Psychosocial factors associated with the public's willingness to pay for genetic testing for cancer risk: a structural equations model

Kwadwo Bosompra1, Takamaru Ashikaga1,2, Brian S. Flynn2–4, John K. Worden2–4 and Laura J. Solomon2,5

Abstract

An adaptation of Andersen’s behavioral model of health services utilization is used to examine the psychosocial and socio-demographic factors that directly and indirectly influence the likelihood of undergoing genetic susceptibility testing for cancer, and the amount of money that individuals would be willing to pay out-of-pocket for such a test. Apart from willingness and likelihood, the model also included perceived benefits and barriers, perceived susceptibility, dispositional optimism, information seeking, family history of cancer, socioeconomic status (SES), and age, and explained 30.3% of the variation in willingness. We found as hypothesized that likelihood of undergoing such tests was central to understanding willingness to pay. Being aware of genetic susceptibility testing for cancer, and talking and seeking information about it was directly associated with an increased chance of being willing to pay more, independent of other indirect associations (effects). Interventions targeting those with a family history of cancer and those with a higher SES should generate more awareness about the potential positive and negative consequences to one's family of testing, and the interface between family history of cancer and perceived susceptibility. Interventions should also motivate people to talk and seek more information about genetic testing for cancer risk to enable them take well-informed decisions.

Introduction

Genetic screening technology has developed dramatically in recent years, but very little is known about the public's willingness to pay for those tests and the psychosocial determinants of such willingness. The ‘willingness to pay’ construct has drawn increasing attention in the field of health economics where it has been used as a method of assessing, among other things, the value that individuals place on health care interventions [see, e.g. (Gafni, 1991, 1997; Miedzybrodzka et al., 1994, 1995; Donaldson et al., 1995, 1997; Dranitsaris, 1997)]. However, the focus has understandably been on economic factors, largely ignoring the potential impact of psychosocial factors. ‘Willingness to pay’ is used in the present context to refer to specific amounts of money that individuals would agree to pay out-of-pocket for a genetic test to assess their cancer risk.

Relative to genetic testing, studies about costs have usually focused on cost-effectiveness analyses of general population screening programs as compared to restricted screening among families at high risk for a specific disease [e.g. (Brown and Kessler, 1995; Coley et al., 1997)]. These studies too, do not address the psychosocial factors that may directly and indirectly influence willingness to pay for genetic testing to assess cancer risks among individuals with no known risk.

Cost(s), monetary or otherwise, has traditionally been conceptualized in health behavior models

1Department of Medical Biostatistics, 2Vermont Cancer Center, 3Office of Health Promotion Research, 4Department of Family Practice and 5Psychology Department, University of Vermont, Burlington, VT 05405, USA

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like the Health Belief Model (HBM) (Rosenstock, 1974; Strecher and Rosenstock, 1997) as a barrier to the performance of a behavior of interest and has been treated as an independent variable impacting that behavior. Studies on cancer screening applying the HBM perspective have consistently found cost(s) to be a major barrier to screening for breast cancer (Stein et al., 1992; Champion, 1994; Urban et al., 1994; Miller and Champion, 1997), colorectal cancer (Thomas and Clarke, 1998; Vernon, 1997) and cervical cancer (Burak and Meyer, 1997).

Thus there are virtually no studies that have examined the psychosocial determinants of out-of-pocket costs of genetic testing for cancer risk. In one of the few studies that examined out-of-pocket costs relative to genetic testing, 96% of 484 diagnostic mammography patients said they would accept a free genetic susceptibility test for breast cancer, but only 68% said they would pay more than $25 for such a test (Chaliki et al., 1995). Among a second group of 498 obstetrics/gynecology patients, 93% said they would take a free test, but only 53% said they would pay more than $25.

In bivariate analysis, willingness to pay more correlated positively with age, concern about getting breast cancer, perceived susceptibility to breast cancer, family history of breast cancer, belief that mammography effectively detects breast cancer in its early stages and belief that breast cancer was curable at that early stage (Chaliki et al., 1995). Although Chaliki and her colleagues did not examine the role of socioeconomic status (SES), the amount of $25 they used appears to be too low and might not accurately reflect the current level of costs for such tests [see, e.g. (Garber, 1999)].

Most people who do not have a family history of cancer and are at neither actual nor perceived high risk for cancer might not be at all ready or willing to pay for genetic testing, but at the same time it is conceivable that for-profit corporations might try to create felt needs. Some questions that arise then are which sections of the general population would be willing to buy the service? How much would people be willing to pay out-of-pocket for the service? How would various psychosocial characteristics impact individuals’ willingness to pay? The responses should be of value to public health practitioners planning interventions to help the public make well-informed and balanced decisions about whether or not to undergo genetic testing for cancer risk and how much to pay for such a test.

Thus we isolate the cost issue—amount of money that individuals are willing to pay—as our primary dependent variable and this is what sets our study apart. In particular, our study focuses on the psychosocial and sociodemographic factors that may directly and indirectly influence willingness to pay among individuals with no known risk for any specific cancers. Our theoretical framework is adapted from Andersen’s behavioral model of health services utilization (Andersen and Newman, 1973; Aday and Andersen, 1974; Andersen, 1995), a model that has been previously used in other cancer-related studies (Phillips et al., 1998; Glanz et al., 1999; Bosompra et al., 2000). The adapted model included measures derived from the HBM and the psychological construct of dispositional optimism. See Figure 1.

The primary outcome variable for the study is willingness to pay for genetic testing to assess cancer risk. The other variables are likelihood of undergoing genetic testing for cancer risk; perceived benefits of, and perceived barriers to, genetic testing for cancer risk; perceived susceptibility to cancer; dispositional optimism; information seeking about genetic testing for cancer risk; family history of cancer; age; and SES.

In a preliminary study, a number of these variables were employed in a structural equation model to examine the proximal and distal influences on the likelihood of undergoing genetic testing for cancer risk (Bosompra et al., 2000). It was found that while perceived benefits, perceived barriers, perceived susceptibility and pessimism might directly impact likelihood, they might also mediate the effects of age, SES, family history of cancer and awareness of cancer genetic susceptibility testing, on the likelihood of undergoing a genetic test. The present study represents an important
extension of the previous research by introducing willingness to pay as the primary dependent variable, and focusing on the proximal and distal psychosocial influences on the likelihood of undergoing a test and the amount of money that respondents are willing to pay for such a test. Additionally, we utilize an information-seeking construct based on an awareness question and two information-seeking behaviors about genetic testing for cancer risk in contrast to a simple awareness measure used previously. We also use an enhanced set of manifest variables to assess the likelihood of obtaining a genetic test for cancer risk. Finally, the operationalization of SES is enhanced as well.

**Literature review and hypotheses**

We assumed that after respondents have made a choice to undergo or not undergo testing, the next step would be to consider how much to pay for the test. Additionally, we expected all model constructs to impact willingness to pay only through likelihood (intention) of undergoing a test and we also expected the association between the two to be positive.

Our assumption that expressing a preference for testing or not testing precedes the issue of an acceptable level of payment is entirely consistent with economic theory (Deaton and Muellbauer, 1980; Laidler, 1981; Leftwich and Eckert, 1982; Hyman, 1986; Hirshleifer, 1988; Kreps, 1990; Browning and Zupan, 1999). Consumer theory describes consumer behavior in the context of ‘constrained optimization’. At the first stage, consumers choose an objective function, i.e. something to maximize (or minimize if it is a ‘bad’ as opposed to a ‘good’), and this is generally represented in the form of a ‘utility function’. At the second stage, consumers confront their objective function with the constraints, generally in terms of costs and a budget, and the optimal solution is the one that maximizes the objective function while remaining within the limits of the budget and the costs.

In the context of the present research, this means that consumers decide whether undergoing testing generates value or satisfaction to them. Then given that a consumer actually decides that testing ‘enters in his/her utility function’ (i.e. yields value or satisfaction) he/she will look into the costs of doing so. The potential of circularity in the relationship between choice and price should be noted since prices are determined by choice aggregated over many individual consumers and, simultaneously, prices constrain those choices (Kreps, 1990).
Previous research on likelihood of undergoing genetic testing for cancer risk has shown that likelihood is positively related to perceived benefits, perceived susceptibility and having a generally pessimistic outlook on life and negatively related to perceived barriers (Bosompra et al., 2000). Psychosocial research on breast cancer issues indicates that interest in obtaining a genetic test for breast cancer risk is positively associated with perceived likelihood of being a gene carrier and perceived risk of breast cancer (Lerman et al., 1994; Struewing et al., 1995; Jacobsen et al., 1997), and also with having positive attitudes towards genetic testing for breast cancer risk (Tambor et al., 1997). Additionally, high levels of interest in genetic testing for breast–ovarian cancer risk have been reported in first-degree relatives (FDRs) of breast–ovarian cancer patients (Lerman et al., 1994, 1995; Struewing et al., 1995). Similar research on genetic testing for colon cancer susceptibility confirm that perceived risk of colon cancer and having a family history of colon cancer are positively associated with interest in obtaining a test (Croyle and Lerman, 1993; Smith and Croyle, 1995; Lerman et al., 1996; Graham et al., 1998).

Results from other studies on genetic testing for general or breast cancer risk indicate that family history is positively related to perceived susceptibility, awareness and age (Mouchawar et al., 1999; Bosompra et al., 2000). Awareness, in turn, is related positively to income and education (Tambor et al., 1997; Hughes et al., 1997; Mogilner et al., 1998; Bosompra et al., 2000), while perceived susceptibility is negatively related to age (Bosompra et al., 2000).

Dispositional optimism refers to the generalized expectation that good versus bad outcomes will occur in one’s life (Scheier and Carver, 1985). The construct has been consistently found to be positively associated with health outcomes (Friedman et al., 1992; Scheier and Carver, 1992; Carver et al., 1993; Andersson et al., 1995; King et al., 1998; Fournier et al., 1999; Scheier et al., 1999; Bosompra et al., in preparation).

Evidence from previous research suggest that the dispositional optimism construct can sometimes produce a two-factor solution with ‘optimistic’ and ‘pessimistic’ items falling into separate groups (Scheier and Carver, 1985; Mook et al., 1992; Marshall et al., 1992; Lai, 1994; Fournier et al., 1999; Bosompra et al., 2000, in preparation). Bosompra et al. (Bosompra et al., 2000) reported such a two-factor solution with the optimism component being of less importance, and the pessimism component being significantly and positively related to susceptibility, perceived barriers and likelihood, and also showing a tendency to be positively associated with perceived benefits, although not statistically significant.

Based on the results of our preliminary analyses (Bosompra et al., 2000) and a review of the relevant literature, we surmised that the prospect of cancer may be perceived as threatening and orientations such as an optimistic versus pessimistic outlook on life may counter the threat. Consistent with the HBM, we expected that the individual’s perception of susceptibility to cancer and the benefits and barriers associated with undergoing testing would determine the likelihood that an individual would take a preventive action like undergoing a test in the face of the cancer threat and this would in turn influence his/her level of willingness to pay for the test. Additionally, we expected that having a family history of cancer, being aware of and seeking information about genetic testing for cancer risk, and sociodemographic factors like age and SES would indirectly influence willingness to pay by impacting optimism, pessimism, perceived susceptibility, benefits and barriers.

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**Methods**

**Study sample**

The target population consisted of adults (18–75 years) in Glens Falls and surrounding towns, an area in northern New York State. The area’s population was similar to that of the US with respect to median household income ($28 995 versus 30 056 in the US), median age (34 versus 33 years) and high school graduation rate (76
versus 75%). However, it was less educated in terms of proportion with 4 years of college or technical school (15 versus 20%), less ethnically diverse (97 versus 80% whites) and more rural (50 versus 25%) (CACI Marketing Systems, 1991).

The survey was conducted by the University of Vermont Biometry Facility between March and July 1996. Experienced interviewers were trained for 2 days using the survey instrument and the Ci-3 computer-assisted telephone interviewing system (Hutchinson, 1994). A systematic sample of about 4000 numbers was selected from residential listings in blocks using multiple random start techniques. Selected numbers were called at least eight times at different times and days of the week. When a number was verified as a household in the target study communities, an eligible adult was selected by a randomization method if more than one was available. A total of 681 interviews were completed with a 70% response rate among those eligible. Out of the 681 interviews, 59 respondents had a personal history of cancer and were dropped from the analysis since they were not asked about their perceived susceptibility to cancer. Thus the final sample size for the study was 622.

Measures
Introductory remarks were inserted in various sections of the survey instrument to provide respondents with the necessary background information for making informed responses to the questions. For instance, remarks preceding the question on awareness noted that:

The next section asks your opinion about some new types of medical tests. Doctors now can use a small sample of blood to look at patterns of genes inside a person’s cells. These tests provide information about a person’s chances of getting cancer in the future. These are called genetic tests for cancer risks.

More remarks were introduced in the section dealing with the benefits and barriers to testing.

Additionally, questions about likelihood of undergoing testing and willingness to pay for the test appeared in the latter half of the survey instrument long after questions dealing with information seeking, benefits, barriers and some health-related constructs not dealt with in this paper had been asked. Thus the structure and content of the survey instrument itself helped respondents to give well-informed responses.

Willingness to pay for a genetic test for cancer risk
The willingness to pay construct was defined with three items assessing respondents’ willingness to request a genetic test if it would cost them $500, $1000 and $1500, respectively. All three items had five-point Likert-type response scales ranging from ‘definitely no’ to ‘definitely yes’. Due to low frequencies at both ends of the scales, the two categories at either end were merged to obtain a three point response scale: ‘definitely or probably no’, ‘possibly’ and ‘definitely or probably yes’. A higher score implied a higher degree of willingness to pay for a genetic test to assess cancer risk.

Likelihood of undergoing genetic testing for cancer risk
The likelihood construct was measured with three items assessing the chances that a respondent would choose to undergo a genetic test for cancer risk if it were free of charge, undergo a test over the next 6 months if it were available and undergo a test if it were recommended by their physician. All items had five-point Likert-type response scales ranging from ‘definitely no’ to ‘definitely yes’. A higher score meant the respondent indicated a greater likelihood of choosing to undergo a genetic test for cancer risk.

Benefits of and barriers to genetic testing for cancer risk
The initial benefits and barriers constructs were measured with a set of 20 items assessing utilitarian advantages for self, self approval and their parallel ‘disadvantages’ categories which we labeled the ‘self’ perspective, and a second set of 12 items assessing utilitarian advantages for others, social approval and their corresponding ‘disadvantages’
categories, labeled the ‘other’ perspective (Janis and Mann, 1977). The ‘disadvantages’ categories reflected mainly concerns about family members’ reaction, potential employment or insurance discrimination and the inability of genetic testing to prevent cancer if the test result was positive. None of the items specified the monetary cost or dollar amount that an individual might have to pay and whether such an amount was high or low.

In preliminary analyses not reported in this paper, the ‘other’ perspective \( \chi^2 (9) = 11.11, P = 0.268; \text{BBNNFI} = 0.990; \text{CFI} = 0.994 \) had stronger fit indices than the ‘self’ perspective \( \chi^2 (9) = 31.02, P < 0.001; \text{BBNNFI} = 0.898; \text{CFI} = 0.939 \) and was therefore selected to represent the consequences of genetic testing. The construct consisted of three positive and three negative consequences to one’s family of undergoing genetic testing for cancer risk. The former reflected respondents’ perceptions of the positive impact of genetic testing on their families and was labeled ‘benefits’, while the latter reflected the negative impact and was labeled ‘barriers’.

Perceived susceptibility to cancer
Perceived susceptibility was assessed with one item about what a respondent considered to be his/her chance of getting cancer in his/her lifetime. The question was: ‘On a scale of 1 to 10, where 1 means “not at all” and 10 means “extremely likely”, how likely are you to get cancer in your lifetime?’. A higher score indicated a higher level of perceived susceptibility to cancer.

Dispositional optimism
A two-factor solution (reflecting optimism and pessimism) from the eight-item Life Orientation Test (LOT) (Scheier and Carver, 1985) was used in this study. Items utilized a five-point Likert-type response scale ranging from strongly disagree to strongly agree. A higher score on the optimism or pessimism construct indicated that a respondent had greater optimism or pessimism, respectively.

Information seeking about genetic testing for cancer risk
This was measured by three items assessing whether respondents had ever heard about genetic testing for cancer risk, engaged in interpersonal communication about it (with family, friends, doctors and others), and sought information about it from the mass media and other sources. All three responses were recorded as yes or no. The measure could thus be seen as ranging from a low ‘passive’ level of never or merely hearing about it to a higher ‘active’ level of seeking information.

Family history of cancer
Family history of cancer was assessed by asking respondents to indicate with a yes or no whether any of their FDRs (parents, brothers, sisters or children) had been diagnosed with cancer. A higher score corresponded to a greater number of family members with a cancer history.

SES
This was assessed with two indicator variables: educational attainment and gross household income. Educational attainment was operationalized as the highest level of education completed and was subdivided into four categories: not high school graduate, high school graduate, some college or technical school and college graduate or higher. Gross household income was measured by asking respondents to sum up the 1995 pre-tax income of all household members and indicating whether they fell into one of four pre-specified categories: less than $10 000, between $10 000 and $25 000, greater than $25 000 but not exceeding $40 000, and greater than $40 000.

Age
Age at last birthday was recorded, and categorized into 18–35 years, over 35 but less than 50 years and 50–75 years to obtain roughly equal groupings.

Data analysis
The EQS structural equation modeling (SEM) program, version 5.5a (Bentler, 1993; Bentler and Wu, 1995) was used to analyze the data. In all cases the maximum likelihood estimation option with the covariance matrix was used. The robust option (Bentler, 1993) was also utilized to examine the possible impact of any distributional problems. For most variables, missing data were present in
less than 10% of the total number of cases and a univariate modal substitution method was used. However, a linear regression substitution was utilized in the case of perceived susceptibility because it was found to correlate highly with the indicator variables for family history of cancer (Afifi and Elashoff, 1969).

Each measurement model was explored and validated using factor analysis methods with a direct oblimin rotation and in all cases except for family history of cancer, only those items that had a final factor loading $\geq 0.4$ were retained. Since we defined family history of cancer with reference to FDRs, all four items in the measurement model for that construct were retained irrespective of the magnitude of their factor loadings.

The SEM approach was employed because most quantitative psychosocial research on genetic testing for cancer risk has utilized general linear models (Lerman et al., 1994, 1995; Smith and Croyle, 1995; Struwing et al., 1995; Andrykowski et al., 1997; Jacobsen et al., 1997; Tambor et al., 1997; Codori et al., 1999; Dury et al., 1999; Glanz et al., 1999; Petersen et al., 1999; Vernon et al., 1999) but that approach limits any examination of the influences of direct and mediating factors and also assumes that the measures were obtained without error. Using SEM overcomes these limitations by enabling the simultaneous analysis of direct and indirect effects of latent and manifest variables, and thus specifying the processes by which the primary outcome variable is influenced by the various psychosocial and socio-demographic factors of interest to the study.

Model fit was ascertained using the $\chi^2$ goodness of fit test ($\chi^2$ reported with its corresponding degrees of freedom in brackets), the comparative fit index (CFI) (Bentler, 1990), the Bentler–Bonnet non-normed fit index (BBNNFI) (Bentler, 1990) and the root-mean-square error of approximation index (RMSEA) (Jöreskog and Sörbom, 1993). A CFI/BBNNFI score $\geq 0.90$ was considered an adequate fit (Bentler, 1992; Crowley and Fan, 1997; Musil et al., 1998). RMSEA values less than 0.05 indicated an excellent or good fit, between 0.05 and 0.08, a moderate fit and from 0.08 to 0.10, a fair fit (Musil et al., 1998; Schumacker and Lomax, 1996). Overall model parsimony was achieved by the use of Lagrange multiplier statistics. The final SEM was characterized by examination of direct and indirect effects of upstream explanatory factors relative to amount of money that individuals would be willing to pay to obtain genetic tests to detect cancer risks.

**Results**

**Summary characteristics**

Women constituted 59% of the sample, 40% of respondents were at least 50 years old, 57% had family incomes of $40 000 or less and 52% had at least some college or technical education (Table I). Family history of cancer appeared to be related to generation; among FDRs who had ever had cancer, parents were mentioned more often (38%) than siblings (7% of brothers and 8% of sisters) or children (1.1%). Over half of all respondents had ever heard about genetic testing for cancer risk, 20% had ever engaged in interpersonal communication about the topic, while 14.0% had sought information from various sources.

Respondents tended to be more optimistic than pessimistic expressing agreement with the optimistic statements more often than the pessimistic ones. They also tended to endorse the benefits of testing more often than the barriers. The majority of respondents did not perceive themselves to be at risk for cancer. On a scale of 1–10, where 1 meant ‘not at all likely’ and 10 meant ‘extremely likely’, over 40% of respondents classified themselves in the 1–3 range as compared to only 8% in the 8–10 range, while 28% placed themselves at the perceived mid-point (5) of the scale.

Over 50% said it was possibly, probably or definitely likely that they would undergo genetic testing for cancer risk within the next 6 months if it were available; almost 90% would do so if their doctor recommended it and almost 80% would do so if it were free of charge. However, most respondents were not willing to pay high dollar amounts for testing. About 29, 15 and 12% said...
<table>
<thead>
<tr>
<th>Item descriptor</th>
<th>Score (%)</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Age</strong></td>
<td></td>
</tr>
<tr>
<td>18–35 years</td>
<td>25.1</td>
</tr>
<tr>
<td>36–49 years</td>
<td>34.7</td>
</tr>
<tr>
<td>50–75 years</td>
<td>40.2</td>
</tr>
<tr>
<td><strong>Education</strong></td>
<td></td>
</tr>
<tr>
<td>&lt; High school grad</td>
<td>8.0</td>
</tr>
<tr>
<td>High school grad</td>
<td>40.0</td>
</tr>
<tr>
<td>Some college</td>
<td>28.8</td>
</tr>
<tr>
<td>College grad and up</td>
<td>23.2</td>
</tr>
<tr>
<td><strong>Income</strong></td>
<td></td>
</tr>
<tr>
<td>&lt;$10000</td>
<td>5.5</td>
</tr>
<tr>
<td>$10,000–25,000</td>
<td>23.6</td>
</tr>
<tr>
<td>$25,000–40,000</td>
<td>27.5</td>
</tr>
<tr>
<td>&gt;$40,000</td>
<td>43.4</td>
</tr>
<tr>
<td><strong>Family history of cancer</strong></td>
<td></td>
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<tr>
<td>Parents</td>
<td>38.3</td>
</tr>
<tr>
<td>Brothers</td>
<td>6.8</td>
</tr>
<tr>
<td>Sisters</td>
<td>8.0</td>
</tr>
<tr>
<td>Children</td>
<td>1.1</td>
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<tr>
<td><strong>Information seeking about genetic testing for cancer risk</strong></td>
<td></td>
</tr>
<tr>
<td>Heard</td>
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<td>Discussed</td>
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<tr>
<td>Looked for information</td>
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<td><strong>Optimism</strong></td>
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<tr>
<td>Disagree</td>
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<td>Neutral</td>
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</tr>
<tr>
<td>Agree</td>
<td>58.7</td>
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<tr>
<td>Strongly agree</td>
<td>14.5</td>
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<tr>
<td><strong>Pessimism</strong></td>
<td></td>
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<tr>
<td>Strongly disagree</td>
<td>8.2</td>
</tr>
<tr>
<td>Disagree</td>
<td>49.8</td>
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<tr>
<td>Neutral</td>
<td>11.6</td>
</tr>
<tr>
<td>Agree</td>
<td>25.2</td>
</tr>
<tr>
<td>Strongly agree</td>
<td>5.1</td>
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<tr>
<td><strong>Perceived Susceptibility:</strong></td>
<td></td>
</tr>
<tr>
<td>1</td>
<td>17.1</td>
</tr>
<tr>
<td>2</td>
<td>8.8</td>
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<tr>
<td>3</td>
<td>15.0</td>
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<td>0.7</td>
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<tr>
<td>9</td>
<td>1.9</td>
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<tr>
<td><strong>Perceived benefits of genetic testing for cancer risk</strong></td>
<td></td>
</tr>
<tr>
<td>Give useful information to family about their cancer risk</td>
<td>94.9</td>
</tr>
<tr>
<td>Help family make better decisions about health care</td>
<td>93.4</td>
</tr>
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<td>Help family decide whether to get tested themselves</td>
<td>95.0</td>
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<td><strong>Perceived barriers to genetic testing for cancer risk</strong></td>
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<tr>
<td>Family might have trouble getting health insurance coverage</td>
<td>63.5</td>
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<tr>
<td>Might have large financial impact on family</td>
<td>69.9</td>
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<tr>
<td>Family might get upset with me</td>
<td>21.1</td>
</tr>
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Table I. Continued

<table>
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<tr>
<th>Likelihood of undergoing genetic testing for cancer risk</th>
<th>Definitely no</th>
<th>Probably no</th>
<th>Possibly yes</th>
<th>Probably yes</th>
<th>Definitely yes</th>
</tr>
</thead>
<tbody>
<tr>
<td>If genetic testing were available to you now, would you be likely to get it the next 6 months?</td>
<td>19.6</td>
<td>29.3</td>
<td>31.0</td>
<td>12.2</td>
<td>7.9</td>
</tr>
<tr>
<td>How likely would you be to agree to genetic testing for cancer risk if your doctor recommended it?</td>
<td>4.8</td>
<td>7.4</td>
<td>25.9</td>
<td>32.3</td>
<td>29.6</td>
</tr>
<tr>
<td>How likely would you be to ask for a test if it were done free of charge?</td>
<td>6.4</td>
<td>15.1</td>
<td>22.2</td>
<td>25.4</td>
<td>30.9</td>
</tr>
</tbody>
</table>

Willingness to pay for genetic testing for cancer risk

<table>
<thead>
<tr>
<th>Willingness to pay for genetic testing for cancer risk</th>
<th>Probably or definitely no</th>
<th>Possibly or definitely no</th>
</tr>
</thead>
<tbody>
<tr>
<td>How likely would you be to ask for a test if it cost you $500?</td>
<td>70.6</td>
<td>20.6</td>
</tr>
<tr>
<td>How likely would you be to ask for a test if it cost you $1000?</td>
<td>85.5</td>
<td>11.3</td>
</tr>
<tr>
<td>How likely would you be to ask for a test if it cost you $1500?</td>
<td>88.1</td>
<td>9.3</td>
</tr>
</tbody>
</table>

they would possibly, probably or definitely be willing to pay $500, $1000 and $1500, respectively, for the test.

Empirical model: total, direct and indirect effects

The initial model which hypothesized that all influences on willingness to pay for genetic testing will occur via likelihood of undergoing testing, fit the data fairly well. Although the $\chi^2$ statistic was significant [$\chi^2 (411) = 787.93, P < 0.0001$] this was not unexpected given the large sample size [see (Crowley and Fan, 1997), p. 515]. The other fit statistics were adequate (BBNNFI = 0.913; CFI = 0.923; RMSEA = 0.038) and the model explained 19.9% of the variance in willingness to pay for genetic testing for cancer risk. However, some hypothesized direct paths were not statistically significant. Therefore, to improve model parsimony we used the Wald multivariate tests for dropping variables to fix those paths to zero. The dropped paths were: from optimism to each of benefits, barriers and likelihood; from susceptibility to benefits and barriers; from pessimism to benefits; and from age to SES.

Additionally, the Lagrange multiplier test recommended introducing a direct path from information seeking to willingness to pay, perhaps reflecting the importance of information seeking regarding the monetary costs of genetic testing. The improvement in fit between the two models was statistically significant [$\chi^2 (5) = 44.21, P < 0.0001$]. The $\chi^2$ statistic remained significant for the final model [$\chi^2 (416) = 743.72, P < 0.0001$] and the other fit indices were adequate (BBNNFI = 0.925, CFI = 0.933; RMSEA = 0.036) while all remaining direct paths were significant. In addition, the reduced model accounted for 30.3% of the variance in willingness to pay as compared to the 19.9% of the initial model.

Since some of the manifest variables showed signs of being non-normally distributed, the final model was re-ran using maximum likelihood robust estimators (Bentler, 1993) and fairly similar indices were obtained [Satorra-Bentler scaled $\chi^2 (416) = 725.25, P < 0.0001$; Robust CFI = 0.927]. More importantly, all significant paths and effects remained significant and there were no substantive changes in the results. Thus only the maximum likelihood estimates are presented in this paper. See Figure 2.

The decomposition of total effects (i.e. direct and indirect effects) is presented in Table II. The study being reported here is not longitudinal but cross-sectional in design, and although the discussion of direct and indirect ‘effects’ might appear to suggest causal relationships, it should be noted that the relationships are correlational. The results indicated that willingness to pay $500, $1000 or $1500 for the test was most strongly and positively
influenced by likelihood of undergoing genetic testing ($\beta = 0.444$). The perception that one’s family stood to benefit from the decision to undergo testing (perceived benefits) indirectly increased the chances ($\beta = 0.187$) that one would be willing to pay by directly increasing the likelihood of undergoing testing ($\beta = 0.421$). Similarly, perceived susceptibility indirectly increased the chances that one would be willing to pay ($\beta = 0.075$) by directly increasing the likelihood of undergoing testing ($\beta = 0.169$). On the other hand, the perception that one’s family would be negatively impacted by such a decision (perceived barriers) indirectly reduced the chances that one would be willing to pay ($\beta = -0.155$) by directly reducing the likelihood of undergoing testing ($\beta = -0.349$).

Pessimism directly increased the likelihood of undergoing testing and also increased it indirectly via a higher perception of susceptibility, but these
### Table II. Decomposition of total effects for final model using standardized coefficients ($\beta$)

<table>
<thead>
<tr>
<th>Variables</th>
<th>Behavioral variables</th>
<th>Mediating variables</th>
<th>Background variables</th>
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<tbody>
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<td></td>
<td></td>
<td>Information seeking</td>
<td>Optimism</td>
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<tr>
<td>SES</td>
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<tr>
<td>Family history of cancer</td>
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<tr>
<td>Information seeking</td>
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<td>Optimism</td>
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<td>Pessimism</td>
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<td>Perceived susceptibility</td>
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<td>Perceived barriers</td>
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<td>Likelihood</td>
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<tr>
<td>Willingness to pay for testing</td>
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</table>

For each variable in the first column, the standardized coefficients ($\beta$) for the total effects model appear along the same row. Thus model equations are read horizontally and model effects are read vertically, e.g. for the third row, the standardized solution for the total effects of Information seeking is: Information seeking $= 0.173 \times$ Age $+ 0.565 \times$ SES $+ 0.358 \times$ Family history. To examine the total effects of perceived barriers, read down column 10 as: barriers had negative total effects on both likelihood ($\beta = -0.349$) and willingness ($\beta = -0.155$). Direct and indirect effects can be determined with reference to Figure 2. A dash (−) means there are no direct or indirect paths linking the corresponding variables. Since the coefficients are standardized and reflect both direct and indirect effects, no probability levels are indicated. Coefficients with (a) indicate that the corresponding unstandardized coefficients ($\hat{\beta}$) for indirect effects were statistically significant and (b) implies corresponding unstandardized coefficients for structural and measurement equations (direct effects) were statistically significant, e.g. age had direct and indirect effects on perceived susceptibility, both of which were statistically significant.
effects appear to have been counteracted by pessimism’s direct effect on perceived barriers ($\beta = 0.347$) which in turn reduced the likelihood of undergoing testing. Although pessimism’s total effect on likelihood ($\beta = 0.088$) was statistically significant, its overall effect on willingness to pay ($\beta = 0.039$) was not. Optimism, on the other hand, reduced the perception of susceptibility to cancer ($\beta = -0.235$) which in turn led to a reduced likelihood of undergoing testing ($\beta = -0.040$) and thus reduced the chances of one being willing to pay for the test ($\beta = -0.018$).

Apart from likelihood, information seeking had the strongest effect, both direct and total, on willingness to pay. Information seeking directly influenced perceived susceptibility ($\beta = 0.327$), optimism ($\beta = 0.445$) and pessimism ($\beta = -0.335$), and through them also impacted perceived barriers and likelihood, but their effects seemed to have cancelled out one another since the total effect on likelihood was only marginal ($\beta = 0.008$) and their ultimate indirect effect on willingness was minimal ($\beta = 0.004$). It was the direct effect of information seeking on willingness ($\beta = 0.323$) that made all the difference and increased the total effect ($\beta = 0.327$).

Additionally, having a family history of cancer indirectly increased the likelihood of undergoing testing by increasing information seeking ($\beta = 0.358$) and perceived susceptibility ($\beta = 0.461$), and also reducing perceived barriers ($\beta = -0.042$). Simultaneously, it affected likelihood in the opposite direction (reduction) by increasing optimism ($\beta = 0.159$) and reducing pessimism ($\beta = -0.120$). The net effect of these contending impacts was a positive and statistically significant effect on likelihood ($\beta = 0.070$), and an even stronger positive effect on willingness to pay for genetic testing for cancer risk ($\beta = 0.147$).

Similarly, SES had a significant indirect effect on willingness ($\beta = 0.169$) by increasing information seeking ($\beta = 0.565$) and perceived susceptibility ($\beta = 0.043$) while reducing perceived barriers ($\beta = -0.202$), on one hand, and increasing optimism ($\beta = 0.251$) and reducing pessimism ($\beta = -0.581$), on the other. Age had a positive total effect on willingness to pay ($\beta = 0.043$) but this was not statistically significant. However, it had a significant positive impact on family history ($\beta = 0.484$), and significant negative impacts on perceived susceptibility ($\beta = -0.154$) and likelihood ($\beta = -0.030$). Perceived benefits of testing indirectly impacted willingness ($\beta = 0.187$) by directly affecting likelihood ($\beta = 0.421$), although it was itself not influenced by any other factor.

Thus, overall, the strongest effects on willingness to pay were, in descending order of magnitude, likelihood of undergoing testing ($\beta = 0.444$), information seeking ($\beta = 0.327$), perceived benefits ($\beta = 0.187$), SES ($\beta = 0.169$), perceived barriers ($\beta = -0.155$), family history of cancer ($\beta = 0.147$), perceived susceptibility ($\beta = 0.075$) and optimism ($\beta = -0.018$). Neither the direct or indirect effects of age and pessimism on willingness were statistically significant.

**Discussion**

In evaluating the results of our model, some qualifications should be noted. As acknowledged in the previous section, the data and resulting structural equation model are from a cross-sectional sample and therefore the ‘effects’ are not necessarily causal. Secondly, formal testing of relationships would require manipulations directed at specific paths and comparison with a similar group of subjects not exposed. Thirdly, the respondents were drawn from a rural population with little minority representation—future studies need to extend the results to larger and more diverse populations.

In relation to the test itself, there is currently no single, global ‘genetic test for cancer’ that anyone could undergo and pay for. Rather, each test has a different cost and can only look for one mutation at a time with different mutations having different implications for cancer risk. Thus the study essentially asked a hypothetical question about a hypothetical test for general cancer risk as has been done in previous studies [e.g. (Hietala et al., 1995; Andrykowski et al., 1996, 1997; Aro et al., 1997)].

Also, the ‘willingness to pay’ construct could
Willingness to pay

have been better operationalized by asking respondents to indicate the specific amounts they would be willing to pay for the test. Additionally, perceived susceptibility was assessed with only one item and this might not have captured the construct fully. It would have been helpful to include other indicator variables like perceived susceptibility relative to peers or with reference to a particular time span. However, perceived susceptibility was significantly associated with both willingness (indirect effect) and likelihood, and could therefore be said to have proved adequate for the model’s purposes.

Forty-five percent of respondents indicated that they had not heard about genetic testing for cancer risk, but this may not necessarily imply that genetics is an entirely new area. Media coverage of new developments like the human genome project and genetic engineering activities related to food production and animal husbandry have all contributed to raise public consciousness about genetic testing in general. Although knowledge about genetic testing for cancer risk may be specific in nature, the survey questions about the pros and cons of genetic testing and other topics were quite general and respondents could conceivably use their knowledge of the broad field of genetics to address those questions.

This paper does not take a position for or against genetic testing for cancer risk in the general population. Rather it examines the proximal and distal psychosocial influences on willingness to pay money out-of-pocket for such a test. Taking note of this caveat, the observed results confirm that likelihood of undergoing testing is central to willingness to pay for the test. Therefore factors impacting likelihood need to be seriously studied to ensure the success of any future public education efforts.

Perceived benefits, perceived barriers, perceived susceptibility and pessimism had significant direct effects on likelihood, while pessimism, optimism, family history of cancer and age had significant indirect effects. These results suggest that the potential effects of testing on one’s family, including both positive and negative consequences, should be clearly explained to the public. Perceptions of susceptibility should also be addressed by, among other things, explaining how exactly having a family history of cancer puts one at risk for getting cancer. Since optimism and pessimism had, respectively, negative and positive direct effects on susceptibility, it would be useful to integrate programs aimed at enhancing optimism and reducing pessimism into any interventions that target perceived susceptibility.

Next in importance to likelihood was information seeking. Being aware of genetic testing for cancer risk, talking to family, friends, personal physician and others about it, looking for information about it from the mass media and other sources substantially and directly increased chances of being willing to pay for genetic testing. This is due to the impact of factors antecedent to information seeking in our model, specifically having a family history of cancer and having a higher SES. Both factors had total effects of increasing the chances of willingness to pay but more importantly they both had strong, positive and direct effects on information seeking.

The results appear to suggest that people with a family history of cancer are more likely to be aware of cancer; to talk to family, friends, family physicians and others about it; and more likely to look for information about it. Ultimately all of these tend to make them more willing to pay for the test independent of any indirect effects through other factors in the model. Being of a higher SES increases the chances of a higher level of willingness, which in itself is not surprising: one has to have the necessary resources to activate any level of willingness.

Age had positive and negative associations respectively with family history of cancer and perceived susceptibility, while family history of cancer had a positive association with susceptibility. This could be due to older respondents having had more ‘opportunities’ to know more relatives who developed various cancers and also younger respondents not having any children who would have developed cancer. Similarly, it may be reason- able to infer that older individuals who have not
contracted cancer may tend to believe that they are less susceptible to it. Also, those with more extensive family histories of cancer tended to believe they were more susceptible to cancer.

An important aspect of the marketing objectives of for-profit corporations is to know how much individuals are willing to pay for a service at the point of consumption (Gafni, 1991). It thus seems that due to the greater chance that they would be willing to pay $500, $1000 or $1500 for a genetic test for cancer risk, people with a family history of cancer and those with higher SES are more likely to be targeted by for-profit corporations. Those with a family history of cancer may have a genuine concern and public health interventions targeting them should stress all the specific details about why having a family history of cancer is a risk factor for cancer. Interventions targeting higher SES individuals should provide information on the importance of having or not having a family history of cancer and also stress the potential negative consequences to one’s family of undergoing genetic testing.

From another perspective, the model suggests that for-profit corporations aiming to increase likelihood of undergoing genetic testing for cancer risk and willingness to pay higher amounts for such tests could seek to minimize inhibitions about family reactions to testing, trumpet the benefits to one’s family of testing and fan higher perceptions of susceptibility. They would focus on those with a family history of cancer and those with a higher SES, and provide more information on the benefits and barriers. Public health activists and their allies would need to take note of this possibility.

Noting the numerous unresolved psychosocial and ethical issues relating to whether to undergo genetic testing for cancer risk (Codori, 1997; Croyle et al., 1997), it is obvious that this is not a ‘normal’ health education situation of increasing or decreasing the performance of a behavior. Rather the role of health education should be to try to help people arrive at the best possible decision for themselves in an ethically unbiased manner. At the minimum, it should generate more awareness about the potential positive and negative consequences to one’s family of undergoing these tests, specify details about how a family history of cancer factors into the equation and motivate people to seek more information about genetic testing for cancer risk.

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