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ORIGINAL RESEARCH

Parents' Communication with Siblings of Children Affected by an Inherited Genetic Condition

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Abstract The objective of this study was to explore parents' communication about risk with siblings of children affected by an inherited genetic condition, and to ascertain what level of support, if any, is required from health professionals. Semi-structured interviews were conducted with affected and unaffected children and their parents. Families were affected by one of six genetic conditions representing different patterns of inheritance and variations in age of onset, life expectancy and impact on families. Interviews were analysed using constructivist grounded theory and informed by models which focused on three different aspects of family communication. Interviews with 33 families showed that siblings' information and support needs go largely unrecognized by health professionals and sometimes by parents. Some siblings were actively informed about the genetic condition by parents, others were left to find out and assimilate information by themselves. Siblings were given information about the current symptoms and management of the genetic condition but were less likely to know about its hereditary nature and their own potential risk. When siblings were fully informed about the condition and included in family discussion, they had a better understanding of their role within their family, and family relationships were reported to be more harmonious. The information and support needs of siblings can be overlooked. Parents with the responsibility for caring for a child affected by a genetic condition may require support from health professionals to understand and respond to their unaffected children's need for more information about the genetic condition and its implications for the children's own future health and reproductive decision-making.

Keywords Sibling · Inherited condition · Genetic risk · Family communication · Support · Genetic counseling

Introduction

There is a body of literature considering the experiences of siblings of a disabled or chronically ill child, which suggests that these children face potential difficulties (Houtzager et al. 2004, 2005; Sharpe and Rossiter 2002; Williams 1997). Fewer studies have focussed on the impact for siblings of having a brother or sister affected by a genetic condition. Such studies suggest this can affect siblings' behavior (Cowen et al. 1986) and lead to worry and jealousy or resentment towards the affected child (Phillips et al. 1985; Bluebond-Langner 1991; Derouin and Jessee 1996; Hutson and Alter 2007), and resentment of disruption to family activities (Derouin and Jessee 1996; Foster et al. 2001; Hutson and Alter 2007). Siblings have to come to terms with their brother or sister's illness and its effects on family life but with genetic conditions, they may also be at risk of being affected. That is, they may also develop the disease if it is later onset [e.g. Duchenne muscular dystrophy (DMD), familial adenomatous polyposis (FAP) or Huntington's disease (HD)] or they may be a carrier of the gene, which

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will have consequences for their future reproductive decision-making [e.g. in families with cystic fibrosis (CF) or haemoglobinopathies (HbO) and sisters of children affected by DMD)].

Giving children information about a genetic condition and associated risk, at a level appropriate to their developmental maturity, is likely to be more beneficial to them than trying to protect them by withholding information from them (Tercyak et al. 2002; Hern et al. 2006; Metcalfe et al. 2008). Communication within families about genetic conditions is not, however, uniform (Gaff et al. 2007), and siblings are a group whose information needs may be overlooked (Gallo et al. 2009). This has been highlighted in recent small-scale studies with siblings at risk of being a carrier of a specific recessive or X-linked condition, which suggest their information needs about genetic risk are often neglected (Hutson and Alter 2007; McConkie-Rosell et al. 2009; Wehbe et al. 2009).

Previous studies have shown that withholding genetic risk information from children can have profound consequences for the individual child and the family unit (Fanos et al. 2001; Fanos and Puck 2001; Sobel and Cowan 2003; McConkie-Rosell et al. 2009). Poor communication with siblings about an affected child's genetic condition can lead to confusion about the risk to themselves and their potential offspring (Hutson and Alter 2007). This confusion may result in poor reproductive decisions, not attending genetic counseling services (Fanos et al. 2001; Fanos and Puck 2001), and suffering lowered self esteem (Metcalfe et al. 2011). It can also lead to reduced family cohesion, resulting in diminished care and support for affected individuals over the longer term and into adulthood (Fanos et al. 2001; Fanos and Puck 2001; Sobel and Cowan 2003).

We recently undertook a large qualitative study to explore communication processes between parents and their children about genetic risk information. We interviewed 96 members of 33 families. We included all family members (aged 8 years and above) who wanted to take part, irrespective of their position in the family. Families were affected by one of six genetic conditions. This paper focuses on the experiences of siblings, their roles in family communication and the consequent emotional and behavioral outcomes. The "siblings" are those children who are not currently affected by the genetic condition. They may however carry a gene mutation for a late onset condition or may carry a recessive gene that puts their future offspring at risk.

Methods

Theoretical Framework

All stages of the study design and execution were informed by three inter-related family communication models. Family systems theory (Segrin and Flora 2005) views family members as elements that combine to create a family unit that is greater than the sum of the individual members. This theory addresses individual family members' roles and interactions within and outside of the family unit. Family systems theory is relevant to this paper in terms of considering the roles of unaffected siblings in relation to the genetic condition and affected families member(s) and how far they are included or excluded in management of, and communication about the condition. The theory also considers their interactions, particularly in relation to support, with those outside of their immediate family. Symbolic interaction theory (Blumer 1969) describes how family members communicate and relate to each other and attends to the language used in family communication about the genetic condition. Social learning theory (Bandura 1977) is a behavioral theory that focuses on outcomes. This theory was used to consider how children and young people coped with and adapted to the genetic condition and the information they received about it. Models informed the design of the interview schedule and were used as part of the grounded theory development for analysis.

An advisory group of parents and young people were consulted about the conduct, findings and recommendations of the study. This group included members of seven families that were affected by a genetic condition, and had responded to an invitation sent via support groups.

Participant Recruitment

Potential participant families were identified via voluntary and National Health Service support groups in England and Wales. Families affected by one of the following six genetic conditions were included:

- Cystic fibrosis (autosomal recessive: onset from birth)
- Duchenne Muscular Dystrophy (X-linked recessive: onset of symptoms in early childhood)
- Familial Adenomatous Polyposis (autosomal dominant: onset in adolescence)
- Haemoglobinopathies (autosomal recessive: onset anytime in childhood)
- Huntington's Disease (autosomal dominant: usually adult onset, often middle age)
- Neurofibromatosis (NF) (autosomal dominant: onset from birth, wide variation in signs and symptoms)

These conditions were chosen to encompass variations in inheritance pattern, in age of disease onset, and in age at genetic testing for conditions where this is relevant. Some are life limiting, and the impact on individuals and families in terms of illness duration and management also differs. This ensured that a wide range of views was represented.

Families were either given written information about the study or they witnessed presentations at conferences or support group meetings. They were asked to contact the research team if they wished to take part. Parents and all children, whether affected, at risk, or unaffected, were invited to take part. Children were only included when their parents: (1) confirmed they had personally explained the genetic condition and its risks to the child, (2) understood that the act of taking part may prompt questions from the child, and (3) expressed a willingness to answer the child's questions.

The study was approved by the Liverpool Children's Research Ethics Committee (REC No: 07/Q1502/16). In accordance with their stipulation we only interviewed children aged 8 years and above (or 16 years and above for Huntington's disease).

Data Collection

Semi-structured interviews were conducted with all family members who elected to take part; parents, children (8–12 years), young people (13–17 years) and adult children (18+ years). Interviews with children and young people were conducted by one of two researchers who are experienced in interviewing this group, and who have health or social care backgrounds. The parents were interviewed by one researcher experienced in genetic risk communication research and also from a health professional background. Parents and children were interviewed separately, except when a child wanted a parent present. This was rare and never required throughout a whole interview. Siblings were offered the choice of being interviewed together or separately. Only three families of siblings chose to be interviewed together. Consent was obtained to audio record interviews in all cases except one. Extensive notes were taken during this interview. Recorded interviews were transcribed verbatim.

Interview schedules were developed from the literature review and discussion with the advisory and steering groups and were informed by the family communication models. They were constructed to elicit information about family members' roles in discussing the condition and associated risks, the language used, how individuals coped with and adapted to the condition and its risks, and the impact of these factors on individual and family behavior. Interviews were responsive to the flow of individual participant's responses. Art materials were offered to all children and young people to help to create a more comfortable environment for discussing personal and sensitive information (Coad et al. 2009). They were asked to create an image of their family if they wished. Some did this and it aided discussion. Others declined, but used the materials to occupy them while talking, giving them, for example, the choice of when they wished to make eye contact. Others preferred to just sit and talk.

Analysis

Transcripts were analysed using social constructivist grounded theory (Corbin and Strauss 2008). Using interview transcripts from five families, affected by different genetic conditions, separate frameworks were developed for the analysis of parents' and children's interviews. This enabled management of a large quantity of data and kept children's and adults' perspectives separate at this stage. Two researchers developed each framework. They independently identified phenomena within each interview and developed concepts and a list of categories and then worked together to agree an overall list for the framework. As part of developing the framework the concepts were considered in terms of family communication models; for example the processes and roles involved in communication, and the behavioral outcomes reported by family members. The identified concepts were used to establish categories with clearly defined properties and dimensions. Once developed, each framework was rechecked for reliability and applicability against the first five interviews, plus the interview transcripts from two additional families that had been chosen randomly. The framework underwent modification to incorporate new phenomena and concepts as they emerged. Interviews that had been analysed previously were revisited to ascertain whether similar phenomena had been overlooked.

Data were coded into these frameworks using the software package NVIVO 7. The data were then examined using a series of grounded theory questions, which had been developed throughout all stages of analysis, from fieldwork to coding. These related to the research questions and inductive ideas and were informed by the family communication models. Each researcher examined data specific to two genetic conditions. The findings generated were randomly checked by a second researcher, and any disputes discussed by all three researchers until consensus was reached.

Finally, the findings from the family groups for each of the six conditions were compared using a constant comparative analysis (Corbin and Strauss 2008). This method allowed for exploration of the level of consensus between family members, in terms of how children learned about the genetic condition and risk, how much they knew and understood, and the level of openness and the shared language used in discussions.

Results

We interviewed a total of 52 parents (birth parents, guardians or step parents) and 44 children and young people from 33 families. The interviewees included 24 of

the 53 children and young people who were siblings of affected children or at risk of developing or carrying an inherited genetic condition. Twenty-two of the other 29 children were either too young or grown up and had left the family home. The remaining seven were excluded by their own choice or because their inclusion was inappropriate, usually because parents had made an active decision to withhold genetic risk information from them. Key aspects of the demographic profile of participants for the overall study are provided in Table 1.

Within the overall findings many similarities were observed across conditions. Where findings pertinent to siblings relate only to particular conditions this is specified. When transcripts were examined in family groups there was usually a strong level of consistency between reports about how, what and when children and young people had been told about a genetic condition and its risks. Shared language was also evident within families, and the views of parents were often reflected in the explanations and descriptions given by their children.

Parents Talking to Siblings About the Genetic Condition

Affected and unaffected children of all ages (8 years upwards) consistently said they wanted to receive information about the genetic condition from a young age. A few suggested an age, in one case as young as 3 years, but most said the condition should be discussed with children as soon as they were able to notice its effects on the family. Children wanted to be able to discuss this information within their families throughout childhood. They felt this facilitated a process of on-going realization and understanding. When directly asked when children should be given information about a genetic condition in their family typical responses were:

> "Like my age, not like my age now, but like seven,"
> (Young person affected by NF)

> "…it will help if they're young cause then they'll grow up already knowing"
> (Young person affected by HbO)

> "I think if a kid turns around and goes to the parents 'Mum or dad, what's wrong with you? Have we got anything in the family?' I think they should just be open and tell them… kids are more resilient to finding stuff like that out at an early age and being able to process it over years …I think they should just start talking about it at five or six when generally kids start to develop their own sense of who they are, what they want to be and stuff." (Young person at 50% risk for HD)

None of the participants (adult or child) suggested any regrets that information had been disclosed. In terms of what parents told children about the condition, there were, however, clear differences in most families between affected children and their siblings. In families with DMD, siblings were generally told more than affected children, whereas in families with other conditions they were told less.

For CF, HbO, and NF, parents sometimes gave siblings less information than affected children about the manifest aspects of the condition, but they were particularly less likely to tell them as much about heredity, and thus carrier risk and reproduction. In general these omissions appeared to result from making assumptions about siblings' knowledge or not recognising their information needs, rather than a deliberate strategy. Siblings were often thought to know about the

Table 1 Key aspects of the demographic profile of participants

Individuals Interviewed	Age range of children in years (gender)	Ethnicity of families	Genetic condition affecting families	Status of children in relation to genetic condition
52 Parents–34 Women and 18 Men	8–11 (5♀4♂)	5 African–Caribbean or Black British	4 Cystic Fibrosis	20 affected or tested positive for genetic condition
33 Children (aged <18 years)	12–14 (3♀7♂)	3 Asian or British Asian or Middle East origins	6 Duchenne Muscular Dystrophy	11 at risk of being affected or carriers
11 Adult Children (aged 18+ years)	15–17 (7♀7♂)	25 White and from the UK	6 Familial Adenomatous Polyposis	1 known carrier
	18+ (9♀2♂)		6 Haemoglobinopathies 7 Huntington's Disease 4 Neurofibromatosis	12 unaffected siblings

The families represented a wide spectrum of religious beliefs and included: Atheism, Christianity, Church of England, Hindu, Humanist, Methodist, Muslim, None, Pentecostal, Roman Catholic and Sikh. There was also widespread variation of parents in terms of age and educational background. Demographics relating to age, ethnicity etc. for each individual condition have not been given to ensure confidentiality for participant families

This table is only intended to be read as columns, not as rows

condition because they were present in the household at the time of diagnosis, and/or because they witnessed the day to day effects of the condition and treatments. A parent of a child affected by HbO, when asked what information he had given his unaffected child, replied:

> "…he was involved in all this procedure because you know, he was seven and he could understand and he was living in the house so he could see us every day, you know, being in the hospital"

Data from children's interviews however did not support these assumptions. Older siblings could not always remember their brother or sister being diagnosed with the condition. Many confirmed that being with the affected child day-to-day meant they knew about symptoms and treatments, but this did not lead to information about less tangible aspects like disease progression, heredity and their own potential risk. In several interviews parents suddenly realized that siblings may not know as much as they had assumed, or that they had not discussed specific aspects of the condition. They often expressed surprise that they had not realized this sooner. A parent of a child affected by HbO and an older child who is a carrier of the gene said:

> "I think I did a lot of talking in the early days… I don't know what happened there, [laughs], I really don't know what happened there, but when this research came about I realized [older son] doesn't know as much….I need to test it out with him to find out. He doesn't live at home now, but he's living with a girl, and you know, you don't know what can happen, you don't know if she's got the trait. So I've realised I've got to spend some time with him to get him to understand that he has got the trait, you know, and what the risk factors are…."

Failure to give unaffected children information about genetic risk was often a deliberate policy, rather than omission based on assumptions about a child's existing knowledge. A number of parents said they would not raise the issue of a sibling's own risk with them unless they asked about it. Others felt children did not need to know about potential risk until their parents thought they were likely to have sexual relationships or were old enough to make their own reproductive decisions. Parents of a child with DMD, when discussing their daughter's potential carrier risk, said:

> "she doesn't know any of that at the moment, because it's irrelevant when there's no point until she…it's an as need to know basis. And then when she's obviously old enough to be in that position then we'll explain to her to say 'These are your choices' and… that sort of thing."

Similarly parents had often not reassured unaffected children who were not at risk of developing the condition or passing it on to their own future children.

In a family affected by NF, an unaffected young adult sibling explained their confusion at 8 years old when a second sibling was born with NF, and they began to realize that it was an inherited condition:

> "…then obviously when my brother was born as well…it was hard to understand why if my dad's got it, my sister's got it, my little brother's got it, why haven't I got it"

From a parental perspective, in another family affected by NF, there was no recognition of the need to explain the lack of risk to unaffected siblings who were of a similar age to the young adult above when she first realised it was an inherited condition.

> "… I don't think we have ever sat down and said, 'Now these are the following things that you mustn't worry about, you know…your children have no more chance of having Neurofibromatosis than the persons across the road'"

In a very small number of families (CF, NF), parents said they had tried to talk to unaffected siblings about the condition that affected their brother or sister, but they had not wanted to discuss it.

Families with DMD demonstrated different communication processes, because most parents found it extremely difficult to talk to their affected child about the condition, particularly the expected progression and life limitation (Plumridge et al. 2010). They tended to talk to them only about imminent treatments and healthcare appointments. Most parents, however, did discuss the condition more with siblings of the affected child. Similar to the recessive conditions (CF and HbO) they rarely told their daughters about their potential carrier status, but unaffected children were more likely to be told about limited life expectancy and given more information about the likely progression of the condition than the affected child. This meant that siblings were often entrusted to keep potentially devastating secrets from the affected child. They felt this could be difficult to cope with, and most said they did not talk to their brother about the condition at all, in case they inadvertently revealed something.

Understanding of Genetic Risk and Information Given by Parents

In the overall study, patterns emerged in children and young people's understanding at different stages of development (Metcalfe et al. 2011). From as young as 8 years they began to understand the notion of heredity in terms of the condition being "passed down" through the family. However, although they understood the day-to-day impact of the condition by 8 to 11 years, most could not describe heredity patterns or accurately quantify risk until they were 15 to 16 years or

older. In particular, young people had difficulty explaining recessive and X-linked conditions, which is especially relevant for siblings who may potentially be carriers. A young person affected by HbO demonstrates the typical confusion:

> "But if my mum had it and my dad didn't, there would be a 50% chance I would—no 25% chance I would have the full blown, 25% chance I would not have it all. There'll be another 25% chance I would have the trait…"

Adolescent girls in families with DMD, including two 15 year olds, were not aware of their potential carrier status. Similarly, some young adult children in families with recessive conditions (CF, HbO) said they, or other young people in their families had only learned about their own potential carrier status when they were tested, or when they had found out from an external source at around 16 years old. In some cases, this occurred because parents thought children did not need to know about their potential carrier risk until they were likely to have sexual relationships, or were old enough to make their own reproductive decisions, usually at around 16 years. Also, in most families, conversations with children focused, by necessity, on management of the condition, so that heredity was relatively neglected. When young people were tested or they found information from other sources, these events prompted them to ask parents for more information. Indeed, learning about hereditary and potential risk often happened as a result of triggers external to the family, such as school, the Internet, or information leaflets from support groups. One young person, a carrier of the CF gene, told the interviewer:

> "…and I was tested when I was 16 for it. I'm a carrier, so you've got a one in four chance of having a CF child if…when both um parents are CF carriers…"

When the interviewer asked if they could remember when they understood that risk rate, they replied

> "Um, I think that was about 16, I think that was from reading a leaflet."

Even when external triggers prompted parents to talk to their children, conversations tended to concentrate on genetic testing, because they thought that information about implications of being a carrier only became relevant if the result was positive.

Children, including siblings, were often uncertain in gauging the level of genetic risk in relation to themselves, and sometimes misinterpreted their risk (Metcalfe et al. 2011). With dominant conditions young people thought that if a sibling had tested negative they were going to test positive, because a 50% risk meant one out of two children in the family would carry the gene mutation. Some adult children (aged 18 years plus) who understood the risk ratios explained that they still found it difficult to not think in this way. Siblings who were at risk of being carriers did not usually have any idea how many people in the population might be carriers, and thus what the chance might be of them meeting a partner who was also a carrier. Some young people who had been through a process of genetic testing were vague about that process, and not sure about the exact reason for which they had been tested. A young person who had been tested for FAP, and got a negative result said, for example:

> "I think they just, what I remember is that they said this blood test is gonna confirm whether you have… I think polyps, or not, and the results will come through and then when we do, we'll tell your parents and then they'll tell me afterwards…"

Across conditions, affected and unaffected siblings reported rarely discussing the genetic condition or associated risks with each other.

Emotional and Behavioral Outcomes for Siblings

Parents and children reported that siblings were not generally expected to help with care activities. In a very few families with a single parent, some help was needed from siblings, and parents expressed regret about this. Generally, when siblings talked about helping, it was within their normal day-to-day activities (for example helping a brother or sister get into the car or cut up food) rather than additional tasks that eroded their spare time or made them feel resentful. When they talked about helping, siblings were willing and often proud to do this.

The presence of the condition in the family could erode sibling relationships, but it could also enhance closeness. Some children found their affected brother or sister's behavior or disability embarrassing, and many were aware that the condition impacted on their lives by, for example, taking up time or finances that could otherwise be spent on family activities. The way children responded to these effects, however, often related to how well they understood the condition.

Children in families where the genetic condition was not openly discussed, were more likely to show resentment or withdraw from the affected child. They were also more likely to express uncertainty about how the condition affected them, in terms of day-to-day life as well as in terms of potential genetic risk. Children in families where the condition was discussed more openly still expressed uncertainties and concerns, but they also demonstrated understanding for their affected sibling and a better understanding of their role within their family.

A parent of a child with DMD explained how difficult her daughter found the situation initially and how this affected her behavior towards her brother:

> "…my daughter was very angry. She wasn't aware of what was happening and all the attention was going on [affected brother]…her behavior was very irritable she blamed him for everything…"

She felt that her daughter needed to understand what was happening and that talking within the family and support from a counselor eventually led to a change in the daughter's behavior towards her mother and brother:

> "…she helps me now, and helps [affected brother], when I go out shopping, she'll look after him really nicely… its a good family unit now; we all get on really well. So yeah, she does know everything now."

Young people who had a good understanding of the condition and associated risk said they chose not to dwell on the condition but to "get on with life." A young person at 50% risk for HD told us:

> "… at end of the day I can't do anything about my genetic heritage, and to be honest I wouldn't want to! cause it's knowing about it and having it in my family's made me who I am… generally I just think well it's not happening to me now… cause in the end, if I do, then keep packing your life in as much as possible and if I don't, then don't need to change and I'll still keep packing my life in as much as possible [laughs]"

Unaffected children and young people were most upset about the effects of the condition on their siblings or parents, particularly limited life expectation, and they were more likely to worry about these effects than about their own genetic risk. They expressed frustration at their inability to do anything about the condition and its impact on their family member(s). It was only in families with FAP, that unaffected children sometimes described feeling a sense of guilt when their sibling was affected. A young person who had tested negative for FAP explained how he felt when he received this result:

> "Well, relieved that I didn't have it. Er…gutted my brother had it… to me it was a bit of a weird feeling because it, you kind of felt a bit powerless, because there's nothing you could possibly do, it was just something that was there, you've got to deal with it."

With the other conditions, children expressed sorrow that their sibling was affected but relief that they were not. Several well informed siblings, however, said they chose to give extra time and attention to their affected sibling in an attempt to compensate them for everything they missed out on in their lives. A young person with one brother affected by DMD and one unaffected brother explained:

> "I try to give him more attention than my little [unaffected] brother because, me and my little brother always go out, [affected brother] is always at home on his own, so, try to, kind of, when I'm at home, try and stay with him most of the time, play games with him and stuff"

Several children and young people said they had worried about whether they or their children would have the condition but had not discussed this with their parents because they did not want to upset or worry them. This was sometimes the case for children who were at no risk of developing or carrying the condition as well as for some who were at risk. The fact that children who were not at risk worried about their potential to develop or carry a condition highlights an important issue, given that parents did not generally plan to discuss genetic risk with siblings until they asked about it. Additionally, several young people were not certain about their own negative risk status even when they had been told (NF, HD), and commented that they would have appreciated further discussion or reassurance.

The Importance of Family Communication for Siblings

The nature of what siblings were told, and the support they received within their families, were particularly important because they often received little support from elsewhere. Few siblings discussed the condition with extended family members or with their peers, often feeling that they would not understand sufficiently. In families with HbO and some families with HD, all children and young people had equal access to support groups aimed at young people, and in one family with DMD, the siblings received support from a hospice. However, most families thought there was very limited support for siblings from health and social care services. A few siblings had been offered access to support groups when they were teenagers; but young people and parents thought this was too late.

> "They [friends] always ask if [affected brother] is ok, but I don't think I could really talk to them (laugh) about it that well …cause I don't really think they understand"
> (Young person, DMD, sibling untested for carrier status)

> "…and [sibling] would need a lot of psychological support…the siblings do need it. Because I think the person with the disease automatically gets everything in a way…It's the siblings that are left all on their

> own and there's no help for them at all … [local support group] hadn't started a sibling group. When she became a teenager, they did. It was too late, she didn't want to go then." (Parent DMD)

Many parents recognized that siblings were not always given as much attention as their child who was affected by the genetic condition. The parents commented that care demands restricted time they could spend communicating with and supporting the unaffected siblings.

Discussion

Using well established family communication models, this study explored the interaction and communication of families affected by a number of different genetic conditions, with different impacts on family functioning and genetic risk. The results provide a critical insight into the experiences of siblings within families with genetic conditions, indicating the type of information they regard as important and some of the reasons they might not have received this information. The participants were from families who were active in support groups, and thus possibly more willing to discuss the genetic condition. Moreover they had talked to at least some children in their family. It is therefore possible that in other families siblings receive even less information.

The present findings suggest siblings are treated differently in terms of information provision, depending on the treatment needs and life expectancy outcomes of the condition. Across conditions however, both in terms of what they are explicitly told and the information they may implicitly assimilate through being part of the family, siblings are more likely to learn about the condition and its current manifestation and management and less likely to learn about genetic risk. Many siblings are not given sufficient information to comprehend their own risk which has repercussions for their futures. If children are not informed about potential carrier status until they reach an age to be tested or they began sexual relationships, they may already have been worrying about this for a number of years. They may not have raised such concern with parents for fear of upsetting them (James et al. 2003; Metcalfe et al. 2011). They may also have little time to absorb and consider the information before it becomes relevant to them. Their reproductive decision making may therefore be impeded, as observed in studies involving other genetic conditions (Fanos and Puck 2001; Fanos et al. 2001), where young people lacking accurate information based their reproductive decisions on spurious assumptions. Conversely children who are not at risk may not have understood this to be the case, and parents may not have realized that their children have considered and worried about the possibility of being at risk.

Similar to previous research findings (James et al. 2003; Tercyak et al. 2001), the present results suggest that siblings are open to receiving information, and that ongoing discussion within the family is important to make sure siblings understand the implications for themselves. Unaffected siblings sometimes subjugated their own feelings and needs, and many parents had not recognized this when they were so involved in the care of an affected child or partner. Siblings were unlikely to discuss the condition with their brothers and sisters, peers or wider family. So if parents do not encourage siblings to discuss the condition, they could become very isolated in coping with it. Additionally unaffected siblings were much less likely than affected family members to receive support from health and social care services.

Practice Implications

The findings presented provide health professionals with insight to enable them to support parents to ensure they are alert to siblings' specific support and information needs. Improving the support of children and parents in this way is an important part of the overall process of enabling families to cope and adapt more effectively to living with an inherited genetic condition. Parents need to specifically consider siblings in terms of their information needs and inclusion in family discussion. They need to tell them they are open to discussing the condition with them, and continue talking to them as they grow up. Results from the overall study highlight parents' need for support from health professionals in talking to their children about genetic conditions and risk (Metcalfe et al. 2011). Genetic counselors and other health professionals should support parents, and the whole family as a unit, in coping with the condition. This support includes helping parents to realize the importance of including unaffected siblings in this coping process. If gene testing is to be increasingly incorporated into healthcare, health professionals need to be able to advise, facilitate, and support family communication at different stages of children's development and disease progression. Interventions of this sort will help parents explain genetic risk information, and assist their children to cope and make informed decisions. Current policies that discourage gene testing of children for most conditions may inadvertently lead parents to believe their children do not need information about the condition and risk until they are grown up (usually around 16 or 18 years). Health professionals need to be aware of, and address this. When necessary, they should also recognise that parents might need additional support to realize the need to question their own assumptions about their children's knowledge and coping.

Study Limitations and Research Recommendations

This study was not specifically focused on siblings, and recruitment criteria were designed for the whole family. Consequently there was not an equal representation of sibling views across genetic conditions. It is particularly regrettable that due to limitations imposed by the ethics committee, we were often not able to include the views of the youngest member(s) of a family. For the study in general, participants were recruited via support groups, and it was a requirement of recruitment that they had talked to at least some children in the family about the genetic condition. Families that participated may therefore be those who are more willing to engage in conversations about the genetic condition whereas siblings in other families may receive even less information.

Qualitative findings of this sort are not necessarily generalizable to the population of interest, and these specific limitations suggest the benefits of further work. However, the similarities in experiences and opinions of siblings across conditions, suggest it is reasonable to assume that many issues are shared. This assumption is supported by Biesecker and Erby 2008, who suggest that over half of all families experience similar issues related to coping with living with a genetic condition.

Conclusion

The information needs of siblings in families with a genetic condition can be overlooked. Siblings often receive relatively little support outside of the family, and need information to help them understand both affected members' and their own role within their family. Those at risk of carrier status need timely information to help them make informed decisions about their reproductive choices. Genetic counselors and other health professionals need to enhance parents' understanding of siblings' information and support needs at different stages of their development and of the disease progression.

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