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ORIGINAL RESEARCH

Genetic test reporting of *CDKN2A* provides informational and motivational benefits for managing melanoma risk

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Abstract

A *CDKN2A/p16* mutation confers 28%–67% lifetime melanoma risk, a risk that may be moderated by ultraviolet radiation exposure. The aim of this study was to test whether melanoma genetic counseling and test disclosure conferred unique informational, motivational, or emotional benefits compared to family history-based counseling. Participants included were 114 unaffected members of melanoma-prone families, ages 16–69, 51.8% men, 65.8% with minor children or grandchildren. Carriers (n = 28) and noncarriers (n = 41) from families with a *CDKN2A* mutation were compared to no-test controls (n = 45) from melanoma-prone families without an identifiable *CDKN2A* mutation. All participants received equivalent counseling about melanoma risk and management; only *CDKN2A* participants received genetic test results. Using newly developed inventories, participants rated perceived costs and benefits for managing their own and their children's or grandchildren's melanoma risk 1 month and 1 year after counseling. Propensity scores controlled for baseline family differences. Compared to no-test controls, participants who received test results (carriers and noncarriers) reported feeling significantly more informed and prepared to manage their risk, and carriers reported greater motivation to reduce sun exposure. All groups reported low negative emotions about melanoma risk. Parents reported high levels of preparedness to manage children's risk regardless of group. Carrier parents reported greater (but moderate) worry about their children's risk than no-test control parents. Women, older, and more educated respondents reported greater informational and motivational benefits regardless of group. Genetic test results were perceived as more informative and motivating for personal sun protection efforts than equivalent counseling based on family history alone.

Keywords

Familial melanoma, *CDKN2A/p16*, Genetic counseling and testing, Ultraviolet radiation exposure, Sun-protection behavior, Skin self-examinations

Implications

Practice: Genetic testing with genetic counseling for patients with a family history of melanoma and known gene mutation promotes education about risk factors and management strategies while empowering them to adhere to prevention recommendations to minimize cancer risk.

Policy: Genetic testing should be offered to at-risk patients in the context of genetic counseling, which should include review of personal and family history to ensure appropriateness of genetic testing, education, informed consent, and tailored management recommendations.

Research: Future research should be aimed at (i) understanding why melanoma genetic test results seem to be impactful over and above the effects of counseling based on family history alone and (ii) identifying strategies to personalize counseling for unaffected individuals who have a significant family history of cancer but no identifiable gene mutation in affected relatives in order for this cohort to reap similar benefits of personalized care as those who can be offered clinical genetic testing.

INTRODUCTION

Predictive genetic testing has the potential to alert members of high-risk families to their highly elevated risk for disease. For hereditary cancers like breast and colon cancer, genetic testing promotes accelerated screening and prophylactic surgery to reduce or eliminate cancer risk [1, 2]. There are mixed findings as to whether and under what conditions predictive genetic testing may increase motivation and performance of regular prevention behaviors that may reduce cancer and other disease risks [3–5]. These efforts have focused on a small set of cancer syndromes (familial melanoma, lung cancer) in which elevated genetic risk may be amplified by either personal behavior or environmental exposure. For example, the *CDKN2A/p16* mutation, which accounts for 20%–45% of familial melanomas [6, 7], confers up to 76% lifetime risk to U.S. residents [8]. Geographic variations in the penetrance of *CDKN2A* suggest that ultraviolet radiation exposure may contribute to these risks [8]. Thus, in addition to regular monthly skin self-examinations (SSEs) and annual professional total body skin examinations (TBSEs), members of melanoma-prone families are advised to reduce sun exposure.

In the case of familial melanoma, initial evidence suggests that genetic test reporting of *CDKN2A* paired with counseling about risk and its management may promote improvements in both photoprotective and screening behaviors. A study randomly assigning patients and family members to *CDKN2A* and high-risk *MC1R* allele testing versus a usual care condition (mailed brochure about skin cancer prevention) indicated more recent SSEs and a smaller decrease in protective clothing use in the testing group 4 months later [9]. However, of the 20 participants who underwent testing, only 5 carriers were identified (only 3 of whom had *CDKN2A* mutations), limiting conclusions about the impact of a positive test result. In a prospective study of melanoma genetic test reporting in a larger cohort of *CDKN2A* family members, unaffected carrier family members reported improvements in daily routine sun protection, including increased reports of protective clothing use and decreased sunburns over a 2-year period [10]. Following test reporting, unaffected carriers also reported SSE behaviors comparable in frequency and thoroughness to family members who had a personal history of melanoma [11]. Qualitative accounts of participant-reported costs and benefits assessed 1, 6, and 12 months after test reporting were similarly encouraging, with 95% of participants reporting one or more positive aspects of learning their genetic test results in the year following test reporting [12]. Specifically, the most frequently reported benefits were increased knowledge about melanoma risk and its management (78.3% of carriers, 95.2% of noncarriers) and improved health behaviors or plans to improve prevention and/or screening behaviors (65% of carriers, 38.1% of noncarriers). Emotional benefits of test reporting, including feeling more at ease about one's own or one's children's risk, were more likely to be reported by noncarriers (71.4%) than carriers (26.1%). Importantly, reports of negative outcomes of test reporting were low, with only 15.9% of participants reporting a negative aspect at any assessment (typically, discouragement, frustration, or insurance concerns). These findings concerning the empowering effects of genetic testing are similar to qualitative accounts provided by unaffected family members undergoing genetic testing for breast and ovarian cancer [13, 14].

The present study

Melanoma genetic testing has only recently entered clinical practice [15–17]. Although results to date suggest several benefits and few costs of melanoma genetic testing, the beneficial impact of the test result itself on perceived informational, motivational, and behavioral benefits has yet to be distinguished from the detailed education about melanoma risk and its management that must ethically be provided with it. Given the importance of demonstrating a benefit of genetic testing beyond counseling alone in motivating prevention behaviors, the primary purpose of the Utah BRIGHT (Behavior, Risk Information, Genealogy, and Health Trial) Project was to identify participant-reported costs and benefits uniquely associated with receiving a melanoma genetic test result. BRIGHT focused on unaffected members of melanoma-prone families, as they report low levels of adherence to sun protection and screening [18, 19]. We used a nonexperimental control group to compare genetic counseling outcomes among unaffected members of families known to carry a *CDKN2A* mutation (*carriers* and *noncarriers*) who received genetic counseling paired with *CDKN2A* test reporting to outcomes among no-test controls. *No-test control participants* were unaffected members of high-risk families (defined as those with at least three affected first- or second-degree relatives [FDRs and SDRs]) known not to carry a *CDKN2A* mutation who received nearly equivalent counseling based on family history alone. At 1 month and 1 year following genetic counseling, we examined whether melanoma genetic counseling and test disclosure conferred unique informational, motivational, or emotional costs or benefits compared to counseling based on family history alone.

We predicted that genetic counseling for familial melanoma risk accompanied by a *CDKN2A* test report would be perceived as more informative to unaffected members of melanoma-prone families than counseling based on family history alone. A genetic test result is both objective and highly personalized; moreover, it provides a concrete, irrefutable explanation of *why* one is at elevated risk [20]. Receiving an explanation for one's risk has been found to increase the acceptance of health risk information, and our initial findings suggested that participants receiving a melanoma genetic test result reported greater understanding of their risk, decreased derogation of the accuracy of the risk information, and greater perceived personal applicability of prevention recommendations compared to no-test controls [20]. For the same reasons, provision of genetic test results may also improve motivation to perform protective and screening measures. One might expect that genetic counseling accompanied by a positive genetic test result would be more distressing than equivalent risk counseling based on family history alone; however, there is little evidence for sustained distress following genetic test reporting for melanoma or other hereditary cancers [1, 2, 12, 21]. Therefore, we evaluated multiple potential positive and negative emotional outcomes, both short-term (1 month after counseling) and longer-term (1 year after counseling).

We also sought to extend prior research on the impact of genetic counseling and test reporting for familial cancer by developing an inventory of perceived costs and benefits for the management of children's risk. Learning about one's children's

risk is a primary motive for pursuing genetic testing [22–24]. We were specifically interested in parents' and grandparents' efforts to manage familial risk [25]. Sun-protection behaviors in children and teens are especially important, as childhood and adolescent sunburns are thought to play a major role in the etiology of melanoma [26, 27]. Further, monthly SSEs are recommended to start as young as age 10 in high-risk families [28]. Parallel analyses were, therefore, performed to evaluate perceived costs and benefits of genetic counseling for managing children's or grandchildren's risk.

Thus, this study is the first to directly examine the motivational and informational benefits that may distinguish high-penetrance melanoma genetic test reporting from receipt of comparable risk education without an accompanying genetic test result. Further, we developed a new inventory of perceived costs and benefits that can be used in future research examining the effects of melanoma genetic counseling and test reporting.

MATERIALS AND METHODS

The Utah BRIGHT project

Inclusion criteria and participant recruitment and retention

BRIGHT was designed to provide a comprehensive assessment of the impact of melanoma genetic test reporting and counseling on health cognitions and prevention behaviors among unaffected members of melanoma-prone families compared to equivalent counseling based on family history alone. Unaffected members of two kinds of melanoma-prone families (those known to carry a *CDKN2A* mutation and those with a significant family history of melanoma but no identifiable mutation) were recruited to a study with four visits over a 13-month period. Inclusion criteria were ages 16–70, no prior melanoma genetic testing or counseling, and no personal history of pathology-confirmed melanoma or melanoma in situ [20]. Participants were compensated $50 per visit with additional compensation for travel costs. BRIGHT was approved by the University of Utah IRB.

Families were identified through Familial Melanoma Research Clinic and Family Cancer Assessment Clinic at Huntsman Cancer Institute. They were offered participation if a *CDKN2A* mutation had been identified in the family or if there were three or more cases of melanoma in the family and genetic testing on an affected member had ruled out a *CDKN2A* mutation. While there are other lower-penetrance genetic factors associated with melanoma risk, only families with *CDKN2A* mutations and those with comparably high risk of melanoma based on family history were included in order to ensure that the two kinds of families were as similar as possible and could receive equivalent counseling about risks and management. Eligible, unaffected relatives were identified through referrals from relatives affected with melanoma, positive for a *CDKN2A* mutation or previously enrolled in research. Of the 167 unaffected family members offered participation, 130 (77.8%) enrolled and attended the initial baseline visit. Of these, one declined to learn his or her genetic test results (participant was already seeing a dermatologist and believed that the test result would not influence his or her care) and one withdrew from the study due to unrelated health issues but received results by phone. Three additional participants withdrew prior to the 1-month follow-up due to either time constraints or an out-of-state move, and one did not attend. Of those who attended the 1-month but not the 1-year follow-up, two withdrew due to time constraints, one withdrew due to unrelated health issues, and seven did not attend. Thus, follow-up surveys concerning perceived costs and benefits of genetic counseling were completed in the clinic by 124 participants (95.4%) 1 month following genetic counseling and by 114 participants (87.7%) 1 year following counseling.

Genetic counseling and test-reporting procedures

All participants received individual genetic counseling from one of two Certified Genetic Counselors working from a structured protocol [20]. For members of *CDKN2A* families, counseling was provided in two visits: a pre-test counseling session to review basic information regarding melanoma and site-specific genetic testing and to obtain consent for genetic testing and a post-testing session in which results were reported. All testing was performed in a Clinical Laboratory Improvement Amendments-certified laboratory. For members of no-test control families, all counseling took place in a single visit (their second) as no consent for testing was needed. All counseling sessions included a brief review of participants' self-reported medical and family health history and education about contributors to melanoma risk including environmental and phenotypic factors as well as high-risk genes. With the exception of the more specific risk estimates afforded by genetic testing results, all information about melanoma risk and its management provided to participants was identical. Carriers were counseled that they had tested positive for a *CDKN2A* mutation and thus had a 70 in 100 risk for melanoma in their lifetime, while non-carriers were counseled that they tested negative for the *CDKN2A* mutation but still had a moderately increased lifetime risk of 2 in 100 based on their family history and other risk factors. No-test controls were provided a range of lifetime risk for melanoma of "30 in 100 to 70 in 100" based on their family history. Detailed recommendations concerning sun protection (avoidance of peak ultraviolet radiation hours, sunscreen, protective clothing, shade-seeking) and screening (monthly SSEs and annual TBSEs) were presented to all participants. Carriers

of *CDKN2A* mutations were also informed about the associated pancreatic cancer risk and options for screening. Few patients met the International Cancer of the Pancreas Screening consortium guidelines for screening [29] based on age or family history at the time of the study; therefore, analysis of outcomes focused on those associated with melanoma risk.

Measures

Perceived costs and benefits of genetic counseling for management of melanoma risk

Structured inventories were developed based on the findings of prior studies [12–14, 22, 30, 31] to capture participants' ratings of four distinct sets of costs and benefits of melanoma genetic counseling for the management of (i) their own melanoma risk (35 items) and, as applicable, (ii) their children's and grandchildren's melanoma risk (24 items, 1 = *not at all true*, 5 = *very much true*). Items were written to assess i*nformational/preparedness benefits*, *motivation to reduce sun exposure*, *motivation to perform skin screening*, and *emotions about melanoma risk*. All items and their mean endorsement across the sample at each assessment are shown in Tables 1 and 2.

Propensity score calculations

To strengthen the validity of the comparisons in this nonexperimental control group design by accounting for measured baseline differences between *CDKN2A* and no-test control families, we calculated propensity scores [32] based on 18 variables that might (i) differ between families at study entry and (ii) influence responses to melanoma risk counseling. All subsequent analyses of group differences statistically controlled for these scores. Included in the propensity score calculations were standard demographic factors (age, gender, education, household income), family medical history (FDR and SDR with melanoma, earliest age of onset among FDR and SDR), phenotypic factors linked to melanoma risk (total nevi greater than 2 mm, clinician-rated Fitzpatrick skin type), prior behavior relevant to melanoma risk and prevention (prior biopsy, prior TBSE, blistering sunburns before age 20, clinician-rated photodamage at study entry), baseline behavior with respect to photoprotection and screening, perceived risk, prioritization of risk, and family communication about melanoma risk and its management. Of note, prior to adjustment with propensity scores, *CDKN2A* families and no-test controls differed on only two of these variables: *CDKN2A* families had an earlier age of melanoma onset (29.4 years old vs. 41.1, $F(1,126) = 30.89$, $p < .001$), and no-test control families had a greater approximate number of moles greater than 2 mm as determined by a TBSE at baseline ($M = 54.04$ vs. 33.82, $F(1,126) = 16.34$, $p < .001$). Following adjustment with propensity scores, there were no significant differences between *CDKN2A* and no-test control families, and all standardized mean differences between the families were less than 0.10 [33].

Overview of analyses

We first describe the factor analytic procedures used to refine the scales for assessing perceived costs and benefits of melanoma genetic counseling. Then, we examine whether participants who received *CDKN2A* test results reported greater costs or benefits than no-test controls who received equivalent counseling based on family history. Analyses of personal costs and benefits were conducted with the 114 participants who provided complete data at both assessments; analyses of costs and benefits for the management of children's and grandchildren's risk were conducted with the 59 parents and 16 grandparents who completed both assessments. Each of the cost or benefit scales was subjected to a repeated-measures analysis of covariance (ANCOVA) with Participant Group (carriers, noncarriers, no-test controls) as a between-subjects factor and Time of Assessment (1 month, 1 year after counseling) as a within-subjects factor. Each analysis was statistically controlled for propensity scores and selected covariates (age at study entry, gender, years of education, annual household income, number of FDR with melanoma, and total nevi greater than 2 mm), and all reported means and effect sizes were adjusted for these covariates. Planned comparisons of the adjusted means for carriers versus no-test controls assessed whether the receipt of a positive genetic test result was associated with increased motivation to reduce sun exposure or perform screening or with any emotional costs or benefits compared to equivalent counseling based on family history alone; all adjusted means comparisons report two-tailed tests. With a single exception noted in the text, there were no significant main effects of Time or interactions of Group × Time on any outcome, suggesting that perceived costs and benefits reported at 1 month were maintained at the 1-year follow-up.

RESULTS

Participant characteristics

Seventy-five members of *p16* families (31 carriers and 44 noncarriers) and 49 members of no-test control families completed the 1-month follow-up survey, and 114 participants (28 carriers, 41 noncarriers, and 45 no-test controls) completed the 1-year follow-up. As shown in Table 3, average age at study entry was 35.82 years, 51.8% were male, and nearly all were White (99.1%), with the majority (79.8%) having Fitzpatrick Skin Type II. Mean education was 14.59 years or "some college." Median annual household income was $60,000–69,000. Nearly two-thirds indicated that they had either children (51.8%) or grandchildren under the age of 18 (14.0%). Participants reported an average of 0.82 FDRs and 1.28 SDRs with melanoma. As noted earlier, prior

Table 1 | Scales, Factor Loadings, and Scale Reliabilities for Ratings of Perceived Costs and Benefits of Melanoma Genetic Counseling for Managing Personal Melanoma Risk at the 1-Month and 1-Year Follow-up

	Factor loading on subscale		Mean (*SD*) of item	
	1 month	1 year	1 month	1 year
Feeling better informed and prepared to manage risk				
Eigenvalue at 1 month = 6.49, 1 year = 6.87; alpha at 1 month = .92, 1 year = .95				
I feel better informed about my melanoma risk.[a]	.85	.88	4.30 (0.98)	4.16 (0.97)
I understand more about why I am at risk for melanoma.	.84	.83	4.27 (0.91)	4.06 (1.09)
I feel better prepared to manage my melanoma risk.	.82	.89	4.09 (0.95)	4.03 (0.98)
I feel I can do something positive about my melanoma risk.[b]	.81	.84	4.21 (0.91)	4.13 (0.95)
I have more accurate information about melanoma.	.77	.75	4.40 (0.82)	4.24 (0.93)
I know more about behaviors that I can do to protect myself in the sun.	.76	.85	4.15 (0.93)	4.04 (1.05)
I feel better informed about the early detection of melanoma.	.74	.88	4.21 (0.97)	4.13 (1.01)
I feel I have the tools to make decisions that will influence my future.[c]	.74	.84	4.19 (0.85)	4.07 (1.05)
I feel better prepared emotionally to deal with my melanoma risk.	.74	.65	3.85 (1.19)	3.69 (1.26)
I feel more certain about my melanoma risk.[b,d]	.73	.65	3.86 (1.12)	3.66 (1.17)
I am more confused about my melanoma risk.	−.30	−.05	1.12 (0.40)	1.15 (1.60)
Motivation to reduce sun exposure				
Eigenvalue at 1 month = 3.10, 1 year = 2.88; alpha at 1 month = .90, 1 year = .87				
I protect myself more from the sun.	.86	.82	3.70 (1.19)	3.90 (1.13)
I am more motivated to reduce my sun exposure.	.86	.87	3.83 (1.21)	3.78 (1.13)
I have made lifestyle changes to reduce my sun exposure.	.84	.81	3.41 (1.26)	3.44 (1.23)
I have a more positive attitude about protecting my skin from the sun.[b]	.79	.67	3.90 (1.16)	3.78 (1.09)
I am less careful about my behavior in the sun.[b]	−.11	−.30	1.15 (0.47)	1.25 (0.60)
Motivation to perform screening				
Eigenvalue at 1 month = 1.59, 1 year = 1.26; alpha at 1 month = .91, 1 year = .89				
I am more motivated to do regular skin self-examinations.	.96	.97	3.73 (1.18)	3.41 (1.27)
I am more motivated to get yearly professional total body skin examinations.	.85	.65	3.86 (1.15)	3.49 (1.22)
I do more thorough skin self-examinations.	.81	.92	3.59 (1.27)	3.36 (1.39)
I am more vigilant about checking my skin for suspicious moles.	.75	.72	3.72 (1.32)	3.58 (1.38)
I have a more positive attitude toward melanoma screening.[b]	.70	.58	4.02 (1.07)	3.85 (1.08)
Items excluded due to loading on a separate factor				
I am less vigilant about doing monthly skin self-examinations.[b]	−.03	−.13	1.48 (0.95)	1.57 (1.01)
I am less vigilant about having annual professional total body skin examinations.[b]	−.01	−.10	1.44 (0.89)	1.62 (1.06)
Negative emotions about melanoma risk				
Eigenvalue at 1 month = 1.48; 1 year = 1.56; alpha at 1 month = .82, 1 year = .79				
I feel discouraged by my melanoma risk.	.80	.81	1.42 (0.87)	1.46 (0.78)
I feel frustrated by my melanoma risk.	.76	.56	1.54 (0.93)	1.60 (0.98)
I feel hopeless about my melanoma risk.	.58	.63	1.28 (0.69)	1.26 (0.67)
Positive emotions about melanoma risk				
Eigenvalue at 1 month = 2.30, 1 year = 2.50; alpha at 1 month = .90, 1 year = .87				
I feel more hopeful about my melanoma risk.	.89	.90	3.28 (1.39)	3.11 (1.22)
I feel greater peace of mind about my melanoma risk.	.89	.76	3.39 (1.41)	3.25 (1.31)
I feel relieved about my melanoma risk.[b]	.81	.85	2.97 (1.43)	3.01 (1.29)
Worry/concern about being in the sun				

(Continued)

Table 1 | Continued

	Factor loading on subscale		Mean (*SD*) of item	
	1 month	1 year	1 month	1 year
Eigenvalue at 1 month = 4.66, 1 year = 4.28; alpha at 1 month = .87, 1 year = .90				
I am more worried about being in the sun.	.87	.95	2.62 (1.38)	2.73 (1.42)
I am more concerned about being in the sun.	.84	.94	3.03 (1.44)	3.11 (1.38)
I feel guiltier about being in the sun.	.78	.69	2.37 (1.29)	2.42 (1.36)
I am more fearful about being in the sun.	.56	.72	2.35 (1.31)	2.51 (1.34)
Fear of screening				
r = .50 at 1 month, *r* = .24 at 1 year				
I am more fearful about doing skin self-examinations.[e]	.49	.70	1.26 (0.74)	1.24 (0.67)
I am more fearful about getting a professional total body skin examination.[e]	.38	.31	1.19 (0.61)	1.30 (0.78)

Items excluded from scales due to low endorsement or low loadings appear in italics. All items were assessed on the following scale: 1 = *not at all true*, 5 = *very much true*. Unless otherwise indicated, items were developed for the present study based on qualitative accounts from participants who received melanoma genetic test results in our initial test-reporting study [12] (N = 124 at 1 month, N =114 at 1 year).
[a]Adapted from the MICRA [31].
[b]Adapted from Lim et al. [13].
[c]From the PPC [30].
[d]Adapted from Kasparian et al. [22].
[e]Adapted from Petersen et al. [50].

to (but not after) adjustment with propensity scores, *CDKN2A* and no-test control families differed on earliest age of onset in the family and total nevi >2 mm but not on any other demographic, medical history, or phenotypic variables (see Table 3).

The participant groups employed to assess differences following *CDKN2A* testing (carriers, noncarriers, no-test controls) did not differ significantly on any of these demographic variables, nor on FDR or SDR diagnosed with melanoma (all *p*s > .10); however, as in our previous study of *CDKN2A* families [34], participant groups differed significantly in approximate number of total nevi greater than 2 mm ($F(2,111) = 15.49$, $p < .001$), such that participants who subsequently received negative test results had fewer such nevi at baseline ($M = 25.46$) than either carriers ($M = 49.21$, $p < .001$) or no-test controls ($M = 55.64$, $p < .001$). There were no group, demographic, family history, or phenotypic differences in attrition at 1 year.

Scale composition and refinement for perceived costs and benefits of genetic counseling

To refine subscale content and groupings, we subjected items comprising each set of potential costs and benefits at each assessment to a separate exploratory factor analysis using a maximum likelihood extraction and oblimin rotation. For each analysis, we examined scree plots to determine the number of factors and inspected factor loadings to determine if any items exhibited low loadings. We retained items with factor loadings greater than .50.

Perceived costs and benefits for the management of personal melanoma risk

As shown in Table 1, 11 items assessed perceptions of being better informed and prepared to manage cancer risk following genetic counseling. Factor analytic results confirmed that this subscale was unidimensional, with all items but one (greater confusion about risk, which received low endorsement) loading greater than .50 at both assessments. The resulting *Better Informed and Prepared to Manage Cancer Risk* scale consisted of 10 items, including being better informed about one's melanoma risk, more emotionally prepared to manage it, and able to do something positive about one's risk.

For *Motivation to Reduce Sun Exposure*, five items assessed greater motivation to reduce sun exposure, implementation of lifestyle changes to reduce sun exposure, and a more positive attitude toward protecting one's skin from the sun. The factor analysis confirmed a unidimensional structure, with all items but one (being less careful about behavior in the sun) loading greater than .50. The remaining four items formed a reliable scale at each assessment.

For *Motivation to Perform Skin Screening*, seven items assessed increases or decreases following genetic counseling. The factor analysis supported a two-factor solution, as items concerning decreased vigilance to SSEs and TBSEs loaded on a separate factor (see Table 1). As the two-factor structure likely reflected method variance (these two items shared a stem—"I am less vigilant"—that was different from other items), these items were not included in the resulting scale. As shown in Table 1, these items also received low endorsement. The remaining five items loaded on a single factor and assessed increased motivation to perform regular and more thorough SSEs and to obtain annual TBSEs.

For emotions about risk and its management, analyses indicated a four-factor solution, with separate

Table 2 | Scales, Factor Loadings, and Scale Reliabilities for Perceived Costs and Benefits of Genetic Counseling for the Management of Children's and Grandchildren's Melanoma Risk at the 1-Month and 1-Year Follow-up

	Factor loading on subscale		Mean (*SD*) of item	
	1 month	1 year	1 month	1 year
Informed and prepared to manage children's risk				
Eigenvalue at 1 month = 3.45, 1 year = 3.75; alpha at 1 month = .89, 1 year = .91				
I feel better prepared to manage my family's melanoma risk.	.92	.96	4.13 (1.09)	4.09 (0.96)
I feel I can do something positive about my children's melanoma risk.[a]	.85	.79	4.23 (0.96)	3.95 (1.00)
I feel better informed about my children's melanoma risk.	.75	.90	4.33 (0.96)	4.19 (1.00)
I feel better prepared emotionally to manage my children's risk.[a]	.74	.77	3.65 (1.36)	3.68 (1.25)
I feel more confused about my children's melanoma risk.	−.07	−.33	1.29 (0.80)	1.33 (0.74)
Motivation to reduce children's sun exposure				
Eigenvalue 1 month = 3.15, 1 year = 3.34; alpha at 1 month = .84, 1 year = .87				
I have made (or my children have made) lifestyle changes to reduce their sun exposure.	.88	.88	3.37 (1.26)	3.40 (1.23)
I am more motivated (or my children are more motivated) to reduce their sun exposure.	.80	.76	3.99 (1.13)	4.01 (1.06)
I protect my children (or my children protect themselves) more from the sun.	.80	.78	3.63 (1.17)	3.83 (1.14)
We have made protection from the sun a family priority.[a]	.67	.75	3.99 (1.06)	4.07 (0.99)
I teach my children more about melanoma prevention.	.50	.66	3.70 (1.42)	3.47 (1.28)
Motivation to screen children				
Eigenvalue at 1 month = 3.45, 1 year = 3.67; alpha at 1 month = .88, 1 year = .90				
I am more vigilant (or my children are more vigilant) about checking their skin for suspicious moles.	.89	.92	3.49 (1.36)	3.31 (1.36)
We have made skin screening a family priority.[a]	.87	.77	3.33 (1.27)	3.08 (1.43)
I am more motivated (or my children are more motivated) to examine their skin regularly.	.84	.94	3.72 (1.33)	3.27 (1.38)
I am more motivated to make sure my children get yearly professional total body skin examinations.	.73	.75	3.42 (1.36)	3.12 (1.29)
I teach my children more about screening.	.57	.68	3.48 (1.39)	3.13 (1.53)
Negative emotions about children's melanoma risk				
Eigenvalue at 1 month = 1.19, 1 year = 2.85; *r* = .75 at 1 month, 1 year = .56				
I am more worried that my children will get melanoma.[a,b]	.95	.56	2.33 (1.45)	2.40 (1.36)
I feel discouraged by my children's melanoma risk.	.69	1.00	1.72 (1.12)	1.80 (1.09)
Positive emotions about children's melanoma risk				
Eigenvalue at 1 month = 2.53, 1 year = 1.56; alpha at 1 month = .81, 1 year = .81				
I feel relieved about my children's melanoma risk.	.86	.50	3.00 (1.60)	2.83 (1.45)
I feel more hopeful about my children's melanoma risk.	.80	.55	3.77 (1.38)	3.67 (1.30)
I am more relaxed about my children's behavior in the sun.[a]	.60	.53	2.43 (1.44)	2.39 (1.37)
Parental reports of children's greater fear of being in the sun				
Eigenvalue at 1 month = 3.18, 1 year = 2.46; alpha at 1 month = .90, 1 year =.85				
My children are more worried about being in the sun.	1.00	.78	2.14 (1.29)	2.27 (1.17)
My children are more concerned about being in the sun.	.83	.79	2.46 (1.32)	2.59 (1.32)
My children are more fearful about being in the sun.	.75	.60	1.89 (1.21)	1.76 (0.98)

Items excluded from scales due to low endorsement or low loadings appear in italics. All items were assessed on the following scale: 1 = *not at all true*, 5 = *very much true*. Unless otherwise indicated, items were developed for the present study based on qualitative accounts from participants who received melanoma genetic test results in our initial test-reporting study [12] (N = 79 at 1 month, N = 75 at 1 year).

[a]Adapted from Lim et al. [13].

[b]Adapted from the MICRA [31].

Table 3 | Baseline Demographic, Family Medical History, and Phenotypic Characteristics of the Study Sample and Comparisons of Participants From *CDKN2A* Families to Participants From No-Test Control Families

	No-test control families (n = 45)	Families known to carry *CDKN2A* (n = 69)	Total sample (N = 114)	p value for difference between types of families[a]
Age	37.84 (14.55)	34.51 (13.82)	35.82 (14.14)	.22
Gender (% men)	46.67%	55.07%	51.75%	.45
Education, years	14.82 (1.95)	14.43 (2.17)	14.59 (2.08)	.33
Household income[b]	6.90 (3.32)	7.38 (3.30)	7.19 (3.30)	.48
Converted value:	$59,000 (23K)	$64,000 (23K)	$62,000 (23K)	
Percent with children or grandchildren under age 18	60%	69.57%	65.79%	.29
FDR with melanoma	0.84 (1.07)	0.81 (0.88)	0.82 (0.95)	.86
SDR with melanoma	1.53 (1.63)	1.12 (1.30)	1.28 (1.45)	.13
Earliest age of onset among FDR and SDR	40.44 (15.03)	29.16 (8.78)	33.61 (12.85)	.001
Total nevi >2 mm[c]	3.44 (0.94)	2.59 (1.13)	2.93 (1.13)	.001
Converted value:	55.64 (27.25)	35.10 (27 .52)	43.21 (29.10)	
Percent with Fitzpatrick skin type II	76.8%	84.4%	79.8%	.17

FDR first-degree relatives; *SDR* second-degree relatives.
[a]These values reflect differences between *CDKN2A* and no-test control families prior to adjustment using propensity scores. There were no significant differences between families when propensity scores were statistically controlled.
[b]The sample for this variable was N = 101 (*CDKN2A* families, n = 61; no-test control families, n = 40) because 13 participants chose not to report (n = 6) or were unsure of (n = 7) their household income. Income was reported in $10,000 intervals (1 = <$9,999, 11 = ≥$100,000), and then converted to an approximate income for ease of interpretation.
[c]Total nevi >2 mm assessed during the clinical skin exam were reported in ranges (0 = 0, 1 = 1–10, 2 = 11–25, 3 = 26–50, 4 = 51–100, 5 = >100) and then converted to an approximate number of nevi for ease of interpretation.

scales for *Negative Emotions about Melanoma Risk* (three items: hopeless, frustrated, and discouraged), *Positive Emotions about Melanoma Risk* (three items: relieved, hopeful, and greater peace of mind), *Worry about Being in the Sun* (four items: worry, concern, guilt, and fear), and *Fear of Screening* (two items: greater fear about doing SSEs or getting a TBSE). As shown in Table 1, the fear of screening items did not meet our criteria for factor loadings (and received low endorsement, *M*s = 1.19–1.30); thus, this scale was dropped from analysis.

Perceived costs and benefits for management of children's or grandchildren's melanoma risk

These procedures were repeated for the items assessing perceived costs and benefits for the management of children's risk. An item assessing increased confusion about children's risk received low endorsement at both visits (*M*s = 1.29 and 1.33) and did not load on the informational benefits factor; thus, it was removed from analyses. As shown in Table 2, the remaining items loaded .50 or greater on their respective factors and were used to create reliable subscales that paralleled those identified for personal costs and benefits. Correlations among the resulting scales are shown in Supplementary Tables 1 and 2.

Group differences in perceived costs and benefits of genetic counseling for the management of personal melanoma risk

We next examined whether there were differences in reported costs and benefits among participant groups when propensity scores and selected covariates were statistically controlled. Correlations between the costs and benefits scales and demographic, family medical history, and phenotypic variables are displayed in Supplementary Tables 3 and 4. Age, gender, education, household income, number of FDR with melanoma, and total nevi greater than 2 mm were significantly associated with one or more outcomes, and thus were statistically controlled in all analyses. Number of SDR with melanoma, earliest age of onset in the family, and skin type were not related to any outcomes and thus not included as covariates.

Feeling informed and prepared to manage melanoma risk

Our primary hypothesis was that genetic testing would have unique informational benefits compared to counseling based on family history alone. As shown in Table 4, the repeated-measures ANCOVA yielded a significant main effect for Group (F(2,104) = 3.54, p < .04). Both carriers (M = 4.29) and noncarriers (M = 4.26) reported feeling more informed and better prepared than no-test controls (M = 3.78, both ps < .03). As shown in Table 4, greater education and income were significant predictors of feeling better informed and prepared.

Motivation to reduce sun exposure

As suggested earlier, melanoma is unique in that preventive behaviors may moderate the impact of genetic risk on disease development. Therefore, the

Table 4 Perceived Costs and Benefits of Melanoma Genetic Counseling for the Management of Personal Melanoma Risk and Children's or Grandchildren's Melanoma Risk as a Function of Participant Group, Age, Gender, Years of Education, Annual Household Income, and Total Nevi >2 mm

	Perceived costs and benefits for the management of personal melanoma risk														
	Carriers (n = 28)	No-test controls (n = 45)	Noncarriers (n = 41)	F group (2,104)	Partial η^2	F age (1,104)	Partial η^2	F gender (1,104)	Partial η^2	F education (1,104)	Partial η^2	F income (1,104)	Partial η^2	F nevi total (1,104)	Partial η^2
Feeling informed and prepared to manage melanoma risk	4.29[a]	3.78[b]	4.26[a]	3.54*	.064	2.72	.025	2.11	.020	4.37*	.040	4.30*	.040	0.00	.000
Motivation to reduce sun exposure	4.15[a]	3.55[b]	3.62[b]	4.06*	.072	10.85***	.094	3.42[+]	.032	6.91**	. 062	0.84	.008	1.27	.012
Motivation to perform screening	3.98[a]	3.49[a]	3.61[a]	2.17	.040	12.71***	.109	4.49*	.041	0.81	.008	1.14	.011	1.81	.017
Negative emotions about melanoma risk	1.78[a]	1.54[a]	1.07[b]	10.26***	.165	2.31	.022	4.26*	.039	0.00	.000	1.40	.013	0.57	.005
Positive emotions about melanoma risk	2.38[a]	2.89[a]	3.93[b]	22.14***	.299	0.50	.005	0.72	.007	0.67	.006	0.23	.002	4.62*	.043
Worry about being in the sun	3.23[a]	2.75[a]	2.18[b]	8.50***	.140	8.67**	.077	5.64*	.051	1.68	.016	0.00	.000	0.54	.005
	Perceived costs and benefits for management of children's/grandchildren's melanoma risk														
	Carriers (n = 17)	No-test controls (n = 27)	Noncarriers (n = 31)	F group (2,65)	Partial η^2	F age (1,65)	Partial η^2	F gender (1,65)	Partial η^2	F education (1,65)	Partial η^2	F income (1,65)	Partial η^2	F nevi total (1,65)	Partial η^2
Feeling informed and prepared to manage children's melanoma risk	4.04[a,b]	3.74[a]	4.35[b]	2.27	.065	0.25	.004	2.60	.038	5.32*	.076	0.09	.001	1.14	.017
Motivation to protect children from sun exposure	3.96[a]	3.49[a]	3.85[a]	1.08	.032	1.15	.017	6.54*	.091	0.49	.007	0.54	.008	0.91	.014
Motivation to screen children	3.72[a]	3.09[a]	3.35[a]	1.15	.034	0.00	.000	4.61*	.066	0.40	.006	1.90	.028	0.05	.001
Negative emotions about children's risk	3.21[a]	2.05[b]	1.43[c]	24.77***	.432	1.38	.021	7.22**	.100	0.07	.001	0.65	.010	0.68	.010
Positive emotions about children's risk	2.21[a]	2.75[a]	4.41[b]	33.36***	.507	0.09	.001	1.00	.015	0.81	.012	0.99	.015	0.29	.004
Parental reports that children are worried about being in the sun	2.34[a]	2.08[a]	2.18[a]	0.27	.008	10.39***	.138	5.51*	.078	0.59	.009	0.59	.009	0.15	.002

Values tabled are the average of reported costs and benefits at 1 month and 1 year (1 = *not at all true*, 5 = *very much true*). All means, F-values and effect sizes reflect the inclusion of propensity scores, age at study entry, gender, years of education, annual household income, number of first-degree relatives (FDR) with melanoma, and total number of nevi >2 mm as covariates in the model. Adjusted means that do not share a superscript are significantly different, $p < .05$. Significant covariate effects for age, gender, education, and income indicate that older or more educated respondents, those with higher income, and women reported greater informational and/or motivational benefits, while those with more nevi reported fewer emotional benefits. Neither propensity scores nor number of FDR with melanoma were a significant predictor of any reported cost or benefit.

$^{+}p < .10$, $^{*}p < .05$, $^{**}p < .01$, $^{***}p < .001$.

impact of melanoma risk counseling on motivation to reduce sun exposure is a primary intervention target. The repeated-measures ANCOVA yielded a significant main effect for Group ($F(2,104) = 4.06$, $p < .02$), such that carriers ($M = 4.15$) reported greater motivation to reduce their sun exposure than either noncarriers ($M = 3.62$, $p < .02$) or no-test controls ($M = 3.55$, $p < .02$). Significant effects of age and education indicated that older and more educated respondents also reported greater motivation to reduce their sun exposure.

Motivation to perform screening

The repeated-measures ANCOVA did not yield a significant main effect for Group ($F(2,104) = 2.17$, $p < .12$); however, planned comparisons yielded a tendency for carriers ($M = 3.98$) to report greater increases in screening motivation than no-test controls ($M = 3.49$, $p < .07$), with noncarriers ($M = 3.61$) intermediate to, and not significantly different from, either group. Women ($M = 3.87$ vs. 3.53 for men, $p < .04$) and older respondents indicated greater motivation to perform screening.

Negative emotions about melanoma risk

Overall reports of negative emotions about melanoma risk were low at both assessments, with the majority of respondents endorsing either 1 = "not true" or 2 = "a little true" for all items (79.82% at 1 month, 83.06% at 1 year). A significant main effect for Group ($F(2,104) = 10.26$, $p < .001$) indicated that carriers ($M = 1.78$) and no-test controls ($M = 1.54$) reported more negative emotions about their risk than noncarriers ($M = 1.07$, ps $< .02$), but did not differ from each other ($p < .18$). Women ($M = 1.59$) reported more negative emotions about their risk than men ($M = 1.35$, $p < .05$).

Positive emotions about melanoma risk

Noncarriers ($M = 3.93$) reported greater hopefulness and peace of mind about their risk than either carriers ($M = 2.38$, $p < .001$) or no-test controls ($M = 2.89$, $p < .001$; main effect for Group: $F(2,104) = 22.14$, $p < .001$). Carriers tended to report lower positive emotions about their risk than no-test controls ($p < .07$). Participants with a greater number of nevi greater than 2 mm were less relaxed and hopeful about their melanoma risk.

Worry about being in the sun

Reported worry about being in the sun was moderate at both assessments (Ms = 2.68, 2.77). The repeated-measures ANCOVA yielded a significant main effect for Group ($F(2,104) = 8.50$, $p < .001$), such that carriers ($M = 3.23$) and no-test controls ($M = 2.75$) reported greater concern about being in the sun than noncarriers ($M = 2.18$, ps $< .05$) but did not differ from each other ($p < .11$). Women ($M = 2.95$ vs. 2.51 for men, $p < .02$) and older respondents reported greater worry.

Group differences in perceived costs and benefits of genetic counseling for the management of children's or grandchildren's melanoma risk

Two-thirds of our study participants reported having either children or grandchildren under 18. There were too few grandparents to permit a formal test of the relation of parental versus grandparental status to perceived costs and benefits, and grandparents were unequally distributed across participant groups (three carriers, four noncarriers, nine no-test controls). However, we conducted an exploratory analysis to examine whether perceived costs and benefits differed for parents and grandparents. As there were no significant main effects or interactions involving parental versus grandparental status, we collapsed across parent status in examining the impact of melanoma genetic counseling on the management of children's/grandchildren's risk, and use the term parents to refer to both parents and grandparents.

Informed and prepared to manage children's risk

As shown in Table 4, parents in all groups reported high levels of preparedness as a benefit of melanoma risk counseling at both assessments (Ms = 4.07, 4.02), with no differences by Group ($F(2,65) = 2.27$, $p < .12$). Those with more years of education reported feeling better informed and prepared to manage their children's melanoma risk, $F(1,65) = 5.32$, $p < .03$.

Motivation to reduce children's sun exposure

Parents reported being somewhat motivated to reduce their children's sun exposure at both assessments (Ms = 3.74, 3.79), with no effect of Group ($F(2,65) = 1.08$, $p < .35$). Women ($M = 4.00$) reported greater motivation than men ($M = 3.46$, $p < .01$) to reduce their children's sun exposure.

Motivation to screen children

Parents reported moderate levels of motivation to screen their children for melanoma at both assessments (Ms = 3.53, 3.25), with no differences by Group ($F(2,65) = 1.15$, $p < .33$). Women ($M = 3.68$) reported greater motivation than men ($M = 3.07$, $p < .02$) to screen their children.

Negative emotions about children's melanoma risk

Carrier parents ($M = 3.21$) reported significantly greater (though moderate) worry and discouragement about their children's risk than either noncarrier ($M = 1.43$, $p < .001$) or no-test control parents ($M = 2.05$, $p < .001$; Group, $F(2,65) = 24.77$, $p < .001$). No-test control parents reported greater worry than noncarrier parents ($p < .05$). Additionally, women ($M = 2.53$) indicated higher levels of worry than

men (M = 1.98, p < .01), though at levels below the scale midpoint.

Positive emotions about children's melanoma risk

A significant effect of Group (F(2,65) = 33.36, p < .001) indicated that noncarrier parents (M = 4.41) reported significantly greater relief and hopefulness about their children's risk than either carrier (M = 2.21, p < .001) or no-test control parents (M = 2.75, p < .001). There was also a marginally significant main effect of Time (F(1,65) = 2.99, p < .09), indicating a trend toward decrease in positive emotions from 1 month to 1 year (3.17, 3.07). These main effects were qualified by a significant Group × Time interaction (F(2,65) = 4.83, p < .02), such that noncarrier parents' reports of positive emotions, while remaining high, decreased significantly from 1 month to 1 year (Ms = 4.71, 4.10, p < .002). Neither carrier (2.24, 2.18, p < .81) nor no-test control parents' positive emotions (2.57, 2.94, p < .12) changed over time.

Parental reports of children's worry about being in the sun

Parental reports that their children were worried about being in the sun were low overall, with responses in the "a little true" range at both assessments (Ms = 2.16, 2.24) and no differences by Group (F(2,65) = 0.27, p < .77). Women (M = 2.46) reported higher levels of children's worry about being in the sun than men (M = 1.91, p < .02). Older parents reported greater worry among their children (F(1,65) = 10.39, p < .002).

DISCUSSION

Consistent with prior findings [9, 12], participants receiving melanoma genetic test results reported high levels of informational and motivational benefits that were sustained over the year following genetic testing. In particular, participants who received positive test results reported being both better prepared to manage their melanoma risk and more motivated to improve their prevention behavior. We extended previous findings by comparing perceived benefits and risks of melanoma genetic counseling between members of melanoma-prone families who received *CDKN2A* genetic test results and members of no-test control families who received equivalent counseling regarding melanoma risk and its management. Compared to counseling based on family history alone, the receipt of a test result (either positive or negative) was associated with significantly greater perceptions of feeling informed and prepared to manage one's own melanoma risk. Of particular importance, carriers reported greater motivation to reduce sun exposure than no-test controls. Additionally, there was a trend for carriers to report greater motivation to perform skin screening than no-test controls.

Although these comparisons suggest the superiority of genetic test reporting paired with counseling to counseling alone in providing specific informational and motivational benefits, the nonexperimental control group design employed here does not permit causal inference. Because participants were not (and could not be) randomly assigned to groups, it is possible that unmeasured differences between the groups accounted for these findings. Propensity scores were used to statistically control for a large set of personal, familial, phenotypic, behavioral, and other contributors to melanoma risk perceptions and beliefs about their management measured at baseline that might differ between *CDKN2A* families and no-test control families, and selected covariates were used to statistically control for demographic, phenotypic, and family medical history factors that were associated with reported costs and benefits. Although a definitive causal conclusion about the impact of melanoma genetic testing awaits a design in which people are randomly assigned to receive results (a design that may not be ethically permissible given professional genetic counseling guidelines [15, 35, 36]), these findings suggest a meaningful and sustained informational and motivational benefit for genetic testing.

Future research should examine *why* a genetic test result seems to be especially motivating and informative to members of melanoma-prone families. These and other findings from studies of reporting results of testing for a high-penetrance melanoma predisposition gene [9–12, 20] are seemingly at odds with reviews suggesting that genetic risk information is ineffective in motivating a wide range of behavioral changes [3–5]. One explanation is that prevention measures for melanoma may be less burdensome and more within an individual's control to change than other behaviors such as smoking cessation. Further, the studies in these previous reviews included the outcomes of genetic reports for lower-penetrance genetic variants delivered to the general public, whereas we delivered counseling about the highly penetrant *CDKN2A* mutation exclusively to participants with extensive family history of melanoma. Receiving genetic information specifically related to risk that had been observed in the family and for which there was specific counseling about the biological link between the impact of the gene mutation (impaired cellular response to DNA damage [37, 38]) and the recommended management behaviors (reduced sun exposure) may have been more impactful than general genetic assessment [39–41]. Specifically, *CDKN2A* is a tumor suppressor gene, and patients were counseled that encoded proteins stop cell growth so that DNA can be repaired or initiate cell death if the damage is too severe to repair. This coherence between the risk-increasing genetic cause identified and the specific risk-reducing behaviors recommended could be one reason that the receipt of a melanoma genetic test result

promoted greater motivation to reduce sun exposure, as well as greater acceptance of prevention and screening recommendations as personally applicable [20]. This link may also explain our previous findings that melanoma genetic test reporting promotes greater perceived control over health outcomes [42].

Much remains to be done to establish whether these informational and motivational benefits may be limited to high-penetrance genetic test results like those for familial melanoma, hereditary breast and ovarian cancer, and Lynch syndrome, or whether genetic testing is similarly uniquely motivating at lower levels of penetrance. Because a genetic test provides an objective, highly personalized risk estimate, we think it is possible that genetic test results could motivate behavioral change in some cases even for lower-penetrance mutations (see, e.g., [43]).

It will also be important to understand socioeconomic, demographic, ethnic group, and cultural factors that may influence responses to genetic testing, as well as properties of the disease risk and specific management recommendations that may make particular costs and benefits more likely [1]. In the present study, greater years of education reliably predicted feeling better informed and prepared to manage one's own and one's children melanoma risk following genetic counseling and greater motivation to reduce one's own sun exposure, while greater household income predicted feeling better informed and prepared to manage personal risk. Greater benefits (and also greater worry about sun exposure) were consistently reported both by women and by older respondents. Because nearly all study participants were white, no generalization can be made to other ethnic groups. Little is known about how Hispanics or Blacks respond to melanoma genetic risk information, although an ongoing study examining Hispanic versus non-Hispanic ethnicity and sociocultural factors as predictors of uptake of and responses to *MC1R* testing will begin to address this question [44]. People of color have been traditionally overlooked in sun-protection research and intervention efforts more broadly, and it is possible that genetic counseling and testing for lower-penetrance genes may help to combat the perception that people with darker skin types are exempt from melanoma risk [45, 46].

As research on the motivational and informational impact of genetic counseling proceeds, it will be important to understand why equivalent counseling based on family history alone does not seem to be as motivating of prevention behaviors. All participants faced an exceptionally elevated risk of melanoma due to their family history and all participants received detailed individual counseling from a Certified Genetic Counselor, yet subsequent motivation to reduce sun exposure was reliably lower among no-test controls than among *CDKN2A* carriers. Future research should examine ways to provide genetic counseling communications about risk and its management in order to motivate high-risk participants for whom clinical genetic testing is not available.

Understanding the emotional costs and benefits of counseling about hereditary cancer risk

Consistent with our prior work on melanoma genetic testing [12], participants who tested positive did not report elevated negative emotions about their melanoma risk either 1 month or 1 year following counseling and test reporting. They did, however, report elevated concern about their children's risk, although these levels of concern were intermediate rather than high. Both carriers and no-test controls reported a greater level of worry and concern about being in the sun than noncarriers, although these levels, too, were intermediate rather than high. Future studies should investigate the relationship between increased worry about being in the sun and the adoption and maintenance of sun-protection behaviors, as worry may prompt people who receive a positive melanoma genetic test result to be more vigilant about sun exposure. In some contexts, cancer worry promotes consistent prevention and screening behaviors [47, 48], and in the present study greater worry about being in the sun was associated with greater motivation to engage in sun protection and screening. Last, it is important to recognize the considerable emotional benefits reported by noncarriers, both with respect to their own melanoma risk and the risks of their children and grandchildren. Although most studies focus on questions of adherence among family members who test positive, greater peace of mind and relief provided to noncarrier family members is also an important intervention outcome [12].

Utility of the new perceived costs and benefits scales

We created reliable multi-item measures of perceived informational and motivational benefits of counseling regarding melanoma risk for the management of one's own and one's children's melanoma risk, along with positive and negative emotions about melanoma risk. These scales may supplement other commonly used measures assessing outcomes of genetic testing [31, 49, 50]. Previous measures have tended to focus on negative outcomes, such as uncertainty, diminished self-worth, stigma, and distress [1]. Our scales—and the present data—suggest that positive outcomes including benefits such as greater knowledge and feelings of empowerment are as common, if not more common, than negative outcomes [30, 49, 50]. Importantly, these measures (like many of those assessing adverse outcomes) were designed based on participants' qualitative reports in prior research [1, 12] rather than solely reflecting researcher-generated concepts. Further, the use of structured inventories allows the collection of participant ratings of the extent to which a benefit or cost is

perceived (rather than whether it was mentioned or not), and these ratings may be obtained from all participants, not just those for whom a particular cost or benefit came to mind in response to an open-ended question. These measures may also prove useful in evaluating the outcomes of different modalities of providing genetic counseling results, as they may be sensitive to different degrees of motivation to perform prevention and screening behaviors following an intervention.

In interpreting our findings regarding perceived costs and benefits of melanoma genetic counseling, it is important to note that the intensive individual clinic-based counseling provided here by Certified Genetic Counselors represents a best-case scenario for the education accompanying melanoma genetic test results. All groups demonstrated low endorsement of items related to confusion about risk or decreased motivation to reduce sun exposure and monthly and annual screening, indicating gains in knowledge related to the educational component of the intervention. It would not be appropriate to conclude that melanoma genetic test reporting that is not accompanied by melanoma genetics education would afford the same benefits. As research proceeds on the effectiveness of other methods of delivering genetic information for more common, moderate risk alleles [43], these scales may be a useful tool in examining melanoma education outcomes in these other less intensive intervention contexts.

Strengths and limitations of the present study

The present study employed a sample twice as large as our initial test-reporting study [18], enrolling 130 unaffected members of melanoma-prone families and retaining 87.7% of them through two waves of follow-up data in the year following genetic counseling. Importantly, participants in the present study also had much less prior engagement with studies in the Familial Melanoma Research Clinic than participants in our prior work, reducing concerns that the benefits reported here reflect decades of investment of time and energy in melanoma genetics research. A nonexperimental no-test control group was used and propensity scores, incorporating 18 potential differences that could be related to subsequent motivational and emotional responses to genetic counseling, were calculated to control for potential baseline differences in measured variables between the two kinds of families. Nonetheless, in any design without random assignment to the intervention versus control arm, it is always possible that unmeasured factors may account for reported group differences. Additional limitations include those inherent to the study of familial melanoma, namely the inclusion of predominantly white participants. High levels of income and education, along with cultural endorsement of science as a tool for improving health among Utah's predominant religious group [51], may limit generalization to other groups. Finally, with respect to understanding how families manage risk, there were too few grandparents enrolled in the study to permit a firm conclusion about whether melanoma genetic counseling has similar effects on parents and grandparents in terms of their management of minor family members' melanoma risk.

CONCLUSION AND DIRECTIONS FOR FUTURE RESEARCH

The present findings suggest that adding a high-penetrance melanoma genetic test result to individualized melanoma genetic counseling provides both informational and motivational benefits to members of high-risk families. Parents reported high levels of preparedness to manage children's risk regardless of group. Understanding how information about genetic vulnerability to cancer informs and motivates prevention behaviors, both for oneself and one's children, is an important future direction for research and intervention. Future studies might examine the ways family discussions and action plans may unfold differently when there is an identifiable contributor to risk (here a genetic mutation) rather than a more abstract sense of family risk. As this work proceeds, it will also be important to test whether similar benefits may be observed for reporting of lower-penetrance genetic risks and in less intensive intervention contexts.

SUPPLEMENTARY MATERIAL

Supplementary material is available at *Translational Behavioral Medicine* online.

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Compliance with Ethical Standards

Statement on any previous reporting of data: Findings reported in this manuscript have not been previously published. This manuscript is not being simultaneously submitted elsewhere. There is no overlap in study outcomes between an initial publication from the BRIGHT study [20] and the present manuscript.

Primary data: The authors have full control of all primary data and agree to allow the journal to review our data if requested.

Statement on any previous reporting of data: Findings reported in this manuscript have not been previously published. This manuscript is not being simultaneously submitted elsewhere. There is no overlap in study outcomes between an initial publication from the BRIGHT study [20] and the present manuscript.

Primary data: The authors have full control of all primary data and agree to allow the journal to review our data if requested.

Conflict of Interest: S.A. Leachman has served in the past on a Medical and Scientific Advisory Board for Myriad Genetics Laboratory and Castle Biosciences Inc., for which she has received an honorarium. She has collaborated with Myriad to test assays as part of an early access program that is unrelated to the research reported here. W. Kohlmann received a $25,000 research grant from Myriad Genetics Laboratory in 2016 to study the psychological and family communication outcomes of multigene panel testing. That work is unrelated to the research reported here. Authors L.G. Aspinwall, T.K. Stump, J.M. Taber, D.M. Drummond, and M. Champine declare that they have no conflict of interest.

Ethical Approval: All procedures involving human subjects were reviewed and approved by the University of Utah's Institutional Review Board. There were no animal participants in any aspect of the research.

Informed Consent: All participants provided informed consent prior to participation.

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