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Giving adolescents a voice: the types of genetic information adolescents choose to learn and why

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Abstract

Purpose: The American College of Medical Genetics and Genomics supports parents' opting in or out of secondary analysis of 59 genes when their child has clinical exome/genome sequencing. We explored the reasons adolescents choose to learn certain types of results and the reasons they want to involve or not involve parents in decision-making.

Methods: Adolescents recruited without clinical indication were offered independent, followed by joint choices with a parent to learn genomic results. After making independent choices, adolescent/parent dyads were interviewed to explore the reasons for their choices. Interviews were audio-recorded and transcribed. The constant comparative method was used to analyze 64 purposefully selected transcripts that included 31 from adolescents who excluded some or all potential results.

Results: Three major themes informed adolescents' choices: (1) actionability of information, (2) knowledge seeking, and (3) psychological impact. Of adolescents who independently excluded some conditions (n=31), 58% changed their initial choices during the joint interview due to parental influence or improved understanding. Nearly all adolescents (98%) wanted to be involved in the decision-making process, and 53% wanted to make choices independently.

Conclusions: Our findings contribute empirical evidence to support the refinement of professional guidelines for adolescents' engagement and preferences in genetic testing decisions.

Keywords

adolescents; genomic sequencing results; carrier testing; preferences; predictive testing

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DISCLOSURE

The authors declare no conflicts of interest.

INTRODUCTION

The American College of Medical Genetics and Genomics' (ACMG's) initial recommendations for the return of secondary results[1] and subsequent updates[2,3] challenged longstanding recommendations for predictive genetic testing in children.[4–6] Deliberation among bioethics and medical communities followed, focusing on protecting children's future autonomy, what results should be returned, how results should be returned, and to whom results should be returned.[7–14] Recent statements of best practice for clinical sequencing call for an engagement process between adolescent, parent, and clinician[6,15] that elicits, considers, and when possible, respects adolescents' preferences and concerns.[6,15] However, there is limited empirical evidence to demonstrate how adolescents want to be engaged in genomic testing decision-making, their preferences for parental involvement,[16,17] or the outcomes of that engagement.[18]

The adolescents who were tested during the study were engaged in predictive testing/genome screening[19] decision-making as they were recruited without clinical indication. While professional statements discourage predictive testing for adult-onset conditions before adulthood,[5,6] they also call for research to inform future best practice statements in the rapidly evolving area of genomic testing.[6,15] Our qualitative methods contribute to a much-needed body of evidence about what adolescents consider when making independent choices and how they consider their parent's perspectives during an engagement process.

MATERIALS AND METHODS

A longitudinal, mixed methods site-specific electronic Medical Records and Genomics (eMERGE) Network Phase III study was approved by Cincinnati Children's Hospital Medical Center (CCHMC) Institutional Review Board (IRB) (2016-3361). Adolescents ages 13 to 17 years old and one of their parents (adolescent/parent dyads) were enrolled following a written consent with parental permission and adolescent assent. Dyads made choices about learning clinically actionable genomic results from the eMERGE III sequencing panel (e3 panel). The e3 panel included genes for adult-onset conditions originally recommended by ACMG[1] and genes for autosomal recessive conditions selected by some of the eMERGE sites.[20]

Adolescents were recruited from the community and select CCHMC clinics with the intent of enrolling adolescents who would likely receive negative results. Adolescent eligibility criteria included having the capacity to make decisions and willingness to sign up for a patient-facing electronic health record portal where results would eventually be viewable.

We purposively sampled eMERGE III participants who enrolled and were interviewed from July 2016 to September 2017 to achieve near equal numbers of adolescents who chose to learn all possible results, and who chose to learn some or none of the possible results. Prior to the informed consent process, potential participants watched a genomics educational video (https://www.youtube.com/watch?v=fnhmveusEOg) that explained the meaning, limitations, and potential benefits and risks of negative and positive genomic results.

Following written informed consent and assent, parents and adolescents were separated and each independently completed a decision tool[16] to choose to learn results for conditions that were preventable, not preventable, or both; treatable, not treatable, or both; to include or exclude adult-onset conditions; and to include or exclude carrier results for autosomal recessive conditions. Adolescents and parents also independently chose whether they wanted to make their choices on their own, with their parent or adolescent, or defer the choice to their parent or adolescent.

After completing the decision tool, participants were given a list of the conditions informed by a subset of genes on the e3 panel determined to be clinically actionable and returnable by CCHMC experts as well as external experts within the eMERGE III Clinical Annotation Working Group. Brief clinical descriptions of the conditions were included. This list also indicated whether or not each condition was preventable, adult onset, and whether carrier status was possible. All conditions on the list were treatable. After reviewing the list, participants could choose to opt in or out of learning specific conditions, and could change the initial choices they made on the decision tool.

After making independent choices, parent/adolescent dyads reconvened to discuss their initial choices and to arrive at a joint decision. A member of the research team trained in qualitative methods facilitated discussions with parent/adolescent dyads. Participants were asked about their individual choices, what they thought about the other's choices, and what they perceived as risks and benefits for learning about the various conditions on the e3 panel. To better capture adolescents' unbiased responses, adolescents were asked to answer all questions before their parent. Dyads were reminded that only results upon which they agreed would be returned. They were encouraged to openly discuss any differences in opinion. After making joint choices, dyads were told they had 2 weeks to change their choices if desired and, as discussed at informed consent, they were free to withdraw from the study at any time.

The joint discussions were recorded, transcribed verbatim, and personal identifiers were removed. Transcripts of 64 adolescent/parent dyad interviews (average time, 17 minutes) were purposively selected from 88 interviews completed at the time of this study and downloaded into ATLAS.ti7. To explore differences between adolescents who initially chose to learn all results and adolescents who made discriminating choices, we selected transcripts from the 31 adolescents who made discriminating choices and the first 33 adolescents of 57 who chose to learn all results. While parent responses were also transcribed, we focused on adolescent responses to achieve our study aim.

Transcripts were analyzed using a constant comparative method.[21] Deductive and inductive codes were used for thematic analysis.[22] Deductive codes were derived from domains in our research questions. As themes emerged inductive codes were created and defined. Two members of the study team independently coded sets of transcripts, then discussed discrepancies until consensus was achieved. All themes, definitions, and sample quotes were reviewed and discussed by the entire study team for credibility.

RESULTS

Adolescent choices

Our study population consisted of 64 adolescents, with a median age of 15 (Table 1). With the exception of one, all adolescents within our sample chose to learn at least some results. Among adolescents whose independent choices excluded some or all results (n=31), the conditions most frequently excluded were conditions that were not preventable (58%) and not treatable (71%). Less commonly excluded were conditions that start in adulthood (35%) and carrier status (29%) (Table 2). After the joint discussion with their parent, 18 (58%) adolescents changed their choices, with the majority (n=14, 78%) selecting to learn more information and 4 adolescents choosing to learn less information.

Three major themes emerged from adolescent discussions about why they made their independent choices: (1) actionability of information, (2) knowledge seeking, and (3) psychological impact. When exploring reasons why adolescents changed their choices after the joint session, (4) parental influence and (5) improved understanding were the two major themes that arose. These five themes are summarized in Table 3 and described below.

Reasons for initial choices

Actionability of information—The ability to use information for some type of action was a common reason among adolescents wanting to learn about preventable, treatable, adult-onset conditions, and carrier status. A spectrum of actionable activities was described by 57 adolescents, the most common being to treat or prevent disease (63%), to prepare/plan for the future (47%), and to make changes (46%). Changes included lifestyle such as diet and exercise, and life choices such as family planning and career.

A clear difference in perception was detected between two groups of adolescents: those who chose to learn results for conditions that were both not treatable and not preventable, and those who chose to exclude those results. Those who wanted to learn results expressed a desire to have the ability to do something beneficial with the information, despite the inability to treat or prevent the disease.

(P44, 16 years old) You can be in control of your health. If you know then you can take steps in your life and in your environment to prevent things from getting worse and to improve the quality of your life.

In contrast, adolescents who chose to exclude results for conditions that were not treatable and not preventable (Table 2) expressed an inability to take action upon those results, and concern for negative consequences such as anxiety and worry. Those who didn’t want to learn carrier status reasoned that learning the information would alter their life choices, which was something they wanted to avoid.

(P66, 14 years old) I just don’t see why I should put myself through all that extra stress, and staying up late, and thinking about it when there is absolutely nothing I can do about it.

(P37, 16 years old) I didn't want [carrier status] to affect my choice to have children or something like that.

Four adolescents initially chose to exclude learning about conditions that were not preventable, but still learn about conditions that were not treatable.

(P56, 15 years old) Well, I did check that if they were preventable, so if there's something that's preventable but not treatable, then there's still something I can do about that if it's preventable.

Knowledge seeking—Adolescents in both groups expressed a desire for knowledge about one's health or to know what to expect. Adolescents who described that learning results would allow them to have a better understanding of their own health status and health risks did not necessarily describe potential actions in response to the awareness.

(P18, 15 years old) … I'd like to have as much information like if I'm going to have it, if I might have it, or if it's a possibility that I have it. Whether it's any sort of knowledge that I can know about myself I guess.

Similarly, adolescents who expressed importance in knowing what to expect in the future did not always follow with statements about actionable responses to the knowledge, despite probing.

(P6, 14 years old) Well, if they're preventable or treatable, then, now, I know that it's genetic or that it's going to happen maybe possibly in the future.

Adolescents considered their family when expressing reasons to learn results, such as a known family history, benefits to other family members, or considerations about future children and spouses.

(P24, 16 years old) …if it was something that I knew wasn't treatable or then because it's connected with my brother and sister and my parents and stuff so then they also can have an idea about something that might happen or a disease they could get.

Others wanted to learn results because they had limited knowledge about their family history and felt the information might help fill gaps.

(P16, 14 years old) We don't know a lot about my dad's side of the family because he was adopted. It's good information to have.

Adolescents who chose to learn all or some results tended to express that they like learning information, and that information is good. In contrast, adolescents who chose to exclude results did so because they didn't value knowing, the information wasn't relevant, or knowing was bad or scary.

(P4, 13 years old) Just to get the broad spectrum. I just like to know everything.

(P45, 15 years old) It's another bad thing, bad things I didn't wanna know about.

Psychosocial impact—When asked to describe the benefits and risks they considered when making their choices, adolescents' responses encompassed a range of responses toward psychological risk (mental well-being), behavioral risk (affecting life choices), and social risk (change in how others perceive them). The range of responses informed the theme Psychosocial Impact. For some adolescents, learning results for all of the conditions would decrease the burden of psychosocial impact and even provide benefits such as hope, relief, and peace of mind. For other adolescents, learning results for certain conditions would have negative consequences such as anxiety, worry, and distress.

(P19, 16 years old)…if you get something back that's positive, but it's treatable, then you're relieved.

(P5, 14 years old) I think [not treatable conditions] would give me anxiety. But I think for other people, it might put peace of mind thinking at least I have so and so much time until it sets in, so, I want to do all of this stuff before that.

It was common for adolescents from both groups to make discerning choices based on their perceived ability to incorporate risks. While they acknowledged that learning information may have negative consequences they felt risks were surmountable and learning information would decrease psychological impact.

(P35, 13 years old) I'd much rather know if I have a higher risk for cancer than for right when I'm 42 and I have five children, suddenly just smack in the face—you've got cancer.

(P48, 15 years old) Well, I know risks are like anxiety, like being really worried and stuff. I know that, but I feel like I have a strong enough mind that I'm gonna be like, "I'm not gonna let that affect me." I'm not gonna be overly worried.

In contrast, adolescents who chose to exclude results more readily expressed that they would feel increased psychological burden or that knowing results would negatively impact social interactions. Risks were perceived as insurmountable and superseded possible benefits.

(P63, 17 years old) Because if I had something that wasn't treatable, I honestly feel like I would just not do anything ever again. I'd just be sad all the time and feel like there's no hope.

Reasons adolescents changed their choices

Among the adolescents who wanted to learn some results (n=31), over half changed their initial choices (n=18). None of the adolescents who chose to learn all results changed their initial choices after the joint interview, even those (n=3) who were discordant with their parent.

Parental influence—Change in choices during the joint interview was largely due to noncoercive parental influence. Hearing the parent's rationale for choices sometimes provided information adolescents were not aware of or a perspective adolescents had not considered.

(P58, 15 years old) Let's include it, just because what you said about my aunt [having breast cancer]. I didn't know that.

(P8, 13 years old) It's just when I listened to what she said; it actually sounded better than what I said in my head.

Improved understanding—Eight adolescents changed their choices during the joint interview due to improved understanding. Adolescents' need for clarification usually occurred when the adolescent was asked to explain their reasons for their choices. Carrier status was the topic that was most often unclear to adolescents. Understanding was improved either through parent or interviewer explanation or the process of the adolescent thinking out loud and changing their mind during their responses.

(P59, 13 years old) I didn't really realize that you could tell you're a carrier for this, but you aren't at a higher risk for it [yourself].

Adolescents' preferences for making decisions

When making initial choices, 98% of adolescents wanted to be involved in the decision-making process. The majority of adolescents who wanted to learn results for all conditions also wanted to make decisions on their own (Table 4). Thirty adolescents made different independent choices than their parents: 3 in the group who wanted to learn all results and 27 of those who did not want to learn results for all types of conditions. Following the joint discussion, 14 parents deferred to the adolescents' decisions, 10 adolescents deferred to their parents' decisions, and the remaining six dyads compromised.

Among the 34 adolescents (53%) who selected to make decisions on their own, two major themes arose for their reasoning regardless of whether they chose to learn all or some results: (1) Exert Autonomy, and (2) Avoid Parental Influence. Reasoning among adolescents who selected to make decisions with their parents was informed by one major theme: (3) To Have a Say (Table 5).

Exert autonomy—Adolescents often expressed attitudes of independence, autonomy, and the capability of taking ownership for health-care decisions when explaining why they wanted to make decisions on their own. While their parent's perspective was recognized, these adolescents believed they should be making the decisions.

(P38, 17 years old) I mean, I think I'm old enough that I can make my own decisions. I mean, I'm gonna be an adult soon. I'm gonna need to learn. Not that I wouldn't want to make them with my mom, because she guides me and she helps me out with a lot. But with this, I don't think I really need too much guiding…

(P8, 13 years old) Well I'm not trying to be mean, Ma, but I wanted to learn about myself and try to figure out by myself and try to get—instead of trying to get help.

Avoid parental influence—When discussing why they wanted to make decisions independently, adolescents considered whether or not their choices might differ from their parent as they wanted to avoid being influenced by their parent.

(P49, 14 years old) …I guess it just depends on who I'm having the discussion with and what we're learning. So, I guess with dad, I would say we could do it together, but with my mom, I wouldn't be so sure about it…it's just I guess they have different opinions than I do and I wouldn't want their opinions to change the outcome of what I can learn about myself.

To have a say—The final theme, informed by adolescents who wanted to make decisions with their parent(s), was wanting to have a say. These adolescents valued their parents' perspective but wanted to have a voice in the decision-making.

(P2, 15 years old) Because it's like a big thing in my life, so, I still would want to have an opinion on it. I know my parents always know best for me. So, they would also probably have a really good opinion on it. And so, like just working it out, put them together.

Only one adolescent initially selected that she wanted her parent to make choices for her and an explanation was not provided. Two other adolescents who initially selected that they wanted to make choices independently voiced during the joint discussion that they preferred their parents make choices for them.

(P53, 14 years old) I feel like I rather have my mom make them for me. I don't really know a lot about the diseases and things like that, so I think it'd just be better if she made them for me.

DISCUSSION

To our knowledge, this is the first study to explore reasoning behind adolescents' preferences when given the option to learn *actual* positive and negative genomic sequencing results without clinical indication. Our findings indicate that adolescents want to be involved in decision-making, are able to articulate their reasons for their decisions, and recognize the value of the parents' perspectives. Finally, our findings ought to be considered when making future revisions to professional society positions that address adolescents' participation in decisions about testing for adult-onset conditions or carrier status.

Nearly all (98%) adolescents wanted to make, or be involved in making, decisions about the type of genomic results they wanted to learn about themselves. These findings support previous research in hypothetical contexts assessing what adolescents want to know and how much they wanted to be involved in making genetic testing decisions.[17,23] Pediatric bioethicists concur that assent ought to be solicited from adolescents in a developmentally appropriate manner and their dissent ought to be respected in the research context.[24–28] We purposefully asked adolescents about their reasons for their choices before engaging the parent to assure adolescents' perspectives were considered during the joint decision.

ACMG recommendations for the return of secondary results within the clinical context[2,3] discuss parents' decisional authority to opt in or out of learning all secondary results on behalf of their child but are silent about adolescent engagement. While the decision to learn secondary results falls under parental jurisdiction, we found that parents respected adolescent preferences and reasoning and often deferred to their adolescents' preferences when parental decisions differed from adolescent decisions. Similarly, Francis et al.[29]

demonstrated parents' willingness to support their adolescents' autonomy and preferences when dyads had discordant willingness to participate in a reproductive health clinical trial. However, differing cultural, religious, and family values may limit some parents' receptivity to their adolescent's engagement.[15,18]

Our results support the recent ACMG statement of best practices,[15] which advocates that adolescents' preferences ought to be independently considered when feasible before parents are asked to give permission about analysis and return of adolescents' secondary clinical or research genomic results. In our study, actionability was a key reason for adolescents' decisions about learning genomic results. However, adolescents' descriptions of actionability were not necessarily consistent with clinically actionable criteria used by ACMG[1,3] as some adolescents excluded treatable conditions that were not preventable but are recommended for return (e.g., Fabry disease and Li–Fraumeni syndrome). Similar to Hufnagel et al.,[17] adolescents in our study considered how their life choices and psychosocial responses might be influenced by anticipating disease onset that may or may not occur. In a different study, providers engaged adolescents simultaneously with their parents but reasoning was not explored when some adolescents offered brief opinions about learning or not learning secondary findings.[18]

Adolescents in our cohort had the capacity to make discretionary decisions based on self-awareness and weighing potential psychosocial risks. They considered whether knowledge of risk for treatable diseases in themselves or their children would result in desired actionability or negatively affect quality of life by eliciting emotions and choices they wanted to avoid. Similar to the adolescents in our study, adults who wanted to avoid learning certain types of results also weighed the potential burden of worry and impact on quality of life.[30,31] Other studies indicate that when given the choice, parents overwhelmingly want to learn their children's carrier results[32] and disease risk results, including those for conditions that are not medically actionable, or treatable in childhood.[33–35]

During the joint decision-making process, adolescents revealed when their choices were based on uncertain knowledge and recognized when their parent's knowledge and perspectives were needed. We found that carrier status was a source of confusion for some adolescents as it has been for adults in other studies.[36–38] While this could be a limitation of how carrier status was described in our decision tool, the joint discussion with their parent provided an opportunity to improve adolescents' understanding. Our joint discussion process was previously recommended by adolescents who recognized that their decisions may differ from those of their parents and that an impartial facilitator might enable the adolescent's different perspective to be considered.[16]

Our small sample size and enrollment from a single academic medical center limit the generalizability of our findings. However, our qualitative methods enabled us to explore the actual choices and perspectives of a largely ignored population for whom genomic health-care recommendations have been published and ethical positions have been deliberated. It is also recognized that our findings were informed by a subset of adolescents whose parents gave permission for their adolescent to undergo genomic testing without a clinical indication. While we gained empirical evidence about adolescents' perspectives when

making choices about the results they wanted to learn, we did not capture how adolescents actually reacted to results that were generated as return of results is underway at the time of this writing.

More studies, which include adolescents from multiple settings and life experiences, are needed to measure the factors that influence adolescent choices and measure adolescents' responses to learning results based on their choices. Yet, our findings that some adolescents choose differently than their parents, and that most adolescents wanted to make choices on their own or at least have a say, ought to give pause and reconsideration of offering parents choices to opt in or opt out of all of their child's secondary results. Findings from our and future studies are especially relevant for discussions about using exome/genome sequencing for newborn screening and storing the data for future interrogation. We demonstrate adolescents' capacity for making reasoned choices, and that facilitated engagement, as recommended in more recent society statements,[6,15] can enable adolescents and parents to reach a joint decision.

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Table 1

Adolescent demographics

Demographics	Learn all results (*N* = 33)	Discriminating choices (*N* = 31)	Total (*N* = 64)
Sex			
Female	23 (70%)	19 (61%)	42 (66%)
Male	10 (30%)	11 (35%)	21 (33%)
Other	0	1 (3%)	1 (2%)
Race			
White	27 (82%)	19 (61%)	46 (72%)
Black	4 (12%)	10 (32%)	14 (22%)
Other	2 (6%)	2 (7%)	4 (6%)
Age (median; range)	15; 13–17	15; 13–17	

Table 2

Types of conditions adolescents excluded during independent and joint choices ($N = 31$)

Types of conditions	Independent exclusions	Joint exclusions
Not treatable	22 (71%)	18 (58%)
Not preventable	18 (58%)	14 (45%)
Adult onset	11 (35%)	7 (23%)
Carrier status	9 (29%)	6 (19%)
Preventable	1 (3%)	1 (3%)
Treatable	1 (3%)	1 (3%)

Table 3

Reasons for adolescents' independent choices and changes during joint choices

Themes	Definition
Reasons for adolescents independent choices *(N* = 64)	
Actionability of information *(n* = 57)	Adolescents describe how learning results for certain types of conditions could or could not be used to initiate an action; this theme is also informed by adolescents who described that their exclusions of certain types of conditions were made to avoid influence on reproductive decisions or life choices
Knowledge seeking (*n* = 61)	Adolescents describe how learning results for conditions is important for the sake of knowledge, to satisfy curiosity, or to benefit family members
Psychosocial impact (*n* = 46)	Adolescents describe psychosocial benefits and risks to learning results for all or certain types of conditions
Reasons for adolescents' changes during joint choices (*N* = 31)	
Parental influence (*n* = 16)	Adolescent indicates a change in choice was because of the parent's reasons for wanting to learn adolescent's results for certain types of conditions
Improved understanding (*n* = 8)	Adolescent's statement or question reveals need for clarification and is followed by adolescent indicating change in their choice was influenced by additional information provided during the joint session

Table 4

Comparison of how adolescents wanted to make decisions between those who did or did not want to learn all conditions

How adolescent wanted to make decision	Learn all conditions (*n* = 33)	Don't learn all conditions (*n* = 31)	Total (*N* = 64)
On their own	20 (61%)	14 (45%)	34 (53%)
With their parent	13 (39%)	16 (52%)	29 (45%)
Parent make decision	0	1 (3%)	1 (2%)

Table 5

Reasons adolescents wanted to make choices on their own or with a parent

Themes	Definition
Make choices on their own ($N = 34$)	
Exert autonomy ($n = 21$)	Adolescent expresses importance of desire for independence, autonomy, and taking ownership of health-care decisions
Avoid parental influence ($n = 10$)	Adolescent describes active avoidance of parent opinions that might cause choices incongruent with adolescent's preferences
Make choices with parent ($N = 29$)	
To have a say ($n = 20$)	Adolescent describes valuing parent perspective but also expresses that they want or have the right to be involved in making choices about the type of conditions to learn