Reproductive decision-making in young female carriers of a BRCA mutation

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**STUDY QUESTION:** How do young women, who were identified as carrying a BRCA gene mutation before they had children, approach reproductive decision-making and what are their attitudes towards reproductive genetic testing?

**SUMMARY ANSWER:** Reproductive decision-making within the context of cancer risk is complex and influenced by personal experiences of cancer. Younger women were not concerned with reproductive decision-making at the time of their genetic test; however, the impact on subsequent reproductive decision-making was considerable and left them with unanticipated dilemmas, such as having children who would be at risk of inheriting cancer predisposition, timing risk-reducing surgery and changing perceptions of responsibility.

**WHAT IS KNOWN ALREADY:** Individuals carrying gene mutations predisposing to hereditary breast/ovarian cancer have concerns about passing on the gene mutation to children.

**STUDY DESIGN, SIZE, DURATION:** Qualitative methodology and thematic analysis.

**PARTICIPANTS/MATERIALS, SETTING, METHODS:** Data were collected through semi-structured interviews with 25 women aged 18–45 who had received a positive result for a BRCA1 or BRCA2 gene mutation while childless.

**MAIN RESULTS AND THE ROLE OF CHANCE:** Analysis revealed four central themes: (i) the impact of cancer on reproductive decision-making; (ii) motivation for genetic testing; (iii) risk management and timing of planning children; and (iv) optimism for future medical advancements.

**LIMITATIONS, REASONS FOR CAUTION:** This study explores the views of female BRCA carriers. Further research should explore the views of couples, men, and include samples with greater ethnic and social diversity.

**WIDER IMPLICATIONS OF THE FINDINGS:** This evidence highlights the need for reproductive decision-making to be addressed at the time of pretest genetic counselling. More information should be provided on reproductive options as well as counselling/support to guide women’s reproductive decision-making and prenatal testing options at the time they undertake genetic testing.

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**Key words:** preimplantation genetic diagnosis / BRCA / reproductive decisions / cancer / ethics
Introduction

Mutations in BRCA1 and BRCA2 breast/ovarian cancer genes place female carriers at a lifetime risk of up to 85–90% of developing breast cancer and 27–65% of developing ovarian cancer (Antoniou et al., 2003; Evans et al., 2008). Cancer development is not inevitably fatal in BRCA carriers, although overall survival is poorer in BRCA1 carriers (Evans and Howell, 2004). Primary cancer prevention strategies (bilateral or contralateral mastectomy and bilateral salpingooophorectomy) provide a significant reduction in risk but are challenging options (Schover, 2008; Rebbeck et al., 2009).

Individuals carrying gene mutations predisposing to hereditary breast/ovarian cancer (HBOC; Staton et al., 2008; Fortuny et al., 2009; Quinn et al., 2009), or familial adenomatous polyposis (Kastri nos et al., 2007) primarily have concerns about passing on the gene mutation to children. BRCA1/2 mutations predisposing to HBOC are inherited in an autosomal dominant pattern, such that there is a 50% chance of passing on the mutation to each child. Women who test positive for a BRCA mutation may be less likely to want additional children than those who test negative (Smith et al., 2004). For individuals at risk of carrying the genetic alteration for Huntington’s disease (HD), a fully penetrant genetic disorder, family planning is often one of the main motivations for seeking genetic testing (Decruyenaere et al., 2007). Reproductive decision-making raises concerns for people at risk of HD relating both to potential children’s risk (individuals rationalize decisions about prenatal genetic testing and having children in order to act in a way they consider is ‘responsible’) and to their own risk (parenting may be compromised through illness and/or premature death; Downing, 2005; Decruyenaere et al., 2007).

Reproductive genetic testing options for individuals carrying a BRCA gene mutation include prenatal diagnosis (PND) with the intent to terminate a carrier pregnancy (TOP; Lancaster et al., 1996) and, more recently, preimplantation genetic diagnosis (PGD) (HFEA, 2006). PND/TOP is physically and emotionally challenging (Sandelowski and Barroso, 2005) and may need to be repeated several times in order to achieve a non-gene mutation carrier birth. Having used reproductive genetic testing for one pregnancy, carrier couples may feel compelled to use it again for future pregnancies (Adam et al., 1993; Downing, 2005; Decruyenaere et al., 2007).

In the UK access to PGD is currently regulated by the Human Fertilisation and Embryology Authority (HFEA) according to whether a particular condition satisfies requirements set out in the Human Fertilisation and Embryology Act (HFEA, 1990). In 2006, the HFEA extended licensing of PGD to include genetic susceptibility to incompletely penetrant, adult onset hereditary cancer, including breast and ovarian cancer (BRCA1/BRCA2) gene mutations (HFEA, 2006). PGD involves ovarian stimulation, egg retrieval and in vitro creation of one or more embryos. From these embryos 1 or 2 cells are taken at the 8-cell cleavage stage of development and tested for the familial mutation (Geraedts and DeWert, 2009). Questionnaire research has found that female BRCA mutation carriers of all ages regard PGD as acceptable for BRCA; however, relatively few (14%) women who had yet to complete their families believed they personally would use PGD for a future pregnancy. Where further information was available, participants cited a good quality of life, their value to society and availability of breast cancer management and treatment options as influencing their perspective of PGD (Menon et al., 2007).

There remains a lack of clarity around the issues impacting reproductive decision-making in BRCA carriers and the current study sets out to explore this specifically. The following research questions are explored using a qualitative methodology: whether and in what ways BRCA status has influenced reproductive decision-making, whether participants have concerns about having children at risk of HBOC, and if so what prenatal testing options they are considering/would consider to avoid transmission of risk. Our study focused on individuals who tested positive for a BRCA gene mutation before having children. Data on specific attitudes towards reproductive genetic testing and the impact on genetic services in this cohort are published elsewhere (Ormondroyd et al., 2012); the current paper reports on reproductive decision-making.

The study was approved by the Royal Marsden NHS Foundation Trust Ethics Committee (07 H0801 106 RMH).

Materials and Methods

Participants

Women and men aged 18–45 who tested positive in the preceding 5 years for a BRCA1 or BRCA2 mutation and, at the time of genetic testing did not have children, were eligible. Participants had undergone diagnostic or presymptomatic BRCA testing in the preceding 5 years at one of two UK hospitals (Royal Marsden London/Sutton and St Mary’s Hospital Manchester), and were recruited between November 2007 and March 2010. Eight participants have had children since receiving their BRCA test result. Participants may have had a personal history of cancer, but needed to be more than 2 years from diagnosis. Those with a personal cancer history were not known to be subfertile or have premature ovarian failure. Those eligible were considered by their clinical team to have no serious mental health contraindications, and were invited at least 6 months or more after their BRCA test result. Fifty-nine eligible people were invited to participate; 55 women and 4 men. Twenty-seven women expressed an interest in participation and 25 women agreed to be interviewed. All eligible males declined. Those who did not wish to participate were not asked for an explanation of their decision.

Data collection and analysis

Interviews lasted between 30 and 120 min and were guided by an interview schedule devised from a literature review. Topics included: how participants found out about their inherited cancer predisposition, why they opted for genetic testing and the effects that genetic test information has had on their life decisions, including planning children, relationships, work and risk management (see Supplementary information). Participants’ own concerns that were not covered by the interview schedule were also explored.

Data were collected using semi-structured interviews at participants’ homes or at one of the hospitals. Participant information sheets carried a short paragraph explaining PND and PGD.

Each interview was audio recorded and transcribed verbatim. An inductive thematic analysis of the transcripts was conducted (Braun and Clarke, 2006). The analysis was open-ended, exploratory and data driven; allowing for themes to be developed that were not predefined questions. This approach focuses on participants’ subjective retelling of their experiences and allows them to share issues that are important to them. Themes were coded inductively in order to build theory that is based on an understanding of how known cancer risk influences reproductive decision-making. In order to ensure reliability in the analysis, members of the team (E.O., L.S.D., C.M. and M.W.) independently
coded a subset of transcripts and met to develop and refine the thematic categories. The remaining interviews were systematically analysed by E.O. and L.D. Emerging themes were compared across the complete body of data. No new themes emerged during the final interviews indicating that data saturation had been achieved.

Results

Demographic details for the 25 participants show (Table I) that 24% of the women had a breast cancer diagnosis more than 2 years or earlier, none had had ovarian cancer. Subsequent to their positive BRCA test, 15 (60%) had undergone risk-reducing bilateral mastectomy and two individuals had undergone risk-reducing bilateral salpingo-oophorectomy, the latter decision being effectively also a reproductive decision. None of the participants had children before they were tested for a BRCA alteration; although eight participants had children since confirmation of their BRCA carrier status (BRCA testing of children is not indicated). One woman had sought PND and one other had terminated a pregnancy because of a personal breast cancer diagnosis.

Analysis revealed four central themes: (i) the impact of cancer on reproductive decision-making; (ii) motivation for genetic test; (iii) risk management and timing of planning children; and (iv) optimism for future medical advancements.

Table I Participant demographics.

<table>
<thead>
<tr>
<th>Demographic</th>
<th>RMH participants (%)</th>
<th>Manchester participants (%)</th>
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<td><strong>Age</strong></td>
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<td></td>
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<td>0</td>
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<tr>
<td>26–30</td>
<td>4 (27)</td>
<td>4 (40)</td>
<td>8</td>
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<tr>
<td>31–35</td>
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<td>36–40</td>
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<tr>
<td>41–45</td>
<td>3 (20)</td>
<td>2 (20)</td>
<td>5</td>
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<tr>
<td><strong>Total</strong></td>
<td>15</td>
<td>10</td>
<td>25</td>
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<tr>
<td><strong>Sex</strong></td>
<td></td>
<td></td>
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<tr>
<td>Female</td>
<td>15 (100)</td>
<td>10 (100)</td>
<td>25</td>
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<tr>
<td><strong>Surgery</strong></td>
<td></td>
<td></td>
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<tr>
<td>Mastectomy</td>
<td>8 (53)</td>
<td>7 (70)</td>
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</tr>
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<td>2</td>
</tr>
<tr>
<td>None</td>
<td>7 (46)</td>
<td>1 (10)</td>
<td>8</td>
</tr>
<tr>
<td><strong>Cancer diagnosis</strong></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Number of children since BRCA test</td>
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<td></td>
<td></td>
</tr>
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<td>0</td>
<td>10 (66)</td>
<td>7 (70)</td>
<td>17</td>
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<tr>
<td>1</td>
<td>4 (27)</td>
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<tr>
<td>2</td>
<td>1 (6)</td>
<td>1 (10)</td>
<td>2</td>
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<tr>
<td><strong>Ethnic background</strong></td>
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<td>White British</td>
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<td>10 (100)</td>
<td>20</td>
</tr>
<tr>
<td>White other</td>
<td>5 (33.3)</td>
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</tr>
</tbody>
</table>

The impact of cancer on reproductive decision-making

For this sample, women’s decisions about having children are deeply embedded in personal and familial experiences of cancer and the reactions of family members to cancer diagnosis and BRCA mutation status. The extent to which HBOC is seen as a ‘manageable’ condition is also influenced by familial experience. The women perceive that older family members experience guilt for transmitting cancer predisposition to their children. For example, participant R03 remains undecided as to whether to have a family; potentially passing on the BRCA alteration is increasingly influential:

> it’s a really difficult decision, but again the more the ovarian cancer risk plays on my mind the more I start thinking ‘I just don’t want to pass this gene on anyway’. You know that’s becoming more and more of a factor in my personal reason for not wanting a family. (R03)

Participants who have had children balance the negative aspects of inherited cancer predisposition against the positive reasons to have a child, based on their own experiences and the value that they place on their own lives and those of their children. This is illustrated in the following quote:

> I accept the fact that she [her daughter] might have the gene as well, and at some point in our future I’ll have to explain to her and make it her decision whether she wants to find out or not. And I hope she does but erm (...) I always think of it (...) as if my mum had had the choice to have all these tests and things and if she decided not to have me. You know, I’ve had a perfectly lovely life up till now and I’ve got the gene so I don’t think that should stop me from having children. (R24)

Despite witnessing the adverse impact of her mother’s treatment, participant R16 carefully weighed the prospect of developing cancer against not having children:

> I just remember her being like she was sick and she was having treatment and she’s come out of it and survived and hmm (long pause) and I just don’t I just don’t sort of see it as being something that would be a reason not to bring a child into the world. (R16)

In contrast participant R20 was close to an aunt who underwent treatment that was ultimately unsuccessful, and is more circumspect about cancer risk in the context of a potential child:

> all the experiences with my auntie changed my life, my approach to life, then having the gene, it does kind of give you a real sense of responsibility… would I have a child then have to know that my child is watching me die and know that that child might have the gene. (R20)

Some of the women, in discussing their thoughts about having children in the knowledge of their cancer risk, mentioned the fact that their own risk is not 100% certain (due to incomplete penetrance of the BRCA mutation), citing this as a reason for not denying themselves and their partners a family. In addition, HBOC is seen as a ‘manageable’ condition by some who have adjusted to their cancer risk and feel confident in their ability to parent despite it; R07 does not yet have children but intends to, believing that her children can learn from her experience by considering risk-reducing surgery should they also carry the BRCA alteration.

> well I guess I think I’ve had a (...) happy life and I hmm can pass on an equally happy life guaranteed up until now I’ll do the best I can hopefully
won’t get run over by a car by the time you reach my age hmm and I’m thinking they’ll be fine until they’re in their mid-thirties and then they’ll have to make the same decisions I make. (R07)

Our data suggest that the degree to which cancer is considered to have impacted on the women’s lives, influences their difficulty with making reproductive decisions. If women perceive HBOC as relatively manageable, decisions about childbearing are less problematic compared with those where the familial cancer has been more aggressive and unmanageable.

**Motivation for genetic test**

For women in this cohort unaffected by a cancer diagnosis, reducing their risk of developing cancer was their primary motivation for undergoing genetic testing. Although some women referred to the incomplete penetrance of *BRCA* gene mutations in the context of having children, others perceived cancer not only as inevitable but also fatal if they did not undergo risk-reducing surgery. As M13 says:

you know people get treated I’ve just never seen it ‘cause I’ve never not seen it but with my personal kind of experience of cancer was people die and that’s my assumption. (M13)

The perceived inevitability of cancer and risk of occurrence at a relatively young age influenced the timing of *BRCA* genetic testing, with a positive result motivating risk management surgery.

whereas if I’d waited and had it done in my 40’s there was a lot higher chance that I would have got cancer at some point. (R14)

**Risk management and timing of planning children**

In addition to the impact of risk management strategies on the timing of having children, some of the women with cancer had been prescribed a 5-year course of Tamoxifen, during which time they were advised against conception.

For others, who were advised to undergo risk-reducing oophorectomy, there was a pressure to have children as soon as possible so that they could complete their childbearing and have preventive surgery at a relatively young age given the possibility of early onset cancer. This sense of urgency was particularly evident for women not yet ready to have children. M30 clearly expressed this tension between planning for children and the recommendation for oophorectomy:

so then so they’re on about taking your ovaries when you’re 35 (.) and you wanna have kids in between it’s like ‘OH MY GOD’. (M30)

This increased temporal pressure to have children is considered by some of the participants to set this group of women apart from their peers:

I wanna have children by the time I’m 35 so that if I have to think about having an oophorectomy you know these things but I have to have children out of the way I can’t delay it longer than other people might. (R07)

Risk management plans and reproductive decisions apparently complicated some relationships—for example, one participant wanted to have children and to talk about her concerns about potential children’s risk and reproductive options with her partner; he already had children from a previous relationship, and was reluctant to engage in discussions. Imperatives to undergo risk-reducing surgery compounded decision-making with regard to the timing of having children that any woman/couple face, such as finding a suitable partner:

my one worry is that like how will I (?!) to have children in [since her relationship break down] … I just I wanted to have them all before obviously having it done … and now that worries me ‘cause I think ‘god I’m 28 years old’ … I’ve always wanted 3 cause I think if she [her daughter] was to go through what we’ve been through without brothers or sisters it would be horrible for her. (R14)

The pressure of time and surgery is linked not only to salpingooophorectomy but also elective mastectomy. For participant R12 the wish to breast feed was a strong factor influencing her decision to have children prior to risk-reducing mastectomy, and imposed a pressure to breastfeed successfully:

I decided to have children before [mastectomy] because I wanted to breast feed … it’s always at the back of your mind slightly I suppose, that anxiety that you’re going to get cancer before you get a chance to have a mastectomy. I was quite uptight about it, he [son] didn’t sort of take, well it was quite hard work in the first few weeks, I found it even more stressful ’cause I knew I’ve got to do this. (R12)

Anxiety about developing cancer, and a feeling of responsibility to do everything possible to prevent it, may be intensified by having children, as R13 discussing her plans for elective mastectomy says:

having had the daughter I feel very emotional you know life becomes much more fragile and much more precious … I recently began to think, this is nuts I could deal with this problem, I’d never forgive myself if I got breast cancer and died of it. (R13)

**Optimism for future medical advancements**

Eighteen of the 25 participants expressed a strong belief in the future development of medical science and its ability to alleviate cancer. Participants reflected on experiences, either personal or of close family members’ cancer treatment. They discussed at length the difficulties in the decision to have a child who may potentially inherit a *BRCA* mutation, but believe that risk is tempered by current and possible future medical advancements, as participant M25 explains:

cause like I say 30 percent of me, before I found out about the gene I probably would have said no I-I don’t think I could put a child through it but you know (.) but then the other 70 percent I think, well medicine’s come on a hell of a lot a hell of a lot since my mum passed away from it you know, and er, you know what it can offer you now. (M25)

Several participants expressed the hope and expectation that, for their children, medicine and science will have developed new treatments and preventive techniques that are more successful than previous generations have experienced:

and if she ever got to be 20 years old then 20 years down the line with cancer research it may be a completely different matter (I: hmm hmn) and if the difference between what I went through, or my mum went through just, you know, not that long before. (R08)

This optimism is often based on relatively recent developments; experiences of older family members for whom less could be offered in terms of treatment and risk management, are contrasted
Discussion

The results of this study highlight the complexity of reproductive decision-making in this group of women. Personal and family history of cancer had a significant impact on reproductive decisions, contributing to perceptions of risk, controllability and severity.

The primary motivation for seeking genetic testing was to inform risk management decisions. Younger women were not concerned with reproductive decision-making at the time of their genetic test; however, the impact on subsequent reproductive decision-making was considerable and left them with unanticipated dilemmas, such as how to rationalize having children at risk, and how to balance risk-reducing surgery with family planning. Findings highlight the need for reproductive decision-making to be addressed early at pretest genetic counselling.

Many of the women, although not all, felt ambivalent about having children in the context of their own, and potential children’s cancer risk. It was notable that the women in this study, who have all under-children in the context of their own, and potential children’s cancer genetic counselling.

for reproductive decision-making to be addressed early at pretest risk-reducing surgery with family planning. Findings highlight the need as how to rationalize having children at risk, and how to balance risk management decisions. Younger women were not concerned with reproductive decision-making at the time of their genetic test, impacting to perceptions of risk, controllability and severity.

The data show that the desire for risk-reducing surgery impacts on family planning. Participants perceived a pressure to have children at a younger age, so that appropriately timed risk-reducing measures can be taken in accordance with clinical recommendations. The desire to act responsibly as a (potential) parent reinforced this imperative to maximize longevity.

When discussing their children’s futures, particularly female children whose cancer risk if they inherit the BRCA mutation is high, participants felt that discoveries in cancer research will mean that the risk associated with a BRCA alteration will be reduced when their own children reach adulthood. This belief apparently mediates reproductive decision-making in this group of individuals.

In this paper, we demonstrate how the decision to have children is impacted by the emotional burden of being a BRCA carrier and the possibility of passing the genetic mutation onto children, perceptions of responsibility of a parent who is also a BRCA carrier, and perceptions of personal risk reduction, as well as belief in the ability of research to reduce cancer morbidity and mortality. Our data strengthen the notion that decision-making is not independent from social values and cultural context (Zeleny, 1982). Indeed, Hastie and Bawes (2010) argue that decision-making is dependent on the individual’s psychological and social assets. Factors such as emotional capacity when making decisions, status of their relationships, the possible outcomes of the decisions and whether they are rationally grounded all have an impact on subsequent reproductive decision-making. Findings highlight the need for reproductive decision-making to be addressed early at pretest genetic counselling.

with currently available options. As R08 says, she has witnessed major developments in breast cancer treatment between her and her mother’s cancers, and her expectation is that this will continue in the future. This view is shared by participant R13:

and hopefully by the time my daughter is my age, the way research is going, it would be an even you know much better position than now. (R13)
effect. This highlights a need for clinicians to take account of these personal factors when providing information and guidance about genetic testing and reproductive decision-making in this group of women.

Limitations and future directions

The study does not provide any information on male BRCA mutation carriers or on those from varying ethnic backgrounds. The Manchester participants are slightly younger, more likely to have undergone surgery and are all White British, and it will be valuable to investigate quantitatively whether and how responses to specific questions, derived from the present study, vary between specific groups. Future work could focus on the experiences of couples undergoing reproductive genetic testing for BRCA, and should also explore how individuals and couples from varying social classes understand, interpret and act on information about their reproductive options, and how professionals impart that information.

For women with BRCA gene mutations, reproductive decision-making is highly complex. Hershberger and Pierce (2010) suggest that couples require reproductive decisional support to help them understand developments in medical science, to appreciate the various implications of reproductive genetic testing, and to explore with them the impact that differing reproductive options can have on their view of their own cancer risk, the risks to their children and the moral impact of family planning that involves reproductive technologies (Offredy et al., 2008).

Conclusions

The study highlights the prioritization of health benefits as a hierarchical process. It builds on the theory of decision-making in that it highlights the importance of the social context including family experience in the genetics arena. It is important that both social and psychological aspects of people’s lives are developed when exploring people’s beliefs and needs.

Our data highlight the need for young women in HBOC families to be aware of the possible impact that a positive BRCA result may have on their future reproductive decisions. Many such women wish to incorporate genetic test information into risk management strategies, but may require additional support when they later consider having children. In some instances, these reproductive decisions appear to be strongly influenced by family experience which suggests that decisional support from genetic clinicians needs to include a discussion of the family background and circumstances leading up to consultations about access to and use of PGD or PND/TOP. In this respect, clinicians need to be aware that individuals are not wholly autonomous in decision-making (Marteau and Dormandy, 2001).

Supplementary data

Supplementary data are available at http://humrep.oxfordjournals.org/.

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Authors’ roles

L.S.D. interviewed the Manchester cohort, transcribed the audio files and jointly analysed all the data. E.O. was responsible for the conception of the study, contributed significantly to the grant application, interviewed the majority of the RMH cohort and jointly analysed all the data. D.G.R.E., R.E. and S.L. are the responsible clinicians and contributed to the grant application. E.B. assisted with recruiting the RMH cohort and compiling the demographic information. C.M. contributed significantly to the grant application, coded a subset of the transcripts and assisted in developing the analysis. M.W. contributed significantly to the grant application, provided overall project management as study PI, coded a subset of the transcripts and assisted with developing the analysis. All authors contributed to writing the manuscript.

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Conflict of interest

None declared.

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cancer quoted should reflect the cancer burden in the family. *BMC Cancer* 2008;8:155.


