



Published in final edited form as:

Soc Sci Med. 2021 February ; 271: 112037. doi:10.1016/j.socscimed.2018.11.017.

A Behavior-Theoretic Evaluation of Values Clarification on Parental Beliefs and Intentions Toward Genomic Sequencing for Newborns

Ryan S. Paquin, PhD¹, Susana Peinado, PhD¹, Megan A. Lewis, PhD¹, Barbara B. Biesecker, PhD¹, Christine Rini, PhD², Myra Roche, MS, CGC³, Rita M. Butterfield, PhD³, Cynthia M. Powell, MD³, Jonathan S. Berg, MD, PhD³, Jr. Donald B. Bailey, PhD¹

¹RTI International, Research Triangle Park

²NC; Hackensack University Medical Center, Hackensack, NJ and Georgetown University School of Medicine, Washington, DC

³University of North Carolina at Chapel Hill, Chapel Hill, NC

Abstract

Decision aids commonly include values clarification exercises to help people consider which aspects of a choice matter most to them, and to help them make decisions that are congruent with their personal values and preferences. Using a randomized online experiment, we examined the influence of values clarification on parental beliefs and intentions about having genomic sequencing for newborns. We recruited 1,186 women and men ages 18–44 who were pregnant or whose partner was pregnant or planning to become pregnant in the next two years. Participants ($N = 1,000$) completed one of two versions of an online decision aid developed as part of a larger project examining the technical, clinical, and social aspects of using exome sequencing to screen newborns for rare genetic conditions. The education-only version provided information about using genomic sequencing to screen newborns for medically treatable conditions. The education-plus-values-clarification version included the same information, along with a values clarification exercise in which participants classified as *important* or *unimportant* five reasons in support of having and five reasons against having their newborn undergo genomic sequencing. We conducted partial correlations, regression analysis, and MANCOVAs with sex, health literacy, and experience with genetic testing as covariates. Participants who completed the decision aid with the values clarification exercise agreed less strongly with four of the five statements against sequencing compared to participants who viewed the education-only decision aid. The groups did not differ on agreement with reasons in support of sequencing. Agreement with four of five reasons against genomic sequencing was negatively associated with intentions to have their newborn sequenced, whereas agreement with all five reasons in support of sequencing were positively associated with intentions.

Correspondence concerning this article should be addressed to Ryan S. Paquin, Center for Communication Science, RTI International, 607 Washington Ave S, #405, Minneapolis, MN, 55415. Phone: 202-728-2041., rpaquin@rti.org.

Publisher's Disclaimer: This is a PDF file of an unedited manuscript that has been accepted for publication. As a service to our customers we are providing this early version of the manuscript. The manuscript will undergo copyediting, typesetting, and review of the resulting proof before it is published in its final citable form. Please note that during the production process errors may be discovered which could affect the content, and all legal disclaimers that apply to the journal pertain.

Keywords

United States; values clarification; decision aids; reasoned action; genomic sequencing; newborn screening; informed choice

Decision aids are used to facilitate informed decision making across a wide range of health-related choices. Their use in research and clinical care focuses on supporting preference-based decision making (O'Connor et al., 2007)—a perspective that emphasizes the consistency between a chosen course of action and what the decision maker values or hopes to achieve. Increasingly, genomic testing is being introduced into health care, and decisions to learn this information need to take into account the beliefs, preferences, and goals of individual decision makers. It is imperative that individuals make informed choices about undergoing genomic testing due to the degree of uncertainty and chances to learn unanticipated results. In this study, we offered parents hypothetical genomic test information about their children to understand how they make decisions about obtaining such information and to assess the usefulness of a theoretically-based values clarification intervention in an online decision aid.

A recent systematic review of outcomes from decision aid research concluded that decision aids reduce decisional conflict, indecision, and passive decision making, and they increase satisfaction with the process of making a decision as well as the final choices made (Stacey et al., 2017). These outcomes highlight how decision aids can prepare individuals to engage in meaningful discussions with health care professionals about treatment options, specifically decisions for which there may be no one best choice (Stacey et al., 2014). Decision aids can also help people form a preference for a treatment option based on personally-relevant values and beliefs (Elwyn et al., 2009). As such, decision aids are an evidence-based approach to support decision making in situations of uncertainty, like genomics.

Despite these noted benefits, it remains unclear how decision aids work to produce these benefits. They typically include five informational components: the decision to be made, background information relevant to the decision, available options, the risks or benefits of the options, and uncertainty about the options. Some also include another core feature: values clarification exercises to promote deliberation about aspects of the decision (Fagerlin et al., 2013; Stacey et al., 2017). Much of the literature examining the benefit of decision aids has focused on the process of values clarification and how it leads to better decisions; however, the underlying mechanism explaining the benefits of decision aids remains unclear. Understanding how values clarification leads to more informed choices, specifically the choice to obtain genomic information, may contribute to more effective decision aid design.

Researchers have continually called for greater conceptual clarity about how decision aids work to produce beneficial outcomes, and more specifically how values clarification supports decision making under conditions of uncertainty (Bekker et al., 2003; Fagerlin et al., 2013; Nelson et al., 2007). This challenge is likely due to the fact that most studies that use decision aids are not guided by an explicit theoretical framework (Winn et al., 2015). The purpose of decision aids is to help people who are facing difficult choices improve the

quality of the decisions they make (Bekker, 2010; Molenaar et al., 2000), with an emphasis on the process of arriving at a decision or the event that represents the culmination of that process. Making sense of decision making points to a model with three basic components: (1) *consequentialism*, or the idea that a good decision is based on an evaluation of expected outcomes for the available choice options; (2) *thorough structuring*, or the belief that decision making is facilitated when decision makers accurately anticipate the relevant consequences of these options both in terms of the likelihood they will occur and their value to the decision maker; and (3) a *compensatory decision rule*, or the notion that trade-offs in the desirability of relevant consequences should be reflected in the decision that is ultimately made (Frisch & Clemen, 1994; see also Fischhoff, 2005; Hastie & Dawes, 2010). Using a theoretical framework to guide decision aid studies could address a significant limitation in this research area and clarify how these components are translated into decision aid design.

The few studies using a theoretical framework to guide decision aid studies have used the reasoned action framework (Winn et al., 2015). This framework provides a logical approach to understand how decision aids may work, because it considers the role of attitudes as a direct antecedent of behavior. Attitudes in this context are defined as the degree to which people evaluate a behavior favorably or unfavorably, and they are formed from the positive and negative beliefs people hold toward the behavior (Fishbein & Ajzen, 2010). Values clarification—the process by which patients consider the desirability of different options or the attributes of those options in a specific decision context—runs parallel to the process of attitude formation with respect to behaviors. The reasoned action framework is consistent with the decision aid literature, which often equates preferences and attitudes (Marteau, Dormandy, & Michie, 2001; Sepucha & Ozanne, 2010; Winn et al., 2015). If decision aids aim to foster choices that are congruent with a decision maker's preferences, then an appropriate theoretic framework would elucidate the factors that lead people to make these choices.

The reasoned action framework is a model of behavioral prediction that integrates the theory of reasoned action (Fishbein & Azjen, 1975), the theory of planned behavior (Ajzen, 1991), and the integrative model of behavioral prediction (Fishbein, et al., 2001). The framework hypothesizes that behavioral intention, which is defined as the subjective likelihood of engaging in a behavior (or in our case making a decision), is the most significant and immediate precursor to whether people will actually enact the behavior (Fishbein & Ajzen, 2010). Behavioral intention stems from the attitudes, perceived norms, and perceived behavioral control individuals hold with regard to the behavior. Individuals holding more favorable views on these dimensions are more likely to engage in the behavior (Fishbein, Hennessy, Yzer, & Douglas, 2003). According to the framework, these direct antecedents of intention derive from a system of beliefs about the behavior under consideration, with qualitative distinctions between the beliefs that underlie each predictor. For example, an attitude toward a particular behavior is determined by *behavioral beliefs*, or the expected consequences of performing the behavior weighted by evaluations of these outcomes.

The model of attitude formation given in the reasoned action framework matches well to values clarification exercises that ask individuals to deliberate on the pros and cons of a particular option or decision (Fagerlin et al., 2013). Generally, the more a parent believes

that learning actionable genomic information about their child will lead to “good” outcomes and avert “bad” outcomes, the more favorable the parent’s attitudes for engaging in the behavior (Fishbein, 2008). With this in mind, we focus on the attitudinal pathway of the reasoned action framework in this study. Importantly, attitudes derive from beliefs about the behavior and should be directly proportional to the summed ratings of the positive and negative consequences that readily come to mind when one considers performing the behavior in question (Fishbein & Ajzen, 1975). To the extent that a values clarification exercise helps people consider and process information related to behavioral beliefs, it can impact the behavior (i.e., choosing to have a child’s genome sequenced) by exerting influence on behavioral beliefs and attitudes. Thus, forming and clarifying beliefs are processes that are likely central to understanding how values clarification exercises may impact decision making.

Making decisions about genomic information is likely a relatively unfamiliar context for most parents, and a situation for which they have little direct experience, making it a preference or values-sensitive decision. Thus, the decision-making context is characterized by uncertainty for parents, and deliberating on beliefs or values could help them form preferences and decisions about whether or not to obtain genomic information about their child. With these considerations in mind about the potential links between beliefs, attitudes, values clarifications and behavioral intentions, we examined the following research questions: 1) Will a values clarification exercise influence beliefs about genomic information? And 2) Will beliefs about genomic information predict behavioral intentions?

Method

We conducted an online experiment to examine the influence of a values clarification exercise on beliefs and behavioral intentions for simulated decisions to have genomic sequencing for one’s child related to actionable genetic conditions.

Participants

We recruited a nonprobability-based U.S. sample via online panels maintained by Qualtrics. Potential respondents were sent an email invitation informing them that the study was for research purposes only, how long the survey was expected to take, and what incentives were available. Eligible participants included women and men ages 18 to 44 who were either pregnant or had a spouse or partner who was pregnant, were actively trying to get pregnant, or were contemplating or preparing for pregnancy within the next two years.

Procedure

Eligible participants completed a consent form and answered questions in an online survey to assess health literacy and experience with genetic testing. They were then randomly assigned to one of two decision aid conditions. In the control condition, participants viewed only educational information, whereas in the experimental condition they viewed the same educational information followed by a values clarification exercise. Participants clicked on a link in the online survey to take them to the decision aid. After completing the online decision aid, they answered questions about the potential outcomes of having genomic

sequencing, decision making self-efficacy, their intention to have the sequencing test for their child, and demographics.

Decision Aid

The decision aid content used in this study was developed for a larger project called the North Carolina Newborn Exome Sequencing for Universal Screening (NC NEXUS) study (Lewis et al., 2018; Milko et al., 2018). The NC NEXUS project aims to evaluate the utility of genomic sequencing as an alternative and extension to the currently administered newborn screening tests, to learn how parents make decisions about obtaining genomic sequencing for their child, and to investigate whether a decision aid would help them make informed decisions about genomic sequencing.

The decision aid was developed to help parents make informed decisions about genomic sequencing for their newborn child. Two versions of the decision aid were created for this study (see Supplementary Material). Both versions included identical educational content and one version also included the values clarification exercise described below. The educational content in both versions of the decision aid includes information about newborn screening, genomic sequencing, and the use of genomic sequencing to identify genetic variants that could lead children to develop conditions for which treatments exist. It also describes how the sequencing test is performed and what to do if the results reveal a child may be at risk for developing a genetic condition. In the education-plus-values-clarification version, this information is followed by a values clarification exercise in which parents sort five reasons for and five reasons against having genomic sequencing for their child by importance. These reasons for and against having genomic sequencing were developed based on input from couples who were pregnant or had recently given birth (Fitzgerald et al., 2016; Moultrie et al., in preparation), suggesting that they represent broadly salient, or accessible, beliefs about having genomic sequencing. Parents completing the decision aid are also given the opportunity to add and sort any additional reasons that were important to their decision making. This task is followed by five questions to further clarify values. In both versions of the decision aid, parents are asked to make a decision about whether they want their child to undergo genomic sequencing. Instructions in this study made it clear that this decision is hypothetical.

Measures

Behavioral beliefs related to genomic sequencing were assessed by asking participants how much they agreed or disagreed with five statements expressing reasons for having genomic sequencing and five statements expressing reasons for not having genomic sequencing. Both sets of reasons were derived from the reasons for and against genomic sequencing included in the values clarification exercise in the decision aid. Reasons for sequencing included statements such as “genomic sequencing will help doctors better understand many genetic conditions” and “a positive result could help my family better plan for the future.” Reasons against sequencing included statements such as “knowing that researchers are studying my child’s genomic sequencing information makes me uncomfortable” and “waiting to learn my child’s genomic sequencing results may cause me to worry or feel anxious.” Responses were given on a 5-point scale from 1 (*strongly disagree*) to 5 (*strongly agree*). For some analyses,

we averaged the reasons for sequencing ($M = 4.21$, $SD = 0.66$, $\alpha = .87$) and reasons against sequencing ($M = 2.95$, $SD = 0.82$, $\alpha = .75$) into separate composite index variables. We used this data reduction strategy for regression analyses to overcome multicollinearity issues encountered when all ten behavioral beliefs were entered as separate predictors.

Behavioral intentions were assessed by asking participants how likely they would be to get the genomic sequencing test described in the decision aid for their child if they could request it ($M = 3.74$, $SD = 1.09$). Responses were given on a 5-point scale ranging from 1 (*extremely unlikely*) to 5 (*extremely likely*).

Health literacy (Morris et al., 2006) was measured by asking participants the following: How often do you need to have someone help you when you read instructions, pamphlets, or other written material from your doctor or pharmacy? Responses were given on a 5-point scale from 1 (*never*) to 5 (*always*) and reverse coded so that a higher score represented better health literacy ($M = 4.40$, $SD = 0.99$).

Experience with genetic testing was assessed by asking a series of five questions about participants' personal experiences, including whether they had ever discussed, made a decision about, or had a genetic test for themselves or their child. We recoded responses into a new dichotomous variable indicating previous experience, 1 (*yes*) or 0 (*no*). Twenty-two percent ($n = 215$) reported having some experience with genetic testing.

Statistical Analyses

We first conducted two MANCOVAs to examine whether there were differences in beliefs between the two experimental conditions (education-only and education-plus-values-clarification). One MANCOVA included the five behavioral-belief statements for accepting genomic sequencing and the other included the five behavioral-belief statements for declining genomic sequencing.

To examine whether behavioral beliefs were associated with behavioral intention, we conducted partial correlations. An examination of the correlations between beliefs and intention can identify beliefs that may underlie attitudes and influence intention. To further apply the reasoned action approach, we tested a regression model predicting behavioral intention that included two index variables aggregating, respectively, the reasons for and against having genomic sequencing as predictors. Finally, we conducted moderation analyses using multiple regression to test whether exposure to the values clarification portion of the decision aid moderated the relationship between beliefs and intention. Building on the regression analysis predicting intention using the two aggregate index variables to represent reasons for and against sequencing, we added interaction terms to form a moderation model. As neither of the interactions between decision aid condition and beliefs were significant, they are not reported below. Gender, health literacy, and experience with genetic testing were included as covariates in all analyses.

Results

Sample Description

Of the 1,186 eligible participants recruited, 186 participants did not complete the decision aid and were excluded from the final sample. Of the 1,000 who completed the decision aid, 72% ($n = 716$) were female. The mean age was 30.2 years ($SD = 5.29$). Demographic characteristics of the analytic sample are presented in Table 1. As a randomization check, we conducted chi-square tests of independence comparing each of the demographic variables by experimental condition. All p values were greater than .05, indicating that randomization succeeded in yielding two groups with comparable demographic profiles.

Did A Values Clarification Exercise Influence Behavioral Beliefs?

Exposure to the values clarification exercise significantly influenced participants' agreement with most of the statements against sequencing, $F(5, 991) = 11.07, p < .001$ (Table 2). Compared to participants who viewed only educational content, those who completed the values clarification exercise expressed weaker agreement with these four statements: "Waiting to learn my child's genomic sequencing results may cause me to worry or feel anxious" ($M = 3.77, SD = 0.97$ vs. $M = 3.37, SD = 1.24$), $F(1, 955) = 34.88, p < .001$; "I would rather wait to see if my child has signs of a genetic condition before having genomic sequencing" ($M = 2.72, SD = 1.21$ vs. $M = 2.51, SD = 1.24$), $F(1, 955) = 5.21, p = .013$; "I am satisfied with knowing that my child will have the standard newborn screening test like all babies" ($M = 3.66, SD = 0.95$ vs. $M = 3.43, SD = 1.08$), $F(1, 955) = 13.83, p < .001$; and "I do not want to learn now if my child is expected to develop a genetic condition in the future" ($M = 2.42, SD = 1.21$ vs. $M = 2.26, SD = 1.17$), $F(1, 955) = 3.98, p = .046$. We found no significant differences between the two decision aid conditions among reasons in support of sequencing, $F(5, 991) = .18, p = .97$.

Did Beliefs Predict Behavioral Intentions?

As an initial exploration of the relationship between beliefs and behavioral intentions, we examined partial correlations. Across all participants, "waiting to learn my child's genomic sequencing results may cause me to worry or feel anxious" was the only belief or value statement that was not significantly associated with behavioral intention ($r_p = -.04, p = .230$). Agreement with the remaining four reasons against genomic sequencing were negatively associated with intention to have genomic sequencing (range $r_p = -.52$ to $-.24, p < .001$), and all five reasons in support of genomic sequencing were positively associated with intention (range $r_p = .32$ to $.56, p < .001$). These reasons for and against sequencing are shown in Table 3 along with statistics detailing their association with behavioral intention.

Because these exploratory analyses revealed that most beliefs were significantly correlated with intentions, we tested a regression model that included the covariates and the two index variables representing aggregate reasons for and against having genomic sequencing as predictors with behavioral intention as the dependent variable (Table 4). When the composite reasons for and against genomic sequencing indices were added to the model with the other predictors, they accounted for an additional 34% of the variance in behavioral intention ($R^2 = .34, F(2, 993) = 265.11, p < .001$). In this model, stronger agreement with

reasons for having genomic sequencing was associated with a greater intention to have genomic sequencing, $B = 0.67$, $SE = 0.05$, $t(993) = 14.97$, $p < .001$. On balance, stronger agreement with reasons against having genomic sequencing corresponded to lower intentions, $B = -0.42$, $SE = 0.04$, $t(993) = -11.39$, $p < .001$.

Auxiliary Mediation Analysis.—Exposure to the values clarification exercise did not have a direct impact on behavioral intentions toward genomic sequencing. In a follow-up test for mediation conducted using ordinary least squares path analysis (Hayes, 2013), we found that the values clarification exercise indirectly influenced behavioral intentions through its effect on the aggregate reasons-against index. On average, participants who completed the values clarification exercise agreed less strongly with the reasons-against statements than those in the education-only arm of the study ($B = -0.18$, $SE = 0.05$, 95% CI $[-0.28, -0.08]$, $p < .001$). Combined with the significant negative association between reasons against having genomic sequencing and intention, we observed a significant indirect effect of the values clarification exercise on intentions that was mediated through the reasons-against index ($B = 0.07$, $SE_{bootstrap} = 0.02$, 95% CI_{bootstrap} $[0.03, 0.12]$, $p < .001$). The values clarification exercise did not have a significant average effect on the reasons-for beliefs ($B = 0.01$, $SE = 0.04$, $p = .889$), and so did not impact behavioral intentions indirectly through the reasons-for index ($B = 0.00$, $SE_{bootstrap} = 0.03$, 95% CI_{bootstrap} $[-0.05, 0.06]$, $p = .889$).

Discussion

We used the reasoned action framework to shed light on how a values clarification exercise embedded in a decision aid influences parental decisions about obtaining genomic information for their children. This framework provides a conceptual approach to help explain how values clarification supports decision making under conditions of uncertainty (Bekker et al., 2003; Fagerlin et al., 2013; Nelson et al., 2007), and starts to address the underlying mechanisms related to values clarification.

This study preceded the larger NC NEXUS trial (Milko et al., 2018), and was conducted to help us understand the role of the decision aid on actual decisions being made in the trial. In NC NEXUS, parents of newborns and parents of young children with a confirmed diagnosis are offered whole exome sequencing. Before deciding to have whole exome sequencing for their child, parents use the online decision aid described in this paper—including both the educational component and the values clarification exercise—and receive in-person counseling. The larger NC NEXUS trial will not allow us to compare versions of the decision aid, as we did here, but will address other research questions. Specifically, sequencing decisions in NC NEXUS are not hypothetical, so we will be able to look at parents' actual behavior in addition to behavioral intentions.

A strength of this study was its experimental design, which allowed us to draw causal inferences about the provision of values clarification versus an education-only approach. Applying the reasoned action framework in this way provides descriptive information about the compensatory decision rule used by parents making sequencing decisions in the larger NC NEXUS study. A similar approach could be used to expand the interactive features of

future decision aids. A potential model for this kind of extension is available in the values clarification portion of DECIDE—a general-purpose decision-support tool for parents making decisions about genome-wide sequencing for children with a suspected genetic condition (Birch et al., 2016). Similar to the values-sorting approach used in the NC NEXUS decision aid, participants using the DECIDE tool review a list of issues related to a genome-wide sequencing decision (e.g., harms and benefits) and select those issues that are personally important to them. Unlike our tool, however, DECIDE users then rate the relative importance of the selected issues and an algorithm produces a choice recommendation based on these responses (Bansback, Li, Lynd, & Bryan, 2014). A similarly re-envisioned values clarification task based on the reasoned action framework might ask participants to rate the expected consequences and outcome evaluations of the behavioral options in question.

The MANCOVA results demonstrate that contemplating the personal importance of reasons against having one's child undergo genomic sequencing weakened agreement with most of those reasons. This may signify that participants who completed the values clarification exercise engaged more deeply with the educational content in the decision aid compared to those in the education-only condition, although research assessing an indicator of engagement would be needed to confirm this possibility. The values clarification task did not attempt to persuade participants to agree or disagree with any of the reasons, but instead asked them to sort the reasons by personal importance. The changes in belief strength relating to completion of the values clarification exercise may be the result of self-generated thoughts elaborating on the decision aid content.

The results of the correlational and regression analyses suggest that most of the reasons for and against having genomic sequencing for one's child that were included in the values clarification exercise were indeed salient to our target population, lending support to an important methodological assumption. We observed moderate to strong bivariate associations between all but one of the behavioral beliefs and intentions. The only belief that was not significantly related to behavioral intentions was the idea that waiting to learn genomic sequencing results would cause worry or anxiety. The remaining reasons against genomic sequencing were all negatively associated with behavioral intentions, but the absolute magnitude of these associations fell within a wider range than the correlations with the reasons for sequencing. Two reasons against genomic sequencing were strongly associated with intentions (i.e., "I would rather wait to see if my child has signs of a genetic condition..." and "I do NOT want to learn now if my child is expected to develop a genetic condition..."). The bivariate associations between all five reasons for genomic sequencing and intentions were positive, fell within a narrower range than the correlations with the reasons against sequencing, and were generally strong. These differences in bivariate associations were also evident in the regression model, where the index representing reasons in support of sequencing emerged as a stronger predictor of intention than the index representing reasons against sequencing. Taken together, the results suggest that participants' attitudes toward having their child undergo genomic sequencing integrates a constellation of behavioral beliefs, representing both reasons for and against sequencing, but influenced slightly more by reasons in support of having genomic sequencing.

Interestingly, an *ad hoc* mediation analysis revealed that exposure to the values clarification exercise had a positive indirect effect on behavioral intentions through its effect on the reasons-against index. Participants who completed the values clarification exercise expressed weaker agreement with reasons against sequencing, which in turn was associated with stronger intentions to have genomic sequencing.

Limitations

Although this study advances what we know about how values clarification works to produce a potential benefit, there are features of the study design that limit our conclusions. The non-probability sampling approach yielded a sample that was more highly educated and less diverse than we would have desired. In addition, all participants were English speaking, further restricting our conclusions about how values clarification might work in non-English speaking groups. Other limitations include the hypothetical nature of the decision-making scenario presented to parents, and the related emphasis on behavioral intentions as the outcome and not actual behaviors. Also, we did not focus on all the concepts that are part of the reasoned action framework, and future studies that include all aspects of the framework can advance this area of research by examining the relative contribution of attitudes, subjective norms, and perceived behavioral control on decisions related to genomic information.

Conclusion

The use of genomic information in research studies, public health, and clinical practice will continue to grow as the technology for generating this information becomes faster, more affordable, and more clinically relevant. With the use of this information in a variety of settings, a greater burden will be placed on individuals to decide if, and what type, of genomic information they want to learn. Our application of the reasoned action framework to understanding the link between values clarification, beliefs, and intentions suggest that belief formation may be a central process through which values clarification fosters the formation of preferences. The simulated decision-making approach and experimental design used in this study provides valuable information about the role of beliefs in values clarification that can be applied to future studies examining actual decisions in research studies and clinical settings. Taken together, the results provide researchers and practitioners behavior-theoretic insight on the function of values clarification tasks in decision aids for genomic sequencing.

Supplementary Material

Refer to Web version on PubMed Central for supplementary material.

Acknowledgements

The research reported here was funded by the Eunice Kennedy Shriver National Institute of Child Health and Human Development/National Human Genome Research Institute grant to Drs Powell and Berg (1 U19 HD077632-01). Funded by the National Institutes of Health (NIH).

References

- Ajzen I (1991). The theory of planned behavior. *Organizational Behavior and Human Decision Processes*, 50, 179–211. doi:10.1016/0749-5978(91)90020-T
- Bansback N, Li LC, Lynd L, & Bryan S (2014). Development and preliminary user testing of the DCIDA (Dynamic computer interactive decision application) for ‘nudging’ patients towards high quality decisions. *BMC Medical Informatics and Decision Making*, 14, 62. doi:10.1186/1472-6947-14-62. [PubMed: 25084808]
- Birch P, Adam S, Bansback N, Coe RR, Hicklin J, Lehman A, et al. (2016). DECIDE: A decision support tool to facilitate parents’ choices regarding genome-wide sequencing. *Journal of Genetic Counseling*, 25, 1298–1308. doi:10.1007/s10897-016-9971-8 [PubMed: 27211035]
- Bekker HL (2010). The loss of reason in patient decision aid research: Do checklists damage the quality of informed choice interventions? *Patient Education and Counseling*, 78, 357–364. doi:10.1016/j.pec.2010.01.002 [PubMed: 20171821]
- Bekker HL, Hewison J, & Thornton JG (2003). Understanding why decision aids work: linking process with outcome. *Patient Educ Couns*, 50(3), 323–329. doi:10.1016/S0738-3991(03)00056-9. [PubMed: 12900106]
- Elwyn G, O’Connor AM, Bennett C, Newcombe RG, Politi M, Durand MA, et al. (2009). Assessing the quality of decision support technologies using the International Patient Decision Aid Standards instrument (IPDASi). *PLOS ONE*, 4(3), e4705. doi:10.1371/journal.pone.0004705. [PubMed: 19259269]
- Fagerlin A, Pignone M, Abhyankar P, Col N, Feldman-Stewart D, Gavaruzzi T, et al. (2013). Clarifying values: an updated review. *BMC Med Inform Decis Mak*, 13 Suppl 2, S8. doi:10.1186/1472-6947-13-s2-s8.
- Fischhoff B (2005). Decision research strategies [Supplement]. *Health Psychology*, 24, S9–S16. doi:10.1037/0278-6133.24.4.S9 [PubMed: 16045425]
- Fishbein M (2008). A reasoned action approach to health promotion. *Medical Decision Making*, 28, 834–844. doi:10.1177/0272989X08326092. [PubMed: 19015289]
- Fishbein M, & Ajzen I (1975). Belief, attitude, intention, and behavior: An introduction to theory and research [PDF version]. Reading, MA: Addison-Wesley Retrieved from <http://people.umass.edu/ajzen/f&a1975.html>
- Fishbein M, & Ajzen I (2010). *Predicting and changing behavior: The reasoned action approach*. New York: Psychology Press.
- Fishbein M, Hennessy M, Yzer M, & Douglas J (2003). Can we explain why some people do and some people do not act on their intentions? *Psychology, Health & Medicine*, 8, 3–18. doi:10.1080/13540021000059223.
- Fishbein M, Triandis HC, Kanfer FH, Becker M, Middlestadt SE, & Eichler A (2001). Factors influencing behavior and behavior change In Baum A, Revenson TA, & Singer JE (Eds.), *Handbook of health psychology* (pp. 3–16). Mahwah, NJ: Erlbaum.
- Fitzgerald TM, Lewis MA, Moultrie RR, Zulkiewicz BA, Rini C, Roche M, et al. (2016). Couple dynamics in decisions about newborn screening via whole exome sequencing 2016 APHL Newborn Screening and Genetic Testing Symposium. St. Louis, MO.
- Frisch D, & Clemen RT (1994). Beyond expected utility: Rethinking behavioral decision research. *Psychological Bulletin*, 116, 46–54. doi:10.1037//0033-2909.116.1.46 [PubMed: 8078974]
- Hastie R, & Dawes RM (2010). *Rational choice in an uncertain world: The psychology of judgment and decision making* (2nd ed.). Thousand Oakes, CA: Sage.
- Hayes AF (2013). *Introduction to mediation, moderation, and conditional process analysis. A regression-based approach*. New York: Guilford.
- Lewis M, Paquin R, Roche MI., Furberg R, Rini C, Berg JS, ...Bailey D (2016). Supporting Parental Decisions About Genomic Sequencing for Newborn Screening: The NC NEXUS Decision Aid. *Pediatrics*, 137(Suppl. 1), S16–S23. doi:10.1542/peds.2015-3731E [PubMed: 26729698]
- Marteau TM, Dormandy E, & Michie S (2001). A measure of informed choice. *Health Expectations*, 4, 99–108. doi:10.1046/j.1369-6513.2001.00140.x [PubMed: 11359540]

- Milko LV, Rini C, Lewis MA, Butterfield RM, Lin F-C, Paquin RS, ... Powell CM (2018). Evaluating parents' decisions about next-generation sequencing for their child in the NC NEXUS (North Carolina Newborn Exome Sequencing for Universal Screening) study: A randomized controlled trial protocol. *Trials*, 19. doi:10.1186/s13063-018-2686-4
- Molenaar S, Sprangers MAG, Postma-Schuit FCE, Rutgers E. J. Th., Noorlander J, Hendriks J, & De Haes HCJM (2000). Interpretive review: Feasibility and effects of decision aids. *Medical Decision Making*, 20, 112–127. doi:10.1177/0272989X0002000114 [PubMed: 10638544]
- Morris NS, MacLean CD, & Littenberg B (2006). Literacy and health outcomes: a cross-sectional study in 1002 adults with diabetes. *BMC Fam Pract*, 7, 49. doi:10.1186/1471-2296-7-49. [PubMed: 16907968]
- Moultrie R, Lewis MA, Fitzgerald TM, Zulkiewicz B, Rini C, Roche M, et al. (in preparation). Values and beliefs important for parental decisions to have genomic sequencing for a child.
- Nelson WL, Han PK, Fagerlin A, Stefanek M, & Ubel PA (2007). Rethinking the objectives of decision aids: a call for conceptual clarity. *Med Decis Making*, 27(5), 609–618. doi:10.1177/0272989X07306780. [PubMed: 17873251]
- O'Connor AM, Wennberg JE, Legare F, Llewellyn-Thomas HA, Moulton BW, Sepucha KR, ... King JS (2007). Toward the 'tipping point': decision aids and informed patient choice. *Health Affairs*, 26(3), 716–725. doi:10.1377/hlthaff.26.3.716. [PubMed: 17485749]
- Sepucha K, & Ozanne EM (2010). How to define and measure concordance between patients' preferences and medical treatments: A systematic review of approaches and recommendations for standardization. *Patient Educ Couns*, 78(1), 12–23. doi:10.1016/j.pec.2009.05.011. [PubMed: 19570647]
- Stacey D, Legare F, Col NF, Bennett CL, Barry MJ, Eden KB, et al. (2014). Decision aids for people facing health treatment or screening decisions. *Cochrane Database Syst Rev*, 1, CD001431. doi:10.1002/14651858.CD001431.pub4.
- Stacey D, Legare F, Lewis K, Barry MJ, Bennett CL, Eden KB, et al. (2017). Decision aids for people facing health treatment or screening decisions. *Cochrane Database Syst Rev*, 4, CD001431. doi:10.1002/14651858.CD001431.pub5.
- Winn K, Ozanne E, & Sepucha K (2015). Measuring patient-centered care: An updated systematic review of how studies define and report concordance between patients' preferences and medical treatments. *Patient Educ Couns*, 98(7), 811–821. doi:10.1016/j.pec.2015.03.012. [PubMed: 25846191]

Highlights

- Decision aids often use values clarification exercises to support decision making.
- Beliefs impact parents' intentions toward genomic sequencing for their newborns.
- Values clarification weakened beliefs that express reasons against sequencing.
- By changing beliefs, values clarification indirectly influenced intentions.

Table 1.

Participant characteristics (N = 1000)

Demographics	n	(%)
Gender		
Female	716	(71.6)
Male	284	(28.4)
Age		
18–30	545	(54.5)
31–44	455	(45.5)
Marital status		
Married	834	(83.4)
Marriage-like relationship or domestic partnership	121	(12.1)
Single, separated, divorced, widowed, or other	45	(4.5)
Educational attainment		
High school graduate or less	249	(24.9)
Trade school or 2-year college degree	127	(12.7)
4-year college degree	385	(38.5)
Graduate or professional degree	237	(23.7)
Income		
Less than \$30,000	114	(11.4)
\$30,000 to \$44,999	145	(14.5)
\$45,000 to \$59,999	149	(14.9)
\$60,000 to \$74,999	147	(14.7)
\$75,000 to \$89,999	140	(14.0)
\$90,000 to \$119,999	173	(17.3)
\$120,000 or more	131	(13.1)
Race/Ethnicity		
White only, non-Hispanic	719	(71.9)
Black only, non-Hispanic	60	(6.0)
Other race, non-Hispanic	123	(12.3)
Hispanic or Latino	98	(9.8)
Health insurance		
Yes	942	(94.2)
No or don't know	58	(5.8)
Genetic testing experience		
Yes	215	(21.5)
No	785	(78.5)

Table 2.

Impact of values clarification on the strength of reasons against sequencing

Behavioral beliefs	Education-only (<i>n</i> = 518)		Education + values clarification (<i>n</i> = 482)		<i>F</i> (1, 995)	<i>p</i>
	<i>M</i>	(<i>SD</i>)	<i>M</i>	(<i>SD</i>)		
Knowing that researchers are studying my child's genomic sequencing information makes me uncomfortable	2.62	(1.19)	2.72	(1.34)	1.51	.220
Waiting to learn my child's genomic sequencing results may cause me to worry or feel anxious	3.77	(0.97)	3.37	(1.24)	34.88	<.001
I would rather wait to see if my child has signs of a genetic condition before having genomic sequencing	2.72	(1.21)	2.51	(1.24)	6.21	.010
I am satisfied with knowing that my child will have the standard newborn screening test like all babies	3.66	(0.95)	3.43	(1.08)	13.83	<.001
I do not want to learn now if my child is expected to develop a genetic condition in the future	2.42	(1.21)	2.26	(1.17)	3.98	.046

Table 3.

Behavioral Beliefs and Intentions

Behavioral beliefs		<i>M</i>	(<i>SD</i>)	<i>r_p</i>
1F	Genomic sequencing could help scientists develop tests that find serious conditions before a child develops them	4.24	(0.73)	.37***
2F	Knowing that a child has a genetic condition may allow parents to get early treatment and support services	4.40	(0.68)	.32***
3F	Genomic sequencing will help doctors better understand many genetic conditions	4.20	(0.79)	.37***
4F	A positive result could help my family better plan for the future	4.23	(0.81)	.43***
5F	It is better to have all possible tests that could tell me about my child's future health even if nothing is found	3.96	(1.00)	.56***
1A	Knowing that researchers are studying my child's genomic sequencing information makes me uncomfortable	2.67	(1.26)	-.24***
2A	Waiting to learn my child's genomic sequencing results may cause me to worry or feel anxious	3.58	(1.13)	-.04
3A	I would rather wait to see if my child has signs of a genetic condition before having genomic sequencing	2.62	(1.23)	-.50***
4A	I am satisfied with knowing that my child will have the standard newborn screening test like all babies	3.55	(1.02)	-.25***
5A	I do NOT want to learn now if my child is expected to develop a genetic condition in the future	2.34	(1.91)	-.52***

Note. F = Reason for genomic sequencing. A = Reason against genomic sequencing.

* $p < .05$,

** $p < .01$,

*** $p < .001$

Table 4.

Regression Analysis Predicting Intention from Reasons For and Against Genomic Sequencing

Predictor	R^2	B	SE	β	95% CI
Constant		2.50***	0.30		[1.91, 3.09]
Step 1	.03***				
Experience with genetic testing ^a		0.28***	0.07	.11	[0.15, 0.42]
Gender ^b		-0.11	0.06	-.05	[-0.23, 0.01]
Health literacy		-0.08**	0.03	-.07	[-0.14, -0.02]
Decision aid condition ^c		0.01	0.06	.01	[-0.10, 0.12]
Step 2	.34***				
Reasons-for index		0.67***	0.05	.40	[0.59, 0.76]
Reasons-against index		-0.42***	0.04	-.31	[-0.49, -0.34]
Total R^2	.37				
Total $F(6, 933)$	95.10***				

Note. $N = 1,000$. CI = confidence interval. Regression coefficients and standard errors from the final model (Step 2) are presented here. Interaction terms crossing the reasons-for and reasons-against indices by intervention condition were added in Step 3. These terms did not

* $p < .05$

** $p < .01$.

*** $p < .001$.

^aReference category is no experience with genetic testing.

^bReference category is males.

^cReference category is the education-only group.