

Brief Assessment of Parents' Attitudes Toward Testing Minor Children for Hereditary Breast/Ovarian Cancer Genes: Development and Validation of the Pediatric *BRCA1/2* Testing Attitudes Scale (P-TAS)

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Objective Predictive genetic testing for hereditary breast/ovarian cancer risk (*BRCA1/2* testing) is not recommended for minor children due to its lack of immediate medical benefit and potential psychological risk. Yet, tested mothers are often interested in learning about their children's cancer risks via pediatric *BRCA1/2* testing, raising a host of bioethical concerns. However, no reliable or valid tool exists to formally gauge parents' interest in such testing. The aim of this study was to develop and evaluate a new measure for use in genetic research and consultation, known as the Pediatric *BRCA1/2* Testing Attitudes Scale (P-TAS). **Methods** After pretest genetic counseling and provision of a blood sample for *BRCA1/2* testing, the P-TAS was administered to 187 mothers of children between 8- and 21-years-old. The measure was also given to 96 of the mothers' nontested co-parents. Analyses of the factor structure and psychometric properties of the measure were performed in mothers and confirmed in their co-parents. **Results** The two factors of the P-TAS, labeled Attitudes and Beliefs (Factor 1) and Decision Making and Communication (Factor 2), accounted for 62.9% of the variance and were reliable (Cronbach's coefficient α s = .70 and .90, respectively); the structure and properties were largely confirmed among co-parents. Validity was indicated through its convergence with related constructs. **Conclusions** This new tool may be integrated into genetic counseling research to better assess parents' attitudes and interests in pediatric *BRCA1/2* testing. Such information may help guide ongoing discussions about the appropriateness of testing in adolescent or young adult children.

Key words bioethics; children; counseling; genetic cancer; genetic counseling; genetic testing; parents.

The completion of the Human Genome Project is ushering in a new era of preventive medicine and the number of predictive genetic tests available for common adult-onset conditions is growing (Collins, Green, Guttmacher, & Guyer, 2003). To date, most predictive genetic testing for such conditions occurs in the area of cancer susceptibility—subsequent to the cloning of the two major breast cancer susceptibility genes, *BRCA1* and *BRCA2* (*BRCA1/2*), in the mid 1990s (Miki et al., 1994; Tavtigian et al., 1996). At the same time, cancer genetic counseling is diffusing into mainstream care and it is estimated that over 240,000 individuals have undergone cancer susceptibility

testing, primarily for mutations in *BRCA1/2* (Myriad Genetics, 2007).

One of the primary reasons that women, and mothers in particular, seek predictive genetic testing for hereditary breast/ovarian cancer risk (*BRCA1/2* testing) is to learn if they may pass along cancer-predisposing gene mutations to their offspring (Lerman et al., 1996; Lynch et al., 1997; Patenaude et al., 2006). Biological children of mothers with *BRCA1/2* mutations have a 50% chance of inheriting these mutations. Thus, female children with a mutation have very elevated lifetime risks of developing hereditary breast and/or ovarian cancer (Chen & Parmigiani, 2007).

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Journal of Pediatric Psychology 34(6) pp. 627–638, 2009

doi:10.1093/jpepsy/jsn033

Advance Access publication April 1, 2008

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Males may also be carriers of *BRCA1/2* mutations and experience increased breast and prostate cancer risks over their lifetimes (Liede, Karlan, & Narod, 2004).

The benefits and risks of testing minor-age children have been extensively debated in the medical literature (Clarke et al., 2005; Cohen, 1998; Duncan & Delatycki, 2006; Elger & Harding, 2000; Pelias, 2006; Robertson & Savulescu, 2001; Ross & Moon, 2000). The primary objection to offering predictive *BRCA1/2* testing to minors is based on the lack of medical benefit in childhood; moreover, there are no clinically accepted medical management options to reduce the likelihood of children developing *BRCA1/2*-linked cancers in adulthood (American Society of Clinical Oncology, 2003; American Society of Human Genetics, American College of Medical Genetics, 1995; Collins, 1996). Nevertheless, it has been suggested that mature minors in particular may desire predictive testing information to inform future decisions about preventive health behaviors and childbearing (Bradbury et al., 2008). By fostering more autonomous decision making, psychological benefits to the child and family unit may be obtained—in part related to the reduction of uncertainty about risk (Bradbury et al., 2008; Robertson & Savulescu, 2001). It has also been argued that preservation of parental authority over decision making is an important consideration as parents are well-poised to gauge the individual benefits and risks of testing to the child (Robertson & Savulescu, 2001). However, despite these potential benefits, there is concern that children may later regret the decision to be tested (i.e., that their autonomy may have been compromised owing to their relative immaturity) and that psychological harm including negative effects on mood, self-esteem, and family relationships may occur (Bradbury et al., 2008; Cohen 1998; Robertson & Savulescu, 2001). In part because of the lack of empirical data to support the benefits or harms of testing, it has been argued that case by case decisions to pursue testing in minors may be appropriate, including an assessment of the child's maturity and understanding of the potential benefits, limitations, and risks associated with predictive genetic testing (American Society of Human Genetics, American College of Medical Genetics, 1995; Borry, Stultiens, Nys, Cassiman, & Dierickx, 2006; Bradbury et al., 2008; Elger & Harding, 2000; Pelias, 2006; Rhodes, 2006).

The aforementioned issues are not suddenly resolved when a child turns age 18, as evidenced by recent case reports (Gaff, Lynch, & Spencer, 2006). It is apparent that, in certain circumstances, medical professionals would be willing to provide predictive genetic testing for

adult-onset disorders to minor children (Borry, Goffin, Nys, & Dierickx, 2008; Campbell & Ross, 2003; Duncan, Savulescu, Gillam, Williamson, & Delatycki, 2005). A survey of clinical geneticists from five nations revealed that 49 adolescents have been tested for adult-onset conditions (including three children age 14 or older who were tested for *BRCA1/2* mutations) (Duncan et al., 2005). Another recent survey of European clinical geneticists revealed that 71% of respondents had provided *BRCA1/2* counseling to a minor younger than age 16, and two geneticists had provided counseling and testing (Borry et al., 2008). Such counseling was provided to older adolescents only, as 96% of study respondents were unwilling to provide *BRCA1/2* testing to a 6-year-old child (Borry et al., 2008). Moreover, respondents were more in favor of providing the testing if the request was made by an older minor child in conjunction with the parents (Borry et al., 2008). Perhaps because of the ethical debate surrounding this issue, published reports of children being tested for *BRCA1/2* mutations remain scarce and no case reports exist to our knowledge. From an empirical standpoint, very little data are available on which to evaluate the validity of these concerns, furthering the lack of clarity on these issues.

Parental requests for predictive genetic testing may be made in response to the child's own interest and inquiry. We (Tercyak et al., 2001a; Tercyak, Peshkin, DeMarco, Brogan, & Lerman, 2002) and others (Hughes et al., 1999; McGivern et al., 2004; Patenaude et al., 2006) have previously noted that at least one-half of mothers who participate *BRCA1/2* testing inform their minor-age children of parents' test results; girls and older children are among the most likely to be informed. The decision to disclose parental test results to children is positively associated with mothers' interest in testing minor children for *BRCA1/2* mutations (Tercyak et al., 2002). This interest may be transmitted to their children, especially among older adolescents. Some children informed of their mothers' *BRCA1/2* test results are prone to worry that cancer may happen to them someday and are interested in knowing about their adult risks for developing cancer—largely so they can take preventive measures later on (Tercyak, Peshkin, Streisand, & Lerman, 2001b). However, in the current climate these youngsters are discouraged from pursuing this opportunity.

The impact on mothers of disclosing *BRCA1/2* test results to children is largely unknown, but it is relevant to consider as the associated feelings of guilt after testing positive or relief after testing negative may influence women's choices about communicating with children.

These feelings have been shown to become intensified as results are communicated with adult family members (van Oostrom et al., 2006b). In addition to reporting feelings of guilt, adults who receive positive *BRCA1/2* test results also report that they worry about their children developing cancer (Lynch et al., 2006). These worries may affect mothers' desires to share information about their test results with children, and may cause them to consider having their children tested sooner rather than later. Disclosure to children may be instrumental in both the short- and long-term psychological adjustment of mothers and possibly their children as well. One of the risk factors for women's distress during *BRCA1/2* genetic testing appears to be whether or not their own mothers were affected with breast cancer when these women were children (van Oostrom et al., 2006a). This dynamic may further contribute to women's motivations for testing their children. In addition, it raises questions regarding how knowledge of cancer history and genetic testing in the family impact children's social and emotional functioning as they mature.

Few studies have directly assessed children's attitudes toward predictive genetic testing. However, Harel and colleagues (2003) found that adolescent girls with a positive family history of breast cancer were significantly more interested in genetic testing for breast cancer risk than were girls without a positive family history (Harel, Abuelo, & Kazura, 2003). We (Tercyak et al., 2001b) and others (Cappelli et al., 2005) have also shown that children—especially adolescent daughters—growing-up in high risk breast cancer families are concerned about their inherited breast cancer risks, validating the possibility that both parents and children may become increasingly interested in the possibility of pediatric *BRCA1/2* testing over time. Parents may contemplate testing for their minor-age children and believe that parents possess the authority to make this decision on their children's behalf (Campbell & Ross, 2003; Hamann et al., 2000; Patenaude, Basili, Fairclough, & Li, 1996). In a recent study, Bradbury and colleagues (2008) reported that 31 of 41 (66%) mothers tested for *BRCA1/2* mutations opposed testing of minors, and a higher proportion (75%) would not have been interested in having their own minor children tested. Although that study provided some data about reasons why parents supported or opposed testing in minor children, the literature does not offer much insight into parents' reasoning in this area, or how they view the functional benefits of testing children. It is also unclear if parents fully appreciate how they and their children might respond to learning about the child's inherited adult-onset

cancer risks, including under what circumstances they might or might not share this information with the child and his or her pediatrician. Because the desire to obtain cancer risk information for relatives, especially children, is a strong motivator for women who seek genetic testing (Lerman et al., 1996; Lynch et al., 1997; Patenaude et al., 2006), it is important for clinicians and researchers to better understand and attend to this need. Addressing these and related questions could help to better guide and inform the debate about high risk parents' needs and desires to protect their children's health and to understand the role that genetic testing for *BRCA1/2* mutations plays in this context. Presently, there is no reliable or valid manner in which to assess these issues—but the need for empirical, objective measurement certainly exists.

In light of this, we set out to develop and evaluate a new assessment tool for use in genetic research and consultation with parents undergoing *BRCA1/2* testing to quantify parents' opinions about *BRCA1/2* testing in children, as well as their attitudes toward decision making and communication of genetic testing results to their children. Data collected with this assessment tool may be used to more closely assess parental attitudes about this issue and could help inform the content of genetic consultations with parents undergoing *BRCA1/2* testing.

Method

Participants

The study population consisted of women with one or more children (8- to 21-years-old). All women ($n = 187$) were participating in pretest education, genetic counseling, and testing for *BRCA1/2* mutations at one of three East coast cancer centers (Lombardi Comprehensive Cancer Center, Washington, DC, Mount Sinai School of Medicine, NY, Dana-Farber Cancer Institute, Boston, MA). These centers offer clinical and research-driven programs focusing on the identification and management of adult hereditary cancer syndromes, including hereditary breast/ovarian cancer. Mothers' demographic and clinical information is given in Table I. Mothers ranged in age from 30- to 59-years-old; most were white, college-educated, with above-average household incomes, and living in partnered relationships. In addition to mothers, co-parents (i.e., parents who share in the responsibility for the upbringing of the child; $n = 96$) served as participants as well (Table I); their age range was 29- to 73-years-old and shared similar demographic characteristics as mothers. The vast majority of these co-parents were male (97%), and the children's biological fathers (90%).

Table 1. Participant Characteristics

	Mothers (<i>n</i> = 187)				Co-parents (<i>n</i> = 96)			
	<i>M</i>	<i>SD</i>	<i>n</i>	%	<i>M</i>	<i>SD</i>	<i>n</i>	%
Age	45.6	5.9			47.5	6.8		
Race								
White			152	81.3			80	83.3
Other			35	18.7			16	16.7
Education								
< College			47	25.1			22	22.9
≥ College			140	74.9			74	77.1
Household income								
<75K			45	24.1			21	22.1
≥75K			142	75.9			74	77.9
Married or living as married								
Yes			150	80.2			89	92.7
Ethnicity								
Jewish			67	35.8				
Other			120	64.2				
Personal cancer history								
Yes			109	58.3				
Family cancer history								
Yes			94	50.3				
Proband status								
Yes			142	75.9				
Child age			13.0	3.8				
Child female gender								
Yes			84	52.5				

Procedure

Eligible mothers were approached by their genetic counselor for possible inclusion in this study at the end of their pretest genetic counseling appointment and following the mothers' stated intention to provide a blood sample for *BRCA1/2* mutation analysis, which was done following the pretest session. As part of the informed consent process, mothers were told that the study focused primarily on learning about parents' attitudes, behavioral intentions, and beliefs regarding the possibility of parents' sharing (communicating) maternal *BRCA1/2* test results with their children, as well as their actual communication decisions and related psychosocial predictors and outcomes; informed consent was obtained at that time and the protocol was approved by each cancer center's Institutional Review Board. The study's consent rate was 81%. Participants completed a baseline telephone survey conducted prior to mothers' receipt of their test results, and two additional follow-up telephone surveys took place 1 and 6 months later. Follow-up surveys collected

additional information about parents' communication behaviors, disclosure-related decision making, and measures of psychosocial functioning.

Upon study enrollment, women's children were enumerated (ages, dates of birth, and genders). For mothers with more than one child falling in the appropriate age range (8- to 21-years-old), a computerized random selection algorithm was used to designate a target child of interest within that range. When responding to all survey items, mothers responded to the designated target child only. Target child identification and reporting has been used extensively in family psychology research and has consistently been shown to be reliable and valid when used with parents (Burrows & Kelley, 1983; Siegel, Dragovich, & Marholin, 1976). This method alleviates parental selection of the child to report on and reduces potential discomfort and bias. As shown in Table 1, target children ranged in age from 8- to 21-years-old and approximately one-half were female.

For mothers with eligible co-parents (*n* = 167, 89% of the study total), they were informed at their pretest genetic counseling appointment that a study invitation could be extended to their co-parent as well, but only if mothers were interested in doing so. Mothers were given a study information packet (including a co-parent informed consent form) and permitted several days during which to reach this decision and consult with their co-parent (if desired). In 30 cases (18% of the eligibles' total) mothers declined this invitation without consulting with their co-parent, in 11 cases (7% of the eligibles' total) mothers declined after consulting, and in 20 cases (12% of the eligibles' total) mothers declined but it could not be determined if consultation had taken place. The primary reasons, mothers offered for declining co-parent participation were relationship estrangement, lack of time, and inability to speak English (DeMarco, Peshkin, Valdimarsdottir, Patenaude, & Tercyak, in press). Among the remaining 106 mothers who were interested in having their co-parent participate, co-parents were approached by telephone by the project assistant for their informed consent; verbal informed consent protocols were reviewed and approved by each cancer center's Institutional Review Board. In only six cases (4% of the eligibles' total) did co-parents decline this invitation; in four other cases, co-parents initially consented to study but subsequently declined to complete a baseline telephone survey. This resulted in a study consent rate of 60% among co-parents and an analyzable sample of 96 co-parent participants. Telephone surveys with co-parents were timed to coincide with those of mothers but were conducted independently.

To acknowledge participants' time and effort, a modest incentive was offered for completing study-related telephone calls (i.e., \$10 gift certificates). All procedures were approved by the institutional review boards at each of the participating sites.

Measures

Pediatric *BRCA1/2* Testing Attitudes Scale

The Pediatric *BRCA1/2* Testing Attitudes Scale (P-TAS) is a face-valid, rationally derived measure of parents' attitudes regarding genetic testing for *BRCA1/2* mutations in children. Initially, items for this measure were developed based on the combined expertise, experience, and suggestions of the study authors with backgrounds in oncology, pediatrics, psychology, bioethics, and genetics and based on conventional methods in psychological test construction (Murphy & Davidshofer, 2004; Nunnally & Bernstein, 1994). Potential item content was elicited via: (a) a review of the behavioral oncology, health psychology, genetics, and bioethics literatures relevant to cancer genetic testing and children, and (b) analysis of formative research results from past work on parent-child communication and genetic testing, and examination of issues commonly raised during clinical encounters with mothers undergoing genetic counseling and testing for *BRCA1/2* mutations. Three of the authors [B.N.P., T.A.D., and K.P.T.] then met and discussed core areas emerging as potentially relevant to assess, grouping common core areas together. These included parents' knowledge, attitudes, and beliefs about the risks and benefits of pediatric *BRCA1/2* genetic testing, decisions and outcomes of testing children, and the respective roles, shared decision making responsibilities, and autonomy of parents, children, and health care providers in pediatric genetic testing. This information was then used to compose an initial pool of items, which was then reviewed by the multidisciplinary team. The 16 items formed a measure with items written as statements with which respondents could strongly disagree (1), neither agree nor disagree (3), or strongly agree (5) along a 5-point scale. All items subsequently underwent cognitive pretesting with a select sample of mothers and co-parents. Five of the items attempting to assess respondents' knowledge about genetics were subsequently dropped due to parents' self-reported difficulties interpreting these items and in responding using the items' scaling. The remaining 11 items were retained to form the final measure and administered at baseline only; higher scores indicate stronger attitudes in favor of pediatric *BRCA1/2* testing. The measures described subsequently were used to further assess the validity of the P-TAS. As this is a new area of research, study-specific measures and items were

relied upon for this purpose. These assessments were derived from the genetic testing literature focusing on parent-child communication of *BRCA1/2* test results.

Child-focused Testing Motive

BRCA1 and *BRCA2* testing studies consistently suggest that a desire to learn about one's children's risk of developing cancer is an important and highly rated benefit of testing (Lerman et al., 1996; Lerman, Daly, Masny, & Balshem, 1994; Lerman, Seay, Balshem, & Audrain, 1995). A single item developed and validated by Lerman and colleagues (1994) was used to assess this construct. Parents' responses were given along a 3-point Likert scale (1 = Not at all important, 3 = Very important). Higher scores on this item indicate a stronger child-focused testing motive, and higher ratings of importance have been significantly associated with decision making about genetic testing (Lerman et al., 1995).

Family Cancer Communication History

Prior work indicates that how often mothers' have talked with their children in the past about cancer in the family and maternal health is positively related to disclosure (Tercyak et al., 2002). This finding is based on the analysis of a 5 item summary scale of the frequency of such communication (1 = Not at all, 4 = Often) that has been found to be reliable (Tercyak et al., 2002); higher scores indicate greater family communication. In the present sample, internal consistency of this measure was adequate (Cronbach's coefficient $\alpha = .66$).

Test Result Disclosure Intentions

Mothers were asked to rate, on a 4-point scale (1 = Not at all likely, 4 = Definitely), how likely they were to disclose a test result to their children in light of a negative (meaning the absence of deleterious gene mutations) and a positive (meaning the presence of deleterious gene mutations) test result. These scores were summed together to yield an overall score, where higher scores reflect stronger intentions to disclose. Prior work suggests that disclosure intentions are a reliable and valid predictor of parent communication behavior regarding *BRCA1/2* genetic test results (Tercyak et al., 2002). In the present sample, negative and positive test result disclosure intentions were highly correlated ($r = .67, p < .0001$).

Results

Factor Structure and Internal Consistency of the P-TAS

A factor analysis of the P-TAS, using principal components extraction method with varimax rotation of factors,

Table II. Exploratory Factor Analysis of the Pediatric *BRCA1/2* Testing Attitudes Scale (Mothers)

Factors and items	Factor loadings	
	1	2
Factor 1: Attitudes and Beliefs		
5. In favor of gene testing for children	.91	.24
3. Even though adult cancer	.88	.24
7. Even though no prevention, treatment, or cure	.88	.27
9. I want my child tested	.82	.18
1. Children should have the opportunity	.77	.24
11. The benefits outweigh the risks	.67	.37
Factor 2: Decision Making and Communication		
10. Noncarriers should be told	.05	.82
4. Children should be involved	.31	.67
6. Carriers should be told	.45	.66
2. Parents should decide	.26	.53
8. Pediatrician of carriers should be told	.11	.44

Boldface factor loadings signify items primarily associated with that factor.

was undertaken on the sample of mothers. The number of factors to be extracted was determined with the assistance of the Scree plot; items with factor loadings >0.4 were considered significant and items were placed on factors with the greatest loading. This yielded a two-factor solution utilizing all 11 items. The solution was determined to be acceptable as it accounted for a total of 62.9% of the variance, resulted in a parsimonious structure, and had interpretable results (Table II). The two factors that emerged were Factor 1: Attitudes and Beliefs (6 items; Cronbach's coefficient $\alpha = .93$) and Factor 2: Decision Making and Communication (5 items; Cronbach's coefficient $\alpha = .70$). Both of these factors appear to capture mothers' considerations in pediatric *BRCA1/2* testing. The overall reliability of the maternal P-TAS was strong, as evidenced by the high internal consistency of the entire scale (Cronbach's coefficient $\alpha = .90$). On Factor 2, it is noteworthy that Item 6 loaded somewhat highly on both factors, and that Item 4 also approached a moderate load statistic on Factor 1, as did Item 11. Given their relatively higher correlations with Factor 2, these items were placed on that factor.

Scores on the P-TAS

Mean (*SD*; *Mdn*) scores for Factor 1 (Attitudes and Beliefs) were 14.61 (7.07; 13.00) and 16.60 (4.70; 17.00) for Factor 2 (Decision Making and Communication). Factors 1 and 2 can be summed together to derive a total P-TAS score, with a sample mean (*SD*; *Mdn*) score of 31.22 (10.57; 31.00). Factors 1 and 2 and total scores tended to be relatively symmetrical (skewness = .52, -.23, and .27, respectively), normally distributed (kurtosis = -.74, -.42,

Table III. Confirmatory Factor Analysis of the Pediatric *BRCA1/2* Testing Attitudes Scale (Co-Parents)

Factors, labels, and items	Factor loadings	
	1	2
Factor 1: Attitudes and Beliefs		
5. In favor of gene testing for children	.90	.14
7. Even though no prevention, treatment, or cure	.88	.17
3. Even though adult cancer	.86	.15
11. The benefits outweigh the risks	.82	.29
1. Children should have the opportunity	.82	.14
9. I want my child tested	.78	.26
Factor 2: Decision Making and Communication		
10. Noncarriers should be told	.23	.79
4. Children should be involved	.49	.51
6. Carriers should be told	.13	.84
2. Parents should decide	.07	.17
8. Pediatrician of carriers should be told	.04	.53

Boldface factor loadings signify items primarily associated with that factor.

and $-.65$, respectively), and utilized the full range of response (score range = 6–30, 5–25, and 11–55, respectively). The correlation between P-TAS Factor 1 and 2 scores was moderately high ($r = .60$, $p = .00$).

Confirmatory Factor Analysis

Co-parents were not incorporated into the factor analysis as they were not undergoing *BRCA1/2* genetic counseling and testing themselves. However, they are important relatives of the mothers participating in that process and share in the responsibility of parenting one or more of these mothers' minor-age children. A confirmatory factor analysis (CFA) was conducted on P-TAS responses from all available co-parents. The purpose of this analysis was to confirm the factor solution realized from the sample of mothers; a two factor solution utilizing all 11 items was assumed. As shown in Table III, the pattern of results of the co-parent CFA are virtually identical to those of mothers. The root mean square error of approximation (RMSEA) is commonly used to estimate the fit of CFA models (Browne & Cudeck, 1993), with RMSEA values less than 0.10 indicating good to reasonable fit, and values less than 0.08 indicating very good fit (Nelson, Aylward, & Steele, in press). The comparative fit index (CFI) is another way to examine the goodness of CFA models, with values greater than or equal to .95 suggestive of very good fit (Nelson et al., in press). In our sample, the RMSEA was .097 and the CFI was .93, suggesting adequate fit. The two factor solution accounted for 61.3% of the variance. Again, the two factors were Attitudes and Beliefs (Factor 1; Cronbach's coefficient $\alpha = .93$) and Decision Making and Communication (Factor 2;

Table IV. Correlations of the Factors of the Pediatric *BRCA1/2* Testing Attitudes Scale with Testing Motive, Communication History, Disclosure Intentions, and Medical and Demographic Information

Measures	Factor/label	
	1 Attitudes and Beliefs	2 Decision Making and Communication
Child-focused testing motive	.21**	.27***
Family cancer communication history	.15*	.23**
Test result disclosure intentions	.24***	.30***
Medical		
Family cancer history (1 = yes, 0 = no)	-.05	-.04
Personal cancer history (1 = yes, 0 = no)	-.02	.01
Proband status (1 = yes, 0 = no)	-.07	-.01
Demographic		
Maternal age	-.10	-.01
Maternal race (1 = White, 0 = other)	-.20**	-.10
Maternal education (1 = ≥ college, 0 = < college)	-.43***	-.32***
Household income (1 = ≥ 75K, 0 = < 75K)	-.25***	-.18*
Child age	.13	.17*
Child gender (1 = female, 0 = male)	.01	.10

* $p < .05$. ** $p < .01$. *** $p < .001$

Cronbach's coefficient $\alpha = .61$). The overall reliability of the entire P-TAS among co-parents also was strong (Cronbach's coefficient $\alpha = .87$). It is important to note that Item 2 showed a low factor loading upon confirmatory analysis and that Items 4 and 11 again appear to load on both factors, though were more highly associated with Factor 2. This pattern of results suggests some caution is warranted when interpreting the P-TAS at the factor versus total score level.

Correlations of the P-TAS with Other Measures

Factor scores derived from the 11 P-TAS items were examined in relationship to mothers' child-focused testing motive, history of family cancer communication with their children, and mothers' intentions to tell their children about their *BRCA1/2* test results. The correlation matrix is displayed in Table IV. Among the validity indicators, correlations ranged from $r = .15$ to $r = .30$ (all p 's $< .05$)—indicating reasonable convergence with related constructs. None of the cancer-specific medical variables assessed [family cancer history, personal cancer history, first person in family to undergo *BRCA1/2* testing (“proband”)] were related to P-TAS factor scores, suggesting that these scores are likely independent of their influence. With respect

to demographic information, maternal race, education, and income and child age were significantly associated with factors scores on the P-TAS. Specifically, non-White mothers, less educated mothers, those with lower family incomes, and mothers with older children were more likely to hold more favorable attitudes toward pediatric *BRCA1/2* testing.

Discussion

We set out to develop and validate a brief assessment tool of parents' attitudes toward testing minor children for hereditary breast/ovarian cancer genes. We believed that such an assessment tool would be a valuable addition to the medical literature as it could be used to more closely assess parental attitudes about this issue and help inform the content of genetic consultations with parents undergoing *BRCA1/2* testing. The results suggest that the 11 item P-TAS appears promising for this purpose, and the use of a dimensional measure is important in this context given the complexity and range of issues potentially associated with it (Cohen et al., 2006; La Greca & Lemanek, 1996). Among tested mothers, its two factors (Attitudes and Beliefs, Decision Making and Communication) accounted for a high proportion of the total measured variance (62.9%) and the internal consistency of the factors and total scale ranged from acceptable to high (.70–.93). These results were similar to those obtained from a paired sample of nontested co-parents, indicating that the measure functions somewhat equally across both members of parenting dyads. Though the degree of fit of the factor model was reasonably confirmed, it is potentially important to recognize the inherent differences between the maternal and co-parent samples. Specifically, mothers underwent extensive genetic counseling prior to completing the P-TAS and co-parents did not. Mothers were also being tested for *BRCA1/2* mutations themselves whereas co-parents were not. Mothers are potentially at risk for developing a *BRCA1/2*-linked breast and/or ovarian cancer and co-parents are not. In light of these fundamental differences, the relative degree of consistency between parents' factored results is encouraging, and any variations between them may be due to these other influences. Nevertheless, standard CFA model fit indices do suggest some caution is warranted when interpreting P-TAS results among co-parents.

When the validity of the P-TAS was assessed by examining its association with related constructs, promising results were also evidenced. Maternal P-TAS scores were higher (indicating more favorable attitudes toward pediatric *BRCA1/2* testing) among mothers participating in

BRCA1/2 testing out of a stronger desire to learn about their children's risk of developing cancer. Additionally, mothers who had more often talked with their children about cancer in the family, and those who were more inclined to disclose their genetic test results to their children, were more in favor of pediatric *BRCA1/2* testing.

Although none of the cancer-specific medical variables analyzed in this research were related to maternal P-TAS scores, several pieces of demographic information were. Specifically, non-White mothers, those who were less educated, and those with lower family incomes were more likely to be interested in this possibility. In our sample, maternal race, education, and household income were confounded with one another, making it difficult to disentangle their individual effects and the representation of these groups in the study sample was modest. The cancer genetic testing literature does contain reference to racial differences in attitudes toward *BRCA1/2* testing in general (Hughes et al., 1997; Lerman et al., 1999). Our results complement these differences, and may reaffirm the need for culturally tailored genetic counseling to more fully address patient interests (Charles, Kessler, Stopfer, Domchek, & Halbert, 2006). That child age was positively related to the P-TAS Decision Making and Communication factor is most likely evidence of parents' anticipation that, among their older children, cascade testing would proceed in the family in light of a positive test result and that their children would be informed of this opportunity.

As noted earlier, there have been considerable discussions in the medical literature challenging current position statements and guidelines advising against testing minor children for adult-onset genetic diseases. However, there has not been a systematic method of assessing parents' opinions about such testing, and what factors might be associated with their attitudes. The current measure helps to fill that gap and could be integrated into genetic counseling research programs to better understand parents' attitudes and preferences about *BRCA1/2* testing in children, and how these may change over time. Findings from this research may also inform future studies that aim to assist parents with making *BRCA1/2*-related decisions, including communicating with minor children about a parent's *BRCA1/2* test results, sharing such information with a child's pediatrician, and (especially for older children) pursuing additional information or consultation in preparation for testing the child (e.g., as a young adult).

Limitations of this study include the highly select nature of the population studied, in that the majority of participants are members of high risk kindreds and may

not be representative of the population of individuals who seek *BRCA1/2* testing in clinical practice. In addition, the sociodemographics of the population studied are also skewed toward Caucasian, educated, and financially secure households. Little is known about how attitudes toward pediatric genetic testing may differ in families of more varying sociodemographic backgrounds. Further, we administered the P-TAS after mothers had participated in an initial (pretest) genetic counseling session, and did not readminister the P-TAS after mothers learned of their *BRCA1/2* test results (posttest). It is possible that administering the measure after genetic counseling had taken place heightened respondents' awareness of certain issues, including affecting their attitudes about pediatric *BRCA1/2* testing. A purer index of parental attitudes could be gained by assessment prior to pretest genetic counseling, though such responses in and of themselves are less meaningful. This is because the purpose of pretest counseling is to provide an informed basis of understanding about empiric cancer risks, and the risks and benefits associated with genetic testing for a particular individual. These risk estimates cannot be determined in advance without detailed family history data and analysis—both of which take place during the course of counseling. It would be more interesting to know if parents' attitudes change in light of their test results, such as if parental attitudes grow stronger in the presence of a confirmed risk-conferring mutation. To evaluate these issues, repeat administrations of the P-TAS would be necessary (i.e., prior to pretest genetic counseling, following pretest genetic counseling but prior to posttest genetic counseling, and following posttest genetic counseling). This would presume the sensitivity of the P-TAS to change over time and its resistance to practice effects, neither of which has been evaluated. In general, more research on the reliability and validity of the P-TAS would be helpful to better determine its potential as an aid to psychosocial genetic counseling.

In sum, this new tool may be useful in assessing parents' attitudes toward pediatric *BRCA1/2* testing and identifying those subgroups of parents who are highly motivated to have their minor-age children tested. These parents may require additional education and counseling, including information about the risks of such endeavors and in light of current prohibitions against such testing in children. Pediatric psychologists are well-poised to be involved in the conduct of this work, contributing expertise in children's developmental and psychological needs.

As greater numbers of predictive genetic tests for adult-onset conditions become available, more and more

parents will be faced with issues such as those currently facing *BRCA1/2* testing participants. As a field, child health psychology has an opportunity to make more significant contributions to informing these issues, particularly through its research and practice focus. This includes the development and evaluation of dimensional, disease-specific psychosocial measures, and the integration of these measures to promote evidenced-based practices and policies in new and emerging areas of preventive and genomic medicine.

Acknowledgments

This research was supported by a grant from the National Human Genome Research Institute at the National Institutes of Health (NIH) (HG002686); additional support was provided by NIH grants CA091831 (to K.P.T.), CA082346 and CA108933 (to M.D.S.), a grant from the American Cancer Society (TURSG02246) (to H.B.V.), and the resources of the Jess and Mildred Fisher Center for Familial Cancer Research at Georgetown University. We gratefully acknowledge the assistance of Marilyn Sampilo,

Lara Wilson, and Lauren Wine in conducting this research and Rusan Chen, PhD for statistical consultation.

Conflict of Interest: None declared.

Received October 18, 2007; revisions received February 27, 2008; accepted March 11, 2008

Appendix 1

Pediatric BRCA1/2 Testing Attitudes Scale (P-TAS)

INSTRUCTIONS: We are interested in learning about your attitudes toward *BRCA1* and *BRCA2* testing for minor-age children, privacy, and your rights as a parent. As you may know, *BRCA1/2* genetic testing for hereditary breast/ovarian cancer risk is generally not available to minor-age children due to several medical, social, and psychological reasons. Please indicate your agreement with each of the following statements using the scale below.

	Strongly Disagree	Disagree	Neither Agree nor Disagree	Agree	Strongly Agree
1. Children under age 18 should be given the opportunity to be tested for the <i>BRCA1</i> and <i>BRCA2</i> gene alterations	1	2	3	4	5
2. Parents should decide if their children are allowed to have a <i>BRCA</i> test or not, even if a doctor disagrees	1	2	3	4	5
3. Even though the cancers associated with <i>BRCA</i> alterations do not affect children until they reach adulthood, children should still be offered <i>BRCA</i> testing	1	2	3	4	5
4. Children should be involved in making the decision about whether or not they participate in <i>BRCA</i> testing	1	2	3	4	5
5. I am in favor of <i>BRCA1/2</i> gene testing for children	1	2	3	4	5
6. If children are tested and they turn out to carry a <i>BRCA</i> alteration (that is, they test positive), they should be told about their test result immediately	1	2	3	4	5
7. Even if there is no known prevention, treatment, or cure for the cancers associated with <i>BRCA</i> alterations, children should still be offered <i>BRCA</i> testing	1	2	3	4	5
8. If children are tested and they turn out to carry a <i>BRCA</i> alteration (that is, they test positive), then this information should be shared with the child's pediatrician	1	2	3	4	5
9. I want my child to be tested for <i>BRCA1</i> and <i>BRCA2</i> gene alterations before age 18	1	2	3	4	5
10. If children are tested and they turn out not to carry a <i>BRCA</i> alteration (that is, they test negative), they should be told about their test result immediately	1	2	3	4	5
11. The benefits of children participating in <i>BRCA</i> testing outweigh the risks	1	2	3	4	5

Scoring:

Factor 1 (Attitudes and Beliefs) Sum all ODD-numbered item responses:

Factor 2 (Decision Making and Communication) Sum all EVEN-numbered item responses:

Total:

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