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Adolescent and Parental Attitudes about Return of Genomic Research Results: Focus Group Findings Regarding Decisional Preferences

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Abstract

Opportunities to participate in genomic-sequencing studies, as well as recommendations to screen for variants in 59 medically-actionable genes anytime clinical genomic sequencing is performed, indicate adolescents will increasingly be involved in decisions about learning secondary findings from genome sequencing. However, how adolescents want to be involved in such decisions is unknown. We conducted five focus groups with adolescents (2) and parents (3) to learn their decisional preferences about return of genomic research results to adolescents. Discussions about decisional preferences centered around three themes: feelings about receiving genomic risk information, adolescent involvement and capacity to participate in decision-making, and recommendations for parental vs. collaborative decision-making. We address the contested space between parental duties to act in their children's best interests when choosing which results to return and adolescents' desires to make autonomous decisions. A collaborative decision-making approach is recommended for obtaining consent from adolescents and their parents for genome sequencing research.

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INTRODUCTION

Genome sequencing can identify genomic variants that may not be related to research study aims. Incidental or secondary genomic findings may have health, reproductive, or personal importance for research participants. However, there can be psychological, economic, and social risks associated with learning incidental or secondary findings.

A significant body of scholarship has emerged in the last decade to explore the ethical, legal, and social implications of returning incidental or secondary genomic findings to research participants and patients. This literature has focused on the potential benefits and harms of genomic information ranging from health, psychological, familial, social, and moral implications (Borry, Goffin, Nys, & Dierickx, 2008; Botkin et al., 2015; Green et al., 2013; Mand, Gillam, Delatyck, & Duncan, 2012; Ross, Rothstein, & Clayton, 2013). There is clear evidence that the public (Bergner et al., 2014; Bollinger, Bridges, Mohamed, & Kaufman, 2014; Bollinger, Scott, Dvoskin, & Kaufman, 2012; Daack-Hirsch et al., 2013; Haga, O'Daniel, Tindall, Lipkus, & Agans, 2011; Murphy et al., 2008; O'Daniel & Haga, 2011; Yu, Crouch, Jamal, Tabor, & Bamshad, 2013), pediatric patients' parents and adult patients (Bergner et al., 2014; Fernandez et al., 2006; Harris et al., 2012; McGowan, Glinka, Highland, Asaad, & Sharp, 2013; Sapp et al., 2014; Ziniel et al., 2014), and research participants (Facio et al., 2012; Wynn, Martinez, Duong, & . 2016) want the option to learn actionable genomic research results. They value results for serious health conditions that can be treated or prevented and having a choice over which results to receive (McGowan et al., 2013). They also perceive participation in the decision-making process to be critical (Bergner et al., 2014; Bollinger et al., 2012; Clift et al., 2015; Dressler et al., 2012; Kaufman, Murphy, Scott, & Hudson, 2008). In a pediatric setting, parents value learning about genetic variants for conditions treatable or preventable in childhood. Although parents report more variation in their desire to learn carrier status and susceptibility for conditions not actionable in childhood (Christensen et al., 2017; Sapp et al., 2014), the majority want to learn results for all conditions for their children in both clinical and research settings (Christensen et al., 2017; Harris et al., 2012; Levenseller et al., 2014; McGowan et al., 2013).

There has been little exploration of how children's values and preferences regarding the return of secondary or incidental findings should be accommodated in a research context, and even less on how a child's age or maturity ought to be considered in enrolling minors in genomic research. There is also limited data on adolescents' preferences for learning incidental and secondary findings. For instance, in online discussion groups with adolescents aged 13-17 with one of three conditions for which sequencing might be offered, most adolescents reported they would hypothetically want to receive all results from whole exome sequencing, including carrier status and adult-onset untreatable conditions (Levenseller et

al., 2014). In addition, a cross-sectional survey of 282 adolescents in grades 7-12 found that 83% of those surveyed wanted to learn hypothetical incidental findings from genomic sequencing that were not medically actionable in childhood. Moreover, the majority (76%) felt they were capable of making the decision about receiving incidental findings and most (73%) felt they should have a role in the decision-making process (Hufnagel, Martin, Cassedy, Hopkin, & Antommaria, 2016). Despite the limited research on adolescent preferences regarding the return of incidental and secondary findings, opportunities for pediatric populations to participate in genomic research are increasing. Large-scale genomic sequencing research networks and nationwide collaborative studies, including the All of Us Research Program (previously the Precision Medicine Initiative), fuel scientific and public expectations that individually-relevant genomic research results will be returned to participants (Juengst, McGowan, Fishman, & Settersten, 2016). With initiatives such as BabySeq and PediSeq, growing numbers of participants in genomic sequencing studies are enrolling as children and adolescents (Rehm, 2017; Walser et al., 2017). Thus, guidelines for consent processes that consider the adolescent perspective and facilitate understanding about the possible consequences of genome sequencing results are needed.

To fill this gap, we designed a qualitative study to explore parents' and adolescents' decisional-preferences about their values and involvement in choices about return of genomic sequencing results. By assessing parents' and adolescents' perspectives on returning genomic sequencing results this study is uniquely positioned to inform and guide the practices of genomic researchers working with adolescent populations. This study also provides a window on the ways in which genomic researchers can learn from the debates regarding adolescent decision-making in pediatric ethics, which has largely emerged from the clinical ethics context.

METHODS

To facilitate hypothetical decision-making, we modified an existing decision tool designed to allow parents to choose which genomic results to receive about their children based on the severity and preventability of possible conditions (Bacon et al., 2015). None of the current authors were involved in the development of the original decision tool. The original tool was modified for the current study and provided context for conversations about decision making in iterative focus group discussions. The tool included choices about return of results based on condition characteristics: severity, age of onset, preventability, and treatability. Participants could exclude developmental delay, neurodegenerative conditions, adult onset conditions with no actionability in childhood, and carrier status (Myers, McGowan, & Prows, 2017). Details about tool development and implementation will be the subject of a future publication.

With the IRB approval from the authors' institution, we recruited adolescents (13 – 18 years) and adult parents to participate in focus groups through an email to medical center employees, study flyers posted around the medical center and on the medical center website, and an advertisement in an institutional newsletter to local community members. The only inclusion criteria were age of the adolescents and parental status. Parents' and adolescents'

written informed consent and assent were obtained in-person immediately prior to study participation.

We held focus groups with parents and adolescents in March-April 2016. Focus group moderator guides for parent and adolescent focus groups were developed and reviewed by the research team for face validity. Focus groups were conducted at a full-service nonprofit pediatric academic medical center and were moderated by two members of the research team skilled in conducting focus groups. Separate focus groups were conducted with adolescents and with parents. Focus groups ranged in size from 5 to 8 participants and lasted approximately 90 minutes. Prior to initiation of the focus group discussion, a genetic counselor gave a presentation to focus group participants to provide background information about genomic sequencing that would allow them to answer the facilitators' questions. Content of the presentation included an overview of genes, types of genetic variants within genes (e.g. polymorphism, risk conferring, or disease causing), methods of genetic testing (e.g. targeted to one gene or many genes), and limitations of genetic testing. Any questions from focus group participants about the content of the presentation were answered before the focus group discussion commenced. Topics covered in focus groups included perspectives on adolescents' roles in research-and health-related decision-making, and adolescent participation in decision-making regarding the return of genomic research results. Reaching theoretical saturation was the criterion of ending focus group recruitment (Merriam & Tisdell, 2015).

Each focus group discussion was audiotaped, transcribed, and analyzed using a process of inductive and deductive thematic analysis (Fereday & Muir-Cochrane, 2006). With a thematic analysis approach, the intention is to systematically identify meaningful themes and patterns around a specific area of interest (Gale, Heath, Cameron, Rashid, & Redwood, 2013; Polit & Beck, 2010). To familiarize ourselves with the data, members of the research team independently reviewed the transcripts to identify important themes and ideas. A constant comparison method was used to examine and refine the themes by comparing and contrasting information within and across the focus groups (Boeije, 2002; Corbin & Strauss, 1990).

RESULTS

Three focus groups were held with parents and two with adolescents to gain prospective genomic research participants' perspectives on decision-making about return of genomic research results for adolescents. A total of 33 individuals participated in these focus groups, 18 parents and 15 adolescents. Six parents had one child participate in a separate focus group; three parents had two children participate in a focus group; and nine parents participated without their children, including two husband/wife pairs. Twelve adolescents, including three sibling pairs, had one parent participate in a separate focus group and three adolescents had no parent participate in a focus group. Participant demographics can be found in Table 1. Discussions about adolescent and parental decisional preferences centered around three themes, each of which will be discussed in turn: feelings about receiving genomic risk information, adolescent involvement and capacity to participate in decision

making, and recommendations for parental vs. collaborative decision-making between parents and their adolescent children.

Receiving Genomic Research Results

Across the focus groups, there were mixed feelings about learning genomic results based on condition characteristics conveyed through the decision tool. Some participants felt that knowledge of genomic risk information would be burdensome, while others felt that knowing would help with future planning, preventive efforts, and treatment, if necessary.

Adolescents as a group expressed a strong desire for as much genetic risk information as possible. One adolescent explained:

> I would rather know than not know overall, just like...even if it's not preventable I think it's good to know like what's coming (Adolescent Focus Group 1)

Focus group participants often framed the implications of decisions about return of genomic risk results to adolescents in terms of how it might impact other members of the family. There were references both to hereditary disease risk results that could be medically relevant for the adolescents' parents or siblings, and recognition that adolescents may not be so far off from making reproductive decisions for which carrier results could be relevant:

> Other people in my family may want to know [my results]. If I'm old enough I may want to have kids, I may want to figure out if they have a chance of having [a genetic risk factor] too. Like if I'm having kids with someone and they have a similar gene there's a pretty good chance my kids are going to have it too and it would make me reconsider some things like adoption. (Adolescent Focus Group 1)

In contrast, each focus group discussed concerns that they had about the existence of personal genomic information about adolescents. These issues were raised spontaneously in most focus groups, and moderators queried directly about any concerns that parents of adolescents might have in the remaining groups. Parents expressed concerns more frequently than adolescents about potential risks that genomic research results could pose, such as negatively impacting a child's insurability and the possibility that legal and privacy implications of genomic information could change dramatically throughout the recipient's lifetime. As one parent elaborated:

> I think this goes back to all the privacy issues and there's a lot of little murky slippery slope stuff that until we work out some of that stuff legally, that is going to be tough because I'm going to make a decision based on what I know in 2016 and that may have very different ramifications in 2036 and that's in that [medical] record. (Parent Focus Group 3)

Parents also raised concerns about how and when to disclose results to an adolescent, and that genomic risk information might negatively impact their perceptions of their child or the child's sense of self or their future. Adolescents similarly commented upon the potential for genomic risk information to influence their sense of their future and their life chances. The following parent's comment illustrates the doubled-edged quality of the receipt of genomic risk information:

> With awesome power comes awesome responsibility. I think we all kind of touched on that, that knowing that information it, how do you look your kid in the face and not see the horrible stuff. I mean if you know that, what lies ahead for them and you look at them and try to be positive for them every day eventually you are going to break and you don't want to break in front of your kid. So, I think knowing these things…I want to know everything, I would like to know because I'm a planner and I like to plan ahead but I think that it would be unfair too. I mean you like kind of both sides of the sword there. It would be unfair to know all that because then your demeanor changes but at the same time you can prepare and plan and work together with the family unit to proceed ahead and see what you can do together. (Parent Focus Group 1)

Despite mixed feelings across focus groups about the implications of learning genomic test results, most participants agreed that if they had decided to participate in a genomic research study, they would most likely want to know all (or most) of the possible test results that a study offered to return.

Adolescent Involvement and Capacity to Participate in Decision Making

The most emphasis and engagement across all focus groups was the degree to which adolescents should participate in decision making about choosing to learn genomic research results. As this parent's comment shows, there were some parents who felt that adolescents should enter into genomic research with a clear sense of what all it could entail, even if the adolescent would not be the ultimate decision-maker:

> If this is going to affect their health I want them to be an active participant, I want them to, not necessarily have the end-all-be-all say on whether or not we should do the testing or we should find out certain things, but I want them to be aware of what that entails. I want them to know 'hey we could find something out that could really drastically change the way you live from now on and I want you to know that by just agreeing to having the testing done you are putting yourself at jeopardy of finding out something that you might not have wanted to know.' (Parent Focus Group 1)

However, unlike this participant, some parents felt that most adolescents would not be able to understand or cope with receiving the genomic results and that there are some genetic risk susceptibilities about which adolescents shouldn't have to know. However, parents still felt an adolescent should be an active participant in decisions about choosing what kinds of results to learn. In contrast, adolescents noted that they would be able to understand and cope with genomic research results, but that an adolescent's age and maturity had to be taken into consideration.

Despite their variable attitudes towards the degree to which adolescents should be involved and have input regarding choosing the specific results to have returned, all groups strongly agreed that adolescent participation in decision-making about the return of individual genomic research results should depend on the age, maturity level, and personality of the adolescent. Attitudes about this matter were consistent, regardless of whether the participant was a parent or adolescent, as the following quotes illustrate:

> The difference between a 13 year old and a 17 year old's emotional maturity is huge. (Parent Focus Group 2)

> How I would have interpreted it when I was 13 and how I would interpret the results now are very different. (Adolescent Focus Group 2)

> I would make very different choices based on their personalities, very different. (Parent Focus Group 3)

The consistency of this finding suggests that blanket recommendations about which genomic research results to return to adolescents would be difficult to ascertain, and that a more individualized approach may be necessary to facilitate informed decision-making with an adolescent population.

Models for decision-making

Overall, parents strongly believed that they should have the final say when making decisions about return of results. Some parents felt that only their own perspective should matter since they are responsible for the health and well-being of their child. Though it was equally common to hear parents remark that they valued their child's input, even if the parent would be the ultimate decision-maker. As the following parent articulated:

> I would want to be with her to have the conversation like 'cause I would want to know her desire before I made the decision. I would want to know what she wanted to know, so I would want to be doing that together, not separately. (Parent Focus Group 2)

Similarly, some adolescents felt only their own perspective should matter since the results would be about them. When taking this kind of stance, focus group participants tended to draw upon existing exceptions to parental consent, such as in the case of sexual or reproductive health where (at least in some states) adolescents are free to make medical decisions without parental involvement. Some participants referenced other types of decisional autonomy afforded to adolescents regarding things that would impact their bodies or their futures, such as getting piercings or choosing a career path. For instance, one 14-year old who had just started high school argued that adolescents are told:

> [They] are mature enough to start their path onto their career choice that they want. I mean, why can't they have a say or strong voice in the genetics that they want to learn about themselves? 'Cause, I mean, if we are allowed to make our own decision on whatever our [course] electives are going to be and what do we want to be when we grown and already researching what colleges and what degree fields we want to learn about why can't we have a voice and say this and the parents should have a voice but I think ultimately it should be up to the child with the input of their parents. (Adolescent Focus Group 1)

While it was common for adolescent focus group participants to express the desire to make their own decisions about return of genomic research results, they did seem to understand that they were legally considered minors, and they would need their parents' permission to learn specific results. Illustrating this acknowledgement, one adolescent said:

> Idealistically I think it should be my decision. But I think when it comes down to it I am still a minor, and I am still under the care and responsibility of my parents until I am 18. And so I think when it comes down to kind of like legally it's not really my decision even though I would want it to be. (Adolescent Focus Group 2)

Many adolescents felt their decisional preferences would differ from their parents'. In this case, they felt that a discussion would be necessary to help both the parents and the adolescents feel comfortable with the final decision. To navigate any incongruence, the adolescent focus group participants proposed a model in which a representative of the health care or research team could serve as an advocate and educator regarding adolescents' preferences by facilitating a discussion between parents and their child:

> Have [the parent and adolescent] fill it out separately and then the doctor can see if they have any differences which they may not because a lot of parents and children tend to think the same way but if they do they could have them discuss it a little more. (Adolescent Focus Group 1)

This proposal, articulated in various forms in each of the adolescent focus groups, informed the approach that we have implemented in a subsequent, ongoing study to enroll adolescents in genomic research.

DISCUSSION

The findings of this study may inform the development of best practice models to facilitate consent processes and shared decision-making about return of incidental or secondary findings from genome sequencing studies involving adolescents. Our participants' responses contribute insight into ways in which the enrollment of adolescent participants may present unique challenges and opportunities for decision-making processes. Of interest are how these empirical findings prompt us to address and reconcile lessons from the bioethics literature on return of results and testing in pediatric populations.

Adolescents and parents in our study had both hopes and fears about the potential for genomic research to reveal impactful information about research participants and their families. Despite these concerns, the majority of participants wanted to have as much knowledge available to them as possible. This finding is consistent with the literature on prospective and early users' attitudes towards clinical and direct-to-consumer genomic testing, and may be a characteristic more likely seen in early user populations (McGowan, Fishman, & Lambrix, 2010; McGowan et al., 2013). As early experience with return of hypothetical genome sequencing research findings has taught us though, the enthusiasm that prospective parents and their children may have for the return of individual results to inform life planning may be overstated, as few genomic research participants will have actionable results that can be offered for return (Dorschner et al., 2013). Previous research and findings from the current study also point to the continuing concern that prospective research participants and patients have about their genomic information being vulnerable to privacy and confidentiality breaches. In particular, concerns about the potential for their personally identifiable genomic information to be shared in the medical record, with insurance

providers, or with legal entities persist (McGowan et al., 2013). The current study suggests that like adults, adolescents may feel genomic information can transform one's sense of self.

To address the kinds of hopes and fears that adolescents and their parents may have about genomic research results, researchers ought to carefully counter misconceptions about the transformative potential of genomic risk information with the accumulating empirical evidence that recipients of genomic information are able to incorporate this information into their sense of self with minimal psychosocial impact (Bradbury et al., 2016; Fernandez et al., 2014; Fishman & McGowan, 2014; Kleiderman et al., 2014; Levenseller et al., 2014; Shkedi-Rafid, Dheensa, Crawford, Fenwick, & Lucassen, 2014; Wade, Wilfond, & McBride, 2010; Wakefield et al., 2016). Lessons from previous research and the current study also suggest that researchers enrolling adolescents in genome sequencing studies ought to be mindful of the ways in which they present the possibility of return of individual results. Whenever possible, examples of the types of results that would be most likely to be returned should be given to afford prospective participants a robust sense of the range of possible outcomes of their participation. Further, researchers ought to address privacy and confidentiality considerations in the informed consent process, being attentive to the risks of personally identifiable genomic research data making its way into medical, legal, or insurance environments.

In addition to illuminating adolescents' perceptions of the value and risks of obtaining personal genomic information, this study also provokes longstanding questions in pediatric ethics regarding predictive genetic testing and disclosure of genetic test results to children. Professional societies and the pediatric ethics literature have long advocated for delaying predictive genetic testing until individuals have the capacity to make an informed decision about their genetic knowledge preferences (Committee on Bioethics, Committee on Genetics, & The American College of Medical Genetics and Genomics Social Ethical and Legal Issues Committee, 2013; Davis, 1997; National Society of Genetic Counselors, 2017). In most cases this has effectively deferred predictive genetic testing and return of genetic and genomic test results until adulthood. Stances recommending this conservative approach toward predictive genetic testing in children rely on paradigmatic cases of genetic testing for autosomal dominant traits, and may not adequately take into consideration the ways in which predictive genomic information can be generated in contemporary research, clinical, public health and direct-to-consumer contexts (Meagher, McGowan, Settersten, Fishman, & Juengst, 2017). More recently we have seen equivocation on the issue of delaying genetic testing until the age of majority, with the American College of Medical Genetics and Genomics putting forward recommendations that secondary or incidental findings from genome sequencing ought to be returned to patients, citing a professional duty to warn these individuals about their risks for developing diseases informed by 56 (and later 59) genes (Green et al., 2013; Kalia et al., 2016). Such disparate recommendations raise the question as to how protectionist approaches to genomic information (as both of these examples illustrate in different ways) ought to be applied when enrolling adolescents in genomic research. Ought adolescents enroll in genome sequencing research at all, if we hold up the principle of a child's right to an open future (Davis, 1997)? Or ought adolescents enroll in genomic research with an understanding that they will only have access to individual results that have been deemed worth returning by genomics experts (Green et al., 2013)? Or is there

a third option, where adolescents can assert a degree of decisional autonomy despite their status as minors, which might draw upon guidance from clinical ethics?

While there is an ethical and legal obligation to obtain parental permission for medical interventions and research involving minors, assent ought to be solicited from minors, as developmentally appropriate in both clinical practice and research. (Committee on Bioethics, 1995; Katz, Webb, & Committee On Bioethics, 2016; V. A. Miller & Nelson, 2006). Parents who participated in focus groups recognized their responsibility to make decisions that would be in the best interests of their children and that all decisions pertaining to genomic information are relational – they could have implications not only for the individual adolescent but for other family members as well. What might be best for one child may have different implications for another; a child's age, maturity, and personality all need to be taken into consideration when deciding whether to enroll in genome sequencing research and which and when results ought to be returned to a minor. The many factors that need to be considered for adolescents suggests that both mandating the return of specific results based on a duty to warn and barring the return of results based on a child's right to an open future may be flawed ethical norms, and that acting in a child's best interests – whatever that may be – may better serve parents and adolescents considering participation in genomic research (Garrett et al., under review). Further, we need to take into consideration the voices of adolescents themselves, who in the case of this study strongly advocated for establishing processes where their own decisional preferences could be accommodated and addressed if dissenting from their parents. Others have recommended that as minors become more mature their values, preferences, and assent or dissent ought to be given more weight in clinical decision-making (Katz et al., 2016). Results of our study suggest that this clinical ethics recommendation ought also apply to the genomic research context.

A model of decision-making that integrates minors' values and preferences leads to the question of adolescent capacity to participate in decision-making about return of genomic research results. The return of results literature has been relatively silent about adolescents, instead treating children as a uniform population and leaving aside clinical ethics guidance regarding graduated models of assent (Committee on Bioethics, 1995). Study participants gave a wide range of examples of the types of decisions afforded to adolescents, ranging from sexual and reproductive health care to decisions about career paths, so the question becomes whether receipt of personal genomic information ought to be treated similarly or differently from the other types of decisions that adolescents can make. There is little consensus in the United States about the degree to which adolescents should have autonomy to make decisions about their own sexual or reproductive health, and our study participants' remarks, as well as debates in the ethics literature suggest that there would be similarly disparate viewpoints on the degree to which we ought to treat individual genomic information as so personally significant and impactful to exist outside of parental purview. Certainly knowledge of one's sexually transmitted infection status would have different public health implications than knowledge of one's risk to develop an adult onset genetic disorder, but there are some similarities that can be drawn between access to private and confidential sexual and reproductive health counseling, and access to one's carrier status, as both could have implications for one's family building and future. Ethicists tend to treat genomic information as exceptional (McGowan et al., 2017), but ought decisional autonomy

to personal genomic information be treated akin to seeking reproductive and sexual health care? This study was not designed to answer this ontological question, and there was a high degree of variability between adolescent and parental perspectives on decisional autonomy, but the examples given by both parents and adolescents to address how decisions regarding genomic information is like other types of decisions provokes questions for further exploration in future research.

Even if adolescents wanted the final word over the decision, many of our focus group participants acknowledged that they would not be making this decision privately and that their parents would need to give approval as their legal guardians. This and previous research with adolescents indicate that they want to be involved in the decision-making about enrollment in genomic and biobank research studies (Geller, Tambor, Bernhardt, Fraser, & Wissow, 2003; Murad, Myers, Thompson, Fisher, & Antommaria, 2017) as well as the return of incidental findings (Hufnagel et al., 2016; Levenseller et al., 2014). The findings of the study prompt us to address the contested space between parental duties to act in the best interests of their children regarding choosing which results ought to be returned and adolescents' desires to be autonomous in their decision-making. The most coherent proposal to come from the focus groups was from adolescents advocating a shared decision-making model, with a third party to mediate any disagreements between parents and their adolescent children. This proposal is aligned with ideas published in the literature to elicit adolescent perspectives on participation in research. For instance, involvement of a researcher in solicitation of the child's questions and concerns during the assent process and letting the child know that s/he is central to the decision, may increase the child's feeling of decision self-efficacy and result in more favorable perceptions of the decision-making process (Victoria A. Miller, Feudtner, & Jawad, 2017). Similar to our findings, previous research shows that both parents and adolescents have suggested that involvement in the decision-making process about genomic research may vary depending on the child's age, maturity level, and personality (Geller et al., 2003; Hufnagel et al., 2016; Levenseller et al., 2014). However, also as in our findings, parents' and adolescents' have varying perceptions as to the amount of decision-making autonomy they would like the other to have (Geller et al., 2003; Levenseller et al., 2014; Victoria A. Miller, Reynolds, & Nelson, 2008). For instance, focus groups with parents and children ages 10-17 from families at increased risk for heart disease or breast cancer found that the more mature the child, the less risk was associated with the research, and the more open the family's communication style, the greater the likelihood of joint decision-making (Geller et al., 2003). Parent-child collaboration processes that afford children a voice is an important aspect in decision making, particularly as children mature developmentally and cognitively (Lipstein et al., 2014; Victoria A. Miller et al., 2008). However, consideration of the family context and multiple stakeholders is needed (Lipstein, Brinkman, & Britto, 2012). Facilitation of collaborative decision making by a health care provider or researcher, as recommended by adolescents in our study, may increase the likelihood that both parents and adolescents are engaged and involved in decisions about research participation and return of genomic research results (Victoria A. Miller et al., 2017). The proposal to have a third party discuss, mediate, and reconcile differences of opinion between parents and adolescents would certainly have resource implications in the context of genome sequencing research, but

would ensure a more robust informed assent process and comfort for participation in research within families for researchers eager to enroll adolescents in their research.

Limitations

This study involved focus groups with parents and adolescents from a single academic medical center, and may not be generalizable to other sites. Participants in this study were healthy volunteers who generally expressed interest in genomic research – enough so that they were interested in participating in a hypothetical study about genome sequencing research. This may or may not be consistent with adolescent patients who are invited to enroll in genome sequencing research to address health concerns of unknown origin. These considerations suggest that further research with adolescent patients and their parents enrolling in genome sequencing research is warranted.

BEST PRACTICES

As these results and discussion illustrate, adolescent decision-making in the context of genome sequencing research has been understudied in the return of results literature, resulting in all minors being treated similarly in existing recommendations. Incorporating empirical findings from focus groups with parents and adolescents regarding their decisional preferences into best practices guidelines may foster enthusiasm for adolescent participation in research and attend to hopes and concerns that parents and adolescents have about participation in genomic sequencing research. Facilitation of collaborative decision making by a researcher or health care provider may increase the likelihood that adolescents and their parents are engaged and involved in decisions about learning genomic research results.

RESEARCH AGENDA

Implementation of a collaborative decision-making process in a research setting where negative and positive genomic results are being returned to adolescents and parents is underway. Outcomes to be studied include understanding, choices made by adolescents and parents, and risks and benefits of return of results following a collaborative decision-making process. Findings from our focus groups may be relevant and have different resource allocation implications in the clinical context where results are returned for clinical exome and genome sequencing. Given the absence of adolescent perspectives in the most of the clinical and research genomic literature informing best practice guidelines, the similarities and differences between return of results to adolescents in research and clinical contexts ought to be addressed in future research.

EDUCATIONAL IMPLICATIONS

Our findings suggest researchers should engage adolescents in choices about return of genomic research results using a collaborative decision-making approach between adolescents and parents. Resources to train researchers in collaborative decision-making processes may be needed. In addition, it is recognized that participants will need some baseline knowledge to inform their choices and that researchers facilitating decision-making may have varying experience and knowledge about genetic testing. For this reason, we

created an online video that we are using to augment the informed consent process. The video, Genome Testing: Expectations and Results (https://goo.gl/4siQXK) is freely available on YouTube for other investigators to use.

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Table 1

Focus Group Participants

	Parents 3 focus groups (n=18)	**Adolescents 2 focus groups (n=15)**
Mean age (range)	45 (31-58)	15 (13-18)
Gender		
Male	3	4
Female	15	11
Race		
White	15	13
Black	2	0
Asian	1	3*
Hispanic		
Yes	1	0
No	17	15
Highest Level Education		
Post HS Training or Associates Degree	4	N/A
Bachelors Degree	8	
Masters or Doctoral Degree	6	
Grade		
6-8	N/A	6
9-12		9
Household Income		
$15,000-$29,999	0	N/A
$30,000-$44,999	1	
$45,000-$59,999	3	
$60,000-$89,999	5	
$90,000-$149,999	5	
$150,00 or more	4	
Marital Status		
Married	14	N/A
Divorced	1	
Never Married	2	
Living with Partner	1	
Participated in other research studies	9	6
If yes, was there a genetic component	0	0
How familiar are you with genetics or DNA?		

	Parents 3 focus groups (n=18)	**Adolescents 2 focus groups (n=15)**
Not at all familiar	1	1
Not very familiar	1	2
Somewhat familiar	13	11
Very familiar	3	1

* one participant checked both White and Asian