Journal of Community Genetics (2020) 11:321–329
https://doi.org/10.1007/s12687-020-00453-9

ORIGINAL ARTICLE

Parent and child perspectives on family interactions related to melanoma risk and prevention after CDKN2A/*p16* testing of minor children

Yelena P. Wu[1,2] · Lisa G. Aspinwall[3] · Bridget Parsons[2] · Tammy K. Stump[4] · Katy Nottingham[2] · Wendy Kohlmann[2,5] · Marjan Champine[2,6] · Pamela Cassidy[7] · Sancy A. Leachman[7,8]

Received: 11 September 2019 / Accepted: 14 January 2020 / Published online: 18 January 2020

Abstract

Predispositional genetic testing of children for adult-onset health risks is typically only used when prevention and screening measures have utility during childhood. Little is known about how children and their parents may use predispositional risk information, including whether it changes their interactions around risk-reducing prevention and screening behaviors. The current study examined perspectives on family interactions around skin cancer prevention and control practices through 1 year after test reporting and counseling among children who received melanoma predispositional genetic testing and their parents. Eighteen children (50% carriers, 56% male, mean age = 12.4 years) and 11 parents from 11 families participated in semi-structured interviews 1 month and 1 year after receiving the child's test result. Both parents (73%) and children (50%) reported making changes to family skin cancer prevention and control practices after receiving the test result. Parent- and child-reported discussions about melanoma prevention increased over time (36% parents and 61% children at 1 month, 73% parents and 67% at 1 year). One-quarter (27%) of parents and no children reported having conflicts about sun protection or screening 1 year after test reporting. A majority of parents (63%) reported treating their child differently at the 1-year follow-up, especially among carriers. Predispositional genetic testing for melanoma was associated with reported changes to plans for and discussions about sun protection, and high levels of parent-child collaboration to implement child sun protection. Future work could seek to identify child and parent factors and interactions that predict improved prevention and screening behaviors following pediatric predispositional genetic testing.

Keywords Predispositional genetic testing · Children · Melanoma · Prevention · CDKN2A/*p16*

✉ Yelena P. Wu
Yelena.Wu@utah.edu

[1] Department of Dermatology, University of Utah, Salt Lake City, USA

[2] Huntsman Cancer Institute, 2000 Circle of Hope, Rm 4509, Salt Lake City, UT 84112, USA

[3] Department of Psychology, University of Utah, Salt Lake City, USA

[4] Department of Preventive Medicine, Northwestern University Feinberg School of Medicine, Chicago, USA

[5] Population Health Sciences, University of Utah, Salt Lake City, USA

[6] Ancestry DNA, Lehi, USA

[7] Department of Dermatology, Oregon Health & Science University, Portland, USA

[8] Knight Cancer Institute, Oregon Health & Science University, Portland, USA

Predispositional genetic testing can inform individuals of their risk for one or more diseases, and provides a context within which disease-specific preventive recommendations are provided. Predispositional genetic testing of children for adult-onset diseases is generally only recommended when positive test results have direct implications for preventive services or behaviors that can be implemented during childhood (Botkin et al. 2015; Ross et al. 2013). For example, predispositional genetic testing has been used to provide information on risk for early-onset cancers or tumors, such as in the case of familial adenomatous polyposis, retinoblastoma, and multiple endocrine neoplasia type 2 (Brandi et al. 2001; Leoz et al. 2015; Rao et al. 2008). Recently, we reported the first study of provision of CDKN2A/*p16* (referred to as "*p16*" hereafter) testing to minors (Stump et al. 2018).

Mutations in *p16* are associated with significant increases in certain cancer risks, including a 28–76% lifetime risk for

melanoma (Begg et al. 2005; Bishop et al. 2002). Strategies to prevent melanoma, such as use of sun protection (e.g., sunscreen, protective clothing) that decreases ultraviolet radiation (UVR) exposure, are important for *p16* mutation carriers, due to evidence that environmental factors (e.g., UVR exposure) are associated with increased melanoma penetrance (Bishop et al. 2002). In addition, melanoma prevention strategies are particularly important to implement during childhood because UVR exposure and the occurrence of severe sunburns early in life are the primary modifiable risk factors for melanoma later in life (Balk 2011; Williams et al. 2011).

The literature suggests that children who receive predispositional genetic testing for a variety of health risks are not, on average, distressed as a result of the testing, and could experience benefits (Botkin et al. 2015; Stump et al. 2018; Wakefield et al. 2016). For example, children who received *p16* testing and their parents reported that children had low levels of anxiety, depressive symptoms, and cancer worry, and improvements in use of sun protection and lower sunburn occurrence regardless of mutation status (Stump et al. 2018). Prior prospective studies have generally focused on children's psychological adjustment to test results in the early months after testing. Much less is known about how parents and children use information on the child's genetic risk, and how family interactions may change over time as a result of knowing the child's genetic risk for a future disease (Wade et al. 2010; Wakefield et al. 2016; Wu et al. 2016). A few studies have examined children's perspectives on family interactions following testing (e.g., for Duchenne muscular dystrophy and other diseases with symptoms presenting during childhood), and these results indicate that the majority do not perceive changes, such as being treated differently by parents or siblings (Järvinen et al. 2000a, b). In the case of pediatric testing for mutations in *p16* where the health condition for which children are at risk is not expected to present until adulthood, it is currently unknown what family interactions and practices related to risk-reducing behaviors may change in the months following receipt of test results. For example, it is possible that families may discuss prevention and screening behaviors more frequently after receiving test results, parents may be more vigilant about the child's prevention and screening behaviors which could lead to increased parent-child conflicts, or parents may choose to share childrens' test results with other family members in order to garner support in implementing child sun protection strategies.

Better understanding parents' and children's perspectives on consequences associated with pediatric predispositional genetic testing for adult-onset conditions could inform development of family-focused approaches to providing information on children's disease risks and to promote healthy adaptation to knowledge of this risk. The current study focused on a qualitative exploration of parent and child perspectives on changes to, family interactions about, and future plans related to skin cancer prevention and control practices through 1 year after children and their parents received information on the child's *p16* status.

Methods

Participants and procedures

Children were eligible to participate in the current study if they (1) had a parent with a confirmed *p16* mutation, (2) were between the ages of 10–15 years, (3) did not have a personal history of melanoma, and (4) had not already received melanoma genetic testing. Potentially eligible children were identified using research records from studies involving melanoma-prone families.

Families were asked to participate in an in-person baseline visit that included pre-test counseling, an in-person counseling and test disclosure visit 2–4 weeks post-baseline, a 1-week phone follow-up (1-week post-counseling and test disclosure), an in-person 1-month follow-up visit 1-month post-counseling and test disclosure, and an in-person 1-year follow-up, a 1-year post-counseling and test disclosure. Of the 23 eligible children, 20 (87% recruitment rate) enrolled in the current study along with a parent and completed the baseline visit. One family (two children and their parent) withdrew prior to the 1-month follow-up due to their busy schedule. The 18 remaining children, from 11 families, were included in the current analysis and participated in all assessments reported in the current analysis.

Additional details on recruitment and study methods are provided elsewhere (Stump et al. 2018). In brief, the genetic counseling protocol included two sessions with a Certified Genetic Counselor. At the pre-test genetic counseling session, all children enrolled from the same family along with at least one parent met with a genetic counselor to review information on melanoma and *p16* genetic testing. This information was provided to families along with an opportunity to ask questions, so that families could make a decision about whether the children would undergo *p16* testing. All families participating in the pre-test genetic counseling decided to proceed with *p16* testing. The post-test genetic counseling session and test disclosure was completed separately for each child along with at least one parent. Genetic counselors briefly reviewed information on melanoma and *p16* testing and provided the child's test result. Information reviewed included an overview about genes/*p16* (e.g., everyone has the *p16* gene, some people have one copy of the gene that is not working which results in a higher chance of developing melanoma). If participants tested negative, the information given included that they do not have the non-working gene, and their chance of getting melanoma is about the same as other children. Regardless of their mutation status, all children received information on how to reduce

sun exposure and check their skin. Children who tested positive also received a recommendation to obtain an annual total body skin examination by a healthcare provider. All materials were created to be developmentally appropriate for children.

Children and parents were provided with gift cards in appreciation for their time as well as travel compensation if the family resided more than 50 miles away from the cancer center. All study procedures were approved by the relevant Institutional Review Board.

Demographic measure

Parents were asked at the baseline assessment to report on childrens' and the family's demographic information, including the child's age, sex, race/ethnicity, and color of untanned skin, the parent's sex and personal melanoma history, and household income. Parents were also asked during the screening process to identify first- and second-degree relatives diagnosed with melanoma.

Interview assessment

Parents and children were separately asked to complete in-person semi-structured interviews with a research assistant 1 month and 1 year after genetic test reporting and counseling. Responses to the interview questions were grouped by carrier status. For parents, if at least one of their children was a *p16* mutation carrier, the parent was considered in analyses to have children who were carriers. Due to the limited sample size, descriptive statistics (e.g., frequencies and proportions) were used to quantitatively summarize the results of the measures below.

Changes to skin cancer prevention and control practices One month after genetic test reporting and counseling, parents and children were asked about changes in children's use of sun protection ("Have you made plans to do things differently after receiving risk counseling?"). For analysis, responses to this question were dichotomized (Yes/No).

Family discussions and collaboration on sun protection One month and 1 year after test reporting, parents and children were asked closed-ended questions about their family discussions related to sun protection and screening: "Now that you have received risk counseling, does your family talk about wearing sunscreen and protective clothing more, less, or about the same amount?" and "Now that you have received risk counseling, does your family talk about conducting skin self-exams more, less, or about the same amount?"

One-month and 1-year post-test reporting and counseling, participants were asked how involved parents were in the child's sun protection habits. Participants were asked to select one of four response options, adapted from prior studies focused on children with diabetes (Berg et al. 2013): Supportive (parent is indirectly involved in their child's sun protection habits), Worked Together (parent is very involved and works together with their child to help with child sun protection), Took Charge (parent takes charge of their child's sun protection habits), or Uninvolved (parent allows child to have complete responsibility for sun protection habits).

Parental treatment of children after test result disclosure One month and 1 year after testing, parents were asked whether their child was treated differently after receiving their melanoma risk information ("Do you or other members of your family treat the child differently than other people in your family because of their melanoma risk?"). For analysis, responses to this item were dichotomized (Yes/No). Parents were also asked, "Are there instances in which you might have been more or less protective of your child since they received their genetic test result risk counseling than you are of your other children?" These responses were categorized into an increased level of protectiveness, decreased level of protectiveness, and no change in level of protectiveness.

Family conflicts One year after genetic test reporting and counseling, parents and children were asked about their conflicts related to sun protection or screening since risk counseling and genetic testing ("Since you/your children have received risk counseling and genetic testing, have you or another family member had any conflicts or disagreements with your child about sun protection, behaviors or screening?") and responses were dichotomized into the presence or absence of conflicts.

Sharing risk information and future plans to discuss sun protection and screening One year after genetic test reporting and counseling, parents were asked about their conversations with other adults about children's risk information, sun protection, and screening ("Since your children received risk counseling and genetic testing, have you had any conversations related to the risk information, sun protection behaviors or screening with another adult?") and their responses were dichotomized (Yes/No). Parents were also asked about their plans for sun protection and screening discussions with their children in the future, including intentions to continue these discussions ("How much do you plan on continuing to discuss sun protection and screening with your children as they get older?" Responses were categorized into "More frequent conversations," "Less frequent conversations," or "Same amount as current") and expectations for whether these discussions would change as children aged were dichotomized into Yes/No.

Results

In total, 18 children (mean age = 12.4 years, SD = 1.9) and 11 parents from 11 families participated in the current study

(Table 1). Approximately half (56%) of children were male. All participating parents were mothers and 46% had a personal history of melanoma. Half of children (50%) were carriers and the other half were noncarriers.

Changes to family skin cancer prevention and control practices 1 month after genetic test reporting and counseling

One-month post-genetic test reporting and counseling, the majority of parents (73%, $n = 8$) and half of children (50%, $n = 9$, 8 of whom had a parent who also reported making a change) reported that as a family, they had made changes to their implementation of sun protection or screening strategies after receiving the child's *p16* test result (Table 2). For example, parents described that they were more careful to apply and re-apply sunscreen for their children and to bring their child to see a dermatologist for screening, and children reported that their families brought umbrellas more frequently on trips outdoors and that they bought new swimsuits that provided more skin coverage. A larger proportion of parents with at least one child who was a *p16* carrier and children who were carriers reported making these new sun protection or screening plans. Among parents, 83% with at least one child who was a carrier reported making family plan changes versus 60% with only noncarrier children. Among children, 67% of carriers reported making family plan changes versus 33% of noncarriers (Table 2).

Family interactions around melanoma prevention and control strategies over time

Discussions about melanoma prevention and control strategies Both parents and children reported increased frequency of family discussions about sun protection after genetic test reporting and counseling (Table 2). For example, across the entire sample, 36% of parents ($n = 4$) at the 1-month follow-up and 73% of parents ($n = 8$) at the 1-year follow-up reported that their families talked about sun protection more frequently. A relatively small proportion (17%) of parents who had at least one child who was a carrier reported discussions about sun protection at 1 month whereas 60% of parents whose

Table 1 Participant demographic characteristics

Parent reported ($n = 11$)[a]		
Child age	Mean (SD)	Range
	12.4 (1.9)	10–15
Child sex	*n* (%)	
Male	10 (56)	
Female	8 (45)	
Child *p16* carrier status		
Carrier	9 (50)	
Noncarrier	9 (50)	
Color of child's untanned skin		
Very fair	2 (11)	
Fair	9 (50)	
Olive	6 (33)	
Light brown	1 (6)	
Child health insurance status	*n* (%)	
Yes	18 (100)	
Child race/ethnicity	*n* (%)	
White/Non-Hispanic	18 (100)	
Parent sex	*n* (%)	
Female	11 (100)	
Parent melanoma history	*n* (%)	
Yes	5 (46)	
Number of relatives with melanoma	Mean (SD)	Range
	3 (1.9)	0–6
Household income	Median	Range
	$90,000–$99,000	$30,000–$100,000+

[a] Parents with more than one child participating in the study responded to these items individually for each child who participated

Table 2 Summary of participant responses from qualitative interviews

		Parent report								Child report							
		1 month, *n* (%)				1 year, *n* (%)				1 month, *n* (%)				1 year, *n* (%)			
Made plans to do things related to sun protection differently after testing	Overall sample	8 (73)				–				9 (50)				–			
	Carriers	5 (83)				–				6 (67)				–			
	Noncarriers	3 (60)				–				3 (33)				–			
Family talks more about wearing sunscreen and protective clothing	Overall sample	4 (36)				8 (73)				11 (61)				12 (67)			
	Carriers	1 (17)				4 (67)				6 (67)				7 (78)			
	Noncarriers	3 (60)				4 (80)				5 (56)				5 (56)			
Family talks more about skin self-exams	Overall sample	7 (64)				6 (55)				9 (50)				5 (28)			
	Carriers	4 (67)				3 (50)				3 (33)				4 (44)			
	Noncarriers	3 (60)				3 (60)				6 (67)				1 (11)			
		U[a]	S	W	T	U	S	W	T	U	S	W	T	U	S	W	T
Parent-child collaboration on child sun protection	Overall sample	1 (10)	2 (18)	6 (55)	2 (18)	0 (0)	3 (27)	8 (73)	0 (0)	0 (0)	9 (50)	8 (44)	1 (5)	0 (0)	10 (56)	7 (39)	1 (5)
	Carriers	0 (0)	1 (17)	4 (67)	1 (17)	0 (0)	2 (33)	4 (67)	0 (0)	0 (0)	4 (44)	5 (56)	0 (0)	0 (0)	4 (44)	4 (44)	1 (11)
	Noncarriers	1 (20)	1 (20)	2 (40)	1 (20)	0 (0)	1 (20)	4 (80)	0 (0)	0 (0)	5 (56)	3 (33)	1 (11)	0 (0)	6 (67)	3 (33)	0 (0)
Parent or family treats child differently after genetic testing[b]	Overall sample	1 (6)				5 (28)				–				–			
	Carriers	1 (11)				4 (44)				–				–			
	Noncarriers	0 (0)				1 (11)				–				–			
Parent is more protective of child after genetic testing[b]	Overall sample	5 (28)				3 (17)				–				–			
	Carriers	4 (44)				3 (33)				–				–			
	Noncarriers	1 (11)				0 (0)				–				–			
Parent is less protective of child after genetic testing[b]	Overall sample	0 (0)				1 (6)				–				–			
	Carriers	0 (0)				0 (0)				–				–			
	Noncarriers	0 (0)				1 (11)				–				–			
Parent or another family member had conflicts with child about sun protection or screening	Overall sample	–				3 (27)				–				0 (0)			
	Carriers	–				2 (33)				–				0 (0)			
	Noncarriers	–				1 (20)				–				0 (0)			
Parent had conversation related to risk information, etc. with another adult	Overall sample	–				9 (82)				–				–			
	Carriers	–				5 (83)				–				–			
	Noncarriers	–				4 (80)				–				–			
		–				11 (100)				–				–			

Table 2 (continued)

		Parent report		Child report	
		1 month, *n* (%)	1 year, *n* (%)	1 month, *n* (%)	1 year, *n* (%)
Parent plans to continue discussing sun protection and screening with child regularly as they get older	Overall sample				
	Carriers	–	6 (100)	–	–
	Noncarriers	–	5 (100)	–	–
Parent believes conversations about sun protection and screening with child will change as they get older	Overall sample	–	8 (73)	–	–
	Carriers	–	5 (83)	–	–
	Noncarriers	–	3 (60)	–	–

Carrier status for parent-report is positive if at least one child tested positive

Percentages were calculated out of the total sample of respondents who were carriers or non-carriers (for parents, carriers $n = 6$, noncarriers $n = 5$; for children, carriers $n = 9$, noncarriers $n = 9$)

–Indicates question was not asked for this group or timepoint

[a] Scale options: Uninvolved, Supportive, Worked together, Took charge

[b] Parents with more than one child participating in the study responded to these items individually for each child who participated, rather than for all their children collectively; so for these items $n = 18$, carrier $n = 9$, noncarrier $n = 9$

children tested negative reported having these discussions at 1 month. The majority of parents (64% at 1 month, 55% at 1 year) and a smaller proportion of children (50% at 1 month, 28% at 1 year) reported that the family talked more frequently about conducting skin self-exams (Table 2).

Parent-child collaboration on child sun protection Across the sample, the vast majority of parents and children reported that they were either "supportive" or "worked together" to implement child sun protection strategies (parents: 1 month = 73%, 1 year = 100%; children: 1 month = 94%, 1 year = 95%; Table 2). The lowest proportion of individuals who reported that they "worked together" on child sun protection were children who were noncarriers (1 month and 1 year = 33%; Table 2). The highest proportion of individuals who reported that they "worked together" on child sun protection were parents of noncarriers at 1 month (80%).

Changes in how parents treat their children after genetic test reporting and counseling

At 1-month post-test reporting and counseling, few parents reported treating their child differently (overall: $n = 1$, 13%; carriers: $n = 1$, 11%; noncarriers: $n = 0$, 0%; Table 2). The one parent who reported treating his or her child differently stated that they now used "more sunscreen than anyone else" with their child who was a carrier. A larger proportion of parents reported treating their child differently at the 1-year follow-up, especially among carriers (overall: $n = 5$, 63%; carriers: $n = 4$, 44%; noncarriers: $n = 1$, 11%; Table 2). In particular, parents of children who were carriers reported that they were more vigilant about implementing sun protection, particularly for their child who was a carrier. For instance, one parent with multiple children noted, "We are more apt to pour sunscreen on him (child who was a carrier) because his risk is higher. We do it to all of them but we are more aware with him." Only one parent of a noncarrier reported a change in how they treated the child, and expressed that they were "less worried" about the child who was a noncarrier than their child who was a carrier.

When asked whether they were more or less protective of their child at the 1-month follow-up after receiving the child's genetic test result, 28% of parents reported that they were more protective of their child and 67% ($n = 12$) endorsed no change ($n = 1$ missing; Table 2). At 1-year post-genetic test reporting, 17% of parents ($n = 3$) reported that they were more protective of their child, 78% ($n = 14$) endorsed no change, and one parent (5%) expressed mixed levels of protectiveness such that they were less worried about their child who was a noncarrier than their child who was a carrier but due to the education received through the study, they were "more worried about all of my kids."

Family conflicts about sun protection or screening

One year after genetic test reporting and counseling, approximately one-quarter of parents (27%, $n = 3$) reported that they or another family member had any conflicts with children about sun protection or screening. These conflicts included child complaints about screening, child attempts to evade thorough sunscreen application, and child expression that sunscreen is inconvenient or takes too much time. Of the parents who had one or more child with a *p16* mutation, 33% ($n = 2$) reported presence of such conflicts with their children, while 20% ($n = 1$) of parents of children who were noncarriers reported presence of such conflicts. No children (0%) reported having conflicts with parents or other family members about sun protection or screening.

Sharing risk information and parents' future plans 1 year after testing reporting and counseling

The vast majority of parents (82% overall, $n = 9$; 83% of parents of carriers, $n = 5$; 80% of parents of noncarriers, $n = 4$) reported that they had had conversations about the child's risk information, sun protection, or screening with another adult, including spouses, immediate and extended family members, and co-workers (Table 2). For example, parents described that they shared the child's test results with the child's grandparents, and that they shared general sun protection recommendations (e.g., type of sunscreen to use, frequency of application) with individuals including the child's other parent, grandparents, siblings, extended family, and neighbors.

All parents (100%), regardless of their child's test status, reported that they planned to continue discussion about sun protection and screening with their children regularly and continuously as they get older. At the 1-year follow-up, 73% of all parents ($n = 8$) reported that they believed that their conversations about sun protection and screening with their children would change as their children got older (parents with at least one child who was a carrier: $n = 5$, 83%; parents of only noncarriers: $n = 3$, 60%; Table 2). For example, four parents reported that they foresaw that they may need to have conversations with their children as they got older about how implementing sun protection would increasingly become the child's responsibility.

Discussion

Children who received *p16* predispositional genetic testing and their parents reported several notable changes to their sun protection and screening plans, discussions, and interactions within the family context. After receiving information on children's *p16* test status, the majority of families reportedly made new plans for sun protection, discussed sun protection frequently, and parents and children collaborated as a unit to implement sun protection strategies for children. These changes appeared to be sustained over the 1 year following test reporting and counseling. Although the modest sample size precluded formal statistical comparisons, the observed changes may have been more pronounced for children who had a positive test result. A small proportion of participants reported that parents were more protective of the child after test reporting or that there were parent-child conflicts about sun protection. In addition, the vast majority of parents reported sharing information about the child's melanoma risk and risk reduction behaviors with other adults, which contrasts with prior findings among first-degree relatives of patients with sporadic melanoma showing that discussions about melanoma occurred relatively less frequently (Harris et al. 2010). Increased family discussions about sun protection, plans for sun protection, and parent-child collaboration on sun protection could account for the reported improvements in sun protection previously documented in this pediatric sample (Stump et al. 2018).

This study is one of the first to examine reported family interactions, including interactions related to preventive health behaviors, among children who received predispositional genetic testing and their parents. Overall, children and their parents reported beneficial changes to their interactions that may have facilitated children's use of sun protection. However, several limitations are important to note. The study did not include a baseline assessment of family interactions and thus precluded examining prospective changes in family interactions over time. While the current study was the first to examine post-test reporting family interactions among children tested for *p16* mutations, the sample size was small, and therefore the results should be replicated in larger samples in future studies. Data from larger samples could also be analyzed using formal tests of statistical significance to explore quantitative changes over time and to stratify analyses by carrier status. Carrier status will be important to examine in future test reporting studies given evidence that non-carriers from families with a known *p16* mutation may still have higher risks for melanoma than the general population (Helgadottir et al. 2017). Test reporting studies will want to consider how best to present information on elevated risk for melanoma among non-carriers, as well as the role of other risk factors such as skin type. The current study also was unable to compare children who received *p16* testing with children who were not tested.

Future efforts to describe the effects of pediatric predispositional genetic testing in this and other high-risk populations could seek to understand individual parent and child characteristics (e.g., understanding of risk information, child age and gender, parent's own carrier status), parent-child interactions (e.g., modeling of desired preventive health behaviors, nature of parent-child collaborations to implement

preventive health behaviors), and sibling interactions that contribute to changes in the implementation of preventive health behaviors (Wu et al. In press). Larger studies could also formally test whether changes to such family interactions related to the receipt of risk information may mediate changes to preventive health behavior implementation.

Families with children who receive predispositional genetic testing reported changes to their family interactions around preventive health behaviors. Post-test counseling or other family interventions could include provision of anticipatory guidance to parents about discussing preventive and screening behaviors with their children and others in the context of children's test results as well as about how to manage skin cancer prevention and control behaviors with their children as they become more independent with age or as new challenges arise.

Funding information This work was supported by the National Institutes of Health (K07 CA196985 to YW, R01 CA158322 to LGA, SAL, PC).

Compliance with ethics guidelines

Conflict of interest Dr. Leachman served on a Medical and Scientific Advisory Board for Myriad Genetics Laboratory, for which she received an honorarium. She has collaborated with Myriad on a project to validate an assay that is unrelated to the research reported here.

Ms. Kohlmann has consulted for Myriad Genetics Laboratory in the past on unrelated projects and received a research grant from Myriad Genetics Laboratory to study the psychological and family communication outcomes of multigene panel testing. That work is unrelated to the research reported here.

Ms. Champine has been compensated for serving on the Genetic Counseling Advisory Board for Invitae, which is a for-profit genetic testing laboratory.

Yelena Wu, Lisa Aspinwall, Bridget Parsons, Tammy Stump, Katy Nottingham, and Pamela Cassidy declare that they have no conflict of interest.

Ethical approval All procedures followed were in accordance with the ethical standards of the responsible committee on human experimentation (institutional and national) and with the Helsinki Declaration of 1975, as revised in 2000 (5). Informed consent was obtained from all patients for being included in the study.

Disclaimer The content is solely the responsibility of the authors and does not necessarily represent the official views of the National Institutes of Health.

References

Balk SJ (2011) Ultraviolet radiation: a hazard to children and adolescents. Pediatrics 127:e791–e817

Begg CB, Orlow I, Hummer AJ, Armstrong BK, Kricker A, Marrett LD, Millikan RC, Gruber SB, Anton-Culver H, Zanetti R, Gallagher RP, Dwyer T, Rebbeck TR, Mitra N, Busam K, From L, Berwick M, Genes Environment and Melanoma Study Group (2005) Lifetime risk of melanoma in CDKN2A mutation carriers in a population-based sample. J Natl Cancer Inst 97:1507–1515

Berg CA, Butner JE, Butler JM, King PS, Hughes AE, Wiebe DJ (2013) Parental persuasive strategies in the face of daily problems in adolescent type 1 diabetes management. Health Psychol 32:719–728

Bishop DT, Demenais F, Goldstein AM, Bergman W, Bishop JN, Bressac-de Paillerets B, Chompret A, Ghiorzo P, Gruis N, Hansson J, Harland M, Hayward N, Holland EA, Mann GJ, Mantelli M, Nancarrow D, Platz A, Tucker MA, Melanoma Genetics Consortium (2002) Geographical variation in the penetrance of CDKN2A mutations for melanoma. J Natl Cancer Inst 94:894–903

Botkin JR, Belmont JW, Berg JS, Berkman BE, Bombard Y, Holm IA, Levy HP, Ormond KE, Saal HM, Spinner NB, Wilfond BS, McInerney J (2015) Points to consider: ethical, legal, and psychosocial implications of genetic testing in children and adolescents. Am J Hum Genet 97:6–21. https://doi.org/10.1016/j.ajhg.2015.05.022

Brandi ML, Gagel RF, Angeli A, Bilezikian JP, Beck-Peccoz P, Bordi C, Conte-Devolx B, Falchetti A, Gheri RG, Libroia A, Lips CJ, Lombardi G, Mannelli M, Pacini F, Ponder BA, Raue F, Skogseid B, Tamburrano G, Thakker RV, Thompson NW, Tomassetti P, Tonelli F, Wells SA Jr, Marx SJ (2001) CONSENSUS: guidelines for diagnosis and therapy of MEN type 1 and type 2. J Clin Endocrinol Metab 86:5658–5671. https://doi.org/10.1210/jcem.86.12.8070

Harris JN, Hay J, Kuniyuki A, Asgari MM, Press N, Bowen DJ (2010) Using a family systems approach to investigate cancer risk communication within melanoma families. Psychooncology 19:1102–1111. https://doi.org/10.1002/pon.1667

Helgadottir H, Olsson H, Tucker MA, Yang XR, Hoiom V, Goldstein AM (2017) Phenocopies in melanoma-prone families with germ-line CDKN2A mutations. Genet Med 20:1087–1090. https://doi.org/10.1038/gim.2017.216

Järvinen O, Hietala M, Aalto AM, Arvio M, Uutela A, Aula P, Kääriäinen H (2000a) A retrospective study of long-term psychosocial consequences and satisfaction after carrier testing in childhood in an autosomal recessive disease: Aspartylglucosaminuria. Clin Genet 58:447–454. https://doi.org/10.1034/j.1399-0004.2000.580604.x

Järvinen O, Lehesjoki AE, Lindlof M, Uutela A, Kaariainen H (2000b) Carrier testing of children for two X-linked diseases: a retrospective study of comprehension of the test results and social and psychological significance of the testing. Pediatrics 106:1460–1465. https://doi.org/10.1542/peds.106.6.1460

Leoz ML, Carballal S, Moreira L, Ocana T, Balaguer F (2015) The genetic basis of familial adenomatous polyposis and its implications for clinical practice and risk management. Appl Clin Genet 8:95–107. https://doi.org/10.2147/tacg.s51484

Rao A, Rothman J, Nichols KE (2008) Genetic testing and tumor surveillance for children with cancer predisposition syndromes. Curr Opin Pediatr 20:1–7. https://doi.org/10.1097/MOP.0b013e3282f4249a

Ross LF, Saal HM, David KL, Anderson RR (2013) Technical report: ethical and policy issues in genetic testing and screening of children. Genet Med 15:234–245. https://doi.org/10.1038/gim.2012.176

Stump TK, Aspinwall LG, Kohlmann W, Champine M, Hauglid J, Wu YP, Scott E, Cassidy P, Leachman SA (2018) Genetic test reporting and counseling for melanoma risk in minors may improve sun protection without inducing distress. J Genet Couns 27:955–967. https://doi.org/10.1007/s10897-017-0185-5

Wade CH, Wilfond BS, McBride CM (2010) Effects of genetic risk information on children's psychosocial wellbeing: a systematic review of the literature. Genet Med 12:317–326. https://doi.org/10.1097/GIM.0b013e3181de695c

Wakefield CE, Hanlon LV, Tucker KM, Patenaude AF, Signorelli C, McLoone JK, Cohn RJ (2016) The psychological impact of genetic information on children: a systematic review. Genet Med 18:755–762. https://doi.org/10.1038/gim.2015.181

Williams LH, Shors AR, Barlow WE, Solomon C, White E (2011) Identifying persons at highest risk of melanoma using self-assessed risk factors. J Clin Exp Dermatol Res 2:129. https://doi.org/10.4172/2155-9554.1000129

Wu YP, Aspinwall LG, Conn BM, Stump T, Grahmann B, Leachman SA (2016) A systematic review of interventions to improve adherence to melanoma preventive behaviors for individuals at elevated risk. Prev Med 88:153–167. https://doi.org/10.1016/j.ypmed.2016.04.010

Wu YP et al (in press) A pilot study of a telehealth family-focused melanoma preventive intervention for children with a family history of melanoma. Psychooncology. https://doi.org/10.1002/pon.5232