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# Talking to Children about Maternal *BRCA1/2* Genetic Test Results: A Qualitative Study of Parental Perceptions and Advice

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## Abstract

Family communication is the primary, initial means of educating the next, at-risk generation about hereditary cancer risk. In this study, in-depth parent narratives provided self-report of motivations, planning, satisfactions and regrets associated with sharing or not sharing maternal *BRCA1/2* test results with young children and advice for parents considering disclosure and for genetic counselors. Interviews were conducted with 32 mothers tested for *BRCA1/2* with children ages 8–21 years and 24 of their co-parents; interview narratives were analyzed qualitatively. Parents were concerned with both protecting and educating children about hereditary cancer risk. They expressed confidence that parents can constructively convey genetic information to minor children. Telling relieved most parents and satisfied a sense of parental duty. Parents strongly advised child-specific, age-appropriate tailoring of genetic information and emphasized conveying the positive, preventive utility of genetic information to children. Immunizing effects of disclosure were viewed as providing forewarning about and preparation for possible later family cancer diagnoses. Parents choosing not to tell children were advised to consider future disclosure.

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Narratives about parental sharing of *BRCA1/2* test results with minor children support the feasibility of parental discussion of maternal genetic test results to the next at-risk generation. Results suggest development of intervention tools for parents would support decision-making and family communication and potentially reduce parental worry and regret. Recommendations are made for more active involvement by genetic counselors with tested parents around the topic of delivery of genetic information to children.

**Keywords**

*BRCA1/2*; genetic testing; family communication; hereditary breast/ovarian cancer; prevention

## INTRODUCTION

In genetic medicine, unlike traditional medicine, findings about one individual have direct health implications for their offspring. A woman's positive genetic test for the cancer predisposition genes, *BRCA1* and *BRCA2*, implies not only her own high risk of both breast and ovarian cancer but also informs her of her offspring's 50% chance of having inherited these cancer risks (Ford et al..1994; Easton et al. 1995; Ford et al., 1998; Struewing et al. 1997). In this area of personalized medicine, parent-child communication is the primary way risk information is conveyed to the next generation of at-risk family members. This information is heavily relied upon to ultimately guide offspring to genetic cancer risk assessments where counselors or physicians can educate them about genetic counseling, testing and screening. The way in which the information is communicated and received may emotionally color much of the way the child feels about hereditary cancer risk in the years to follow and may also affect the tone of future parent-child communication about this topic. Hence, the communication of a mother's *BRCA1/2* test result to her child carries special importance, as there are implications for the future health of the mother and child (even when results are negative or uninformative).

Between 5 and 10% of all cases of breast and ovarian cancer are thought to be due to mutations in *BRCA1* or *BRCA2* (Offit, 1998). Once confined to families with striking histories, *BRCA1/2* testing has become more common among individuals with moderate and even weak histories of breast and ovarian cancer. Since the inception of genetic testing for hereditary breast/ovarian cancer, over 800,000 individuals have been tested for mutations in the *BRCA1/2* genes (Myriad Genetics, personal communication, July 2012) and the annual number of tested individuals is rising. A recent study of community oncology practices found that 41% of patients had sufficient risk factors to merit testing of deleterious *BRCA1/2* mutations (Eisenbraun et al. 2010).

Research on family communication of *BRCA1/2* genetic test results has mostly focused on communication between adult relatives (MacKenzie et al. 2009). Studies are emerging about how parents make decisions about the sharing of *BRCA1/2* genetic test results to children, though we do not yet have a complete understanding of how children come to know, understand, and act on such information during childhood, adolescence and adulthood. Research suggests that many women tested for *BRCA1/2* shared positive and negative results with their young children, often within a few weeks or months after learning this information themselves (Tercyak et al. 2001, Tercyak et al. 2002; Tercyak et al. 2009; Bradbury et al. 2009; Segal et al. 2004); Patenaude et al. 2006; Bradbury et al. 2007). In a recent study, among 224 tested mothers, 63% disclosed results to their children under age 21 within a month of receipt of test results, with disclosure to children reaching 68% by 6 months post-disclosure (Tercyak et al. 2009). While older children are told more often than younger children, Patenaude and colleagues (2006) found 37% of tested mothers shared

results within 4 months with children 6–13 years of age, and 85% of 14–17 year old children. Age and maturity appear to be more potent determinants of disclosure to minors than gender (De Marco et al. 2009).

Parents report considerable preoccupation and worry about their children's hereditary cancer risks, including how to tell their children about their or their spouse's or partner's *BRCA1/2* test result (Miesfeldt et al. 2003; Peshkin et al. 2010; Tercyak et al. 2001; Tercyak et al. 2002; Tercyak et al. 2007; Clarke et al. 2008; MacDonald et al., 2010). Paradoxically, parents seldom seek advice from health care professionals regarding decisions about disclosing results to their children (Bradbury et al. 2007). Professionals have few resources to draw upon to guide parents who are deliberating about how to discuss test results with their minor children. There is an absence of first-hand reporting of parental experience of disclosure to minor children. Guidelines concerning parental notification of children about genetic test results have not been established. Data are also lacking on the utility of evidence-based tools to support parental decisions and actions. These gaps may, to date, have discouraged provider communication with tested parents on this important topic.

When specifically queried, parents report a need for enhanced information about how to talk to children beyond what is offered during routine cancer genetic counseling (Tercyak et al. 2007; Sharff et al. 2011). In a 2007 study in which mothers tested for *BRCA1/2* mutations were asked what resources they would like to have to support their decision making about speaking with minor children about test results, nearly 80% mentioned wishing they could speak to "previous participants", i.e. parents who had already confronted this decision (Tercyak et al,. 2007). Peer support is typically an important source of normative information and valued encouragement, especially in situations where the circumstances are rare, isolating, and/or sensitive. The present study uniquely responds to this need and aims to fill a critical gap in the literature by reporting on parents' experience and advice for other parents regarding disclosure of maternal results to young children.

## METHODS

### Participants

This qualitative study was conducted with 32 mothers and 24 co-parents (all but one were male) of children ranging in age from 8 to 21, recruited as part of a larger, prospective, mixed-methods investigation of decisions and outcomes of parent-child communication about maternal *BRCA1/2* test results. The larger study involved 213 mothers and 104 co-parents. Specifically, the study focused on learning about parents' attitudes, behavioral intentions, and beliefs regarding communicating maternal *BRCA1/2* test results with children, as well as actual communication decisions and related psychosocial predictors and outcomes. The methodology of the larger study is detailed elsewhere (Sharff et al. 2011). Informed consent was obtained for all participants, and the protocol was approved by each cancer center's Institutional Review Board.

For the qualitative interviews, this subgroup consisted of mothers and co-parents where the mother had been tested for *BRCA1/2* and received results; mothers and co-parents were invited to detail their experiences surrounding telling or not telling their child and the impact of the decision they made. Quota systems were established to invite equal numbers of male and female target children (5 each), children under and over age 13 (a typical age cutoff for assuming that children may be able to understand informed consent documents as well as adults) (5 each), similar numbers of true negative and uninformative mothers (4 each) and a slightly higher number of positive mothers (5 each) and equal numbers of children whose mothers had and had not had cancer (5 each). The demographics of interviewed participants are reported in Table 1.

### The Interview

The qualitative, semi-structured interviews were conducted by telephone by a clinical health psychologist with experience interviewing parents and training from project leaders in the conduct of this interview schedule. The interviewer was aware of the maternal test result and the ages of the family's children, but was not otherwise involved in the development of the interview guide or the study recruitment procedures nor did she have prior contact with participants. One-time interviews were conducted approximately six weeks after post-test genetic counseling and were audio-recorded, assigned identification numbers and transcribed. Coding utilized ATLAS-ti (Muhr, 1991) qualitative data analysis software. The code book was developed by the project investigators. Two graduate-level research assistants double-coded 3 sets of interviews, repeatedly discussing discrepant codings with the lead investigator (AFP) until mutually acceptable definitions were achieved. A final comparison of codings from 10% of the interviews yielded 86% agreement. Narrative analysis followed Weiss' (1994) techniques of local integration and inclusive integration. Local integration refers to the organization and integration of observations into those which are the most common and those which are variants or exceptions. Inclusive integration is the development of a coherent, overarching framework for organizing the important findings from analysis of the narratives into a meaningful report, allowing for the drawing of relevant conclusions and recommendations for future work.

Through responses to the interview questions and prompts, parents 1) described decision making about disclosure and, if they opted for disclosure, reported their disclosure experiences, 2) discussed reactions they observed in their children following disclosure, 3) told of the impact on themselves individually and as a couple and 4) offered advice to other parents considering disclosure. The analysis of the interviews identified factors in parental satisfactions and regret and in what disclosing and non-disclosing parents thought were important issues for parents to consider.

Narrative data presented below delineates parent reports and advice. Quotes are identified by speaker, ("M") for mothers and ("CP") for co-parents and code number ("#") and denoted by maternal *BRCA* test results as being either carriers ("BRCA+") or noncarriers ("True Negative", i.e. negative for a known familial mutation) or uninformative ("UI"), i.e. negative results in the absence of a known familial mutation, negative for the Ashkenazi Jewish panel of 3 common mutations, or variants of unknown significance.

## RESULTS

From the larger study population of eligible participants, 84% of mothers and 74% of co-parents indicated they would be willing to complete a qualitative interview if invited to do so. Of these, 33 mothers and 24 co-parents were selected using the previously described quotas; 97% (32) of selected mothers and 100% of the 24 selected co-parents completed a qualitative interview: their characteristics were similar to those of the larger study sample. Interviews were typically 30 minutes in length.

Disclosure to the target child was reported as 54% among families with a mutation-positive mother, 50% among true negative mothers, and 60% among mothers who had received an uninformative result. In one family (True Negative result), parent and co-parent were discrepant in their reports, with the mother reporting disclosure to the 11-year old daughter and the co-parent reporting non-disclosure. In the Uninformative result group, one co-parent said he didn't know if his wife had told their 12 year old daughter and in a second family in that group, parents discrepantly reported, the mother saying the child had been told and the co-parent reporting the child had not.

### Telling the Child: Timing and Triggers

Parents reported spending considerable time thinking and talking together about when and how to communicate the mother's *BRCA* test result to their at-risk children. Many parenting couples discussed at some length the pros and cons of telling their young children. For other couples, there was little discussion as the conclusion seemed pre-ordained to both members of the couple, either that the child was too young to be told or that the children would all be told.

The telling was most often triggered by the mother's receipt of her own test result. Many children were told the same day as the results were received, still more within the week or month that followed. It was relatively rare for a parent who wanted to tell their child the mother's result to wait longer than a month to disclose.

**Spontaneous disclosure—**Despite prior plans for joint telling of their children, disclosure of the maternal test result sometimes occurred differently than parents planned. This was most frequently because one parent seized an apropos moment and spoke unilaterally to their children rather than having a more formal meeting with both parents present. At least four parents told their children while driving in a car, a time when the parent had control of the child's attention in a place where they often shared conversations. Spontaneous telling by one parent was viewed by the other as unsettling, but not of long-term consequence. Flexibility and trust in the co-parent's judgment was the predominant experience reported. Spontaneity was viewed favorably as taking advantage of a good time for discussion rather than as a deviation from a formerly-planned script.

**Impact of prophylactic surgery on telling minors—**Another common trigger for discussion of genetic test results was the mother's pending, risk-reducing surgery, either a prophylactic mastectomy or oophorectomy. The mother's absence from the home had to be explained and disclosure answered the question, at least partially, about why the mother was undergoing surgery at this time.

> *The kids know if you have to have the surgery, they're going to know something and you have to tell them something.*(M#20, True Negative, 9 year old daughter) (Mother talking hypothetically about need for surgery had she been positive).

**Not telling is a Decision too—**Parents who felt strongly that their children did not need to be told, at least not at present, offered a variety of reasons for their decision.

> *I mean when I had cancer, I had to tell my son [not the target child] about that. He was only 5 then. But, that was really happening. But this isn't something she can see or that is going to do anything. I really don't see a reason for her to know at all right now.* (M#24, *BRCA*+, 8 year old daughter)

However, subjects in our study thought parents who had decided not to tell their children the maternal result now should talk with their genetic counselor and consider all the options about disclosure.

> *Get them to be thinking, you know, at a conscience level about whether or not they intend to talk to them, to be free with them… and why they may have decided one way or the other. And get them to think about the advantages and disadvantages of deciding one way or the other. Well, you know, you have some people who may have decided that there's no way they're going to talk to their kids about this or anybody else for whatever reason, and they may not even-it may be just, you know, it may be just the gut level thing and, umm, they really haven't thought through it. And they haven't, may never even occurred to them to think through it…Of course*

*they'd be free to-to take it from there or just leave it.* (CP#22, BRCA+, 8 year old daughter)

Age was not the only consideration. One mother who had previously had cancer said she was leaving information in her will about the *BRCA1/2* mutation, in case she died before she thought it appropriate to tell her (older) children.

*I felt at this age, she wasn't mature enough to take in this information. They* [the children] *would be just too emotional. They could know later rather than now. I will tell them at an appropriate time, certainly by 33, as long as I'm still around to tell them. So I have put* [it] *in my-the envelope- with my will. I put all the information I got from the hospital and a note, my reasoning, why I have not told them prior.* (M#7, *BRCA*+, 19 year old daughter)

Parents who opt to delay disclosure still seem to find value in planning for telling at a future time. Having some idea of either the age a child might be told or of the likely triggers to future telling (child asks, child has taken course and learned some basic genetics concepts, etc.) seems to give parents some closure on discussion about disclosure to children.

### Impact on Parents: Parental Reactions to Telling Maternal Result to Children

Those who shared the news with their children mainly reported experiencing relief in the disclosure. Parents disclosing negative or inconclusive results were most "happy" about the telling.

*I told them that I didn't have the gene, and that my chances of getting ovarian cancer was the same as any other person that didn't have the gene. I had like a two percent chance of getting ovarian cancer, some time in my life. But that I wasn't going to get it as early as Aunt S. did or had to worry about it ever again as far as my family history… And that they didn't have to worry because if I didn't have the gene, then they didn't have it. And that we're just really lucky.* (M#18, UI, 8 year old female)

Parents with positive test results nonetheless reported a sense of accomplishment in telling their children. It was not only telling the child the facts about the testing, but also helping the child to put the information into a positive life frame which parents felt good about. Parents seemed to see this as an extension of their general responsibility to guide children about which occurrences were life events which could be usefully dealt with, hoping the child would embrace the positive stance they themselves took in their reaction to the information.

*I felt very positive about it. I felt that I had accomplished something that would very possibly protect my child in the future so it was a positive experience.* (M#23, *BRCA*+, 20 year old daughter)

*I sort of give that whatever you got, you got. So what are you going to do with it? You live with it. You flow. You enjoy it. You only got one life to live..so if that's where you are, that's where you are and there is no room for sulk. I don't have that in me.* (CP#14, *BRCA*+, 10 year old son)

Many parents mentioned feeling unburdened, i.e. "It is not a secret anymore". Secrecy about the test result had worried parents who thought their child might learn about the result inadvertently by overhearing them in conversations with other adult family members before they were ready to tell their children themselves.

*I honestly think that's the best thing, because if they ever heard a conversation on the phone when you thought they were sleeping or got, you know, just heard anything at a family gathering, it'd be distrust. In my particular experience I think*

*it's best to keep the lines of communication open and be very honest.* (M#2, UI, 9 year old daughter)

A few parents reported feeling numb or "neutral" about the telling, as they were focused on other health-related matters. One co-parent didn't himself remember what the result was, only that it had been good news. Overall, parents reported that telling their child the mother's *BRCA* test result felt like an accomplishment, a way of clearing the air, ending parental worry about how and when to convey results.

**Regrets and disappointments—**In a few instances parents, in retrospect, expressed regret about some aspect of talking with their children about the mother's *BRCA1/2* result. Some regretted telling too much, others of talking too little. None experienced only regret about telling. One mother felt she should have asked more about her daughter's feelings after she conveyed her information. Several felt the configuration in which they told their children was not optimal, that it would have been better to tell them separately rather than together, allowing each child a better chance to "express what she feels". One such mother wished she had separately told a younger (15 year old) and an older (33 year old) daughter. She had not realized that the older daughter would feel much more pressured about approaching the age at which risk-reducing surgery would be recommended, while for the younger daughter this was a distant concern. Another mother felt she had told her children mostly for her own sake, not theirs, and wished she had waited to talk to them until they brought up related questions later on. She worried she had introduced a fear the children had not previously had. A mother wished she had had "a bigger discussion" with her child but did not define what that would have involved. A co-parent regretted not having made the disclosure a more formal discussion instead of it taking place while he was cutting onions in the kitchen. Another parent wished she had told her child in a less formal way, sitting on his bed or on the floor. Yet another mother was disappointed that she and her son could not talk more in-depth about the emotions aroused by the disclosure.

Lack of sufficient specificity in consideration of each child's need was the most common reason for regret, either in not configuring the disclosure conversation so there was sufficient room for each child to be able to express their emotions and ask questions or in having introduced too much of what the parent later thought was their own anxiety into what the parent said out of their own anxiety. The major conclusion parents came to, however, was that having their expectations of the disclosure met was less important than their ability to respond empathetically to each child, appreciating the child's cognitive understanding of the information received and encouraging the child to express his/her emotional reactions.

**Couple impact—**Most parents said they felt either the same or closer to each other as a result of disclosure to their children. However, for some parents where the maternal result was positive or uninformative, telling the child reactivated their own fears about possible future maternal breast cancer. Overall, however, the vast majority of parents remembered having come to quite similar conclusions about questions of whether, when and how to tell their children, even if, ultimately in some cases, the experience of telling did not mirror their plan.

### Child Impact as Perceived by Parents

Many of the parents felt their children generally had considerable fear about the cancer risks their mothers and other relatives faced, based on prior family experiences with maternal cancer and cancer treatment or cancer or cancer deaths in other relatives.

> *I think she has more questions about death than most children do and she give more contemplation to it than other, than most other children do.* (CP#18, UI, 8 year old daughter)

The co-parent of a 15 year old daughter whose mother tested positive, when asked if his daughter seemed worried after the discussion said,

> *Who wouldn't be? Yes, certainly, she-children-will worry.* (CP#28, BRCA+, 15 year old daughter).

He went on to say he thought his daughter worried about both her mother and herself. The father of a 16-year old son with a mutation-positive mother said his son's worry focused on the potential loss of his mother and also on the effect such a loss would have on his father.

Parents reported a range of initial child responses to the disclosure ranging from silence and withdrawal (in about a quarter of the cases) to considerable subsequent discussion of the children's questions and reactions. It tended to be parents where the mother was positive who reported children receiving the results in silence or not asking questions. However, one parent whose result was uninformative said her son left the room as soon as he "heard it was negative." Many children, according to their parents, also reported satisfaction with being told. Children whose parents had reported the mother did not carry a deleterious mutation were typically pleased, but also sometimes confused, especially if the mother had previously had cancer.

> *Oh, she'll say, "Mom, why did you get breast cancer if you don't carry this gene?" and I'll say to her, "There's no reason why". Or she'll say, "Just because you don't carry the gene, does that mean I won't get breast cancer?" And I'll say, "No, but there's less of a chance of you getting it" …I think she stews in her mind a little."* (M#17, UI, 12 year old daughter).

Clearly, in families heavily affected by cancer, genetic testing disclosure does not erase all the cancer-related concerns young people have. The discussion itself may bring up many concerns, though, from what most parents said, these were not new concerns, but ones which were the result of the family history of cancer. If the mother is not a carrier, the disclosure can be viewed as a source of relief. In many breast cancer families thought to possibly have a *BRCA1/2* mutation, the heightened additional risks of ovarian cancer associated with *BRCA1/2* are much feared and the absence of a positive maternal result brings relief of that fear for parents and knowledgeable, typically older, adolescent, children. Genetic information about *BRCA1/2* results is complex in its implications, however, and children, older and younger, may require several discussions to understand the implications for themselves and their mothers of a maternal result.

## Parental Advice

As pioneers in the discussion of *BRCA1/2* test results with next-generation, at-risk children, parents were asked what advice they could offer to other parents deciding about talking with their children. Parents were glad to offer advice, stating they'd often wished for some help prior to telling their own children.

**You Know Your Child—**The queried parents offered reassurance that parents are, after all, experts on their own children. While advice is desired from genetics professionals (see below), parents advised others to remember how well they know their own children and to take into consideration both the advice from others and the qualities and needs of each child in making decisions about disclosure. They encouraged other parents to think about each child's maturity, anxiety (especially about health-related issues), temperament, readiness to hear about the maternal result and other current stressors, i.e. to utilize their considerable

knowledge of their children to decide when and how it would be best to tell each child and only then, to formulate a plan for the timing and nature of telling all their children.

> *I think it really depends on the age of the child, that's so big in terms of their level of understanding what they could possibly absorb. And also, just, knowing the character of your child; how well they take -- how analytical they are or how worried they can get. You know it - it sort of takes - it does take thought. It can't just be blurted out probably if you're positive. I think if you're negative, you know, it's - OK, it's two things, age of child and, of course, is it positive or negative?* (M#19, True Negative result, 11 year old daughter)

> *The point is you have to, in some way, be able to guess how this is going to affect the children; how is it going to affect the future family life and if it's going to be devastating.* ( CP#4, BRCA+, 16 year old son)

**Maturity: A Personal Judgment—**Maturity was frequently mentioned as being important to a parent's disclosure decision, although there was considerable disparity in the ages at which parents considered a child mature enough to be told their mother's test result. Some thought 13 or 14 year olds would be mature enough to receive this information, while others thought the early 20s was when children are mature enough to understand this information responsibly. One mother advised telling at the earliest possible point so that children could use the information to make long-term plans about education or professional choice.

> *Tell them when they're old enough to understand--as soon as you can, especially girls. Because I think it has to factor into how they look at things. You know a girl who decided she was going to put off childbearing until she went through becoming an analyst, which you do by the time you're 40, you might want to change that plan and get her children finished before she turns 40 and has a hysterectomy.* (M#28, *BRCA*+, 15 year old daughter)

**Honesty and Openness—**While honesty was frequently praised as being critically important, several parents also made clear that they didn't think this necessitated giving all of the information to children before they needed it or were ready for it. What was stressed was that children need to know parents are secure in their beliefs that the genetic information being disseminated could be useful, that positive outcomes could result from the child's knowing the mother's mutation status, and that parents can be relied on to convey additional information if it becomes available.

> *A teenager does not need to know more than the basic. It depends on the nature and the personality of the child and how mature the child is, but I don't think a child has to know from A to Z about the whole test results and so forth…As time goes on, I will eventually share that information, just so they should know that they have to go for screenings and so forth. I really think you have to disseminate that information on a need-to-know basis rather than just saying, "We don't know and we are worried about this and we are worried about that" and* [so forth]. *Children don't need that. They need to know that the parents have some sort of plan.* (M#4, *BRCA*+, 16 year old son)

Similarly, another mother talking about her 16-year old son (who was not the target child) described him as believing:

> *I think he trusted me just to be in charge. If there was something else I need to tell him, I would.* (M#6, UI, 8 year old daughter).

**Take Time: Get Help if Uncomfortable—**Parents advised other parents to "take their time" and to think through who would benefit from telling their child--the child or themselves.

> *I think the first thing is to tell them to calm down about the time frame-that they don't have to decide this immediately. And, at the same time, about the resources that can help them reach a decision, one being the professionals.* (M#31, *BRCA*1/2+, 14 year old son)

If parents felt uncomfortable in the decision-making about telling their child, they were advised to first "find somebody to talk to who knows more than you." Some had talked to other relatives whom they felt had a better understanding of the facts related to *BRCA1/2*. One woman suggested talking to a pastor or priest. Genetic counselors were mentioned more frequently than physicians as possible sources of assistance in the telling of children. One mother imagined a mental health professional whose sole job it would be to offer parents advice about result disclosure to their children.

**Balance Fear and Hope; Emphasize Benefits—**Balancing provision of honest information and protection of children emotionally was what parents stressed for discussions with young children.

> *Just to break it to them gently, that's all, so that they are not afraid. Just give it to them as clear as possible, but as least fearful as possible because children deal with more than you actually think they are able to deal with. Just give it to them comfortably. They'll work with it. (M#14, BRCA*+, 10 year old son)

Parents said messages given to young children about maternal test results and genetic risk should be constructive and effective.

> *There are so many negatives in the world. Why would you want to put another negative in their lives? You know? Positively say, "Aren't we lucky that we have this gene? And if we do have it, well, this is what they recommend and these are the studies that show that"…gosh…and then try to go through it with a great attitude.* (M#26, *BRCA*+, 12 year old daughter)

Some advised putting the protective or preventive aspects of genetic information in the context of other ways families try to prevent other bad things from happening to them or their homes. These parents wanted to convey to the children that there were positive outcomes of testing, that if the mother was a carrier there were special approaches to preventing or finding cancer that would be available that were not open to her otherwise. Thus, the emphasis was on the news that things might be better in the future for knowing this information, even if it seemed a bit scary at present.

> *And the other thing is to talk about prevention…where to put the burglar alarms and fire alarms and when you have your oil changed... And you have to have preventive maintenance for your body and, luckily, in America today, we have tests that work. And proof of it is that your mother went through and it was found. Thank God we found it, because now we know what to do.* (CP#28, *BRCA*+, 15-year old daughter)

Subjects conveyed that parents needn't fear overwhelming their children if they pay attention to the cues children give through silence or active questioning about how open they are in the moment to receiving further information. Some reported that their children were freer to ask questions after the maternal result was disclosed; others felt children were silent or withdrew, suggesting that it had been difficult to hear about the mother's result. In conclusion, parents felt strongly that genetic information could be empowering for children

if the fear of being singled out genetically could be balanced by hope for better future health of family members.

**Telling can be Immunizing—**Some parents said giving children information about the presence of a hereditary cancer predisposition in the family, one which may impact their mother's health, could help prepare them if the mother were to develop cancer in the future. They felt it was important to slowly and carefully introduce the idea that this information did mean the parent was at increased cancer risk as a forewarning to help the child prepare for an uncertain future.

> *I think they should tell their children. Whatever the news. Whether it is good news or bad news. They can prepare their child. I think they should tell them. As long as the child can understand. If it is good news, the child will know and feel better and know how to handle it. If it is bad news, maybe it is not good at the time, but the child will have to know and prepare.* (M#12, UI, 21 year old son)

> *If I say that I maybe I will get cancer next year, he knew about this. He has been prepared for it.* (M#21, BRCA+, 12 year old son)

These parents felt that genetic risk information could offer immunization or stress inoculation against the shock of a future maternal cancer diagnosis.

**Advice About How Genetic Counselors Can Help Parents—**Parents and co-parents offered advice as well about how genetic counselors could help parents make decisions about telling their children the mother's genetic test results and carry out the disclosure. They conveyed that some parents do not even think about this issue and that counselors could usefully introduce the topic and offer a discussion of the pros and cons of telling children. The child's maturity, age and interest should be considered. Some difference of opinion was noted between the majority of subjects who suggested genetic counselors offer a non-judgmental listing of pros and cons ("it is not a right and wrong answer"), allowing parents to make their own decision, and others who thought the counselor should convey that parents needed to protect their children by informing them and that the only decision to be made was when to tell.

> *Knowledge is important. A child should have that.* (M#4, BRCA+, 16 year old son)

> *That it isn't just about the parents. If it is a positive result, it is about the kids, too, and they are just as affected as the parent is.* (CP, Uninformative, 8 year old son)

Parents hoped genetic counselors could offer statistics about how many parents disclose, how many do not, etc. and about real outcomes experienced by other parents when they have told their children or when they have not. They thought counselors could stress that even a child with a mutation does not have a 100% chance of getting cancer and that the prevention possibilities could be emphasized to parents. Counselors could also give parents room to consider their own emotional reaction before discussing their result with their child.

> *I think they should just stress to the parent that the parent has to be able to handle the information themselves before passing it on to their child.* (M#14, BRCA+, 10-year old son).

The value of telling children was something parents felt counselors could stress, even going so far as to say that children told their result would know it and could utilize it in the future in case the parent died.

> *You might be doing your children a favor by sharing information with them…and making it available in their health record, it's something that they might want to consider being tested for at a later time….When she's 21 or 18 or whatever, 25, I'm*

> *not sure exactly the age of, but as a young adult, that would be a topic of discussion at that point in time.* (M#18, UI, 8 year old daughter)
>
> *I think children can absorb a lot of information and they can actually sort through, if you present it to them the right way. They can not panic and then they can have this piece of information. And then, God forbid you die, at least they kind of remember the results of this.* (M#19, True Negative, 11-year old daughter)

Overall, parents thought it would be helpful if genetic counselors had an even larger voice in educating parents and preparing them for the challenges of talking to their children about hereditary cancer and genetic test results.

## DISCUSSION

Overall, importantly, this study reveals valuable, first-hand experiential reports of parents' thoughts and actions in deciding whether or not to disclose a maternal *BRCA1/2* test result to their children and in carrying out their decisions. These narratives support and extend earlier work offering information about the degree and timing of parent-child communication about a mother's result (Tercyak et al. 2001, Tercyak et al. 2002; Tercyak et al. 2009; Bradbury et al. 2009; Segal et al. 2004; Patenaude et al. 2006; Bradbury et al. 2007). Parents in our study report being largely relieved by the telling and the vast majority felt that their children did not respond adversely. A few questioned the depth of understanding young children had about the subject even after disclosure. Parents also made clear that some children showed little response to the initial telling and recommended later follow-up discussions. These particulars and, especially, the high level of parental satisfaction conveyed, support that parents can and do make decisions about disclosure to their children, the next generation at high risk, about genetic test results, often in close proximity to when the parents learned the result themselves. However, concern about this disclosure and the parents' call for greater involvement by genetic counselors in supporting parents suggests that this is a remediable source of anxiety tested parents experience.

The balance of protecting and educating children about potential harms associated with genetic predisposition was viewed as a classic parental responsibility. Parents saw the genetic information as a double-edged sword, full of both frightening (higher, multiple cancer risks for many family members) and hopeful (prevention possibilities) messages which it was their responsibility to convey accurately (though not necessarily fully) to their children. Subjects advised conveying the information in ways which stressed potential benefits of the knowledge gained by testing. Parents' views were predictable in some ways, citing the child's maturity and anxiety as factors in deciding when and how to tell or not to tell. Strikingly, however, the age which parents decided the child had sufficient "maturity" varied drastically, with parents of some children 16 or 18 years old still not considering them mature enough to tell the result to and other parents clearly deciding their 8-year old children should know. This strongly suggests the need for individualized discussion with parents about each of their offspring.

The children of our subjects are the first generation to grow up with parents who have personalized genetic information about their own risk. The parents we interviewed said they had wanted help in thinking about disclosure to their children, as they had viewed these conversations as difficult and different from anything they had previously discussed with their children. The novelty and complexity of genetic information have given professionals as well as parents pause in considering how best to convey it (MacDonald et al. 2010; Bennett et al. 2007; Brierly et al. 2010). The themes parents emphasized [see Table 2] mirror concerns voiced early in the era of Genetic Medicine by scientists and ethicists about whether tested adult individuals would react negatively or fatalistically to receipt of their

own genetic information, resulting in increased depression or anxiety (Lerman & Croyle, 1996; Codori 1997; Stiefel et al. 1997), which have generally not proven to be the case (Meiser 2005). Parents in our study offer hope to other parents that it is feasible to give children genetic information in fulfillment of parental responsibilities in ways that empower, protect, and prepare the children. The reassuring voices of our subjects suggest that parents can experience a deep sense of satisfaction in conveying familial genetic information to children, even when the test result is mutation-positive. The work, however, also supports the need for future research which directly assesses children's reactions so that we can better understand first-hand the impact of parental genetic testing on children. Both areas of research would usefully inform the development of formal psycho-educational interventions to aid parents and children in communicating about sensitive, genetic information. Our findings suggest these resources would be highly valued by tested parents.

The concept of immunizing children by providing forewarnings about possible future cancer diagnoses of a mother or other family members at a time before it is a reality has not been previously reported and deserves further investigation. It speaks to the resilience and empowerment which parents hope to induce in their children by successfully conveying the genetic information they possess, even if it also brings up the possibility of future medical challenges for family members. Parents see their children as benefitting from these discussions, especially if parents can frame the discussion in terms of the long term advantages of having and using genetic information.

While further research is needed, our findings suggest that genetic counselors have much they can offer to parents by bringing up the typical concerns parents face in thinking about talking to their children. Our data suggest it would be helpful for counselors to outline for parents making decisions about when or whether to tell the child the maternal result that they:

- Consider each child's maturity, anxiety (especially related to health issues), family history of illness, awareness about genetics, concurrent stressors and concern about the health of the mother and him/herself.
- Discuss among themselves their wishes, motivations and hopes for disclosure to each child,
- If they decide on telling the child (and most parents agree about whether or not to tell), that they discuss the ideal setting for telling the child, but also discuss if it is acceptable for either parent to convey the information when an opportune time presents itself.
- Discuss the actual words they want to use to convey information about hereditary cancer risks, genetic testing, and the genetic test result to their children. This can be surprisingly more complicated than expected and is an area where many parents would like guidance.
- Determine if they have enough uncertainty about how to inform children to seek professional or peer counsel either in further consultation with the genetic counselor or geneticist, in talking with a mental health professional or by speaking with other parents who have made and carried out their decision regarding disclosure. Connections might be made with other parents through liaison by the genetic counselor or by contacting online advocacy groups like Facing Our Risk of Cancer Empowered (FORCE), or by accessing relevant, online psycho-educational interventions as they become available.

- Recognize that they do not need to tell each child everything at the initial discussion, but can see how the child reacts and answer the child's questions (if asked), which will help guide future discussions.
- Acknowledge that the most important outcome is that the parents keep open the line of communication and questioning with their child about cancer and hereditary cancer risks so that any misconceptions or fears can be addressed.
- Know that most parents find telling to be a relief; parents who do not tell initially are advised to consider when and how they might convey this information to their children in the future.

While non-directedness has guided much of the field of genetic counseling, counselors have a storehouse of information of relevance to parents which can be helpful in resolving the dilemmas which arise about the sharing of familial genetic information. Raising relevant issues with parents who may not even know which questions to ask can certainly still be done within a non-directed structure. Creativity may be needed in the planning of genetic services to determine how and when best to aid parents coping with the questions which take on immediacy soon after result disclosure about whether, when or how children will be informed of the parent's result.

Limitations of this study include the prior selection of subjects from the larger sample based on predetermined characteristics, restriction to only discussion of maternal test results, and small sample size. This was a pilot effort to report parents' own views on telling children maternal test results.

More research is needed on the parental disclosure decision and the impact of inter-generational discussion on children's health behaviors. By engaging parents in this open discussion of their experiences we have significantly enlarged what is known about how parents fare in deciding about talking to their children about maternal test results and in carrying out their decisions. We have also learned where there continue to be gaps and needs, i.e. what parents would like to have better preparation for in this critical translation of information to the next at-risk generation. This information will be useful in the development of family communication interventions for parents, but it is also important that genetic counselors receive additional training about how best to prepare and support parents in these matters. Information gained from such studies will help us further refine knowledge of the impact of genetic information on parents and children and also help to define larger roles for genetic counselors in future decades, as they care for increasing numbers of families affected by hereditary cancers.

## Conclusion

Peer narratives offer support for primary care providers, oncologists, geneticists, genetic counselors, and mental health professionals to more confidently and actively advise parents about whether, when, and how to inform young children about parental test results. Decision tools developed as adjuncts to genetic counseling could further help parents anticipate important issues and model ways to frame discussions to emphasize strengths of each child. Future research which includes the voices of the children themselves will help us understand how parents' discussions with minor children affects children's perceptions of cancer risk, well-being, and attitudes towards future genetic testing and cancer screening. This knowledge will inform the future practice of genetic counseling, especially the genetic counseling of the next generation of those at risk for *BRCA1/2*-associated cancers.

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## References

Bennett C, Burton H, Farndon P. Competencies, education and support for new roles in cancer genetics services: outcomes from the cancer genetics pilot projects. Familial Cancer. 2007; 6(2): 171–180. [PubMed: 17520353]

Bradbury AR, Dignam JJ, Ibe CN, et al. How often do *BRCA* mutation carriers tell their young children of the family's risk for cancer? A study of parental disclosure of *BRCA* mutations to minors and young adults. J Clinical Oncology. 2007; 25(24):3705–3711.

Bradbury AR, Patrick-Miller L, Pawlowski K, et al. Learning your parent's BRCA mutation during adolescence or early adulthood: a study of offspring experiences. Psychooncology. 2009; 18(2): 200–208. [PubMed: 18702049]

Brierley KL, Campfield D, Ducaine W, Dohany L, Donenberg T, Shannon K, Schwartz RC, Matloff E. Errors in delivery of cancer genetics services: implications for practice. Connecticut Medicine. 2010; 74:413–423. [PubMed: 20806621]

Clarke S, Butler K, Esplen MJ. The phases of disclosing *BRCA1/2* genetic test results to offspring. Psychooncology. 2008; 17:797–803. [PubMed: 18646247]

Codori AM. Psychological opportunities and hazards in predictive genetic testing for cancer risk. Gasteroenterology Clinics of North America. 1997; 26:19–39.

DeMarco, T.; Sharff, M.; Peshkin, BN., et al. Qualitative analysis of parents' reasons for and against disclosing maternal *BRCA1/2* test result to their minor-age children. Abstract for the 28th Annual meeting of the National Association of Genetic Counselors Education Meeting; Atlanta, GA. November 2009; 2009.

Easton DF, Ford D, Bishop DT. and the Breast Cancer Linkage Consortium. Breast and ovarian cancer incidence in *BRCA1/2* mutation carriers. American Journal of Human Genetics. 1995; 56(1):265–271. [PubMed: 7825587]

Eisenbraun A, Wenstrup R, Hellerstedt B, et al. Hereditary breast and ovarian cancer testing: integration and outcomes within community oncology practices. Community Oncology. 2010; 7:75–81.

Ford D, Easton DF, Bishop DT, Narod SA, Goldgar DE. Risks of cancer in *BRCA1/2* mutation carriers. Lancet. 1994; 343(8899):692–695. [PubMed: 7907678]

Ford D, Easton DF, Stratton M. and the Breast Cancer Linkage Consortium. Genetic heterogeneity and penetrance analysis of the *BRCA1* and *BRCA2* genes in breast cancer families. American Journal of Human Genetics. 1998; 62(7):676–689. [PubMed: 9497246]

Lerman C, Croyle RT. Emotional and behavioral responses to genetic testing for susceptibility to cancer. Oncology. 1996; 10(2):191–195. [PubMed: 8838261]

MacDonald DJ, Sarna L, Weitzel JN, et al. Women's perceptions of the personal and family impact of genetic cancer risk assessment: Focus group findings. Journal Genetic Counseling. 2010; 19(2): 148–160.

MacKenzie A, Patrick-Miller L, Bradbury AR. Controversies in communication of genetic risk for hereditary breast cancer. The Breast Journal. 2009; 15(Suppl 1):S25–32. [PubMed: 19775327]

Meiser B. Psychological impact of genetic testing for cancer susceptibility: an update of the literature. Psychooncology. 2005; 14(12):1060–74. [PubMed: 15937976]

Miesfeldt S, Cohn WF, Jones SM. Breast cancer survivors' attitudes towards communication of breast cancer risk to their children. American Journal Medicine Genetics C Seminars in Medical Genetics. 2003; 119C(1):45–50.

Muhr T. ATLAS. ti - A prototype for the support of text interpretation. Qualitative Sociology. 1991; 14:349–371.

Offit, K. Clinical Cancer Genetics: Risk Counseling and Management. Wiley-Liss; New York: 1998.

Patenaude AF, Dorval M, DiGianni LS, et al. Sharing *BRCA1/2* test results with first-degree relatives: factors predicting who women tell. Journal of Clinical Oncology. 2006; 24(4):700–706. [PubMed: 16446344]

Peshkin BN, Demarco TA, Tercyak KP. On the development of a decision support intervention for mothers undergoing *BRCA1/2* cancer genetic testing regarding communicating test results to their children. Familial Cancer. 2010; 9(1):89–97. [PubMed: 19609726]

Sharff ME, DeMarco TA, Mays DM, et al. Parenting through genetic uncertainty: Themes in the disclosing of breast cancer risk information to children. Genetic Testing and Molecular Biomarkers. 2011 [Epub ahead of print].

Segal J, Esplen MJ, Toner B, et al. An investigation of the disclosure process and support needs of *BRCA1* and *BRCA2* carriers. American Journal of Medicine Genetics A. 2004; 125A(3):267–72.

Stiefel F, Lehmann A, Guex P. Genetic detection: the need for psychosocial support in modern cancer prevention. Supportive Care in Cancer. 1997; 5:461–465. [PubMed: 9406360]

Struewing JP, Hartge P, Wacholder S, Baker SM, Berlin SM, McAdams M, et al. The risk of cancer associated with specific mutations of *BRCA1/2* and BRCA2 among Ashkenazi Jews. New England Journal of Medicine. 1997; 336(20):1401–1408. [PubMed: 9145676]

Tercyak KP, Hughes C, Main D, et al. Parental Communication of *BRCA1/2* genetic test results to children. Patient Education and Counseling. 2001; 42(3):213–224. [PubMed: 11164320]

Tercyak KP, Peshkin BN, DeMarco TA, et al. Parent-child factors and their effect on communicating test results to children. Patient Education and Counseling. 2002; 47(2):145–153. [PubMed: 12191538]

Tercyak KP, Peshkin BN, Demarco TA, et al. Information needs of mothers regarding communicating *BRCA1/2* cancer genetic test results to their children. Genetic Testing. 2007; 11(3):249–255. [PubMed: 17949286]

Tercyak, KP.; Peshkin, BN.; DeMarco, TA., et al. Parental decisions and outcomes regarding disclosing maternal *BRCA1/2* test results to children. Presented at ASCO Annual Meeting; Orlando, Florida. May 30-June 2; 2009.

Weiss, RS. Learning from Strangers: The Art and Method of Qualitative Interview Studies. Free Press; New York: 1994.

## Table 1

Participant characteristics (*N*=56)

| Characteristics | Mothers (*N*=32) *M* (SD) | *N* (%) | Co-parents (*N*=24) *M* (SD) | *N* (%) |
|---|---|---|---|---|
| Age | 46.4 (6.7) | | 49.9 (9.2) | |
| Gender | | | | |
| Female | | 32 (100.0) | | 1 (4.2) |
| Male | | 0 | | 23 (95.8) |
| Race | | | | |
| Caucasian | | 23 (71.9) | | 22 (91.7) |
| Noncaucasian | | 9 (28.1) | | 2 (8.3) |
| Education | | | | |
| <College | | 8 (25.0) | | 4 (16.7) |
| ≥College | | 24 (75.0) | | 20 (83.3) |
| Household Income | | | | |
| <$75*k* | | 11 (34.4) | | 5 (21.8) |
| ≥$75*k* | | 21 (65.6) | | 19 (79.2) |
| Married/living as married | | 25 (78.1) | | 23 (95.8) |
| Child Age | 12.9 (3.8) | | 11.8 (3.1) | |
| Child Gender | | | | |
| Female | | 18 (56.3) | | 14 (58.3) |
| Male | | 14 (43.7) | | 10 (41.7) |
| Relationship to child | | | | |
| Biological Parent | | 32 (100.0) | | 21 (87.5) |
| Step-Parent | | 0 | | 2 (8.3) |
| Partner of mother | | 0 | | 1 (4.2) |
| *Maternal cancer history | | 21 (65.6) | | 14 (58.3) |
| Breast Cancer | | 17 (53.1) | | 12 (50.0) |
| Ovarian Cancer | | 2 (6.2) | | 1 (4.2) |
| Breast and Ovarian Cancer | | 1 (3.1) | | 1 (4.2) |
| *Maternal *BRCA1/2* carrier status | | | | |
| True negative | | 11 (34.4) | | 9 (37.5) |
| Uninformative | | 8 (25.0) | | 4 (16.7) |
| Positive | | 13 (40.6) | | 11 (45.8) |

*Note: Refers to mothers in study; for co-parents refers to their tested partner

## Table 2

Parental Concerns and Advice Concerning Talking to Children about Maternal *BRCA1/2* Test Result

| **Parental Concern** | **Theme of Parental Advice** |
|---|---|
| Burden on parents of not telling | Assess children's vulnerabilities. There is relief in telling |
| Fear child may overhear test result | Tell at earliest point when parents are comfortable |
| Couple needs to discuss telling children | Though good to plan, also tolerable if one parent spontaneously tells children at opportune moment |
| Prophylactic surgery may prompt telling test result | Children can understand, but, especially daughters may need time to adjust to this news |
| Question own ability to convey proper information | Get help from well-informed relatives or by asking professionals for advice about informing children |
| Couple impact | Test result disclosure may re-arouse parents' fears about maternal illness, which merits discussion between the couple. |
| Indecision about how to tell children | Telling all children together may not recognize differences in strengths, vulnerabilities of each child or differences age makes in consideration of implications for screening or risk-reducing options |
| Desire to be honest, but not overwhelming to children | Initial talk doesn't have to tell all about *BRCA1/2*. Can introduce, ask what questions child has and try to answer, and encourage on- going dialogue. |
| Uncertainty about child's maturity to understand genetic result and implications. | Parents differed widely on what ages they considered children had sufficient maturity to handle receipt of *BRCA1/2* test result |
| Concern about distressing child | Important to stress positive, constructive ways in which genetic information can reduce family illness burden in the future; Assess individually. |
| Children need preparation for possible future maternal cancer diagnosis | Telling about positive or UI maternal test result may immunize children, help prepare them for and reduce shock of future maternal illness. |
| Parental decision is not to tell children | Planning for future telling of children may reduce parental anxiety; some parents make plans for post–death informing of children |