were searched with truncations of genetic and chronic (with illness, disease and condition) and with the following specific conditions: huntingtons disease, familial adenomatous polyposis, hereditary non polyposis colorectal cancer (HNPCC), duchenne muscular dystrophy, cystic fibrosis, neurofibromatosis, sickle cell anaemia. And chronic conditions: arthritis, asthma, cancer, diabetes and epilepsy were searched and also matched with paediatric/pediatric.
Databases searched: Ovid databases (British Nursing Index, Cinahl, Embase, Medline, Psychinfo and Ovid 'in process') using free-text keywords and mapping to subject headings. (**N = 2274 abstracts**)
Web of science, Assia, Sociological Abstracts, Cochrane Library, Pubmed, ERIC, Economic & Social Data Service, ZETOC, Wiley Interscience, Swetswise, Science Direct, Childlink and Sigle. **(N = 5417 abstracts).**
Index to Theses (Europe) and Digital Dissertations (USA) **(N = 971 abstracts).** National Research Register (UK) **(N = 1036 abstracts).**
Total citations identified and abstracts read **N = 9698**
Results for genetic condition **N = 5324**
Results for chronic conditions and cancer **N = 4374**
Potentially relevant citations identified **N = 158**
Inclusion criteria
1. Original peer-viewed research articles
2. Family communication
3. Intergenerational relationships and their communication
Age groups: include
Children as minors (<18 years) – affected children and siblings
Adults referring to their experiences as children
Adults views, belief and experiences of discussing causes and management of inherited diseases with their children (affected child and siblings)
Adults views beliefs and experiences of discussing causes and management of chronic conditions with their children (affected child and siblings)
4. Genetic conditions (categorise by type if possible – eg recessive, dominant)
5. Chronic conditions
6. Cancer
Exclusion criteria
1. Literature reviews undertaken by other researchers
2. Discussions between parents and health professionals
3. Quantitative surveys of attitudes to genetic testing
4. Antenatal and neonatal screening or testing
5. Role of genetic counsellors or other health professionals
6. Health professionals views only
Studies meeting inclusion criteria 1 – 4 and exclusion criteria 1 - 6 and appraised: **N = 19**
Studies meeting inclusion criteria 1 – 3, 5 & 6 and exclusion criteria 1 - 6 and appraised: **N = 11**
Studies omitted following appraisal **N = 2**
Reason: Ambiguous reporting of the results meant none of the 3 reviewers could understand them
All papers analysed separately from genetic conditions and not included in meta-narrative. Reason: To compare findings to ascertain if there were differences in families affected by genetic conditions compared with other types of illness
Studies included in the analysis to produce the meta-narrative on family communication about genetic conditions following appraisal **N = 17**