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Communicating inherited genetic risk between parent and child: A meta-thematic synthesis

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ABSTRACT

Objectives: Communicating genetic risk is a distressing process for families affected by inherited genetic conditions. This systematic review identifies and explores the challenges faced by parents and their (non)affected or at risk children caused by the (non)disclosure of genetic risk information.
Design: Qualitative meta-synthesis and thematic analysis.
Data sources: Ovid databases; Ovid 'in progress', British Nursing Index, Embase, Medline and Psychinfo were combined with searches of EBSCOhost databases; CINAHL and ERIC and Web of science and ZETOC databases using truncations of communication, chronic illness and disease and words relating to family with specific genetic conditions; Cystic Fibrosis, Duchenne Muscular Dystrophy, Familial Adenomatous Polyposis, Hereditary Non-polyposis Colorectal Cancer, Huntington's Disease, Neurofibromatosis and Sickle Cell Anaemia. This was augmented with free Internet and hand searches and an exploration of the bibliographies of all included papers.
Review method: All papers were quality assessed to ascertain their research quality and methodological rigour.
Results: A total of 2033 citations were retrieved. Following the removal of duplicates, irrelevant articles and the application of an inclusion criterion, 12 articles remained. A further three papers were omitted due to poor quality leaving nine papers which focussed on the disclosure of genetic information between parent and child (<18 years). Eight papers were qualitative in design and one used a mixed method approach. Thematic synthesis produced four themes that inform the structure of the paper; disclosure, emotions involved in disclosure, desired disclosure and recommendations.
Conclusion: Disclosure of genetic risk information within families is a highly complex and affective process often resulting in delayed disclosure. This can lead to increased family tensions generated by misunderstanding, blame and secrecy. Early, age appropriate disclosure can better prepare children for future considerations such as care planning and reproductive decision-making. It also contributes to effective coping strategies that promote enhanced adaptation and emotional well being. Early disclosure also reduces parental anxieties concerning disclosure from an unwitting source. Research shows that children and young people want their parents to engage in open and honest discussions about the genetic condition. Therefore to help facilitate effective family communication health professionals should provide family centred care and better emotional and informational support.

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What is already known about the topic?

- In families affected by inherited genetic conditions, parents find disclosing genetic risk information to their children extremely challenging.
- Health professionals are required to provide greater informational support to families affected by inherited genetic conditions, but what and how this information should be delivered remains elusive.

What this paper adds

- Drawing together empirical evidence from a range of qualitative studies, the paper demonstrates that the emotional consequences of (non)disclosure have profound effects on family members' ability to cope and adapt to an inherited genetic condition.
- The review identifies that children and young people create desired disclosures which detail how and what information they want their parents to disclose. These desired disclosures may conflict with their parent's method(s) of disclosure causing family tensions.
- Health professionals need to develop relationships with families affected by inherited genetic conditions to better manage expectations of the genetic counselling process, support individual families' informational and psychosocial needs and provide bespoke disclosure guidance. This will improve the effectiveness of genetic risk communication between parent and child.

1. Background

Inherited genetic conditions (IGCs) are perceived as rare, only affecting a small proportion of the UK. However, it is estimated that 2–3% of the population are affected by an IGC from birth (Davis et al., 1998), and by the age of 25, 3.3 million people will have developed a genetic condition (GeneticAllianceUK, 2012). Whilst some people affected by IGCs may display symptoms from infancy, others may carry an affected gene (autosomal recessive) and consequently, although they may not exhibit symptoms, their future offspring may be at risk. Families affected by genetic diseases therefore face challenges in living with the condition and having to manage the risk implications for present and future generations (Etchegary and Fowler, 2008; Klitzman et al., 2007; Metcalfe et al., 2011).

An important aspect of managing the genetic condition is the communication of risk information to family members, especially children. Despite studies showing that parents want to disclose information (Gallo et al., 2005, 2009; Metcalfe et al., 2011), the communication of genetic risk information, particularly for potentially debilitating or life limiting conditions, is a highly challenging and distressing process, complicated by potential future care giving obligations (Etchegary and Fowler, 2008) and reproductive choices (Claes et al., 2011; Etchegary and Fowler, 2008). For this reason many parents delay or avoid disclosing information to their (non)-affected or at risk off-spring (Cavanagh et al., 2010; Klitzman et al., 2007) hoping to protect them from the "devastating news" and thus prolonging childhood (Etchegary and Fowler, 2008, p. 719). Delayed disclosure has however been shown to weaken family cohesion creating conflict and family breakdown which may lead to lowered self esteem (Metcalfe et al., 2011), poor emotional well being and an engagement in risky behaviours such as self harm (Metcalfe et al., 2011) and (attempted) suicide (Forrest Keenan et al., 2009) in children and young people as well as poorer psychological functioning in parents (Claes et al., 2011; Tercyak et al., 2000).

Consequently, health professionals recommend that parents communicate genetic risk information to their child(ren) from an early age (Cavanagh et al., 2010). However, parents often feel unsupported in this process, becoming overwhelmed by questions about how, when and what information they should impart (Cavanagh et al., 2010; Etchegary and Fowler, 2008; Klitzman et al., 2007). Other parents report that a dearth of support from health professionals and extended family members has repercussions for their child's emotional well being as they are prevented from providing their child(ren) with the effective emotional support required (Metcalfe et al., 2011; Plumridge et al., 2011).

2. Aims

In recent years the importance of communicating genetic risk information has increasingly been recognised, resulting in a plethora of literature focusing primarily on the communication between parent and child. This meta-thematic review therefore builds on an earlier review by Metcalfe et al. (2008), incorporating the latest research and its' emergent findings to enhance our knowledge and understanding in this burgeoning field.

To answer the following questions the synthesis systematically explores and analyses recent qualitative literature exploring communication themes about genetic risk information and illustrating the impact of disclosure on families. The objectives of this review are therefore:

1. What factors influence how, what and when genetic risk information is disclosed within the family?
2. What are the emotional and psychosocial implications of (non)disclosure on families?
3. What information do children and young people want or need?
4. What recommendations would better support family communication?

3. Method

A systematic review of empirical studies was conducted to examine how parents communicate genetic risk information to their children (<18 years). In analysing the existing literature, thematic synthesis was applied to produce a qualitative meta-synthesis (Arai et al., 2007; Dixon-Woods et al., 2006; Mays et al., 2005).

Building upon and updating a meta-synthesis conducted by Metcalfe et al. (2008), a systematic search of all peer reviewed papers published between 2007 and 2012, associated with family communication and genetic risk

was undertaken between June 2001 and December 2011. The search was conducted using Ovid, EBSCOhost, Web of science and ZETOC databases using truncations of communication, chronic, illness with disease and words relating to family with specific genetic conditions; Cystic Fibrosis (CF), Duchenne Muscular Dystrophy (DMD), Familial Adenomatous Polyposis (FAP), Hereditary Non-Polyposis Colorectal Cancer (HNPCC), Huntington's Disease (HD), Neurofibromatosis (NF) and Sickle Cell Anaemia (Hbo). This was augmented with free Internet and hand searches and an exploration of the bibliographies of all included papers (Fig. 1).

A total of 2033 citations were retrieved. Following the removal of duplicates (919), non-genetic conditions (522) and irrelevant articles (533), 59 potentially relevant papers remained. These papers were reviewed for inclusion using the original inclusion/exclusion criteria assigned by Metcalfe et al. (2008) (Fig. 1). Twelve papers were identified that focussed specifically on the disclosure of genetic information between parent and child. These papers were quality assessed using criteria adapted from a pre-established checklist (Mays and Pope, 2000) and inter-rated by the authors (Fig. 2). Three papers were omitted due to poor quality and lack of methodological rigour (Arribas-Ayllon et al., 2008; Branstetter et al., 2008; Demarco et al., 2008) (Fig. 2). The final nine articles (Cavanagh et al., 2010; Etchegary and Fowler, 2008; Forrest Keenan et al., 2009; Forrest et al., 2008; Klitzman et al., 2007; McConkie-Rosell et al., 2009; Metcalfe et al., 2011; Plumridge et al., 2010, 2011) were included in the review and the papers' methodology, key findings and conclusions were scrutinised using a data extraction form (Hawker et al., 2002; Pearson, 2004; Pearson et al., 2007).

The papers' key findings and conclusions were then analysed using a thematic approach (Fereday and Muir-Cochrane, 2006; Joffe and Yardley, 2003; Thomas and Harden, 2008). They were read and re-read and coded by hand to allow primary level sub-themes to emerge from each paper. These sub-themes were then analysed and compared across all papers to produce secondary level themes. The emergent themes were discussed between the authors and synthesised, to develop a thematic framework from which the final four themes were constructed.

4. Results

Eight papers identified are qualitative in design, with one paper using a mixed methods approach, combining qualitative methods with quantitative survey data (Cavanagh et al., 2010). The papers implement semi-structured interviews with parents and (adult) family members only (Cavanagh et al., 2010; Etchegary and Fowler, 2008; Forrest et al., 2008; Klitzman et al., 2007), with children and young people only (Forrest Keenan et al., 2009; McConkie-Rosell et al., 2009) and with families (parent and child) (Metcalfe et al., 2011; Plumridge et al., 2010, 2011). The majority of the papers focus on a single genetic condition HD (Etchegary and Fowler, 2008; Forrest Keenan et al., 2009; Klitzman et al., 2007), CF (Cavanagh et al., 2010), Fragile X (McConkie-Rosell et al., 2009), DMD (Plumridge et al., 2010). However, 3 papers (Forrest et al., 2008; Metcalfe et al., 2011; Plumridge et al., 2011) compare a range of genetic conditions (Fig. 3). All of the genetic conditions investigated have profound consequences for children and young people's future reproductive decisions, and with the exception of Fragile X, serious implications for the child's present or future health and well being, with several of the conditions having a life-limiting prognosis. Studies of family communication for less serious genetic conditions appear to be absent from the literature and should be considered as a topic for future research.

Thematic analysis produced four themes; disclosure, emotions involved in disclosure, desired disclosure and recommendations which form the foundation for this synthesis.

5. Disclosure

Health professionals, such as genetic counsellors recommend that parents communicate genetic risk to their (non)affected or at risk child(ren) in early childhood (Cavanagh et al., 2010). However, whilst this is advised, parents experience lack of support and guidance from health professionals to aid them with this process (Forrest et al., 2008; Klitzman et al., 2007; Metcalfe et al., 2011) and therefore find disclosure a complex and stress-inducing process. Parents may find themselves caught in a moral dilemma between their children's moral and ethical right to know about their risk and their parental obligation and responsibility to inform their child(ren). This maybe compounded by their anxieties about disclosing such information during childhood (Cavanagh et al., 2010; Etchegary and Fowler, 2008; Klitzman et al., 2007). Parents therefore need to make sense of their own belief systems and assess their child's receptivity to the information before deciding whether to disseminate this knowledge (Forrest Keenan et al., 2009).

5.1. Responsibility

Disclosing parents are often compelled by a sense of obligation (Etchegary and Fowler, 2008) and responsibility (Etchegary and Fowler, 2008; Klitzman et al., 2007) to share genetic information, believing that their children have a right to have access to information affecting their future health (Etchegary and Fowler, 2008; Klitzman et al., 2007). Metcalfe et al. (2011) found that both parents and children believe that "parents should be the main people to provide genetic risk information because they [understand] their children best" (Forrest Keenan et al., 2009; Klitzman et al., 2007; Metcalfe et al., 2011, p. 3). However, research indicates that parental disclosure is a "highly gendered task" (Forrest Keenan et al., 2009, p. 1899), with mothers acting as the main "gatekeepers of genetic information" (Forrest Keenan et al., 2009, p. 1899) and therefore more likely to take responsibility for disclosure (Klitzman et al., 2007; McConkie-Rosell et al., 2009; Plumridge et al., 2010) with fathers playing only a minor role in disclosure (Metcalfe et al., 2011; Plumridge et al., 2010). This however is refuted by Klitzman et al. (2007) who did not find women served as gatekeepers any more than men (Klitzman et al., 2007, p. 1846), with fathers

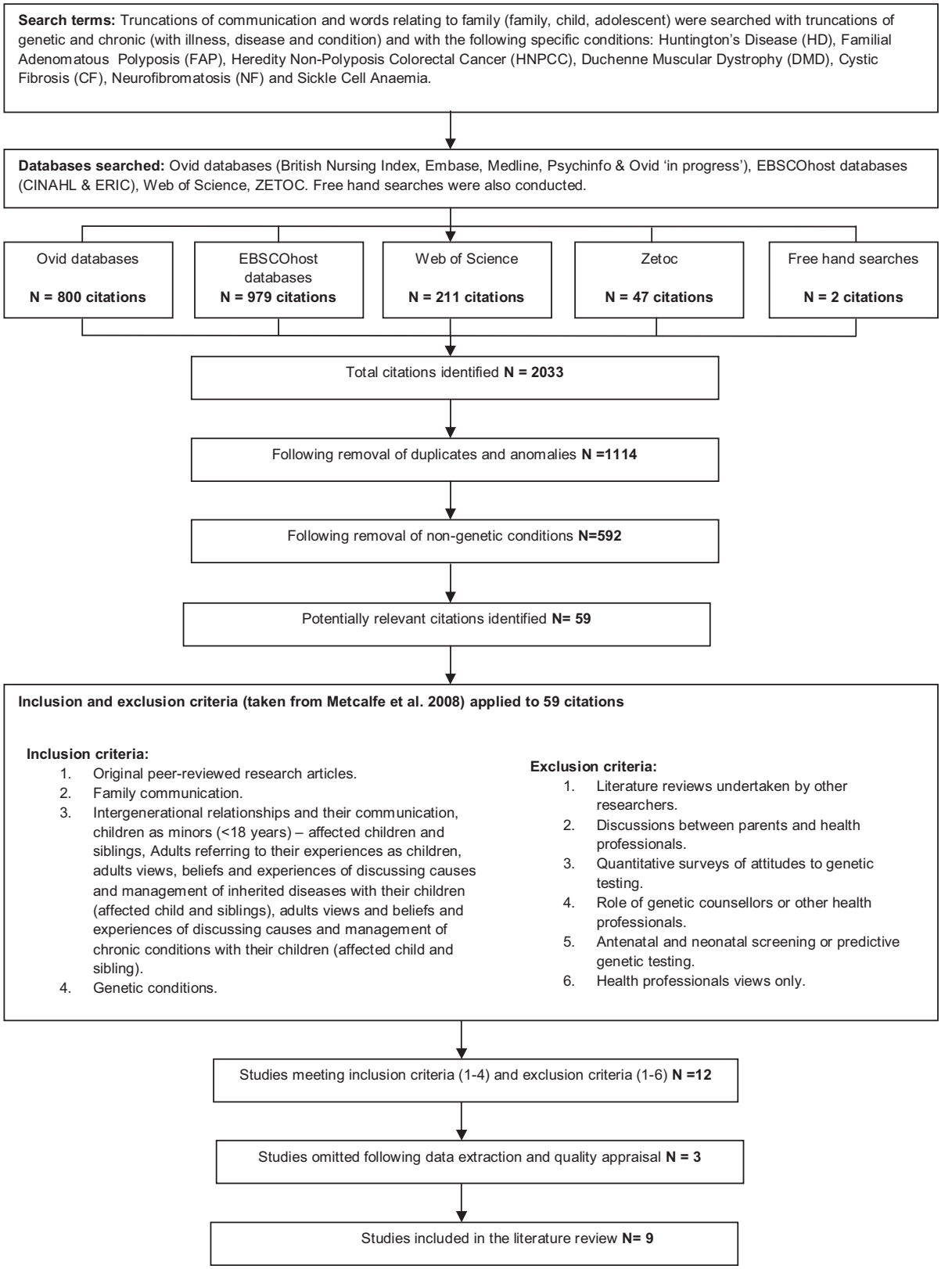

Fig. 1. Flow chart of the literature identification process.

Paper / Criteria for evaluating qualitative research	Arribas-Ayllon et al. (2008)	Branstetter et al. (2008)	Cavanagh et al. (2010)	Demarco et al. (2008)	Etchegary & Fowler (2008)	Forrest et al. (2008)	Forrest-Keenan et al. (2009)	Klitzman et al. (2007)	McConkie-Rosell et al. (2009)	Metcalfe et al. (2011)	Plumridge et al. (2010)	Plumridge et al. (2011)
Clarity of research question?	✗	✓	✓	✗	✓	✓	✓	✗	✓	✓	✓	✓
Appropriate research design?	✗	✗	✓	✗	✓	✓	✓	✓	✓	✓	✓	✓
Is the purpose of research justified?	✗	✗	✓	✗	✓	✓	✓	✓	✓	✓	✓	✓
Is the context of the research adequately described?	✗	✓	✓	✗	✓	✓	✓	✗	✓	✓	✓	✓
Is the sampling frame appropriate?	✗	✗	✗	✗	✓	✓	✓	✗	✓	✓	✓	✓
Are the data collection procedures described?	✗	✓	✓	✗	✓	✓	✓	✓	✓	✓	✓	✓
Is the analysis procedure explicit?	✗	✓	✗	✗	✓	✓	✓	✓	✓	✓	✓	✓
Is the research reflexive?	✗	✗	✓	✗	✓	✗	✓	✓	✓	✓	✓	✓
Are the implications of research clearly defined?	✗	✗	✓	✗	✓	✓	✓	✓	✓	✓	✓	✓
Are the limitations of the research discussed?	✗	✗	✓	✗	✓	✓	✓	✓	✓	✓	✓	✓
Included in the review?	✗	✗	✓	✗	✓	✓	✓	✓	✓	✓	✓	✓

Fig. 2. Quality assessment of literature using criteria for evaluating qualitative research.
Adapted from Mays and Pope (2000).

(Klitzman et al., 2007), grandparents (Forrest Keenan et al., 2009) and other relatives (McConkie-Rosell et al., 2009) sometimes sharing responsibility with the mother.

Whilst parental disclosure is preferred, some children also want health professionals such as genetic counsellors, health care workers and doctors who "[understand] the contemporary issues faced at school, college and the transitions [associated with their] development stage" to supply them with supportive or supplementary information (Klitzman et al., 2007; McConkie-Rosell et al., 2009; Metcalfe et al., 2011, p. 3).

5.2. Why disclose?

Parents believe that early disclosure will allow their children to cope better with the implications of the genetic condition (Forrest et al., 2008) because they will be increasingly better equipped to assimilate the condition into their self identity, thus enhancing their emotional well being. Additionally early disclosure better prepares children for future considerations such as care planning and reproductive choices (Metcalfe et al., 2011; Plumridge et al., 2011) as well as empowering children by giving them greater autonomy over their health care needs (Etchegary and Fowler, 2008) and health behaviours such as diet and exercise (Etchegary and Fowler, 2008; Forrest et al., 2008; Klitzman et al., 2007).

Parents also disclose genetic risk information to their children because they believe that communication will strengthen family relationships (Klitzman et al., 2007; McConkie-Rosell et al., 2009; Plumridge et al., 2010) with some studies reporting greater familial bonding between members of the family affected by IGCs (Forrest Keenan et al., 2009). However, whilst disclosure is reported to improve family cohesion (McConkie-Rosell et al., 2009; Plumridge et al., 2010) other research shows that parents fear that disclosure will weaken family relationships resulting in non-disclosure (Klitzman et al., 2007).

5.3. Non-disclosure

Metcalfe et al. (2011) found that "only a small number of parents expressed a view that their child(ren) had a moral and ethical right to know about a genetic condition affecting their family" (Metcalfe et al., 2011, p. 3). Instead, non-disclosing parents believe that they have a moral right to protect their child from the emotional and psychosocial repercussions generated by the disclosure of genetic risk information such as anxiety and fear. Parents hope that through non-disclosure their children will be able to continue a "normal" childhood (Forrest Keenan et al., 2009, p. 1896; Forrest et al., 2008; McConkie-Rosell et al., 2009; Metcalfe et al., 2011).

As well as protecting child(ren) from harmful information, non-disclosing parents also stated that they choose to avoid or delay disclosure because they are unable to visualise what they might say. The challenge of finding the 'right words' is often intensified by anxieties aroused by their limited knowledge about the condition and its implications to the future health and reproductive choices of their children (Cavanagh et al., 2010). This challenge is further exacerbated by the perceived lack of support and advice from health professionals (Plumridge et al., 2010, 2011), deteriorated information retention since the original diagnosis (Cavanagh et al., 2010) and lack of opportunity to update their knowledge. In response to their perceived dearth of knowledge some parents attempt to establish a "stronger basis from which to explain genetic information" to their family (Forrest et al., 2008, p. 1334) by seeking out information in "scientific articles and accessing any available texts" (Forrest et al., 2008, p. 1333).

5.4. Recipients of genetic risk information

Parents choose whether to disclose information to all the children in the family or to target children according to whether they are affected, not affected or at risk (Metcalfe et al., 2011; Plumridge et al., 2011). This decision is often disease-specific and influenced by the morbidity of the condition (Metcalfe et al., 2011) and/or the child's age and gender (Forrest Keenan et al., 2009).

In families affected by DMD (Plumridge et al., 2010, 2011) and HD (Forrest Keenan et al., 2009) parents report communicating information to non-affected siblings more than the affected child (Plumridge et al., 2010, 2011). In these families a "family view" might exist to which siblings

Paper	Method	Country	Participants	Genetic condition(s)	Aims	Key Findings
Cavanagh et al. (2010)	Telephone interview	Australia	Parents of children identified as CF carriers N=37, female = 32, male = 5. Mean age 43.9, age range 30-55	CF	To determine whether and how parents make use of genetic information about CF received during new born screening.	• Information was disclosed when; children reached a critical age, were mature enough to understand genetic information, before they became sexually active or when they were emotionally ready. • Parents informed children through discussions and diagrams often gaining additional advice from health professionals or the Internet. • Disclosed information included: carrier status, symptoms and screening process. • Some parent's interpreted their child's lack of questions as understanding whilst others believed that questions were caused by a lack of understanding. • Reasons for non-disclosure included: forgotten about it, lacking in knowledge or child's age.
Etchegary & Fowler (2008)	Semi-structured interviews	USA	At risk persons and their family members. N= 24, female = 18, males = 6. Mean age 46, age range 21-73	HD	To understand how patient perceptions of responsibility are experienced, not only in the context of test decisions or reproduction but also future planning and care-giving.	• Genetic responsibility revolved around reproduction, future care-giving and family communication. • Most participants with children fully endorsed their right to know about risk and had communicated a family history of HD to their children. • Families with very young children felt that their children were too young to handle the news, but reported they would talk to them when they were old enough to understand. • There was a need to protect young children from devastating news however at the same time a belief that children should know before critical life junctures. • Children's ability to cope effected motivations for disclosure and non-disclosure.
Forrest et al. (2008)	Semi-structured interviews	Australia	At risk persons and their family members. N= 13, female = 11, male = 2	Adrenoleuko-dystrophy (3), CF (3), Fragile X (1), haemo-chromatosis (1), balanced reciprocal chromosomal translocation (3), Robertsonian chromosomal translocation (1)	To elucidate the process of communicating genetic information in families affected with non-cancer conditions.	• Communication occurred after diagnosis but the focus was on health implications not the families risk status. • Family communication took place over time and is an integral part of family coping mechanisms. • Patients used the Internet to supplement their understanding of their genetic condition. This helped them to disseminate information to wider family members. • Better support is needed during diagnosis and communication to at-risk family members. • Follow up consultations would allow families to have questions about their genetic condition answered and receive up-to-date information and advice on how to communicate information to their children.
Forrest-Keenan et al. (2009)	Semi-structured interviews	Canada	Young people with a family history of HD. N= 33, female = 21, male = 12. Mean age 20.5, age range 9-28	HD	To detail the experiences of young people who live in a family affected by HD and their experiences of disclosure.	• Majority of participants were told early in childhood by a female relative. • Some were told gradually, after realising something was wrong with a family member or becoming more aware of the illness after becoming the main carer. • Sometimes information was kept a secret until the parents felt that the child could understand. • Some children were in complete ignorance about their illness until late 20's. This often resulted in strained relationships with their parents and increased anxiety. Many children had to keep information a secret from their younger siblings. • For others HD was a new diagnosis that caused fear as parents blurted out information in a shocking manner.
Klitzman et al. (2007)	Semi-structured interviews	UK	Parents with risk of HD. N= 21, female = 9, male = 12	HD	To critically examine what, when and to whom to disclose genetic risk information	• Parents struggled with what to tell children as they didn't want to cause distress. • Disclosure often occurred over time. • Children were often given partial and incomplete information generating misunderstanding but it was believed that children needed age appropriate information. • Parents justified non-disclosure by stating they wanted their children to have a childhood. • Disclosures surrounding HD may prove more difficult than other disorders. • Professional training is needed to make health care workers more aware of the difficulties of communicating risk in families.
McConkie-Rosell et al. (2009)	Semi-structured interviews	USA	Young adolescent girls and young women in families with a previous diagnosis of Fragile X. N= 53, female = 53, male = 0. Mean age 17, age range 14-25	Fragile X	To explore how genetic risk information was learned, what information was given and why it was provided to adolescent girls and young women in families with Fragile X syndrome	• Young girls were informed of genetic risk by a relative, normally the mother. • Disclosure styles included; open communication, information seeking and indirect i.e. overhearing. • Information disclosed included: family diagnostics, genetic status of family members, carrier status and reproductive implications. • Information provided should be age appropriate and given in stages. • Families need support and education about the genetic condition as they are the ones primarily informing children of their genetic risk. • Genetic counsellors can provide an environment in which parents can practice what they might say to their children.
Metcalfe et al. (2011)	Semi-structured interviews	UK	33 Families (parents and children) affected or at risk from genetic conditions. N=85, parents = 52, female = 34, male = 18. Children = 33, female = 15, male = 18	NF (4), HD (7), HbO (6), FAP (6), DMD (6), CF (4)	To find out what information children require at different developmental stages and how to provide information.	• Children thought parents should be the main people to provide information and health professionals should support parents to do this. • Mother's were usually the key person to disclose genetic risk to the child. • Siblings rarely discussed genetic condition with affected child. • Factors compromising ability to disclose genetic risk included; shock, increased emotional and physical care giving, experience of grief and the need to protect children. • Affected children were given more information than sibling except for DMD. • There is a greater need for family centred care to support parents in advising and helping them to manage care and maintain family relationships.
Plumridge et al. (2010)	Semi-structured interviews	UK	Families at genetic risk of DMD. N= 19, parents = 11, children and young people = 8	DMD	To provide insight for parents about sharing genetic risk information about DMD within their families and to raise awareness of the advice and support required from health professionals.	• Mothers talked about double shock of the child's condition and their carrier status. • Caring for the affected DMD child was emotionally and physically challenging. • Parents thought that it was their role to talk to their children about their condition. • Affected children were told less than siblings with female siblings not told about their carrier status until 16 yrs. • Parents need to be more proactive in giving children and young people, particularly the affected child information about DMD and health professionals need to support them with this.
Plumridge et al. (2011)	Semi-structured interviews	UK	33 Families (parents and children) affected or at risk from genetic condition. N=96, parents = 52, female = 34, male = 18. Children (<18 years)= 33, female = 15, male = 18, children (>18 years), female = 15, male = 17	NF (4), HD (7), HbO (6), FAP (6), DMD (6), CF (4)	To explore communication processes between parents and their children about genetic risk information. The paper focuses on the experiences of siblings and their roles in family communication.	• Siblings are treated differently in terms of information provision, depending on the treatment needs and life expectancy outcomes of the condition. • Many siblings are not given sufficient information to comprehend their own risk which has future repercussions. • Children misinterpreted their risk because they did not understand probability. • Affected and non-affected siblings rarely discussed the condition and risk with each other. • A genetic condition can erode or strengthen family relationships. • Families with open communication saw more acceptance and understanding of the condition leading to better sibling and family relationships. • Health professionals need to support parents to ensure that they are alert to siblings' specific support and informational needs

Fig. 3. Key findings of included literature.

are expected to abide by (Plumridge et al., 2010, p. 1230). This view may contain facts about the genetic condition as well as the parent's feelings, beliefs and views of what the affected child should know about the condition (Plumridge et al., 2010, p. 1230). Forrest Keenan et al. (2009) demonstrate that this controlled disclosure style can lead to tensions between non-affected and affected siblings, particularly when a non-affected sibling becomes burdened by the secret (Forrest Keenan et al., 2009; Plumridge et al., 2010). This causes siblings to feel guilty believing that their affected sibling "should know more about their condition" (Plumridge et al., 2010, p. 1230).

In contrast Metcalfe et al. (2011) discovered that in families affected by CF, FAP, HbO and NF the "affected child [is] normally given more information than their siblings" (Metcalfe et al., 2011, p. 5; Plumridge et al., 2011). In such families the non-affected child(ren), who are unaware of what is happening, often harbour resentment towards the affected child as they perceive that they are receiving preferential treatment (Metcalfe et al., 2011; Plumridge et al., 2011). However, on receiving an explanation of the affected child's condition the unaffected child's resentment often diminishes (Metcalfe et al., 2011; Plumridge et al., 2011).

Decisions to disclose or conceal genetic risk information from certain family members therefore have significant impacts on family cohesion or the commitment to the long term care of the parents, sibling or affected child (Metcalfe et al., 2011; Plumridge et al., 2011) which can lead to tensions that may result in family breakdown or divorce (Etchegary and Fowler, 2008; Klitzman et al., 2007). However, this is refuted by Forrest Keenan et al. (2009) who showed that in some families whose parents were able to contain their own anxieties about the genetic condition "decisions to withhold information did not have a detrimental impact on family relationships either before or [after disclosure]" (Forrest Keenan et al., 2009, p. 1897).

5.5. When to disclose genetic risk information

Parents are concerned that they may harm their child(ren) if they disclose information too early, or if their child(ren) are not emotionally or cognitively ready to understand the information (Etchegary and Fowler, 2008; Klitzman et al., 2007). However, conversely they do not want to harm their children by communicating risk too late (Klitzman et al., 2007). Parents therefore deliberate over the most appropriate time to disclose information. Research shows that parents prefer to disclose information during childhood (Forrest Keenan et al., 2009; McConkie-Rosell et al., 2009; Plumridge et al., 2010) but that the timing of disclosure is subjective, centred around parental beliefs and the "child's age, developmental stage and maturity" (Klitzman et al., 2007, p. 1843).

Whilst not providing a defined disclosure time, Klitzman et al. state that decisions to disclose are "shaped by two sets of time frames: lifecycle ... and medical course" (Klitzman et al., 2007, p. 1842). Related to lifecycle, parents are more inclined to disclose genetic risk information at "critical life junctures" (Etchegary and Fowler, 2008, p. 719) such as birth (Cavanagh et al., 2010; Metcalfe et al., 2011), first sexual experience (Cavanagh et al., 2010; Plumridge et al., 2011), engagement (Klitzman et al., 2007), marriage (Etchegary and Fowler, 2008; Klitzman et al., 2007; Metcalfe et al., 2011) or reproduction (Etchegary and Fowler, 2008; Klitzman et al., 2007; Metcalfe et al., 2011). Parents are also reported to disclose genetic information following prompts in their child's education and curriculum, for example when "genetics or reproductive systems" are being taught (Cavanagh et al., 2010, p. 205), when the topic naturally comes up in conversation (Cavanagh et al., 2010) or when their child begins to ask questions (Forrest Keenan et al., 2009; Metcalfe et al., 2011). Research shows that parents are ambivalent in response to their children's questions. Whilst some parents "interpreted the absence of questions as a sign that the child clearly understood, ... others equated asking questions with comprehension" (Cavanagh et al., 2010, p. 206). It is however, important for parents to answer their children's questions as accurately as possible to enhance and satisfy children and young people's desire for information.

Associated with medical course, parents are prompted into disclosing information; "rapidly after diagnosis" because they are concerned about their child's health (Forrest et al., 2008, p. 1333), when an older sibling is considering genetic testing (Cavanagh et al., 2010), a close family member is displaying symptoms, or a family crisis (Metcalfe et al., 2011). This often leads to parents "blurting out information in a shocking manner" (Forrest Keenan et al., 2009, p. 1987).

In contrast to the subjective nature of choosing when to disclose information, Cavanagh et al. (2010) and Forrest Keenan et al. (2009) present a "critical age" (Cavanagh et al., 2010; Forrest Keenan et al., 2009, p. 1898) to which parents are more likely to disclose information. This critical age is typically between the ages of 9–10 (Cavanagh et al., 2010; Forrest Keenan et al., 2009; Metcalfe et al., 2011), with Cavanagh et al. reporting a mean age of 9.2 years (Cavanagh et al., 2010, p. 205). Parents in these studies believe that by this age, children are emotionally competent and "mature enough to understand the genetic information" (Cavanagh et al., 2010, p. 205) but innocent enough not to have become sexually active (Cavanagh et al., 2010).

5.6. How to disclose information

Metcalfe et al. (2011), Forrest Keenan et al. (2009) and Forrest et al. (2008) regard "disclosure [as] a process, rather than a single one off event" (Forrest et al., 2008, p. 1333). Open styles of communication (Forrest Keenan et al., 2009; McConkie-Rosell et al., 2009; Metcalfe et al., 2011) allow disclosure to become a process as they encourage parents to "prepare the ground" (Klitzman et al., 2007, p. 1840) and gradually inform their children of their risk over longer periods of time (Forrest Keenan et al., 2009; Forrest et al., 2008; McConkie-Rosell et al., 2009). This style of communication also permits parents to provide their children with developmentally appropriate information (Klitzman et al., 2007) and for children to ask questions enabling them to fully understand the

consequences of the information at their own pace, helping them to "come to terms with the risk" (Forrest Keenan et al., 2009; McConkie-Rosell et al., 2009; Metcalfe et al., 2011, p. 6) and facilitate a "process of on-going realization and understanding" (Plumridge et al., 2011, p. 377).

Learning about a condition gradually (Forrest Keenan et al., 2009; Forrest et al., 2008; Klitzman et al., 2007) causes some children to believe that they have always known about their condition (Forrest Keenan et al., 2009; Forrest et al., 2008). These children rarely recollect the moment when they were told that they are affected or at risk from a genetic condition (Cavanagh et al., 2010; Forrest Keenan et al., 2009; McConkie-Rosell et al., 2009). For these children, the condition becomes embedded into their personal and family history through a genetic narrative that had been incorporated into a story about their birth (Metcalfe et al., 2011).

Conversely, other children are kept in the dark about their condition. These children often have a sense that something is wrong and that their parents and/or siblings are keeping a secret from them (Forrest Keenan et al., 2009; Forrest et al., 2008; Klitzman et al., 2007). In these circumstances children might seek information themselves becoming "active agents in their own learning" (Forrest Keenan et al., 2009, p. 1898), retrieving information from a variety of sources, for example; TV programmes (Forrest Keenan et al., 2009), Internet (McConkie-Rosell et al., 2009; Plumridge et al., 2010, 2011), informational leaflets (Plumridge et al., 2011), over hearing conversations (McConkie-Rosell et al., 2009) or asking their parents direct questions (Forrest Keenan et al., 2009). In seeking out information children prompt their parents to communicate genetic risk information. For some parents their participation in research also prompted disclosure (Metcalfe et al., 2011).

5.7. What information should be disclosed?

The literature indicates that parents disclose broad categories of information, for example their child's potential risk (McConkie-Rosell et al., 2009) or carrier status (Cavanagh et al., 2010; Klitzman et al., 2007; McConkie-Rosell et al., 2009), parent's genetic counselling experience (Klitzman et al., 2007), the new born screening or test process (Cavanagh et al., 2010; Klitzman et al., 2007), symptoms (Cavanagh et al., 2010; Klitzman et al., 2007), the impact to future children (McConkie-Rosell et al., 2009; Metcalfe et al., 2011) or other family members affected by the condition (McConkie-Rosell et al., 2009). However, information within these categories is "selectively communicated" (Klitzman et al., 2007) with the conditions minutiae influenced by their child's developmental stage and/or age (Metcalfe et al., 2011), perceived maturity (Cavanagh et al., 2010; Klitzman et al., 2007), gender (Metcalfe et al., 2011; Plumridge et al., 2010) and the genetic condition's morbidity (Metcalfe et al., 2011; Plumridge et al., 2010). Klitzman et al. (2007) state that parents often engage in partial disclosure to prevent other family members from worrying.

To demonstrate, Plumridge et al. (2010) showed that children affected by DMD "were usually given less information than their siblings". Whilst affected children were told that they had poorly legs or muscles, limited information was provided about the disease trajectory. Reasons cited for selective communication include: the forging of strong emotional and empathetic relationships between mothers and their affected son(s). This prevented mothers from finding the words to communicate the limited life expectancy caused by the progressive nature of the condition. Learning difficulties associated with the condition also caused mothers to believe that their sons' could not cope with the information. However, it was not only affected sons that were party to partial disclosure. Plumridge et al. (2010) also demonstrate that sisters of the affected child, who were at risk of carrying the x-linked gene, were also subject to selective communication, with their carrier status or hereditary risk not being disclosed until they were 16 years old (Plumridge et al., 2010, p. 1229). Similarly, Cavanagh et al. (2010) found that in the majority of children affected by Fragile X, parents discussed the child's carrier status, however they did not necessarily discuss the risk to future generations.

In addition to selective communication Metcalfe et al. (2011) indicate that knowledge gaps between children's age, gender and the inherited genetic condition are not only linked to the condition's morbidity but to the child's maturity. McConkie-Rosell et al. (2009) and Metcalfe et al. (2011) illustrate the complexity of this issue by demonstrating that children and young people's insight into their genetic condition enhances as they develop cognitively and their life priorities begin to change. For example, children as young as 10–11 affected by conditions may understand general information about the condition, such as how the condition is passed through the family. By 12–15 years, children gain a more complex understanding of their own risk and by 15–17 years, young people may be more concerned about the risk that the genetic condition poses to their future children and the implications of the condition to their reproductive choices.

6. Emotions involved in disclosure

Families experience a wealth of emotions associated with the communication of genetic risk information to their (non)affected and at risk children. For many families emotions exist prior to disclosure and continue through to the post-disclosure phase. Prior to disclosure parents may feel overwhelmed by anxiety (Forrest Keenan et al., 2009) and stress (Cavanagh et al., 2010; Etchegary and Fowler, 2008; Metcalfe et al., 2011) about how they might tell their children. This is often compounded by feelings of guilt (Etchegary and Fowler, 2008; Klitzman et al., 2007; Plumridge et al., 2010) for passing on the genetic condition and fear about how family members might react to the information (Metcalfe et al., 2011; Plumridge et al., 2010). Avoiding disclosure also increases anxiety in parents because they often "[live] in constant dread of their children asking questions and [fear] that other people such as teachers or health professionals [will] tell them things, they as parents [do] not yet want them to know" (Plumridge et al., 2010, p. 1230).

While concealing a family history of a genetic condition is stressful for parents (Etchegary and Fowler, 2008, p. 720), children and young people often show ambivalence towards the emotional consequences of their parents "disclosure burden" (Forrest Keenan et al., 2009, p. 1896). Some children accept their parents delayed or disclosure avoidance tactics, believing their parents had their best interests at heart and therefore "accept the reasons why they had not been told earlier" (Forrest Keenan et al., 2009, p. 1897). Despite sensing "parental procrastination, half truths and selected information", these children concede parental authority and do not "seek out external information from other sources" (Forrest Keenan et al., 2009, p. 1896). They therefore continue to "conform to their parents' wishes and remain in the dark about their condition" (Forrest Keenan et al., 2009, p. 1896). However, keeping children in the dark "generates misunderstanding" and confusion (Klitzman et al., 2007, p. 1841), causing children to feel "scared about the risk of inheriting something they [do] not fully understand" (Metcalfe et al., 2011, p. 6). Therefore children use their imaginations to postulate what is wrong with them. Children's speculative imaginations however, are often far worse than the reality, causing unnecessary stress, worry and self blame for the child (Metcalfe et al., 2011). Fear also leads to lowered self esteem contributing to the child being bullied (Metcalfe et al., 2011), suicidal thoughts (Forrest Keenan et al., 2009) or engaging in risky behaviours such as self harm (Metcalfe et al., 2011).

Conversely children who do not accept delayed disclosure tactics believe that their parents made the wrong choice in keeping the information from them. These children suffer from a variety of mixed emotions such as; worry, fear, relief, anger, frustration, anxiety, anger, hurt and disappointment (Forrest Keenan et al., 2009) and as a result struggle to cope with the newly disclosed information and often exhibit rebellious behaviour (Klitzman et al., 2007). Some children with late disclosure occurring in their mid teens and early adulthood are also reported to suffer from depression, insomnia and suicidal thoughts (Forrest Keenan et al., 2009). These children and young people have been reported to desire earlier communication.

7. Desired-disclosure

Children and young people want their parents to disclose genetic risk information from an early age (Metcalfe et al., 2011; Plumridge et al., 2011). They therefore believe that their parents should be knowledgeable about the condition affecting their family (McConkie-Rosell et al., 2009). This desire for well-informed parents assimilates with parents who engaged in educative processes to further their knowledge to better explain the condition to their children (Forrest et al., 2008). Children also require their parents to be erudite because they want to engage in open and honest discussions where they can freely ask questions and have their parents answer them informatively and accurately. Openness provides opportunities for children to use their parents as role models for their own coping with the genetic condition (Metcalfe et al., 2011).

Asking questions is particularly important to children because they want to be "informed and knowledgeable" about the condition affecting their family so that they can better cope with the emotional and psychosocial aspects of the condition (Metcalfe et al., 2011; Plumridge et al., 2011). To further enhance their knowledge children also state that they want the opportunity to meet with health or social care professionals to discuss the genetic condition and gain more insight into their risk (Metcalfe et al., 2011, p. 6).

Corresponding with parental desires for disclosure, the majority of children and young people also believe that it is important that genetic risk information is delivered gradually over time with the content of the information reflecting their developmental stage (McConkie-Rosell et al., 2009; Metcalfe et al., 2008; Plumridge et al., 2011). However, the young people in McConkie-Rosell et al.'s study stated that they did not want their parents to "leave anything out" and that they wanted them to be straightforward and honest (McConkie-Rosell et al., 2009, p. 11). This desire sits uncomfortably against the notion of gradual disclosure and parental desires to implement partial or selective disclosure to protect their child from harmful information. It is however, important to note that this desired-disclosure narrative emerges from research with teenage girls who were carriers of Fragile X syndrome and therefore may not represent the views of all young people but is possibly specific to their age, gender, risk status and the genetic condition.

Finally children wanted risk information to be communicated in a "positive light" (McConkie-Rosell et al., 2009). For many children positive disclosure involves the normalisation of information rather than disclosing genetic risk as "life shattering" (McConkie-Rosell et al., 2009). Cavanagh et al. demonstrate how parents with children affected by CF attempt to normalise the condition by "explaining that everyone possesses disease causing genes or that one of the parents is also a carrier" (Cavanagh et al., 2010, p. 206). Normalisation has been demonstrated to reduce anxiety and a sense of isolation (McConkie-Rosell et al., 2009).

8. Recommendations and conclusions

Whilst health professionals advocate early disclosure, guidance about when, to whom and the content of the information to be communicated remains elusive with disclosure being influenced by the child's status as a (non)affected or at risk child, their age, gender, perceived maturity, and the genetic conditions morbidity (Metcalfe et al., 2011; Plumridge et al., 2011). Disclosure of genetic risk information, therefore remains a highly complex, challenging and daunting process which can result in delayed or non-disclosure. Such disclosure methods may generate anxiety, guilt, blame, secrecy and misunderstanding in both parent(s) and their child(ren) leading to increased family tensions.

In disclosing families, the responsibility to disclose predominately falls to the mother although responsibility

is sometimes shared with fathers, grandparents and occasionally health professionals. Disclosing families typically adopt open styles of communication which allow them to communicate risk information gradually over time. This style of communication is preferred by children and young people who desired disclosure from well informed and knowledgeable parents and preferred gradual disclosure that accommodated their developmental stage and allows them to ask questions.

Age appropriate disclosure has been shown to increase children and young people's understanding of their condition contributing to more effective coping strategies, enhanced adaption and better emotional well being. Early disclosure also better prepares children for future considerations such as reproductive decision-making and care planning and reduces parental anxieties concerning disclosure from an unwitting source.

Following these conclusions the synthesis poses three recommendations for the enhancement of effective and efficient disclosure of genetic risk information for both families and health professionals. First, it is imperative that genetic risk information is disclosed through an open communication style, as this will facilitate more open and honest discussions between parent and child (Metcalfe et al., 2011; Plumridge et al., 2011). Children will therefore have the opportunity to ask their parents questions and have them answered honestly (McConkie-Rosell et al., 2009), without feeling intimidated or worried that they might "upset their parents" (Metcalfe et al., 2011, p. 4; Plumridge et al., 2011). However, concurrently it is notable that whilst children do not intentionally want to upset their parents, it is perhaps unavoidable as the disclosure of genetic risk information is equally distressing for disclosing parents as it is for the children and young people receiving the information. Within the parameters of open communication it is therefore crucial that parents and children address and manage their emotions to better understand each other's feelings and adapt to the information exchanged.

Second, disclosure should not be a single transaction but a continual process where information is disclosed incrementally to reflect and support the child's development, maturity, cognitive and emotional ability as well as the genetic condition's symptoms and future implications to the child's health and reproductive choices. It is therefore important that information is tailored to the child's temporal needs.

Finally, to assist the disclosure process, it is necessary for health professionals to provide greater family centred care creating greater emotional and informational support for parents and children (Metcalfe et al., 2011; Plumridge et al., 2011). To provide better family care three interrelated recommendations are presented by the literature; enhanced professional training, the provision of effective genetic counselling sessions and the development of resources.

Professional training is required to enable health professionals such as genetic counsellors and social workers to understand the difficulty that families face in communicating genetic risk (Forrest Keenan et al., 2009; Klitzman et al., 2007). Being aware of these challenges will allow health professionals to understand that families vary in their ability to communicate and consequently they need to be more responsive to the informational and psychosocial support requirements of individual families.

Within genetic counselling sessions genetic counsellors need to explore parental communication patterns (McConkie-Rosell et al., 2009) by asking the parents probing questions about their disclosure, for example; whether disclosure has occurred (Klitzman et al., 2007), what transpired (Klitzman et al., 2007), what the child might have heard/understood (McConkie-Rosell et al., 2009), their intended future disclosure (Klitzman et al., 2007) and any perceived barriers to communication (Klitzman et al., 2007). These questions will allow genetic counsellors to focus on the family's communication style and the effectiveness of the disclosure so that they can further develop disclosure plans and provide practical advice and guidance to make communication more effective and efficient. It is also important that the counselling sessions provide an environment in which parents can practice what they might say to their children, building confidence and enabling parents to normalise the condition, which will reduce anxiety and fear in their children.

Finally, resources should be developed to provide parents with techniques, diagrams and appropriate language to transfer information more successfully. Resources should support families psychosocial needs providing them with the coping mechanisms to affirm that they can "gain [affective] control over their lives" (McConkie-Rosell et al., 2009, p. 324; Plumridge et al., 2010) and address emotions such as guilt (Plumridge et al., 2010). Parental or family support groups could also be implemented to reduce isolation and encourage peer support (Metcalfe et al., 2011).

9. Limitations

Whilst family communication of genetic risk information is a burgeoning field the systematic review retrieved a paucity of high quality research papers with only 9 papers being accepted as methodologically rigorous since 2007 following quality rating criteria adapted from Mays and Pope (2000). Whilst methodologically rigorous the studies had two principle limitations. First, due to the rarity of the inherited genetic diseases under study, many of the papers have small sample sizes, the smallest consisting of 13 participants (Forrest et al., 2008) and the largest population 96 (Plumridge et al., 2011) which has repercussions for the generalisation of the results. However, the papers were included because their findings and qualitative evidence based experiences contributed greatly to the knowledge and understanding of this field.

Second, only five papers (Etchegary and Fowler, 2008; Forrest Keenan et al., 2009; Metcalfe et al., 2011; Plumridge et al., 2010, 2011) analysed family communication of genetic risk information from an inclusive family perspective. With two papers solely focussing on parental perspectives (Cavanagh et al., 2010; Klitzman et al., 2007) and a further two from the perspective of

children and young people (Forrest Keenan et al., 2009; McConkie-Rosell et al., 2009). Therefore their ability to capture the nuances and complexity of familial communication is limited. Despite the limitations, the systematic review builds and integrates the latest burgeoning findings following Metcalfe et al's (2008) review. It thus develops and strengthens the evidence base by focusing on communication between parents and their child(ren) (<18 years), which has often been neglected in favour of more prevalent research which distillates on communication between adult children (>18 years) and broader familial communication patterns.

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