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Interpersonal Responses Among Sibling Dyads Tested for BRCA1/BRCA2 Gene Mutations

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Abstract

Objective—The familial context plays an important role in psychosocial responses to genetic testing. The purpose of this study was to compare sibling pairs with different combinations of BRCA1/BRCA2 test results on measures of affect, interpersonal responses, and physiological reactions.

Design—Forty-nine sibling dyads with different combinations of BRCA1/BRCA2 test results (i.e., mixed, positive, negative) completed a questionnaire, and 35 of the dyads also participated in a laboratory-based discussion of genetic testing.

Main Outcome Measures—The primary outcome variables included participant reports of supportive actions toward their sibling, state anger and anxiety, perceptions of sibling behavior, and electrodermal responses.

Results—Compared to positive and negative dyads, mixed pairs reported less friendly general support actions, noted more anger, and perceived their sibling to be less friendly and more dominant during the interactions. In comparisons between same-result (i.e., positive, negative) pairs, positive dyads reported more dominant support behaviors and perceived their sibling to be friendlier during the interactions.

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Portions of this research were presented at the annual meetings of the American Psychosomatic Society (2003) and the Society of Behavioral Medicine (2003).

Conclusion—Data suggest that siblings who have different test results may experience more interpersonal strain than siblings who have the same test result. Future research on genetic testing and family relationships can expand upon these findings.

Keywords

genetic testing; siblings; family; BRCA1/BRCA2; cancer

A woman who has a mutation in the BRCA1 or BRCA2 gene carries significantly increased risks for breast (approximately 45%–65% by age 70) and ovarian (approximately 11%–39%) cancers (Antoniou et al., 2003; Miki et al., 1994; Wooster et al., 1995). Among men, gene mutations may also be associated with smaller increases in risk for breast and prostate cancer (Gayther et al., 1997; Thompson, Easton, & the Breast Cancer Linkage Consortium, 2002). Based on associations between family support and psychological adjustment among cancer patients and high risk women (e.g., Coyne & Anderson, 1999; Manne, Taylor, Dougherty, & Kemeny, 1997), researchers have argued that the family context may play a role in psychological reactions among individuals who have undergone genetic testing (e.g., Richards, 1998; Wylie, Smith, & Botkin, 2003). Specifically, since multiple individuals within a family may be tested for BRCA1/BRCA2 mutations, the various combinations of test results among family members may affect psychosocial responses.

A handful of studies have reported on the potentially adverse emotional impact of having family members with opposite test results. Most studies in this domain have focused on individuals who receive genetic testing for Huntington Disease (HD; a typically adult-onset neuromuscular disorder) or breast/ovarian cancer susceptibility (i.e., BRCA1/2). Although genetic testing for both of these conditions provides information about the degree of disease susceptibility, there are important differences in the meaning of the test results. Huntington Disease is a progressive, virtually untreatable disorder; a positive HD test result is associated with a relatively certain lifetime chance of disease (although age of onset and severity are variable; Bennett, n.d.). In contrast, BRCA1/2 mutation carriers have significantly increased, but not certain, risks for cancer, and there are often more risk reduction (e.g., prophylactic surgery) and early treatment options (Offit, 1998). Although these various disease characteristics may differentially affect test responses, the general literature is useful for understanding how different combinations of genetic test results among family members could impact relationships.

In case reports of testing for HD, a subset of noncarriers (i.e., individuals who tested negative for a mutation) were distressed about their test results compared to other family members who were carriers (i.e., had tested positive for a mutation; Huggins et al., 1992; Tibben et al., 1992). Within cancer genetics, Lodder and colleagues (2001) noted that BRCA1 noncarriers with carrier sisters reported higher levels of depressive symptoms than non-carriers without carrier sisters. Smith, West, Croyle, and Botkin (1999) found that noncarrier men with all BRCA1 carrier siblings reported higher levels of test-specific distress than noncarriers without this sibling configuration. Distress among noncarriers with carrier family members has been interpreted as possible “survivor guilt” (e.g., Huggins et al., 1992; Kessler, Field, Worth & Mosbarger, 1987; Smith et al., 1999). For carriers in the mixed result families, there is also some evidence of increased distress in response to comparisons with noncarrier family members. Reports from studies of HD families indicated possible hostility and resentment in a subset of carriers toward their noncarrier family members (Tibben et al., 1992). Smith et al. (1999) noted that distress levels in the group of carrier women with all noncarrier siblings were among the highest in the entire sample.

Relative to opposite-result family members, having family members with the same genetic test result may be associated with better psychological adjustment. Smith and colleagues (1999) found that BRCA1 noncarriers with all noncarrier siblings reported lower levels of test-specific distress than almost all other sibling groups. A similar phenomenon has also been seen among carriers with carrier family members. Tibben and colleagues (1992) noted that a subset of HD carriers reported strengthened relationship bonds with their affected parents posttesting. In the Smith et al. (1999) study, BRCA1 carrier women with at least one carrier sibling reported lower levels of psychological distress than carrier women who were either the first among their siblings to learn their test results or had only noncarrier siblings.

In summary, previous studies suggest that individuals who have opposite-result family members may experience greater psychological distress compared to individuals whose test results match those of family members. However, the evidence is sparse and partly drawn from case studies. It has also focused primarily on measurements of individual distress in which an interpersonal process is inferred but not directly assessed. To date, there have been few examples from the genetic testing literature in which emotional responses and relationship perceptions have been assessed during real-time family interactions in a controlled setting. It is unclear whether test result matches and mismatches are associated with emotional and behavioral differences in actual family interactions.

An Interpersonal Approach to Understanding the Impact of Genetic Testing

The interpersonal tradition in personality, social, and clinical psychology provides a valuable framework for investigating the impact of genetic testing on family relations. The interpersonal approach emphasizes the importance of social behaviors as units of interpersonal communication and posits that the behaviors of one person influence the experiences and responses of other persons (Kiesler, 1996). Within this framework, interpersonal behavior is conceptualized as varying along two dimensions—affiliation (friendliness vs. hostility) and control (dominance vs. submission; Kiesler, 1983; Wiggins, 1979). Related measures comprising the interpersonal circumplex assess such constructs as supportive actions toward partners (Trobst, 2000) and covert responses elicited from partners' behaviors (Schmidt, Wagner, & Kiesler, 1999). Measurements of specific emotions are also important in delineating the impact of these behaviors during interpersonal interactions. Interpersonal theorists argue that interactions are guided by the principle of complementarity (e.g., Kiesler, 1983, 1996), in which the actions of one person “constrict” or pull for certain covert experiences from the interpersonal target.

Goals of the Present Study

Within the context of genetic testing, the interpersonal approach may be useful for understanding differences in distress and behavior based on test result match or mismatch between family members. Based on this framework, it is quite possible that levels of psychological distress (i.e., anger, anxiety) and the degree of affiliation and control during family interactions could vary based on whether family members have same or different test results. The present study focused on adult sibling pairs with three possible combinations of BRCA1/BRCA2 test results (i.e., positive: carrier/carrier; mixed: carrier/noncarrier; negative: noncarrier/noncarrier). First, we compared sibling dyads on measures of general supportive actions toward their sibling. Next, we assessed more specific emotional responses, interpersonal perceptions, and physiological reactions during a laboratory-based discussion of genetic testing. Scales assessing warmth (i.e., friendliness vs. hostility) and control (i.e., dominance vs. submission) were used to measure health-related supportive actions and perceptions of interpersonal behavior during a discussion of genetic testing. Repeated measures of anger and anxiety were utilized to detect changes in affect during the

laboratory discussion. Measurements of change in electrodermal activity (i.e., skin conductance response; SCR) were also gathered in the laboratory as supplemental markers of general emotional arousal and task engagement.

The first set of study predictions focused on differences in affect, interpersonal friendliness, and physiological arousal between dyads. We did not make predictions about interpersonal dominance, but instead treated these analyses as exploratory. Since the existing literature has identified potential conflicts among family members with opposite test results, the primary contrast compared carrier/noncarrier (“mixed”) dyads with the two same-result dyads. It was hypothesized that compared with carrier/carrier (“positive”) and noncarrier/noncarrier (“negative”) dyads, mixed dyads would report less friendly support actions toward their sibling. Similarly, we expected that the mixed dyads would report more negative affect, display more skin conductance responses (SCRs), and perceive their sibling as less friendly while discussing the topic of genetic testing in the laboratory. In a second contrast, we compared the two same-result dyads (positive vs. negative). There is evidence that having family members with the same genetic test result may be associated with better psychological adjustment for both carriers and noncarriers. However, direct comparisons have revealed higher levels of individual distress among carriers than noncarriers (e.g., Croyle, Smith, Botkin, Baty, & Nash, 1997; Lerman et al., 1996). Therefore, we predicted that compared with negative sibling pairs, positive dyads would endorse less friendly support behaviors, report more negative affect, display more SCRs, and perceive their sibling as less friendly during the laboratory session.

A second set of study hypotheses focused on potential differences among carriers and noncarriers within the mixed dyads. Based on case studies identifying anger and resentment among carriers, we hypothesized that carriers in the mixed dyads would report more anger and note less friendly support actions toward their sibling. Reports of poorer psychosocial adjustment and possible guilt among noncarriers with carrier family members led us to hypothesize that compared with their carrier siblings, noncarriers in the mixed dyads would report more anxiety, display more SCRs, and perceive their sibling’s behavior as less friendly.

Method

Participants

Study participants were recruited from a previous study of genetic testing in an extended BRCA1 kindred (K2082; Botkin et al., 1996) and BRCA1 and BRCA2 kindreds from the High Risk Breast Cancer Clinic (HRBCC) at the Huntsman Cancer Institute. Individuals from these sources were eligible for the current study if they were tested for BRCA1/2 mutation(s), received positive (carrier) or “true” negative (noncarrier) results, and had at least one sibling who also received definitive BRCA1/2 results. As part of their inclusion in the previous K2082 and HRBCC studies, participants had undergone comprehensive pretest and posttest genetic education and counseling with subsequent written follow-up. Within this process patients were provided with screening and other medical recommendations, along with information about medical consultation options. More detail about the K2082 genetic counseling protocol in particular can be found in Baty et al. (1997).

Detailed information about study recruitment is presented in Figure 1. To summarize, 254 individuals were identified as eligible for the current study because they had received definitive BRCA1/2 test results and had at least one sibling who had also undergone genetic testing. Consent information was sent to these individuals, and 148 (58.3%) of them agreed to be contacted for the current study. These individuals also completed a brief questionnaire that assessed whether they knew their siblings’ correct test results and approved of each

sibling knowing their own test results. Based on presence of eligible siblings who consented to contact and knew each others' test results, 54 sibling pairs (108 individuals) were successfully contacted for the study. Comparisons indicated that the 108 individuals in eligible sibling pairs were more likely to be female $\chi^2(1, N = 133) = 7.04, p < .01$ and marginally more likely to be younger, $t(131) = 1.75, p < .10$, than the 25 who were contacted but did not have an eligible sibling (15 individuals who were lost to follow-up are not included in these comparisons).

Forty-nine of the 54 eligible sibling dyads (90.7%; 98 individuals) completed the study questionnaire. Among the other five dyads (10 individuals), either one or both siblings did not return a completed questionnaire and did not respond to a follow-up telephone call. There were no significant age or gender differences among the 98 participants compared with the 10 eligible nonparticipants. Of the 49 sibling pairs (98 individuals) who completed the questionnaire, 35 of them (70 individuals) also participated in the laboratory interactions (see Figure 1). There were no significant differences in demographic (i.e., age, gender), medical (i.e., time since testing, cancer history), or personality variables (i.e., neuroticism, agreeableness) between the 35 dyads who completed both the questionnaire and interaction and the 14 dyads who completed just the questionnaire. Therefore, data from all 49 sibling pairs were analyzed together when applicable.

Overall, the 98 participants included 75 women and 23 men, all of whom were Caucasian. Mean age was 47.72 years old ($SD = 12.99$). The majority (90%) were married, 55% had college degrees, and 49% reported yearly household incomes greater than \$50,000. On average, participants had known their genetic test results for 3.54 years ($SD = 1.37$ years), and 28% ($n = 27$) reported a prior history of cancer. Almost 90% of the sample identified a religious membership with the Church of Jesus Christ of Latter-day Saints (LDS or Mormon).

Measures

Dependent Variables

Circumplex measures: Circumplex measures are useful for examining the two dimensions of interpersonal behavior—affiliation (i.e., friendliness vs. hostility) and control (i.e., dominance vs. submission; Kiesler, 1983; Wiggins, 1979). Within this investigation, we were interested in two aspects of social behavior between tested siblings. The first was the extent to which siblings perceived themselves to be generally supportive of one another. This was assessed by the Support Actions Scale—Circumplex (SAS-C; Trobst, 2000), a measure of self-reported support behaviors toward a specific person with a hypothetical “problem.” Since we were most interested in how siblings characterized their support actions toward their sibling in relation to illness, the wording of the SAS-C was modified so that participants rated their own support behaviors toward their sibling with a “health problem.” Second, we were interested in how each individual perceived the interpersonal behavior of their sibling during the laboratory interaction. To assess this aspect, we utilized the Impact Message Inventory—Circumplex (IMI-C; Kiesler, 1983, 1996; Kiesler & Schmidt, 1993; Schmidt et al., 1999), a widely used measure focusing on perceptions of another person's interpersonal behavior. For both the SAS-C and IMI-C, four items per octant (instead of the usual 8) were used for calculate friendliness and dominance factor scores (possible SAS-C range: -14.4 to 14.4 ; IMI-C range: -7.2 to 7.2).

Affect measures: At four different timepoints during the interaction session, both siblings in the dyad rated 12 adjectives from the state form of the Spielberger (1979) State-Trait Personality Inventory (STPI-S). This measure is focused on self-reported anger and anxiety; instructions for the measure asked participants to rate their mood “right now... at this

moment.” Scores from the appropriate items were combined into anger and anxiety subscales; possible scores for each subscale ranged from 6 to 30 with higher scores indicating more negative affect.

Physiological measure: Electrodermal activity (skin conductance) is a well-known measure of sympathetic nervous system activity that has shown to be sensitive to changes in emotion (Dawson, Schell, & Filion, 2000). During the structured interaction sessions, skin conductance was measured continuously for each sibling by constant voltage (0.5 V) Biopac units. Silver-silver chloride electrodes were used to minimize bias potential and polarization. As recommended by Fowles et al. (1981), the electrode paste consisted of sodium chloride electrolyte in a neutral cream medium. Skin conductance values were assessed from the middle 40 seconds of each 60-s interval. The number of significant skin conductance responses (SCRs; > 0.05 microsiemens) within each 40-s time period was calculated and converted into a measure of SCRs per minute.

Control Variables—Each participant’s age was coded as a continuous variable, as was the number of years since participants had received their genetic test results. Since we were primarily interested in between-dyad analyses, gender and cancer history were coded at the level of the dyad. Specifically, each sibling pair was coded as one of two possibilities for gender (two female siblings vs. either male/female or male/male combinations) and cancer history (no cancer history vs. at least one reported cancer diagnosis in the dyad). Personality measures included two scales (Neuroticism, NEO-N; Agreeableness, NEO-A) of the NEO-Five Factor Inventory (NEO-FFI; Costa & McCrae, 1992). Each of the NEO-FFI scales consisted of 12 items, with a possible scale range of 12 to 60. A final control variable included a version of the IMI-C administered prior to the interactions. In this version, participants rated their sibling’s “general” behavior (as opposed to interaction-specific behavior). This measure provided preinteraction indices of interpersonal behavior among the sibling dyads, and factor scores served as covariates controlling general levels of warmth and control in analyses of interaction-specific behavior.

Procedure

Initial Contact and Questionnaire Assessment—Individuals who had undergone BRCA1/2 testing were included in this study if they had expressed interest in study contact, knew the correct carrier status for a sibling who had also consented to contact, and approved this sibling’s having knowledge of their own test results. Once sibling pairs had been identified, they were contacted via telephone by the primary investigator and provided with information about the study. If there were more than two eligible siblings in a sibship, priority in contact was made based on proximity to the university and need for balanced group sizes. Once the two siblings had authorized their involvement, they were scheduled for a session and sent the questionnaire (plus \$15) with instructions to complete it sometime prior to their interaction session. Sibling pairs who were not able to travel to the University of Utah for the interaction but still wanted to participate were given the opportunity to complete only the questionnaire. The questionnaire contained demographic assessments, the Neuroticism and Agreeableness subscales of the NEO-FFI, the version of the IMI-C focused on “general” sibling interactions, and the SAS-C measure.

Interactional Session—Sessions were conducted in a double-occupant chamber with adjoining space for the experimenter and monitoring equipment. All study instructions were audiotaped, and except for brief interactions with the experimenter, the dyads were alone in the chamber. After the informed consent process and preparation for SCR measurement, siblings participated in a minimally demanding task of rating pictures during a 10-min “vanilla” baseline period (Jennings, Kamarck, Stewart, Eddy, & Johnson, 1992).

Participants completed the first (baseline) STPI-S measure and were then given instructions about the first structured discussion (Interaction 1). Specifically, this interaction was framed as a discussion of “both your initial reactions and current feelings regarding your BRCA1/2 test result.” Each participant was provided with a sheet of paper that included open-ended questions to address when speaking (in an effort to standardize the interaction, identical questions were given to each participant). These questions addressed both past reactions (e.g., “What emotions/thoughts did you have when the genetic counselor told you your result?”) and current thoughts (e.g., “What does your test result mean for you now?”) to maximize the chances that participants addressed both of these issues in the discussion. Participants were given 2 min to silently prepare for Interaction 1. Following the preparation period, Sibling A spoke for 1 min, while Sibling B listened and then responded to the comments for 1 min. After this exchange, Sibling B spoke about his or her experiences for 1 min while Sibling A listened and then responded for 1 min. This interaction format was used because it structured the speaking, listening, and response periods into equal intervals for each sibling. After the discussion of their own test results, participants completed a second STPI-S measure.

Following their completion of the second STPI-S, participants were prepped for the second discussion (Interaction 2). This interaction was framed as a discussion of participants’ thoughts and feelings about their own genetic test result compared to their sibling’s test result. It was emphasized that participants should focus only on the sibling with them that day and avoid discussions of other family members’ results. Siblings were given another 2-min preparation time and provided with identical sheets of paper that listed three positive, three negative, and one neutral descriptors of possible reactions to their sibling’s genetic test result (e.g., feeling closer to sibling, feeling jealous, feelings unchanged). Participants were encouraged to discuss their own thoughts and feelings during the interaction, even if they had not been represented in the list. Siblings then participated in a discussion with the same format as the first interaction, except that the order of speaking was reversed. Following this interaction, siblings completed a third STPI-S and then participated in a 5-min, unstructured Free Discussion period in which they focused on emotions or unanswered questions from the previous two sessions. The siblings then completed a fourth STPI-S and the IMI-C measure in which they referenced their sibling’s behavior specifically during the laboratory interaction. Following completion of the measures, the physiological sensors were removed, the participants were debriefed, and they were paid \$25 each for their participation.

Statistical Analyses

Analyses were based on a general framework of a mixed factorial analysis of variance (ANOVA), with dyad type (positive, mixed, negative) as a between subjects factor and each sibling treated as a repeated factor in order to accommodate the dependent nature of observations from members of a sibling pair. In the mixed dyads, the designated member for the repeated measures design was determined based on individual test result (carrier or noncarrier). In the other dyads, the designations were balanced by speaking order. If the dyad-level index of gender (two female siblings vs. female/male or male/male dyads) or cancer history (no cancer history vs. at least one reported cancer diagnosis in the dyad) was significantly related to a given dependent measure, it was added as a second between-subjects variable in that statistical model. For the anxiety and anger inventories during the laboratory session, there were four measurement periods (i.e., baseline, post-Interaction 1, post-Interaction 2, post-Free Discussion). For the SCR measure there were three measurement periods (i.e., baseline, mean of four 1-min periods comprising Interaction 1, mean of four 1-min periods comprising Interaction 2). Skin conductance measurements from the Free Discussion period were not included because of the difficulty in teasing out effects of speech or movement from this less structured period. For each of the SCR measurement

periods, values for the task period (i.e., postbaseline) were highly correlated ($r = .53$ to $.94$, all p s $< .001$). To increase reliability of the dependent variables, mean difference scores for anger, anxiety, and SCR were calculated by averaging the task period timepoints and subtracting baseline values to form change scores. In addition, the baseline value was covaried if there was a baseline difference ($p < .10$) between the contrasted sibling dyad types. In certain cases, continuous personality or demographic variables had also been identified as covariates based on their significant associations with the dependent variable. Since program constraints of Statistical Package for the Social Sciences (SPSS) did not allow analysis of covariance with separate covariates at different levels of the repeated factor (sibling within each dyad; Tabachnick & Fidell, 1996; SPSS 10.1, 2000), residual scores were calculated by regressing the dependent variables on the covariates. This procedure of creating residualized scores is similar to that described by Gillespie and Streeter (1994) in their review of analyzing change in nonexperimental research (see also Cronbach & Furby, 1970, and Llabre, Spitzer, Saab, Ironson, & Schneiderman, 1991, for discussions). The residual values were then converted back into adjusted scale values and included as dependent variables in the mixed factorial equations.

Weights for the first a priori between-dyad contrast compared the mixed dyads (2) with the positive (−1) and negative (−1) dyads. A second contrast compared positive (1) and negative (−1) dyads. Each between-dyad contrast was calculated with harmonic means of the cell sample size and the error term from the overall between-dyad ANOVA equation. A within-dyad contrast compared carriers and noncarriers in the mixed dyads. For the calculation of this contrast, the error term from the repeated factor in the ANOVA equation was utilized. When reporting results, mean values of the dependent variables were adjusted for the appropriate covariates unless otherwise indicated. Estimates of effect size for a priori hypotheses were calculated as eta squared (η^2), presented as the proportion of variance attributable to an effect (Tabachnick & Fidell, 1996). Eta squared values of .04, .25, and .64 were conceptualized as small, medium, and large effect sizes, respectively (Cohen, 1992).

Results

Group Differences

Table 1 presents comparisons of the three dyad groups on demographic, medical, and personality variables. The only significant difference was in gender make-up of the dyads, with an overrepresentation of female/female dyads in the positive group, $\chi^2(2, N = 49) = 7.58, p < .05$. Differences in time since testing almost reached significance, $F(2, 46) = 2.72, p < .10$, with a trend toward longer intervals for negative dyads.

Determination of Covariates

Bivariate correlation analyses identified control demographic (i.e., age), medical (i.e., time since testing), and personality factors (i.e., neuroticism, agreeableness) that were associated with the dependent measures at a level of $r > .25$ or $r > -.25$. Results from the correlation matrix revealed that trait agreeableness was positively correlated with SAS-C friendliness scores ($r = .41$), while age was negatively associated with SAS-C dominance scores ($r = -.28$). Trait agreeableness was negatively correlated with change in state anger ($r = -.38$), and trait neuroticism was positively correlated with change in state anxiety ($r = .29$). For the interaction-specific IMI-C measures, age was negatively correlated with dominance scores ($r = -.35$). To reduce unexplained variance in the dependent measures and provide more sensitive tests of the hypotheses, trait agreeableness, age, or neuroticism was included as a covariate when it correlated with an outcome variable.

In addition to the continuous demographic and personality variables, other potential sources of variability included dyad-level measures of cancer history (i.e., presence or absence of cancer history in the dyad) and gender (i.e., all-female v. female/male or all-male dyads). We also performed a secondary analysis for gender in which individual-level relationships between gender and the outcome variables were assessed. Cancer history was predictive of SAS-C Friendliness scores and both indices (Friendliness, Dominance) of the interaction-specific IMI-C measure. In particular, dyads with a cancer history ($M = 10.35$; $SE = .35$) reported more friendly personal support actions than dyads without a cancer history ($M = 9.37$; $SE = .32$), $F(1, 43) = 4.28$, $p < .05$. Dyads with a cancer history also perceived their siblings to be significantly friendlier ($M = 5.48$; $SE = .20$) and less dominant ($M = -1.16$; $SE = .10$) than dyads without a cancer history ($M_s = 4.75, -0.87$; $SE_s = .18, .09$), $F_s(1, 33) = 7.21$ and 4.65 , respectively ($p_s < .05$). In models for each of these dependent variables, cancer history of the pair was included as an additional independent variable. Dyad-level gender was not related to significant differences in any of the dependent variables. When gender was measured at the individual level, only SAS-C Friendliness and SCR change scores were significantly different ($p < .05$) among women and men (women had higher values for both measures). Therefore, for SAS-C Friendliness and SCR change scores we report on a primary model that does not include gender and a secondary model with its inclusion.

Supportive Actions Toward Sibling

Items from the SAS-C focused on respondents' reports of friendly and dominant support behavior in response to a sibling "health problem." Table 2 lists means by dyad type and a summary of the a priori contrast results. Consistent with our predictions for the first a priori contrast, mixed dyads reported significantly lower levels of friendly support behavior than positive and negative dyads, $F(1, 38) = 4.72$, $p < .05$, $\eta^2 = .09$.¹ However, in the second planned contrast, the difference between positive and negative dyads in friendly support behavior was not significant, $F(1, 38) = 1.37$. The predicted difference between carriers ($M = 9.11$; $SE = .51$) and noncarriers ($M = 9.63$; $SE = .47$) within the mixed dyad was also not detected, $F(1, 38) < 1$.

In the analysis of dominant support behaviors, mixed dyads were not significantly different than positive and negative dyads, $F(1, 42) < 1$. The second a priori comparison revealed more dominant support behaviors among positive than negative pairs, $F(1, 42) = 10.24$, $p < .01$, $\eta^2 = .20$. No significant within-dyad difference was found for the mixed dyads (carriers: $M = 2.70$; $SE = .77$; noncarriers: $M = 2.87$; $SE = .78$), $F(1, 42) < 1$.

Affective Responses

The two subscales from the STPI-S reflected state anger and anxiety during the interactions. Comparisons of baseline anger scores revealed a trend approaching significance ($p < .10$) in which mixed dyads ($M = 8.79$; $SE = .42$) reported higher scores than positive ($M = 8.00$; $SE = .46$) and negative ($M = 7.67$; $SE = .53$) dyads. As a result, baseline anger was added as a covariate to the model for change in anger. As predicted in this model, mixed dyads reported more change (increase) in anger than the positive and negative dyads, $F(1, 29) = 10.66$, $p < .01$, $\eta^2 = .26$. The second between-dyad comparison (positive vs. negative) did not reveal the predicted effect for anger, $F(1, 29) < 1$ (mean values for dyad types are listed in Table 2). In the within-dyad contrast for the mixed group, a weak trend in the direction of the prediction emerged; carriers ($M = 1.44$; $SE = .41$) reported higher increases in anger than noncarriers reported ($M = .48$; $SE = .37$), $F(1, 29) = 2.93$, $p < .10$.

¹When gender was added to the model, this result remained statistically significant.

Results of dyad comparisons revealed no significant differences in baseline anxiety ($M = 9.97$; $SE = .49$), so this variable was not included as a covariate in the statistical model. None of the predicted between-dyad effects were noted for change in state anxiety between mixed dyads vs. positive and negative dyads, $F(1, 29) < 1$, or positive vs. negative dyads, $F(1, 29) < 1$ (see Table 2). No difference was noted among carriers ($M = 1.23$; $SE = .67$) and noncarriers ($M = .64$; $SE = .58$) in the mixed dyads, $F(1, 29) < 1$.

Perceptions of Sibling Behavior During Discussion Task

The IMI-C measure that participants completed at the end of the session contained ratings of friendly and dominant sibling behavior during the interaction sessions. To focus even more specifically on these interactions, appropriate scores from the questionnaire version of the IMI-C, which assessed perceptions of “general” sibling behavior, were covaried in the models. Consistent with our prediction, a primary between-dyad contrast revealed that mixed dyads perceived less friendliness during the interactions than positive and negative dyads, $F(1, 26) = 8.41$, $p < .01$, $\eta^2 = .10$. There was also a significant difference between the positive and negative dyads, but it was in a direction opposite to the hypothesized effect. Specifically, positive dyads reported higher levels of sibling friendliness during the interactions than negative dyads, $F(1, 26) = 5.63$, $p < .05$; $\eta^2 = .07$ (see Table 2). Dyads in which one or both members had a history of cancer continued to perceive their siblings as more friendly than dyads in which neither member had a history of cancer, $F(1, 26) = 11.76$, $p < .01$, $\eta^2 = .15$. In addition, a significant interaction between cancer history and dyad type, $F(1, 26) = 13.65$, $p < .01$, $\eta^2 = .34$, indicated that mixed dyads with no cancer history reported the least sibling friendliness ($M = 3.70$; $SE = .22$) of any other group. Inconsistent with our hypothesis, no significant difference was noted in the within-dyad comparison for the mixed groups (carriers: $M = 4.67$; $SE = .28$; noncarriers: $M = 4.71$; $SE = .23$), $F(1, 26) < 1$.

Mixed pairs perceived more dominant (i.e., less submissive) sibling behaviors than the positive and negative dyads, $F(1, 26) = 6.00$, $p < .05$, $\eta^2 = .16$ (see Table 2). There was no difference in perceptions of dominance between positive and negative dyads, $F(1, 26) = 1.74$. Carriers ($M = -.77$; $SE = .14$) and noncarriers ($M = -.95$; $SE = .14$) within the mixed dyads were also not significantly different, $F(1, 26) < 1$.

Electrodermal Response

There were no significant differences among dyad types in number of baseline SCRs per minute ($M = 1.94$; $SE = .27$), so this variable was not covaried in the model. Results from the first comparison of SCR change between mixed and same-result dyads revealed no significant differences, $F(1, 32) = 1.71$ (see Table 2). The second planned contrast of positive versus negative dyads approached significance, with positive dyads displaying a marginally greater change in SCR than negative dyads, $F(1, 32) = 3.70$, $p < .10$.² No significant within-dyad difference was reported among carriers ($M = 2.58$; $SE = .49$) and noncarriers ($M = 1.90$; $SE = .46$) in the mixed dyads, $F(1, 32) < 1$.

Discussion

The purpose of the present study was to examine associations between genetic test result combinations and the quality of sibling relationships. Specifically, we compared three types of sibling dyads (i.e., mixed, positive, negative) previously tested for BRCA1/BRCA2 mutations on measures of negative affect, interpersonal responses, and physiological

²When gender was added to the model, this result was not statistically significant.

reactions. The interpersonal approach provided a framework by which to conceptualize and measure aspects of social behavior and emotional responses among siblings.

Mixed-Result Dyads

Although not every prediction about differences between mixed and same-result dyads was supported, the pattern of results was generally consistent with our hypotheses. Compared with positive and negative dyads, members of the mixed dyads reported less friendly support behaviors in response to a sibling health problem. During the interaction, they reported more anger and more sibling dominance than the same-result pairs. They also perceived their sibling to be less friendly during the discussions; this was particularly true among the mixed pairs with no history of cancer. Hypothesized differences between the mixed and same-result pairs were not found for state anxiety or skin conductance.

Previous studies (i.e., Lodder et al., 2001; Smith et al., 1999; Tibben et al., 1992) had introduced the notion of negative reactions among mixed-result family members; data from this study suggest a level of anger, interpersonal strain, and controlling behavior during the interaction among mixed dyads that was not seen in siblings who shared the same genetic test result. Interpreting the data within an interpersonal model, the mismatch in test results may have influenced both siblings' behaviors and emotions so that a more negative pattern emerged from the interaction. In general, the results from the mixed dyads support the notion that negative interpersonal effects of differing genetic test results may complicate discussions of cancer risk and other related health issues. Given the current study's focus on interpersonal responses approximately 3.5 years after genetic testing, these issues may be especially relevant in the longer term after test result disclosure. During this later time period, individuals may be more likely to address such issues as cancer risk reduction (e.g., prophylactic surgery), cancer diagnoses and decisions about cancer treatment, testing of adult children, and caregiving for family members with cancer. These are important decisions that may benefit from social support, and more strained interactions related to these issues could undermine otherwise beneficial support relationships.

The finding that mixed dyads without a cancer history perceived the least friendly sibling behaviors may suggest that divergent test results are particularly disruptive when individuals have little personal experience of cancer. This finding is similar to that by Croyle and colleagues (1997), who found that carrier women without a history of cancer or cancer-related surgery reported higher levels of test-related distress than carrier women who did have a history of cancer or related surgery. Within the current study, it is possible that the sibling dyads without a history of cancer were anticipating an "unknown" cancer-related threat which detrimentally affected their interpersonal relationships. Another explanation is that dyads in which one or both siblings had a previous cancer diagnosis may have experienced closer relationship ties as a result of this experience, and these feelings were then reflected in their more positive interpersonal responses during the study. It should be noted that as part of their postresult genetic counseling, participants had received comprehensive information about cancer risks and treatment options. It is possible that this information helped those who developed cancer after testing to feel more prepared and satisfied with their treatment choices, which may have beneficially influenced their family relationships. In general, findings such as these emphasize the potential role that cancer history may have on interpersonal responses to BRCA1/2 genetic testing, and more research is needed to address this issue.

Comparisons of carriers and noncarriers within the mixed dyads indicated that in general, their responses to the interactions did not significantly differ. Overall, this may reflect similar understandings among dyad members of their general supportive actions and responses during the interactions. Although guilt was not measured directly, the lack of

significant differences in anxiety and SCR among carriers and noncarriers within the mixed dyads does not support the notion that noncarriers experience significant “survivor guilt.” This was somewhat surprising given the focus on survivor guilt within some aspects of the genetic testing literature. However, even though the notion of survivor guilt related to genetic testing has been discussed as a theoretical possibility, it is less supported by actual data (Hayden, Adam, & Bloch, 1993). In addition, the majority of discussions about survivor guilt and genetic testing have been in the context of Huntington Disease which, based on its grim prognosis for affected individuals, may elicit more guilt-related feelings among noncarrier family members than does testing for breast/ovarian cancer. We did find a weak trend in which carriers in the mixed dyads reported higher levels of anger than noncarriers. Although this finding should be interpreted cautiously, it may support an assertion by Tibben et al. (1992) that some carriers direct anger and resentment toward their noncarrier siblings.

Same-Result Dyads

Results from the majority of contrasts between positive and negative dyads were not consistent with the hypotheses. For example, it was surprising to find that positive dyads perceived their sibling to be friendlier during the interactions, and we did not find the hypothesized differences in friendly support behaviors, anger, and anxiety. We did find that positive dyads displayed marginally more SCRs than negative dyads, but this effect was weakened when gender was added to the model. Positive dyads also reported more dominant support behaviors in regard to a sibling health issue.

One possible reason for the lack of hypothesis support among same-result dyads is that the predictions were based on data of individual adjustment, which may not reflect the complexity of interpersonal responses. Kessler (1993) emphasized the importance of addressing adjustment to mutation disclosure within the context of the family system, noting that this inclusion adds a dimension not seen in reports of individual adjustment. Overall, the comparisons between positive and negative pairs in our study suggest that positive dyad members were relatively more engaged in their sibling’s health needs and perceived their sibling as warm and supportive when discussing their test results. These results may support the notion of emotional engagement and psychological buffering among carriers who may “share the adversity” with their carrier siblings. Especially during discussions of genetic testing and cancer, carrier siblings may be able to provide each other with social support, share treatment information, and give advice about family issues. This process may be similar to that observed in cancer support groups, in which the shared experience of an adverse event may strengthen social and emotional bonds.

Previous research focused on individual responses (e.g., Croyle et al., 1997; Lerman et al., 1996) had noted reduced distress among individuals who tested negative, so we had expected more friendly interpersonal responses among the negative dyads. However, the overall pattern of results for negative dyads suggests a relatively neutral, less invested relationship pattern than the other groups. Again, this difference may be reflective of interpersonal assessments that capture different responses than do measures of individual reactions. Since cancer risk was less salient for negative siblings, discussions of genetic testing and cancer among them may not have provoked particularly emotional interpersonal responses. They also may not have felt as involved in each other’s health-related issues.

Limitations and Conclusions

Although there were several differences between dyads on the outcome measures, scores suggest that the majority of sibling pairs were experiencing relatively supportive and friendly relationships. Therefore, it is not clear that members of even the most

interpersonally distressed group (mixed dyads) display difficulties that would warrant substantial clinical attention. Nonetheless, siblings' relationship reports and reactions to the laboratory interactions do suggest that divergent genetic test results likely complicate their relationship to at least some extent and could undermine social resources that would otherwise be useful in adaptation. Individuals who have particularly troublesome family relationships may benefit from short-term counseling. One study found that a single-session, client-centered intervention produced significant decreases in family conflict among individuals tested for BRCA1 mutations (McInerney-Leo et al., 2005).

Issues of participant self-selection and the inability to randomly assign groups should also be considered when interpreting the results. As reported in a study by Hughes et al. (2002), the most common reason why women did not divulge test results to sisters was because of strained relationships. By requiring that siblings knew each other's correct result to participate in our study, we likely excluded the most conflicted dyads. Although we covaried certain factors in a subset of analyses, the lack of randomization between the three dyad groups does not allow us to make definitive statements about causal relationships between mutation carrier status and interpersonal relationships. Even though we examined relationships between outcome variables and gender, the potential effects of gender distribution are of particular note since gender was unequally represented among dyad types.

Another limitation of this study is the relatively small sample size, which likely affected the type and strength of conclusions made from the data. First, the sample size may have limited power to detect significant associations between certain variables. In addition to the theoretical issues noted earlier, a portion of hypothesized differences that were not found could be related to insufficient power. Second, the overall sample size and limited sub-sample sizes likely reduced the ability to completely parse out the effects of certain variables, such as gender. Third, the limited sample size may also be of concern given the number of analyses that were undertaken in the study. Since this study explored issues that had not been previously addressed in this context, we wanted to focus on a number of potential comparisons and did attempt to be parsimonious in our predictions and a priori contrasts. However, it should be acknowledged that the number of analyses could limit the interpretations made from the study.

Characteristics of the population should also be taken into account when assessing the generalizability of this study. On average, participants in our sample had known their genetic test results for approximately 3.5 years; it is possible that individuals who were notified of results more recently would respond differently to their siblings. In addition, all participants had received comprehensive genetic counseling as part of their test result disclosure and were also able to address questions with genetics and other medical professionals. It is possible that individuals who do not undergo such extensive counseling would have different types of emotional responses to their own and their family members' test results. External validity may be an issue in that a relatively low percentage of the eligible sample participated in this study. In addition, all participants in our study were Caucasian, many were well-educated, and the vast majority were members of the Church of Jesus Christ of Latter-day Saints (LDS or Mormons). Compared with other populations, Mormons spend more time with extended family members (Barlow & Bergin, 1998) and as such, may have more involved relationships with their adult siblings.

The findings from this study demonstrate that interpersonal processes, affective responses, and at least to some extent physiological activity are related to the combination of genetic test results among siblings. Despite the knowledge that "genetic diseases are family diseases," there is a relative paucity of empirical data in this area of research (Sorenson & Botkin, 2003). Therefore, as genetic testing continues for cancer and other disease

conditions, research on family relationships will become increasingly important in the study of psychosocial processes.

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Figure 1.
Study recruitment flowchart.

Table 1

Demographic, Medical, and Personality Characteristics by Dyad Status

Characteristics	Mixed (<i>n</i> = 20)	Positive (<i>n</i> = 16)	Negative (<i>n</i> = 13)
Age			
<i>M</i>	48.65	47.19	46.96
<i>SE</i>	2.86	3.20	3.55
Time (in years) since testing *			
<i>M</i>	3.48	3.03	4.12
<i>SE</i>	0.28	0.31	0.35
Neuroticism			
<i>M</i>	29.67	29.27	29.12
<i>SE</i>	1.31	1.44	1.54
Agreeableness			
<i>M</i>	48.74	50.78	47.65
<i>SE</i>	1.05	1.15	1.23
Gender **			
Female/female	12	14	5
Other	8	2	8
Cancer history			
Present	10	9	4
Absent	10	7	9

*
p < .10.**
p < .05.

Table 2

Adjusted Mean Scores of Supportive Actions, Affective Reactions, Interpersonal Perceptions, and Electrodermal Responses by Dyad Type

	Dyad type		A priori contrasts	
	Mixed (M)	Positive (P)	Negative (N)	Contrast 1 Contrast 2
SAS-C friendliness	9.37 (.34)	10.02 (.36)	10.61 (.41)	M < (P, N) * P = N
SAS-C dominance	2.78 (.63)	3.52 (.67)	0.48 (.72)	M = (P, N) P > N **
Change in anger	0.96 (.27)	-0.41 (.29)	0.01 (.32)	M > (P, N) ** P = N
Change in anxiety	0.94 (.47)	0.85 (.49)	0.62 (.54)	M = (P, N) P = N
IMI-C friendliness	4.69 (.15)	5.54 (.16)	5.00 (.19)	M > (P, N) ** P > N *
IMI-C dominance	-.86 (.09)	-1.07 (.10)	-1.25 (.11)	M < (P, N) * P = N
Change in SCR	2.24 (.36)	2.17 (.39)	1.06 (.45)	M = (P, N) P = N

Note. SAS-C = Supportive Actions Scale-Circumplex; IMI-C = Impact Message Inventory-Circumplex; SCR = Skin Conductance Response. Standard errors are in parentheses. In Contrast 1, positive and negative dyad types were combined (and compared to mixed dyads). In Contrast 2, positive and negative dyad types were compared to each other.

* $p < .05$.

** $p < .01$.