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The role of cognitive appraisal and worry in BRCA1/2 testing decisions among a clinic population

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Abstract

Previous studies examining decision making in the context of genetic testing for BRCA1/2 gene mutations have been limited in their reliance on cross-sectional designs, lack of theoretical guidance, and focus on measures of intention rather than actual behavior. Informed by the Health Belief Model and other theories of self-regulation, the present study set out to examine the role of cognitive appraisal and worry in BRCA1/2 testing decisions. A total of 205 women completed baseline questionnaires prior to their genetic counselling appointment. Medical charts were audited to determine testing decisions. Bivariate analyses indicated that perceived severity of being a carrier and perceived benefits and barriers to testing were significantly associated with testing decisions. Perceived benefits remained significant in multivariate analyses. Moreover, multivariate analyses revealed a significant three-way interaction between perceived susceptibility, perceived severity, and worry about being a mutation carrier and testing decisions. Among women high in baseline worry, those high in perceived susceptibility but low in perceived severity were significantly more likely to undergo genetic testing than all other susceptibility/severity combinations (80% *vs.* 36.2–42.9% range; Wald test $= 8.79$, $p < 0.01$). These results support the need for researchers and practitioners to consider how interactions between cognition and worry may influence genetic testing decisions.

Keywords: *Cognitive appraisal, Health Belief Model, self-regulation, worry, genetic testing, decision making*

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Introduction

Genetic testing is now available for several hereditary cancer syndromes including breast and ovarian cancer (Eng, Hampel, & de la Chapelle, 2001). The rationale for undergoing cancer predisposition testing stems from its potential benefits: namely, the identification of high-risk individuals to enable increased surveillance, preventive surgery (e.g., prophylactic mastectomy and/or oophorectomy), or chemo-prevention. Based on initial surveys, interest in cancer predisposition testing has been high (Andrykowski, Lightner, Studts, & Munn, 1997; Chaliki et al., 1995; Croyle & Lerman, 1993; Diefenbach, Schnoll, Miller, & Brower, 2000; Lerman, Seay, Balshem, & Audrain, 1995). Actual uptake of various tests has varied depending on several factors including investigator influences (e.g., provision of genetic testing at no cost) and study population (i.e., members of hereditary cancer families *vs.* clinic populations)(Ropka, Wenzel, Phillips, Siadaty, & Philbrick, 2006).

Many studies have been published on predictors of women's decisions to undergo BRCA1/2 testing for hereditary breast and ovarian cancer (Bosompra et al., 2000; Cameron & Reeve, 2006; Durfy, Bowen, McTiernan, Sporleder, & Burke, 1999; Helmes, 2002; Jacobsen, Valdimarsdottir, Brown, & Offit, 1997; Shiloh & Ilan, 2005). Much of this prior research, however, has been limited by the reliance on cross-sectional designs, which focused on interest or intention to test (Wang, Gonzalez, & Merajver, 2004). Yet, it is well documented that behavioral intentions, interest or willingness to test are not always reflective of behavior (Cappelli et al., 1999; Croyle & Lerman, 1999; Kelly et al., 2004). Few studies have prospectively examined predictors of actual BRCA1/2 testing decisions (Biesecker et al., 2000; Kelly et al., 2004; Lerman et al., 1996, 1997). Overall, these prospective studies have identified several predictors of testing including older age, more first-degree relatives with breast cancer, having health insurance, more knowledge of BRCA testing, greater psychological distress, and greater perceived benefits of testing. However, none of these studies were guided by theory to explain testing decisions, and all but one were conducted with high-risk research populations; namely, those individuals who were previously involved in testing for the purposes of genetics research and/or who were from families with hereditary breast and ovarian cancer.

This study had two main objectives: (1) to explore the contribution of the Health Belief Model (HBM) constructs and their interrelationships in predicting testing decisions; and (2) to examine the additional role of worry in testing decisions. The first objective focused on applying theory to help predict decisions to undergo genetic testing for hereditary breast and ovarian cancer (BRCA1/2 testing). The HBM was chosen as a starting point to guide our study. Although the HBM has been widely used in the genetics realm, application of this model in our study allowed for possible clarification of prior inconsistencies in the literature (i.e., association between perceived risk and genetic testing decisions) and replication of findings that were based on cross-sectional studies (Durfy et al., 1999; Glanz, Grove, Lerman, Gotay, & Le Marchand, 1999; Helmes, 2002;

Shiloh & Ilan, 2005; Shiloh, Petel, Papa, & Goldman, 1998; Welkenhuysen, Evers-Kiebooms, Decruyenaere, Claes, & Denayer, 2001). The HBM was also selected because of its cognitive appraisal components; namely perceived susceptibility and perceived severity, which are common to other theories of health behaviour and decision-making including Self-regulation Theory, Protection Motivation Theory, and Transactional Model of Stress and Coping (Lazarus & Folkman, 1984; Leventhal, Safer, & Panagis, 1983; Rogers, 1983; Ronis, 1992).

The second objective focused on examining the role of worry in the decision to undergo BRCA1/2 testing. Worry or affect is commonly excluded from traditional value-expectancy theories of health behaviour and decision-making. However, the manner in which an individual emotionally responds to a threat can have implications for motivating behaviour (Leventhal, 1970; Leventhal et al., 1983; Loewenstein, Weber, Hsee, & Welch, 2001; Redelmeier, Rozin, & Kahneman, 1993). Thus, consistent with theories of self-regulation (e.g., common-sense model), emotional reactions to a stressor are considered partially independent processes from cognitive assessments or appraisals, which interact with one another to produce behavioral outcomes (Leventhal, 1970; Leventhal et al., 1983; Loewenstein et al., 2001; Miller & Schnoll, 2000). Cognitive assessments of a stressor influence emotional responses which, in turn, influence subsequent assessments of the threat. These two processes, however, not only have different determinants, but also lead to differing respective coping strategies to deal with either the cognitive representation or the emotional reaction to the stressor. In the present study, we set out to examine how worry may influence the decision to undergo genetic testing, especially in relation to traditionally studied cognitive factors.

In addition, since the lack of model testing has been a major criticism of HBM research (Janz, Champion, & Strecher, 2002), our study also focused not only on how the constructs depicted by HBM were individually associated with testing decisions, but on the relation between the constructs themselves, and how this relation, in turn, influenced testing decisions. To do so, the role of HBM constructs (and worry) was examined in two ways. First, we analyzed the role of HBM constructs in a manner consistent with the majority of other studies using this model – by examining all the constructs at once (i.e., entering all model-relevant predictors into a logistic regression) to determine the strongest predictor(s) of testing decisions. Second, we set out to separately test the interaction between the cognitive appraisal components of the HBM that are common among value-expectancy theories and worry to determine the interrelation between these constructs. Namely, we focus on the role of worry as a moderator of perceived susceptibility and perceived severity of being a mutation carrier on genetic testing decisions.

We hypothesized that individuals would be more likely to undergo genetic testing if they perceive themselves at greater risk for being a BRCA1/2 mutation carrier, and believe that the benefits of genetic testing outweigh the barriers to testing. Perceived severity of being a gene mutation carrier was hypothesized to be

negatively associated with testing decisions, such that those who perceived the consequences of being a carrier to be more severe (i.e., it will have a greater impact or disruption) would be less likely to undergo genetic testing. This latter hypothesis was based on the earlier work of Becker and co-workers who found an inverse relationship between perceived severity and genetic testing decisions (Becker, Kaback, Rosenstock, & Ruth, 1975).[1]

Prior evidence suggests that perceived susceptibility and psychological distress, such as worry, interact to explain testing decisions – those who perceive their risk to be high and are more worried about the condition are most likely to go ahead with genetic testing (Codori et al., 1999; Shiloh & Ilan, 2005). Moreover, because perceived severity and perceived susceptibility often interact to produce threat perceptions (Weinstein, 2000), it was anticipated that heightened perceptions of severity of being a carrier would modify the relationship between perceived susceptibility and worry on genetic testing decisions. As such, it was hypothesized that women would be most likely to undergo genetic testing if they thought they were at risk, were worried about being a carrier, and perceived the consequences of finding out they are a carrier to be less severe.

Finally, unlike the majority of prior prospective studies in this area, women in the present study were not high-risk women previously enrolled in genetics research, but rather women who were attending a clinic that provided genetic counselling and testing as a service. Women did not have to be of a certain "risk" level to be eligible to attend the clinic, which allowed for greater variability in the actual risk status of women included in this study (i.e., women at low hereditary risk). In addition, both the counseling service as well as genetic testing were not provided free of charge and were paid for either out-of-pocket or via health insurance. It was anticipated that the findings on this clinic-based sample would be more applicable to the general population of individuals interested in genetic testing than many of the prior studies, which have focused primarily on women from high-risk families where mutation status was already known through participation in research and/or where testing was offered without cost (Biesecker et al., 2000; Bonadona et al., 2002; Croyle, Smith, Botkin, Baty, & Nash, 1997; Dorval et al., 2000; Kelly et al., 2004; Lerman et al., 1996, 1997).

Method

Participants

Participants in this study were women who attended the Breast and Ovarian Cancer Risk Evaluation Program (BOCREP) at the University of Michigan Comprehensive Cancer Center. The BOCREP is a clinic that provides breast cancer risk assessment, genetic counseling and follow-up risk management and preventive services. Women can either self-refer or be referred to the clinic by a health care provider. Clinical genetic testing via Myriad Genetic Laboratories in Salt Lake City, Utah, is also available through the clinic. Between November 2000 and December 2002, a total of 205 women had completed baseline

questionnaires and were seen in the clinic. None of these women had prior genetic testing performed. Individuals were not excluded from the study based on their likelihood of having a BRCA1/2 mutation.

Procedures

Women who contacted the BOCREP for an appointment were mailed an initial pre-clinic questionnaire package, which included an informed consent form, family history questionnaire, and patient questionnaire. The patient questionnaire contained items addressing demographic variables and baseline predictors of interest. Women were consented to complete the patient questionnaires and informed that their medical care would not be affected if they chose not to complete the forms. All forms were completed at home and returned by mail to the clinic. A clinic appointment was scheduled once the initial questionnaires were returned.

On the day of the clinic appointment, all women met with a genetic counselor and a medical oncologist. The average counseling session lasted approximately 1.5–2 h. Clinic recommendations for genetic testing (i.e., best candidate for testing) were based on cutoff of 10% risk of familial mutation (Couch et al., 1997; Frank, 1999). Women with less than a 10% chance of harboring a germline mutation in BRCA1/2 were discouraged from testing, but not denied. Based on these criteria, best candidacy for genetic testing was defined as (1) self, (2) other (i.e., affected family member), and (3) not encouraged to test due to low risk. Women who decided to have genetic testing could either have their blood drawn on the day of their initial appointment or at a later date.

All the women included in the present study were also part of a randomized trial to examine the impact of two strategies intended to facilitate the genetic counseling encounter: a CD-ROM computer program and feedback to the genetic counselor on women's prior misconceptions about cancer and genetics (2×2 factorial design). Detailed findings from the randomized trial are presented elsewhere (Wang, Gonzalez, Milliron, Strecher, & Merajver, 2005). In summary, women attending clinic were randomized to one of four experimental conditions (standard care, CD-ROM, feedback to counselor, both CD-ROM and feedback), and may have viewed a CD-ROM program prior to their genetic counseling session. The study found that the CD-ROM, but not feedback to the genetic counselor, had an impact on genetic testing decisions. Specifically, women who viewed the CD-ROM prior to genetic counselling were significantly less likely to undergo testing compared with women who did not view the CD-ROM. Due to the impact of the randomized trial on testing decisions, the experimental group was controlled in the analyses of the present study.

Measures

Demographics. Age, education, ethnicity, marital status, income, and prior cancer history (breast, ovarian, DCIS) were assessed using baseline questionnaires.

The most appropriate candidate for initial testing in the family was assessed by the clinic team.

HBM constructs (see Appendix).   Application of the HBM to BRCA1/2 testing was done by using a reformulation of the model that focused on susceptibility and severity of being a mutation carrier, rather than of disease (i.e., breast or ovarian cancer). This reformulation was adopted for several reasons. First, similar to a study conducted by Becker et al. (1975), our study focused on adults undergoing genetic testing, where individuals often test to determine the risk of disease among offspring (Lerman et al., 1995; Lynch et al., 1997, 1999; Tessaro, Borstelmann, Regan, Rimer, & Winer, 1997). HBM constructs were similarly modified in the Becker et al. study in efforts to better explain genetic testing decisions. Second, assessing beliefs about disease susceptibility or severity likely differs between affected and unaffected individuals, even if one considers risk and severity of disease recurrence among the former. As BRCA1/2 testing often begins with testing in an affected relative, assessing perceived susceptibility and perceived severity of being a mutation carrier was more applicable and appropriate for all individuals included in the study. Perceived benefits and barriers were also examined by focusing on benefits and barriers of undergoing a genetic test and learning one's carrier status. The items used to assess HBM constructs in the present study were adapted from other published measures that assessed HBM constructs in the context of breast cancer screening (Champion, 1999) or assessed similar constructs (i.e., pros/cons) in the context of BRCA1/2 testing (Jacobsen et al., 1997).

Perceived susceptibility.   Perceived susceptibility of being a gene mutation carrier was assessed by averaging the values on two items that were rated on a 5-point Likert scale, ranging from strongly disagree to strongly agree. Internal consistency was high for this measure (Cronbach's $\alpha = 0.89$). Higher scores indicated a greater level of perceived susceptibility.

Perceived severity.   Three items were used to measure perceived severity of being a gene mutation carrier. This measure was also relatively high in internal consistency (Cronbach's $\alpha = 0.81$). A higher mean score indicated a greater level of perceived severity.

Perceived benefits.   Seven items were included to assess the benefits of genetic testing and a total benefits score was obtained by taking the average of the sum of values. The benefits scale had relatively high internal consistency (Cronbach's $\alpha = 0.80$). Higher values indicated greater perceived benefits.

Perceived barriers.   Eight items were included to assess the barriers of genetic testing, and a total barriers score was obtained by taking the average of the sum of values. Internal consistency for this scale was adequate (Cronbach's $\alpha = 0.71$). A higher score indicated a greater level of perceived barriers.

Worry. Worry was examined in the present study by focusing on worry about having a gene mutation, rather than disease, to remain consistent with the HBM reformulation. Worry about being a gene mutation carrier was assessed with a single item that asked, ''As of this moment, how worried are you that you have a gene mutation that may increase your risk for developing breast/ovarian cancer?'' This item was rated on a 5-point Likert scale ranging from not at all worried to extremely worried.

Genetic testing decision. Genetic testing decisions were recorded via a medical chart audit approximately 1 year following the initial clinic visit.

Analysis Plan

Descriptive statistics were first analyzed for demographics, prior cancer history (affected status), best candidacy for testing as indicated by the clinic team, and actual uptake of genetic testing. In addition, descriptive statistics and Pearson product-moment correlations were computed for the psychosocial predictors of interest; namely, the HBM constructs and worry about having a gene mutation. Chi-square tests of association were used to examine the association between psychosocial predictors and genetic testing decisions. Two hierarchical logistic regression analyses were performed to determine (1) the relative importance of the HBM variables and worry on genetic testing decisions, and (2) interactions between cognitive appraisal and worry. All logistic regressions controlled for covariates including age (<50, 50+), best candidacy (self *vs.* other/not encouraged), and experimental group (CD-ROM, Feedback, CD-ROM × Feedback).

Results

Descriptive Statistics

Study participants ranged in age from 22 to 76 years (M = 45, SD = 10.2). The majority of women were Caucasian (93%), married (80%), and reported having at least a bachelor's degree or higher (58%) and a household income greater than $60,000 (66%). In total, 31% of the women had previously been diagnosed with DCIS, invasive breast cancer, and/or ovarian cancer. Clinic recommendations for genetic testing candidacy were as follows: 49.8% (self), 32.7% (other), and 17.6% (not encouraged). In total, 82 out of 205 women (40%) underwent genetic testing.

Predictors of genetic testing: descriptive data and bivariate analyses

Means and Pearson product-moment correlations between HBM variables and worry are presented in Table 1. Bivariate associations between various psychosocial predictors and genetic testing decisions were examined. Significant correlations were noted between worry and several variables including perceived susceptibility, perceived severity, and perceived benefits. Perceived

Table I.   Means, SDs and zero-order correlations for predictor variables of interest.

	Perceived susceptibility	Perceived severity	Perceived benefits	Perceived barriers	Worry about gene mutation
Mean	3.55	2.96	3.74	1.95	3.48
(SD)	(0.99)	(0.95)	(0.70)	(0.57)	(1.11)
Perceived susceptibility	–	−0.051	0.152*	0.007	0.459**
Perceived severity		–	−0.119	0.333**	0.180*
Perceived benefits			–	−0.388**	0.294**
Perceived barriers				–	−0.095

*$p < 0.05$. **$p < 0.001$, two-tailed.

Table II.   Bivariate associations of predictor variables with genetic testing decisions.

Variable	Levels	N (%) tested	ϕ	χ^2	p-value
Baseline					
Perceived susceptibility	Low (<4)	38 (37.3)	0.064	0.84	0.360
	High (≥4)	44 (43.6)			
Perceived severity	Low (<3)	39 (48.8)	−0.140	3.88	0.049
	High (≥3)	41 (34.7)			
Perceived benefits	Low (≤3.6)	25 (26.6)	0.252	12.93	<0.001
	High (>3.6)	56 (51.4)			
Perceived barriers	Low (≤2.0)	52 (46.4)	−0.148	4.44	0.035
	High (>2.0)	29 (31.9)			
Worry about gene mutation	Low (<4)	34 (33.7)	0.127	3.33	0.068
	High (≥4)	48 (46.2)			

barriers were significantly associated with perceived severity and perceived benefits. Finally, perceived susceptibility was also significantly associated with perceived benefits. All predictor variables were then dichotomized according to median splits in order to facilitate presentation of data in figures and logistic regression tables. As shown in Table II, the decision to have genetic testing was associated with higher levels of perceived benefits (51.4 *vs.* 26.6%), lower perceived barriers (46.4 *vs.* 31.9%), and lower perceived severity (48.8 *vs.* 34.7%). Testing decisions also showed a trends towards being significantly associated with higher worry (46.2 *vs.* 33.7%).

Predictors of genetic testing: multivariate analyses

The first logistic regression looked at the four predictors commonly examined within the HBM entered as additive predictors: perceived susceptibility, perceived severity, perceived benefits, and perceived barriers. In addition, the impact of worry about gene mutation on testing decisions was also examined. Covariates including age, best candidacy, and experimental condition were entered first into the logistic regression, followed by the HBM constructs in Step 2 and worry in Step 3. As shown in Table III, best candidacy for testing ($OR = 0.02$, *CI* [0.006–0.048], $p < 0.001$) and randomization to the CD-ROM experimental condition ($OR = 0.61$, *CI* [0.39–0.95], $p = 0.03$) were significantly

Table III. Logistic regression analysis of genetic testing decisions: perceived susceptibility, perceived severity, perceived benefits, perceived barriers and worry about gene mutation.

Variable	Levels	$\Delta\chi^2$	N	OR [CI]	p-value
Step 1: Covariates		115.39*			
Age	<50		128	1.74 [0.70–4.36]	0.24
	50+		61		
Best candidate	Self		95	0.02 [0.006–0.048]	<0.001
	Other/not encouraged		94		
Randomized condition					
CD-ROM				0.61 [0.39–0.95]	0.03
Feedback				0.94 [0.61–1.44]	0.77
CD × Feedback				1.21 [0.78–1.86]	0.40
Step 2: HBM predictors		18.28*			
Susceptibility	Low (<4)		94	0.71 [0.44–1.16]	0.18
	High (≥4)		95		
Severity	Low (<3)		75	1.17 [0.71–1.91]	0.54
	High (≥3)		114		
Benefits	Low (≤3.6)		89	2.75 [1.59–4.75]	<0.001
	High (>3.6)		100		
Barriers	Low (≤2)		104	0.99 [0.59–1.67]	0.98
	High (>2)		85		
Step 3: Other predictors		0.03 ns			
Worry	Low (<4)		94	1.05 [0.62–1.77]	0.85
	High (≥4)		95		

Overall model χ^2 (10) = 133.70, $p < 0.001$.
**$p \le 001$.

associated with genetic testing decisions. Women who were deemed the best candidate for testing and those who were not randomized to the CD-ROM experimental condition were significantly more likely to have genetic testing. The only HBM variable that significantly predicted genetic testing decisions was perceived benefits ($OR = 2.75$, CI [1.59–4.75], $p < 0.001$). Thus, women who perceived the benefits of testing to be high were more likely to undergo genetic testing compared with women who perceived the benefits of testing to be low. The inclusion of worry in Step 3 with HBM variables did not alter the associations between HBM variables and testing decisions, nor was worry a significant predictor of testing decisions.

Next, we focused on testing interactions between the psychosocial predictors of interest. From the HBM, we included the variables of perceived susceptibility and perceived severity, which are constructs common to value-expectancy theories. In addition, worry was also included in this model in efforts to examine how it may interact with cognitive appraisal and influence genetic testing decisions. A separate logistic regression was conducted to specifically test the interactions between perceived susceptibility, perceived severity, and worry about having a genetic mutation. As the theoretical interest was on the three-way interaction, we included two stages of control variables. Step 1 included standard covariates

Table IV. Logistic regression analysis of genetic testing decisions: perceived susceptibility, perceived severity, and worry about gene mutation.

Variable	Levels	$\Delta\chi^2$	N	OR [CI]	p-value
Step 1: Covariates		116.87*			
Age	<50		129	1.72 [0.68–4.32]	0.24
	50+		61		
Best candidate	Self		96	0.16 [0.006–0.047]	<0.001
	Other/not encouraged	94			
Randomized condition					
CD-ROM				0.61 [0.39–0.94]	0.03
Feedback				0.93 [0.61–1.42]	0.72
CD × Feedback				1.22 [0.79–1.88]	0.37
Step 2: Main effects and two-way interactions		5.71 ns			
Susceptibility	Low (<4)		95	0.75 [0.47–1.22]	0.25
	High (≥4)		95		
Severity	Low (<3)		114	1.02 [0.63–1.64]	0.94
	High (≥3)		76		
Worry	Low (<4)		95	1.29 [0.79–2.08]	0.31
	High (≥4)		95		
Susceptibility × severity				0.88 [0.54–1.43]	0.61
Susceptibility × worry				1.55 [0.93–2.59]	0.10
Severity × worry				0.90 [0.59–1.44]	0.66
Step 3: Three-way interaction		4.32**			
Susceptibility × severity × worry				0.59 [0.36–0.98]	0.04

Overall model χ^2 (12) = 126.91, $p < 0.001$.
**$p < 0.001$, *$p < 0.05$.

and Step 2 included all the main effects and two-way interactions. Finally, in Step 3, the three-way interaction term was entered into the logistic regression model (Table IV). Consistent with the first logistic regression, best candidacy ($OR = 0.16$, CI [0.006–0.047], $p < 0.001$) and randomization to the CD-ROM experimental condition ($OR = 0.61$, CI [0.39–0.94], $p = 0.03$) were significantly associated with genetic testing decisions. None of the main effects or two-way interactions were significant in Step 2. However, results from the second logistic regression indicate a significant three-way interaction between perceived susceptibility, perceived severity, and worry ($OR = 0.59$, CI [0.36–0.98], $p = 0.04$). The omnibus test for Step 3 was statistically significant, $\Delta\chi^2 = 4.32$, $p < 0.05$.

Figure 1 shows the three-way interaction and presents the percentage of women who underwent genetic testing by susceptibility, severity and worry. Based on the three-way interaction finding, additional analyses were conducted to examine whether the group of women who perceived both low severity and high susceptibility were different from the other three combination groups among high worriers. Contrast tests revealed that the proportion of women deciding to have testing among the low severity/high susceptibility group was significantly
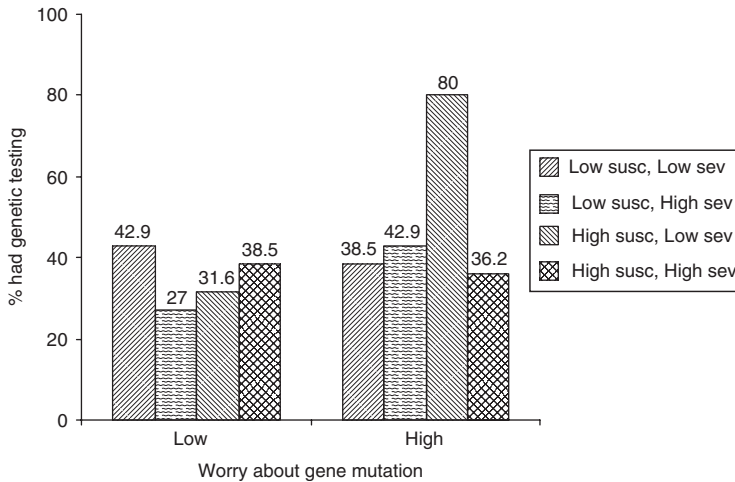
Figure 1. Percentage of women undergoing genetic testing by susceptibility, severity, and worry about gene mutation.

greater than the average proportion of women from the other three groups (80 *vs.* 36.2–42.9% range; Wald test $= 8.79$, $p < 0.01$).

Discussion

This study set out to better understand and predict how women are making BRCA1/2 testing decisions. A central focus of our study was to examine the contribution of the major constructs of the HBM as well as the role of worry in explaining actual genetic testing decisions. Bivariate analysis of predictor variables demonstrated the expected patterns of association between perceived severity, perceived benefits and perceived barriers with respect to genetic testing uptake. In contrast, perceptions of susceptibility and worry were not found to be significantly associated with testing decisions. When all HBM constructs were simultaneously entered into a logistic regression model, only perceived benefits significantly predicted decisions to undergo genetic testing. Other researchers have also found evidence to support the importance of perceived benefits in the decision to undergo genetic testing (Cameron & Reeve, 2006; Cappelli et al., 1999; Lerman et al., 1996). Neither perceived severity nor perceived barriers, which were significant in the bivariate analyses, were significant in the multivariate model. The lack of findings with respect to perceived severity and perceived barriers might be due to the population presenting at the clinic for risk assessment. Perhaps, individuals who have high perceived severity or perceived barriers to testing do not seek out a clinic for genetic counseling.

Although it is useful to understand the extent to which the predictors independently contribute to explaining actual testing decisions, entering all the predictor variables of interest simultaneously into a model does not allow us to address the issue of how these variables affect one another. To address this issue,

we focused on cognitive and affective aspects of the decision to test. There is evidence in the literature that both of these processes are important in motivating decisions and behaviours (Loewenstein et al., 2001; Mellers, Schwartz, & Ritov, 1999; Miller & Schnoll, 2000). Although evidence supporting the importance of cognitive appraisals has been abundant, the evidence for the role of emotions has received less attention. Findings from this study suggest that the importance of the various cognitive and affect components were not due to additive effects, but rather to the interaction between perceived susceptibility, perceived severity and worry. As such, the study findings point to the integration of worry or affect within the context of the HBM and support other cognitive-affective models, such as self-regulation (Leventhal et al., 1983), which have suggested the integration of affective responses to decision making and health behaviors.

The results of this study also provide a possible explanation for prior inconsistencies in the relevance of either cognitive appraisals or emotional responses in understanding behavior. Previous studies have typically not examined the interactions between these variables but rather have reached their conclusions based on either bivariate analyses or multivariate analyses that contain all the predictor variables in the model. If we followed the same approach, we would have concluded that perceived susceptibility and worry were not important in genetic testing decisions. Our findings lend support the recent cross-sectional findings by Shiloh and Ilan (2005), who also argue for the importance of examining interactions between psychosocial predictors of genetic testing decisions.

An important outcome of the present study was replicating an earlier finding of the inverse relationship between perceived severity of being a mutation carrier and decision to undergo genetic testing (Becker et al., 1975). There are several possible explanations for this outcome. First, the items measuring perceived severity would be considered by some researchers as measures of 'anticipated' emotions or expectations of *future* experiences such as anticipated regret or disappointment regarding a decision (Loewenstein et al., 2001). This is in contrast to 'anticipatory' emotions such as worry or anxiety, which are *immediate* and experienced by the individual at the time of decision. Anticipated emotions, such as perceived severity, are part of cognitive appraisal and may have implications both for how an individual perceives they can cope with the threat and for how they emotionally react to that threat. Thus, if high perceptions of severity resulted in reduced confidence in the ability to cope with the threat and increased worry about the threat, then it would be expected that these women would resort to coping with the emotional response to the stressor (i.e., by controlling their fears) and avoid the behaviour (e.g., genetic testing) in question (Leventhal, Leventhal, & Cameron, 2001). Further support for this reasoning comes from research in the area of stress and coping, which has postulated that individuals who do not perceive they can control a situation will resort to more emotion-focused strategies such as avoidance or denial in order to adapt to the stressor (Folkman et al., 1991; Lazarus & Folkman, 1984).

Differences in perceived severity of carrier status may be one explanation for the variability in uptake of genetic testing across disease conditions. Studies have found that the uptake of genetic testing for hereditary cancer syndromes has been greater than for Huntington's disease and have hypothesized that this is due to differences in the availability of treatment options (Marteau & Croyle, 1998). Testing positive for Huntington's disease would likely be perceived as more severe than testing positive for a hereditary cancer syndrome because of the lack of treatment options for the former. When little can be offered in terms of disease prevention or treatment, most people prefer not to know the information (Marteau & Croyle, 1998). Future research should examine contributors to severity perceptions, such as disease controllability or availability of treatment options, in efforts to understand why some individuals choose to forgo genetic testing.

The strengths of this study include the use of theory, the focus on actual testing decisions rather than intentions, and the focus on a clinic population that has not been previously involved in genetics research. However, the generalizability of the findings is limited to a predominantly Caucasian and highly educated group of women who were seeking counseling. Further research is needed to determine the extent to which the study findings can be generalized across various subgroups that were not well represented in the present context. In addition, because all study participants received counseling, this study cannot directly address the question of how genetic counseling may have influenced the psychosocial predictors of interest. Although HBM and worry items were also collected at one week following the genetic counseling session, problems with causal directionality would have occurred in the interpretation of pre–post findings because many women had their blood drawn for genetic testing during the initial visit. As such, assessment of psychosocial variables may have occurred after the women already decided to test and thus may not be reflective of predicting who undergoes genetic testing, but rather the attitudes women may have after making the decision to test. Concerns over response burden precluded all but the bare minimum of questions assessed at immediate follow-up on the day of the initial visit. Readers interested in how our randomized trial influenced some of the predictor variables of interest (i.e., worry) and genetic testing decisions are referred to Wang et al. (2005) for further details.

Another study limitation is the use of a single item to assess worry. However, our single item worry is consistent with several studies that utilize a single item to assess this construct (Diefenbach, Miller, & Daly, 1999; Gramling et al., 2005; Lerman, Kash, & Stefanek, 1994; Lipkus et al., 2000; Williams-Piehota, Pizarro, Schneider, Mowad, & Salovey, 2005). Moreover, our worry item focused on concerns over being a mutation carrier, and not breast cancer disease, to be consistent with how perceived susceptibility and severity were operationalized. We should also note that our single worry item was strongly correlated with the IES (intrusion subscale – focused on thoughts of family history of cancer) among the same group of women ($r = 0.44$, $p < 0.001$). However, IES at baseline was not strongly associated with genetic testing intentions ($r = 0.14$, $p = 0.06$) or actual

testing decisions ($r = 0.04$, ns), whereas worry about having a gene mutation was strongly correlated with genetic testing intentions ($r = 0.32$, $p < 0.001$) and showed a modest trend with actual testing decisions ($r = 13$, $p = 0.07$). Future studies should continue to expand upon existing measures of assessing worry about mutation carrier status (Kelly et al., 2004).

The results of this study may help to identify those women at greater risk for harm from genetic information (Pasacreta, 2003). For example, would women who had high perceptions of severity respond more adversely to receiving a positive genetic test result and thus benefit from additional psychosocial support and intervention? In contrast, are there women who avoid testing due to unfounded fears and perceived inability to cope with the information? Knowledge of the psychological processes involved in testing decisions may be used to inform and tailor the delivery of genetic counselling, which can have implications for counselling outcomes (Wang et al., 2004).

The advances in genetic technologies will continue to bring about new challenges and difficult decisions for patients and the general public regarding testing and treatment options. A better understanding of the factors that make these decisions difficult will hopefully enable researchers to find ways to better facilitate the decision making process.

Acknowledgments

Note

[1] Perceived severity, which is most often operationalized in the literature as severity of disease, is traditionally hypothesized to be positively associated with health behaviours. As perceived severity was reformulated in the present study to focus on severity of being a mutation carrier (i.e., severity of a risk factor), the hypothesized direction was reversed.

Appendix

HBM measures

The following asks you to rate the extent to which you agree or disagree with the statements provided (5-point Likert scale, strongly disagree–strongly agree).

Perceived susceptibility

1. It is likely I carry a gene mutation that increases my risk for breast cancer.
2. The chances that a gene mutation runs in my family are great.

Perceived severity

1. If I found out I carried a gene mutation, it would greatly disrupt my life.
2. Finding out I carried a gene mutation would be very difficult for me.
3. If I found out I carried a gene mutation, I would worry much more about developing breast cancer.

Perceived benefits

1. Genetic testing will help me learn about my children's risk for breast cancer.
2. Genetic testing to learn my risk will help other family members (sisters, daughters) decide whether to undergo testing.
3. Genetic testing will ease my mind, regardless of the test result.
4. Genetic testing will help me decide on the best course of action to take to deal with my cancer risk.
5. Genetic testing will help me reduce uncertainty about the future.
6. Genetic testing will help me make important life decisions (such as getting married, having children).
7. Genetic testing to learn about my risk will give me a sense of personal control.

Perceived barriers

1. I am afraid to undergo genetic testing because I may not be able to cope with the result.
2. I am afraid to undergo genetic testing because I do not understand what will be done.
3. Genetic testing will not help me because I would not do anything different to manage my cancer risk.
4. Genetic testing will have a negative impact on my family.
5. I cannot afford the cost for genetic testing.
6. My family will not be supportive if I undergo genetic testing.
7. Genetic testing will lead to unfair treatment of some people – that is, discrimination.
8. Genetic testing will not tell me anything new about my risk I do not already know.

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