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ARTICLE

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

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In families affected by an inherited genetic condition, parents face a difficult task of having to communicate genetic risk information to their children. A systematic review of all major health and medical research databases was undertaken using current guidelines to identify original relevant research papers from 1980 to 2007, which explore the issues surrounding parents and their children's communication about inherited genetic risk. A total of 9698 abstracts were found of which 158 research papers were reviewed as potentially relevant. A final 17 papers were identified which met predefined inclusion and exclusion criteria. Using a meta-ethnographic approach, all identified studies' findings were analysed as primary data sources by three researchers, who independently identified the key concepts. A high level of congruence emerged between researchers, and agreed concepts were used to examine similarities and differences between papers. The findings informed the development of a narrative framework exploring the issues that related to parents' explanations of inherited genetic risk to their children, the reasons for sharing information, children's understanding of parents' explanations, the emotions evoked for family members and the support and guidance received from health professionals. Providing information, checking understanding, and explaining and managing the emotional feelings that arise were integral to supporting children's coping with genetic risk information. However, many parents struggled with one or more of these components and required more support specific to the child's developmental stage, and family members' transition of readjustment to the impact of the genetic condition.

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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 (children refers to children and young people <18 years. Where we refer specifically to young people, these are 13–17-year olds).

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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 while simultaneously trying to foster a robust self-concept and self-esteem in their child[1] and limit their anxiety. 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. Also, revealing such information to children 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]

Research into family communication about genetic conditions mainly focuses on the reasons for and against genetic testing of children while minors. Receiving 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. With major developments in understanding the inheritance of a wide range of genetic conditions, health professionals are increasingly asked for advice about how to discuss these risks with children in affected families.

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 between parents and their children. The objective of which was to answer the following questions:

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

Methods

Conventional systematic review methodology is ill suited in examining the range of diverse and predominantly qualitative studies[5] produced on family communication about genetic risk information between children and their parents. Therefore, qualitative meta-synthesis was used,[5–8] using a narrative synthesis approach,[9] which is based on guidelines for meta-ethnography.[5] Not all studies were ethnographic, but there is agreement that these guidelines can be applied for synthesising other qualitative and quantitative data.[6,8,9]

Search strategy

Using Centre for Reviews and Dissemination (CRD)[10] Guidelines, eligible papers published between 1980 and 2007 were identified using electronic databases, personal contacts and hand searches. Searches were conducted between May and August 2006 and have been updated by all available alerts in the intervening interval to September 2007 (Figure 1 shows 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 (Figure 1) by the research team prior to commencement of the review and a working definition of what constituted 'family' developed (Figure 2). Nineteen papers were identified, focusing on family communication and genetic conditions and were critically appraised using relevant guidelines[15] by two researchers. Only two papers were omitted because of ambiguously written results.

From papers identified on family communication about genetic conditions (see Supplementary Table 1), the three researchers independently took the findings of original studies and treated these as primary data to identify first-level concepts.[8,9] Using three researchers increased the reliability of judgements about findings and reduced personal bias. These first-level concepts were analysed to produce a secondary level of conceptualisation, the emergent themes (see Supplementary Table 1). By comparing and analysing these different concepts across the papers, similarities and contradictions could be observed and explored. The concepts, interpretations and themes of each researcher were aggregated and examined for similarity and consistency. Consistently derived concepts and themes were synthesised from comparisons across the papers and the findings developed into a narrative,[5,8,16] exploring the themes, identifying different factors involved and examining the relationship between them.[9] This narrative was structured around a framework developed and agreed by the three researchers.

Findings

Seventeen papers were identified which were predominantly qualitative studies but some included quantitative results: eight studies involved parents only, three included children only and six included both parents' and children's perspective. A large degree of congruence

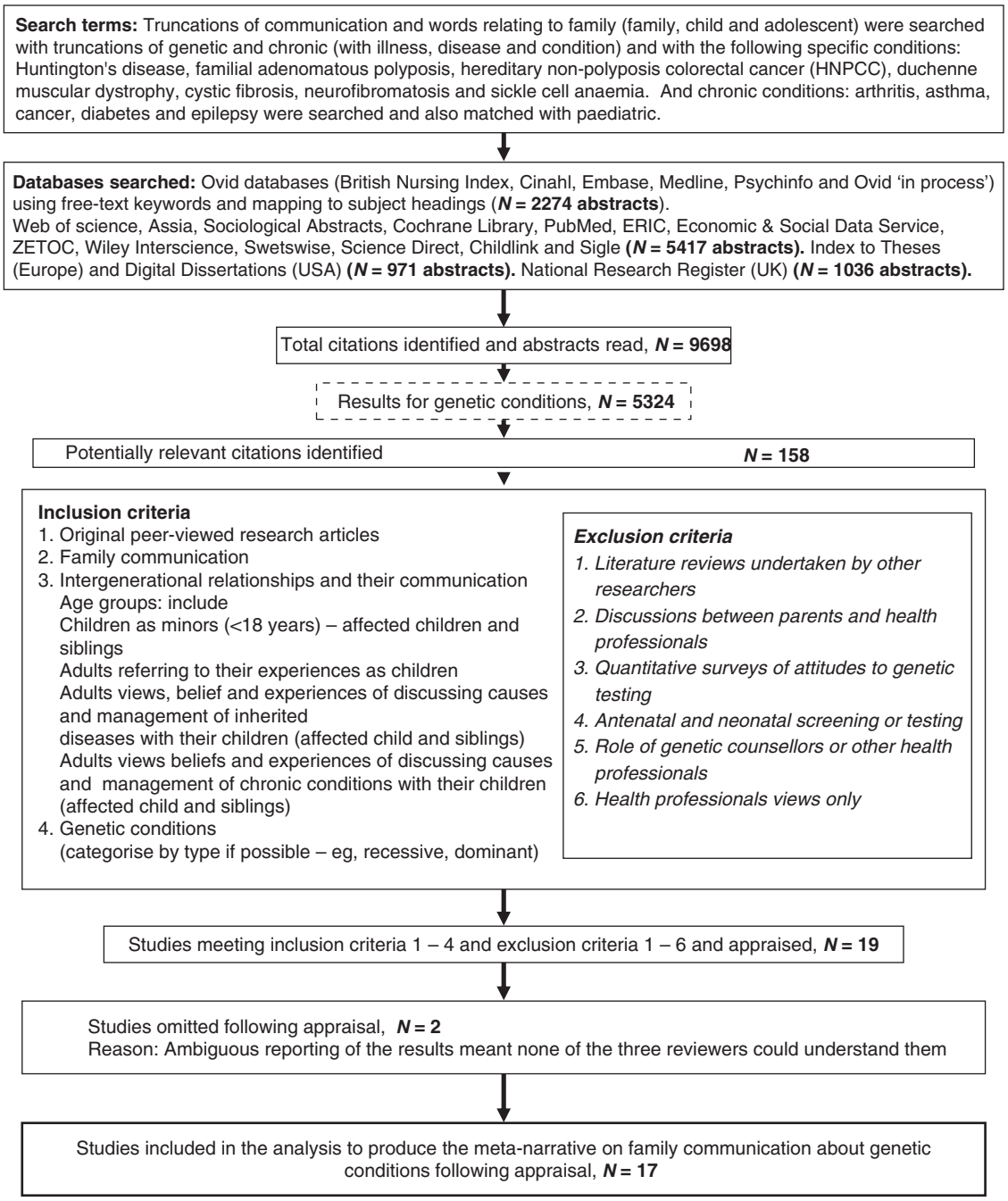

Figure 1 Flow diagram of study identification and selection process.

emerged between the researchers who interrogated the data independently. Four overarching components were identified that allowed the incorporation of the themes into a narrative framework (divided into italicised subcomponents for clarity) (papers informing relevant sections are referenced).

Narrative framework

Parents' explanations of genetic conditions and the risks to their children[17–31]

Decision to share genetic information Parents often struggled with what and when to tell their children recognising they had different concerns and questions

The term 'family' can reflect a different discourse depending on the context in which it is used.[11,12] Our focus is principally on the psychosocial definition of family. Building on suggested definitions of Degenova and Rice (2002)[13] p2, and Koerner and Fitzpatrick (2002)[14], 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.

Figure 2 Definition of family used by the research reviewers.

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 time span of the publications, parents appear to have become increasingly open and honest with their children about genetic conditions affecting their family. However, there was large variation in the information children were given, particularly, in relation to mortality, and in whether they were encouraged by their parents to seek further information.

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, most parents carefully considered: 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.

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

Who should share information 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, it was predominantly women who participated. It is unclear whether the responsibility for communicating genetic risk information is assumed or allocated to women in the family. The 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.

Reasons for discussing and sharing information[17–19,21,22,25,26,29,31,32]

Some parents who emphasised open communication felt a strong sense of responsibility to discuss information about inherited risks, because this 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 difficult and that openness did not lessen the psychological and emotional pain of living with the condition and knowledge of your own possible risk. However, it was felt that openly discussing the condition 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 studies, but, where they were, mothers found that 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 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. Although limited communication protected the individuals initially, the inability to openly discuss problems and issues resulted in tense relationships between family members.

Where parents managed to successfully hide information about a genetic condition until their children were adults, adult children were usually resentful and felt they should have been told. Adult offspring, regardless of whether they personally had knowledge as children about a genetic condition affecting their family, thought retrospectively that having such knowledge was important. It would empower the child in making their life and reproductive choices with time to adjust to the information.

Children and young people's understanding[17,19,21,24–26,28–31,33]

Parents tried to give children information appropriate to their stage of development but no comprehensive descriptions of this process were provided. Further, none of the studies explored children's understanding based on 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 on the potential psychological risks of carrier testing if undertaken at a young age.

Where 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 effect on their choices and psychological health. Where the condition affected another family member and may have risks for them in the future, young people emphasised the value of knowing because they were able to offer support to the affected individual and each other and tried not to worry too much for themselves. In contrast, poor communication led to reproductive choices based on inaccurate information and emotionally driven decision-making. Adult children felt with hindsight, that more information during childhood could have prevented this.

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

In families where there was less open communication, the main concern of siblings if they were to have a child affected by a genetic condition was with regard to the potential impact of this on the well-being of their other children. 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.

Emotions and feelings evoked for parents and children[22–33]

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

Parents' emotions Parents' expressed emotions of anxiety, worry and concern with many relying on 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 reported 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 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 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 or at risk from genetic conditions 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, and 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 by unaffected siblings based on their feelings and behaviour towards their ill sibling including guilt about feeling relieved that they were not affected, and also guilt that they could leave the family home on reaching adulthood.

Some siblings reported intense relationships with an affected sibling and others remoteness. Often 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 and some 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 that 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 objectively assess their risk of carrying X-linked conditions with many guessing whether they were carriers and reported basing their life transition decisions on these suspicions rather than requesting genetic counselling.

Finally, insufficient studies did not allow conclusions to be drawn about variations in how and what information parents share with their children depending on the morbidity and mortality of the genetic condition or the inheritance pattern. None of the studies explored differences in communications between families of different ethnic backgrounds or alternative family structures. Family communication theory was rarely mentioned and not used to comprehensively underpin any of the studies.

Discussion

The findings from this meta-synthesis show that many parents need more help and support to assist them in successful communication with their children about genetic conditions. Parents often had difficulty successfully communicating and often required advice on the provision of developmentally appropriate information, checking children's understanding and encouraging open discussion, and helping their children manage their emotional responses. Despite these difficulties and the complexity of communication about inherited genetic risk, little support from health professionals was available for parents or their children.

Genetic risk 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. Consequently, the implications for health professionals are that advice and support are 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 coping, adaptation and functioning. The findings suggest that good communication of genetic risk information by parents to their children will have long-term consequences, including informed reproductive decision-making and better family cohesion, laying a foundation for the child's future communication with their own family.

This meta-synthesis highlighted that support is required 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; however, few studies highlighted or explored suitable strategies and materials for discussing genetic conditions and their implications based on children's maturity, disease morbidity and outcome or inheritance pattern of the condition. Health professionals need to consider how they can support and advice families in discussions about genetic conditions and associated risks, although we are aware that, at present, there is currently only limited evidence on effective strategies and the wider implications.

We suggest that to inform the effective development of communication strategies and process about genetic conditions and associated risks for the benefit of both parents and children, future work needs to apply family communication theory. This includes an examination of the use of language and symbols to convey genetic risk information between family members and explore how well it is understood, the effect it has on the family system and the psychological outcomes for individuals' coping and adjustment.[34–36] Communication research should go beyond exploring the appropriateness of genetic testing of children but should consider the wider implications and the impact of living in a family affected by a genetic condition.

Limitations

Most studies did not explore the effect of communication across all family members' perspectives: parents, affected child and siblings, and none examined children's understanding of the information their parents had reportedly given them. The majority relies on parental reports or retrospective accounts. Other studies only briefly examined the communication of parents with their

children as a subsidiary component. Therefore, many of our conclusions are based on a particular type of family members' (eg, parent or sibling) perspective.

Several studies had a small sample size but were included because their findings added to the breadth of evidence from other studies, which is acceptable in meta-syntheses of qualitative studies[5,6,8,9] where the quality of research is related to the breadth of participants' experiences.

The benefits and limitations of the meta-syntheses method in qualitative research, most notably, the reproducibility of the findings between researchers, has been discussed at length elsewhere.[5,7,9] We tried to overcome lack of reproducibility by triangulating the findings of three researchers who independently analysed the papers and produced concepts and themes and developed the narrative framework. Findings were only reported where they had been identified by at least two of the researchers. Similar to others,[5,9] we argue that this meta-synthesis approach reflects the inductive nature of qualitative research where variations between researchers on primary data are equally likely. The value of meta-synthesis in qualitative research is that all findings are treated objectively, undue emphasis is not placed on one study above another and the analysis process is charted to demonstrate transparency. Thereby providing greater confidence in the reliability of the findings and conclusions reached.

Conclusion

Parents require greater support from health professionals in helping them communicate genetic risk information effectively. Further research driven by family communication theory is required to examine communication between parents, affected children and their unaffected siblings across a range of genetic conditions. This would inform parents and provide evidence to health professionals in choosing appropriate strategies to promote children's understanding, help them cope with the knowledge and manage the emotions evoked, at different developmental stages.

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