Published in final edited form as:
Soc Sci Med. 2012 May ; 74(10): 1536–1543. doi:10.1016/j.socscimed.2012.02.003.

The Decision-Making Process of Genetically At-Risk Couples Considering Preimplantation Genetic Diagnosis: Initial Findings from a Grounded Theory Study

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Abstract

Exponential growth in genomics has led to public and private initiatives worldwide that have dramatically increased the number of procreative couples who are aware of their ability to transmit genetic disorders to their future children. Understanding how couples process the meaning of being genetically at risk for their procreative life lags far behind the advances in genomic and reproductive sciences. Moreover, society, policy makers, and clinicians are not aware of the experiences and nuances involved when modern couples are faced with using Preimplantation Genetic Diagnosis (PGD). The purpose of this study was to discover the decision-making process of genetically at-risk couples as they decide whether to use PGD to prevent the transmission of known single-gene or sex-linked genetic disorders to their children. A qualitative, grounded theory design guided the study in which 22 couples (44 individual partners) from the USA, who were actively considering PGD, participated. Couples were recruited from June 2009 to May 2010 from the Internet and from a large PGD center and a patient newsletter. In-depth semi-structured interviews were completed with each individual partner within the couple dyad, separate from their respective partner. We discovered that couples move through four phases (Identify, Contemplate, Resolve, Engage) of a complex, dynamic, and iterative decision-making process where multiple, sequential decisions are made. In the Identify phase, couples acknowledge the meaning of their at-risk status. Parenthood and reproductive options are explored in the Contemplate phase, where 41% of couples remained for up to 36 months before moving into the

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Resolve phase. In Resolve, one of three decisions about PGD use is reached, including: Accepting, Declining, or Oscillating. Actualizing decisions occur in the Engage phase. Awareness of the decision-making process among genetically at-risk couples provides foundational work for understanding critical processes and aids in identifying important gaps for intervention and future research.

Keywords

USA; Assisted Reproductive Technology; Decision Making; Decision Theory; Family Planning; In Vitro Fertilization; Prenatal Diagnosis

Introduction

The post-human genome sequencing era has led to a burgeoning number of modern couples who are aware of their procreative ability to transmit known genetic disorders to their future child(ren).The ever-increasing number of national and private genomic initiatives, such as general population preconception testing for cystic fibrosis (American College of Obstetricians and Gynecologists and the American College of Medical Genetics, 2001; Grody et al., 2001), and direct-to-consumer genetic testing initiatives (Ferreira-Gonzalez et al., 2008; Helgason & Stefánsson, 2010; McPherson, 2006), such as web-based *Counsyl* that will test procreative couples for more than 100 genetic disorders for about $350 U.S. dollars (Counsyl Inc., n.d.; Totty, September 27, 2010.), accelerate the number of procreative couples who are aware of their increased risk to transmit inherited disorders that were unseen in prior generations.

Preimplantation genetic diagnosis (PGD) is a cutting-edge, genomically based reproductive testing option that has been available to a growing number of couples for the past two decades. To be expected, there has been a recent upswing in the use of PGD worldwide, particularly among couples seeking to prevent the transmission of known genetic disorders to their future child(ren) (Gutiérrez-Mateo et al., 2009; Harper et al., 2010; Rechitsky et al., 2009; Verlinsky et al., 2004). This increase in use has, in part, resulted in the formation of the first nationwide datasets monitoring PGD use by government and professional groups in the United States (Centers for Disease Control and Prevention, 2008; Ginsburg et al., 2010). Recently published data from the United States indicate about 5,850 PGD procedures were completed nationally in 2009, (Centers for Disease Control and Prevention, 2011) with several private PGD centers reporting that cystic fibrosis, hemoglobin disorders, and muscular dystrophies are among the most common genetic disorders for which PGD is used (Rechitsky et al., 2009; Simpson, 2010).

While emerging datasets provide valuable threshold information about population-based trends, the science examining how couples become aware of and decide whether to use novel genomic biotechnology in the United States, such as PGD, lags far behind these and basic genomic advances, leaving couples with little or no decision-making support. Moreover, society, policy makers, and clinicians are left without understanding the experiences and nuances involved when modern couples are faced with the decision to use PGD (Hershberger & Pierce, 2010; Karatas et al., 2010b; Klitzman et al., 2008).

In the few studies that have examined aspects of decision making surrounding PGD use, investigators worldwide have primarily targeted female partners (Farra et al., 2008; Hui et al., 2002; Karatas et al., 2010a; Miedzybrodzka et al., 1993; Pergament, 1991; Quinn et al., 2009) or have used hypothetical or prospective scenarios (Chamayou et al., 1998; Hui et al., 2002; Kalfoglou et al., 2005; Krones et al., 2005). Other scholars have provided

recommendations or opinions as to how genetically at-risk couples should act toward PGD (Cameron & Williamson, 2003; de Wert, 2005; El-Toukhy et al., 2008; Offit et al., 2006; Watt, 2004). While these studies and recommendations have provided insight, a tremendous gap remains in understanding how genetically at-risk couples make real-world decisions surrounding PGD use. The purpose of this article is to provide the first reported research-based theoretical model of the decision-making process of genetically at-risk couples who were considering PGD use or who had made a decision regarding PGD use within the past three months to prevent the transmission of known genetic disorders to their child(ren).

Methods

The qualitative design and study procedures were reviewed and approved for adequate protection of human subjects by the Institutional Review Board (IRB) at the University of Illinois at Chicago, which is affiliated with the Principal Investigator (PI). After receiving IRB approval, a multifaceted recruitment plan was implemented to obtain the sample of 22 couples (44 individual partners) who were aware of their genetic at-risk status to transmit known single-gene or sex-linked genetic disorders and who were deciding whether to use PGD or had made a decision about PGD use within the past three months. The three-month window from the point of actual decision making was set after careful consideration of those couples who made the decision to proceed with PGD to allow for the intense three-month physiologic, psychological, and clinical burden associated with undergoing in vitro fertilization (IVF) (Gourounti et al., 2011; Greil, 1997; Peddie et al., 2005; Rajkhowa et al., 2006), which is required in order to perform PGD.

Recruitment challenges were heightened by the sensitive nature of the research and because couple dyads (i.e., as opposed to individual participants), who are the focus of the research, further increase the level of difficulty in obtaining an adequate sample (Kenny et al., 2006). Consequently, a multifaceted recruitment plan was implemented and couples entered the study from June 2009 to May 2010. During this time, ~82% (n = 18) of couples were recruited from Internet strategies (e.g., web sites, electronic mailing lists) and ~18% (n = 4) of couples were recruited from traditional strategies (e.g., PGD center, patient newsletter) from 14 states within the United States. Details about the successful multifaceted recruitment plan are reported elsewhere (Hershberger et al., 2011)

After receiving signed informed consent documents from the couples, the PI completed a private, in-depth, semi-structured, digitally recorded phone (n = 28 participants) or email (n = 16 participants) interview with each individual partner within the couple dyad, separate from his or her respective partner. Each partner within the couple dyad was allowed to select their preferred method (i.e., phone or email) for participating in the interview. The majority of participant couples (n = 12) selected phone interviewing exclusively, while a smaller number of couples (n = 6) opted for email interviewing exclusively and the remaining couples (n = 4) were those where the partners opted for mixed interviewing methods (e.g., one partner selected email and the other partner selected phone). In addition, one participant, who opted for email interviewing, also completed a short phone interview, per participant request, to complete the final research questions.

The phone interviews lasted an average of 61 minutes with a range of 38 to 114 minutes and were limited by IRB protocol to 120 minutes to minimize participant stress. Building upon qualitative email procedures (Hunt & McHale, 2007; Meho, 2006), the email interviews were conducted through serial asynchronous procedures where the PI emailed the primary research question to the participant. After receiving the participant's initial response, a series of "investigator probe-participant response" cycles took place between the PI and the participant in order to complete the interview. The investigator probe-participant response

cycles ranged from 4 to 8 cycles with a mean of 4.94 cycles. The length of days to complete the email interviews ranged from 9 to 96 days with a mean of 28 days.

A sample of the Interview Guide, including the primary research question and probes, which were used to clarify or obtain more detailed information, is provided in Table 1. Each couple was sent a $50 honorarium in the form of a gift card to a major store (e.g., Target) after both partners completed the research interview.

Once the interview data were obtained, the digitally recorded phone interviews were transcribed verbatim and checked for accuracy, with discrepancies corrected. The data were then entered into NVivo 8 software (QSR International, Pty Ltd, Doncaster, Victoria, Australia) to assist with organization and retrieval. The analysis followed a grounded theory approach and used the constant comparative method (Charmaz, 2006; Glaser, 1978; Glaser & Strauss, 1967). Coding, the initial process in the analysis, was completed by the PI and began with line-by-line coding on the first four interviews where a code or term(s) that best described the data was applied. Incident coding of each of the remaining interviews was then completed to allow for a more in-depth analysis (Charmaz, 2006). During incident coding, the PI was able to compare each incident in the interviews with incidents in the other interviews. Codes generated were linked directly to the data. Focused coding, where the codes were collapsed into categories and sub-categories, and finally, modified axial coding, a process of reassembling the data to give coherence to the developing theory (Charmaz, 2006; Strauss & Corbin, 1990), was completed with input from the co-investigators.

To add to the overall rigor, the preliminary findings were presented for 1) audience review (Glaser & Strauss, 1967; Patton, 2002) at two research conferences in 2011 (i.e., International Society for Prenatal Diagnosis, International Prenatal Screening Group Congress & Midwest Nursing Research Society Annual Meeting) and 2) member checking (Buchbinder, 2011; Mays & Pope, 2000; Patton, 2002) by three participant couples who represented each of the three theoretical processes generated from the data (one Accepting, one Declining, one Oscillating). This process of obtaining feedback allowed further enhancement of analytic insight into the developing theory.

Findings

Couple Demographics

All 22 couples were in heterosexual marriages. The mean age was 33 years (range = 26 to 45 years) and the majority of the couples were of White race (n = 20) with one couple self-identified as Middle Eastern and one couple of Hispanic ethnicity. The majority of couples (n = 16) indicated congruence in their religious preference; however, in five couples, one partner indicated a religion and the other partner indicated “None” or “Agnostic.” In the one remaining couple, the partners indicated differing religions (i.e., Roman Catholic and Baptist). Given these data, the predominate religious preference indicated by one or both partners was Protestant or non-denominational (e.g., Baptist, Episcopalian, Lutheran, Nazarene) (n = 10 couples), Roman Catholic (n = 6 couples), no religious preference (n = 4 couples), Jewish (n = 2 couples), and Mormon (n = 1 couple).

The sample was well educated (mean = ~16 years of education), had a high income level (mean = ~$86,700 U.S. dollars per couple), and resided in 14 states within the United States. Half of the sample had conceived or were in the process of conceiving by natural conception (n = 11 couples). Other conception options used by the couples, often prior to undergoing PGD, included intrauterine insemination (n = 2 couples), IVF alone to treat infertility (n = 2 couples), and adoption (n = 1). Seven of the couples had undergone at least one PGD attempt prior to completing the research interview. Table 2 provides a succinct reproductive

history of the sample. The couples were at risk for transmitting 14 different genetic disorders that included several variations of muscular dystrophies. One couple was at risk for transmitting two genetic disorders. Because confidentiality was a major concern to many of the couples, demographic details are limited to these aggregate findings. However, to aid understanding and provide context, illustrative quotes that correspond to the gender of the partner and the couple number, which can be correlated to the couple's reproductive history (see Table 2), genetic disorder(s) and decision type (see Figure 2 in Resolve Phase) are provided in the narrative below. Pseudonyms are used, when appropriate, to enhance clarity and understanding.

Decision-Making Process

Procreative couples who are considering PGD move through four active phases of a complex, dynamic decision-making process in which they make multiple decisions over time in a sequential or serial process. Yet, the process is iterative in that couples often revert back and forth, within and between the phases, before reaching an intended and then actualized decision about PGD use. The phases that comprise the decision-making process are: Identify, Contemplate, Resolve, and Engage. Figure 1 depicts a visual theoretical model of the decision-making process that couples undergo to determine whether to use PGD.

The decision-making process was examined through four *a priori* dimensions that delineate decision making: knowledge, concerns, perspectives (i.e., preferences and values), and relationships (President's Commission for the Study of Ethical Problems in Medicine and Biomedical and Behavioral Research, 1982). While these dimensions were fundamental and permeated the decision-making process, couples also described another dimension – experience – which played a vital role in the process. Aspects of these fundamental dimensions, as described by the couples, are presented in each of the phases; however, detailed description of each of the dimensions is beyond the scope of this initial article.

Identify Phase

In the Identify phase, couples embark on the decision-making process by becoming aware of and ultimately acknowledging one or more genetic disorders for which they are at risk of transmitting. Couples actively gain knowledge and accept the meaning and impact that being at risk has on their reproductive future. They also begin to formulate perceptions about the disorder(s).

Couples described various experiences for becoming aware of their genetically at-risk status. In some couples, especially for those with an autosomal dominant inheritance pattern, the at-risk partner typically had thoughts or "suspicions" about his or her increased risk since childhood. Often, these couples discussed genetic risk prior to marriage and would undergo genetic testing to ascertain reproductive inheritance risk. For example, Female 7 said: "Johnny and I had talked about the possibility of he having Huntington's before we got married and the impact that it may have on the possibility of having our own children." Her partner, Male 7 echoed: "That is the reason I was tested so early on for HD – was to see which direction we needed [to go] to have children." For other couples, especially those with autosomal recessive inheritance patterns, knowledge of their risk became apparent after a naturally conceived child was diagnosed with a genetic disorder. Couples from at-risk ethnic groups typically sought out genetic testing prior to conceiving to determine their genetic risk. In one couple, the male partner was diagnosed with cystic fibrosis while undergoing infertility treatment and his wife was identified as a carrier shortly thereafter.

As couples gain knowledge and accept their genetic at-risk status, they begin to formulate perceptions about the genetic disorder. For example Female 4, at risk for transmitting

hypertrophic cardiomyopathy, typically considered an adult-onset genetic disorder (Maron et al., 2003), remarked: "My husband's brother had a stroke in his late 20s because of the disease, and my [28-year-old] husband has a pacemaker because of it. I just couldn't imagine passing along a disease without fighting to have a *healthy baby* to begin with [italics added]."

Couples typically reported consultation with clinicians (e.g., genetic counselors, nurses, physicians, psychologists) at this phase to discuss the genetic disorder. A wide range of information exchange and counseling took place at this encounter that may or may not have included information about inheritance patterns, available reproductive options, or in-depth discussion about PGD. Additionally, the couple or individual partners may or may not have been ready and able to absorb information at this phase, as illustrated by Female 22: "I was so relieved that was my diagnosis [i.e., Charcot-Marie-Tooth disorder] that I gotta be honest, I wasn't listening much [to what the genetic counselor said about PGD]."

Contemplate Phase

In the second phase, Contemplate, couples carefully consider whether to become first-time parents or add (a) child(ren) to their family as they seek out and obtain information about PGD. They also explore, consider, and deliberate among a wide range of reproductive options (e.g., natural conception followed by prenatal testing with or without amniocentesis, donor gametes, adoption) while moving toward a decision about PGD use.

A major focus for couples within this phase is determining whether to become parents. Couples took seriously the responsibilities of becoming parents as noted in this example by Female 22: "Do I consciously decide to have a child when I know that I need so much help taking care of it [the child]?" Although most couples reached a decision about becoming a parent and then moved toward contemplating PGD use, there were couples for whom the progression was iterative or back and forth within the phase.

In their quest to obtain information about PGD, in every couple, either one or both partners reported searching on the Internet. Female 10 described this scenario for how she found out about PGD: "Well, I, I researched it [PGD] on the Internet. And then … well, I went to families of spinal muscular atrophy, and they actually have a forum [on the Internet] … and if you go on there, they have one that's just on PGD. And people go in and ask questions and then, that kind of stuff, and I went in there and I asked questions to find out like where was what, you know, the best places to do PGD." Another participant, Female 16, reported: "… anything I found out [about PGD], I sought out on my own. And it was, yeah, through the Internet, just looking at, I think just looking at doing a simple search on cystic fibrosis, like options during pregnancy or something like that."

Although couples used the Internet to gain information about PGD, some couples acquired information about PGD through family, friends, or clinicians. Overall, couples reported little contact with clinicians during this process, such as Female 22 who said: "We actually haven't been to the doctor together yet over this issue [whether to use PGD]. Because we thought we would wait until we knew what we were gonna do." For the few couples who did discuss PGD with a clinician at this phase, mixed reactions regarding the helpfulness of the consultation were reported. Male 7 wrote: "We talked with [genetic counselor] Pam Smithe and she gave us a great overview of our options." Other couples, including Female 17, expressed situations in which the clinician lacked knowledge or interjected personal opinions about PGD: "I think it was actually the neurologist [who discussed PGD] – who doesn't really know much about it [PGD]. He knows a lot about myotonic dystrophy. But he said something that, like the embryos can be damaged during that PGD process. And he kind of acted like, you wouldn't want to do that, you know, it can damage the embryos."

Couples would construct ideas about the benefits and drawbacks of PGD and other reproductive options. In doing so, couples took another step toward making a decision about PGD by considering and weighing less desirable options while contemplating remaining options. For example, Male 18, who was deliberating the use of PGD versus the use of donor eggs said: "So now we're thinking about doing the PGD and the benefits of the PGD, of course, is that it would be our child genetically, both of our children, you know, it would be both of ours genetically. And the cost is a little bit less, than the other [donor egg option]. The risk though, is that it's [PGD] not, you know, 100%, from what I understand; it's not 100% foolproof in terms of being able to catch this little chromosome or genetic twist that caused the problems with [our son]. It's highly likely but not 100%. So there's that thing in the back of our heads; so that's kind of what we're weighing right now." Illustrating the iterative process within this phase regarding whether to become parents, Male 18 added: "There have been times in this whole process where just thinking about it has been so frustrating where I've said to myself, you know, it would be easier just to not have any kids, frankly."

For some couples, movement through the Contemplate phase was rapid and streamlined and they moved easily into the next phase. Yet for many couples (41%), the process was much more difficult and they remained in the Contemplate phase for up to 36 months after learning about PGD. When probing these couples about what makes the decision difficult, Male 18 summed up many of the couples' concerns when he said: "I don't think the problem is lack of information. I think that the problem is just, we have all the information and we don't know what to do … It's such a monumental thing to bring a child into the world, and you'd like to have, you just wish it could be easy like it is for seemingly everybody else. But with us, there's no, there's like no perfect decision. So, you know, it's not easy to wrestle with all these different factors that we're trying to, trying to keep balanced."

Resolve Phase

When couples establish a clear intent about PGD use, it signals another step in the process and movement into the third phase, Resolve. In the Resolve phase, couples reach an intended decision about PGD use and they emerge into three theoretical processes or decision-type categories: Accepting, Declining, and Oscillating. Couples who accept or decline PGD use represent dichotomous decision-type categories where they voice an intention to use PGD (i.e., Accepting) or not use PGD (i.e., Declining). Couples who oscillate about their intention to use PGD represent a novel decision-type category where they are neither decisively for PGD use nor against it. Figure 2 depicts a visual spectrum of the couples' decision types surrounding PGD use along with the genetic disorders for which they were at risk.

The couples whose decision type was Oscillating described boundaries and experiences that included attempting natural or assisted conception first, then if conception was unsuccessful, accepting PGD use; intending to use PGD, but delaying PGD in the immediate future because of financial concerns; undergoing PGD first but if PGD is unsuccessful, initiating natural conception or adoption; and formulating a complex strategy that foregoes PGD for the first child's conception with future PGD use determined by the genotype of the resulting child or other future children. Female 21 explains her experience with the last option as: "We get pregnant the first time without doing PGD. If it is a girl, from here on out, if we have any more children, we will do PGD to make sure the rest are boys, because this will limit the number of carriers of the gene. If we have a boy, we can reassess."

Although couples reach an intended decision about PGD use at this phase, their decision is dynamic and may change as they move iteratively in either direction through the phases. Illustrating this dynamic process is Female 18 who wrote: "[My husband] and I have

decided to revisit the idea of doing PGD … We both realized we weren't 100% with donor eggs while discussing your study … even though we were all set to start with donor eggs after deciding to do so a couple of months ago."

Engage Phase

The Engage phase is marked by couples initiating behaviors that solidify and carry out their decision regarding PGD use. For example, couples Accepting PGD describe behaviors such as scheduling an appointment with a physician specializing in PGD or undergoing hormonal stimulation in preparation for PGD. Couples Declining PGD initiated other behaviors such as engaging in planned, natural conception for procreation. Couples with the decision type Oscillating reported a wide variety of behaviors that may include two or more simultaneous procreative activities as describe by Female 9: "We just felt the need to go ahead and complete the [adoption] approval process and have that kind of there … So yeah, we're definitely pursuing both [PGD and adoption], I think for now."

Another process occurring in the Engage phase is couples coping with the implications of their PGD decision. This often includes a reevaluation of their PGD decision as noted in this example by Male 19: "Yeah, I mean it's, it's one of those things that if yeah, if everything works out perfect, then it works out and you say that was a great idea, saved 20 grand. Yet any little problem and do you kick yourself for the rest of your life? I don't know. I hope not."

Discussion

To our knowledge, this is the first study delineating the decision-making process surrounding PGD use from a sample composed exclusively of couples who were actively engaged in the decision process and who represent a wide spectrum of decision types regarding PGD use. Thus, the findings have numerous implications. Foremost, this real-world model contributes to our understanding of decision-making process models. For example, decision-making process models typically describe one-time, one-decision processes (Alagoz et al., 2010; Schwartz & Bergus, 2008). However, couples in this study clearly expressed dynamic processes in which the decision surrounding PGD use unfolded over time in a series of choices. Yet, at the same time, the process was often iterative as couples traversed back and forth, between, and within the four phases of the model.

Another descriptive model that posits similar processes and concepts to our model is The Transtheoretical Model (TTM) (DiClemente & Prochaska, 1982; Prochaska et al., 2008). In the TTM, individuals move iteratively, over time through a process that includes multiple phases in order to achieve behavioral change such as smoking cessation. Our model is also similar to TTM as both recognize a key phase, Contemplation, that is followed by an intermediate phase (i.e., Resolve or Preparation) before onset of an action phase -- either Engage (our model) or Action (TTM model). These similar concepts and processes identified in both models may be because the TTM, although examining behavior change, also encompassed aspects of the decision-making process that individuals move through in order to establish a "decision" about implementing changes in their behavior.

Within the Identify and Contemplate phases of our model, we were surprised how little couples reported consultations with clinicians and in particular, physicians, about specific information regarding PGD, despite the findings that many couples (41%) voiced difficulty in reaching a decision about PGD use even after years of contemplation. This may explain why couples accessed the Internet for information about PGD. While it is possible that many couples used the Internet because of familiarity, as the majority of the sample was recruited from the Internet, it is also evident that some couples experienced difficulty processing

information from clinicians or perceived that the information presented during consultations was biased. Other investigators have suggested that Internet use is increased among those who view a treatment decision as significantly risky or uncertain, or in those who subsequently decline treatment (Couper et al., 2010). Our findings support this notion because during recruitment we found it difficult to find couples who were declining PGD use when our initial eligibility criterion required that a physician had consulted with one or both partners about using PGD. When we removed this criterion and replaced it with the criterion that couples "are aware and knowledgeable about PGD," we rapidly found declining couples. This finding is key to understanding the decision-making process and for other investigators who plan to recruit genetically at-risk individuals or couples that are making reproductive decisions. Although there is an increasing number of couples who are aware of their genetic risk, our findings indicate that while couples often interface with multiple clinicians such as genetic counselors, nurses, and physicians they often make difficult reproductive decisions outside of the clinical context.

Another important finding is the identification of the Oscillating decision type, in the Resolve phase, demonstrating the complexity of the decision-making process and providing insight into how couples creatively navigate an increasing array of genomically based reproductive testing options that are available to them. It also indicates that the decision surrounding PGD use is, for many couples, clearly, not simply deciding whether to undergo PGD, but deciding how PGD use fits within an array of prenatal options such as deciding to sequentially pursue PGD and adoption. This finding is significant for clinicians and also policy makers as it suggests that the decision spectrum between Accepting and Declining is blurred for many modern couples. This key finding supports research by Kalfoglou and colleagues (2005), who examined the beliefs and values of 181 demographically diverse individuals regarding the use of new reproductive genetic technologies including PGD, and found that opinions about PGD use did not always align neatly into classic dichotomies, such as "pro-life" and "pro-choice" arguments. The nine couples who comprise the Oscillating decision-type have constructed a novel decision choice that is beyond a classical dichotomous perspective and reflects a more progressive approach.

What is striking and universal about the couples in this study is the genuine care and concern they expressed for their future child(ren) and their understanding of the profound significance of their decision – regardless of their decision type. This may explain the onset of coping strategies reported in the Engage phase to mitigate decision regret. However, because we interviewed couples when they were immersed in the decision process and found the process dynamic, we were unable to ascertain how couples' decisions about PGD may have been actualized – or changed – beyond three months post decision. We are aware that longitudinal study of these couples could be very useful in providing this information and, in preparation for such future study, we did query couples regarding notification of future follow-up studies during the informed consent process and 90.9% of couples (42 participants) have agreed to future contact.

Because we were examining decision making, the qualitative interview process may have impacted the participant's decision regarding whether to use PGD. To compensate, we implemented strategies throughout the design to alleviate investigator and procedural influences regarding couple's decision making. Specific to the interview process, we used non-judgmental probes and avoided leading questions that may indicate investigator preference. In addition, our interview guide included the specific question: "Describe your level of satisfaction with your ability to express your true thoughts and feelings by participating in the way you did" near the completion of the interview to gain insight into any socially acceptable or investigator driven responses by the participant. Based on the participants' responses, we were able to maintain a neutral equipoise; however, the findings

should be considered in view of this possibility. In addition, the process of answering the interview questions may have contributed to greater reflection among the participants, which could have influenced the decision that they ultimately made.

Consistent with evolving qualitative methods (Charmaz, 2006; Hall & Callery, 2001; Lincoln, 1995), we acknowledge that the findings are limited by our own experiences, knowledge, and a collective belief that decision making about PGD use should ultimately rest with the informed couple. Nevertheless, we have presented important findings and have generated multiple nuanced questions for further research, which is also consistent with qualitative research. Foremost, we are curious to know more about underlying decision-making processes within the initial model, such as: When and what strategies do couples engage in to manage conflicting sequential decisions? What are the relationship concerns of couples? How does the use of the Internet for information gathering assist and impede couples in their decision making? Why do some couples struggle to reach a decision about PGD use while other couples describe less difficulty? Where in the process are interventions most needed and what type of intervention (e.g., educational, counseling, decision aid) would be most helpful? Furthermore, because many couples did not engage in formal consultation, how can these couples best be reached for decision support?

The discovery of three decision-types (i.e., Accepting, Declining, Oscillating) lends itself to further inquiry. In this paper we delineated the decision-making process among genetically at-risk couples overall; however, we noted glimpses in the data where couples voiced underlying influences on the decision, such as their perception about the severity of the genetic disorder, PGD success rate, or cost. In the United States, there is limited financial reimbursement for PGD (Henne & Bundorf, 2008; Tur-Kaspa et al., 2010) and questions remain about the extent to which the decision-making process was impacted by cost, especially among the Declining and Oscillating decision types. It is also possible that the amount of time expended by some couples in the Oscillating group to make the decision about PGD was related to their ability to obtain the necessary funds. Lingering questions remain surrounding how social and personal factors such as cost, greater social acceptance, past reproductive history, and religious views impact the decision and ultimately how couples construct preferences and values to determine whether to use PGD. Research that further examines these underlying processes regarding how couples move into each of the decision types, in a larger and more theoretically diverse sample, is needed and would benefit couples, clinicians, and policy makers (Green & Guyer, 2011; Hershberger & Pierce, 2010; Klitzman et al., 2008).

Clinicians can apply the findings through increased understanding and recognition of the decision-making process that couples undergo surrounding PGD use by tailoring counseling, education, and support during the four phases of the model. For couples in the Identify phase, clinicians can recognize that this may be the couple’s primary interface with health care professionals regarding future reproductive decisions. Counseling at this phase should be clear, unbiased, and include information about PGD that couples can access either in print or electronically at a future point in time. Clinicians should also engage in anticipatory guidance by scheduling future reproductive counseling sessions with the couple. In the Contemplate phase, clinicians can discuss the couple’s perspective on parenthood and provide non-judgmental, accurate, and comprehensive information about all reproductive options including PGD. Referral to a clinical psychologist and/or genetic counselor specializing in advanced reproductive options or genetic inheritance patterns may also be beneficial for couples. In the Resolve phase, clinicians can inform couples about the three decision types identified in this study to help couples understand the decision approaches of others. And, beneficial to couples in the Engage phase would be appropriate referrals such

as to physicians at PGD centers or to professionals at adoption organizations who can assist couples in carrying-out their decision.

We have discussed couples' decision making surrounding PGD use in the context of the United States. The novel nature of PGD and the profound implications that it has on the lives of couples and their potential children make it a concern worldwide (Hershberger & Pierce, 2010; Karatas et al., 2010b; Klipstein, 2005; Soini et al., 2006). Research that examines the decision-making process from an international sample, where political, contextual, and societal circumstances differ, would also add to a more complete comprehension of the decision-making process. There is a burgeoning number of couples who will face decisions about whether to use PGD; research in this area is critical to aiding modern couples, clinicians, and policy makers as we navigate the continuing technological advances of the 21st century.

Acknowledgments

We gratefully acknowledge the couples who participated in the study and shared their decision-making experience with us. Katy Drazba, Sara Lake, and Ramya Padmanabhan provided valuable research assistance and Mark Mershon assisted with the Figures. Support for this research was from research grants awarded to Dr. Hershberger by the National Institutes of Health (NIH), National Institute of Child Health and Human Development and the Office of Research on Women's Health (K12 HD055892), National Institute of Nursing Research (R03 NR010351), and the University of Illinois at Chicago College of Nursing Dean's Fund. The content of this article is the authors' responsibility and does not necessarily represent the official views of the NIH.

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Research Highlights

- Examines the decision-making process of 22 couples who were actively considering the use of PGD.
- Identifies four active phases of the decision-making process that are: Identify, Contemplate, Resolve, and Engage.
- Movement through the decision-making process is iterative and includes sequential decisions.
- Couples in the Oscillating decision type formulate a decision that is neither decisively for PGD nor against it.

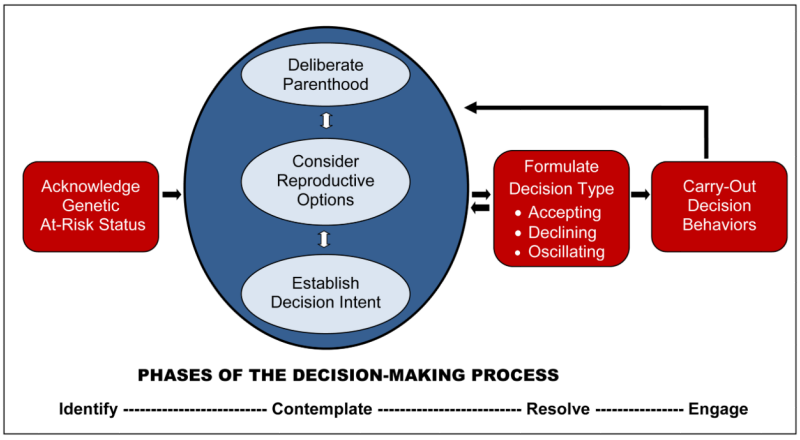

Figure 1.
Genetically At-Risk Couples' Decision-Making Process Surrounding PGD Use

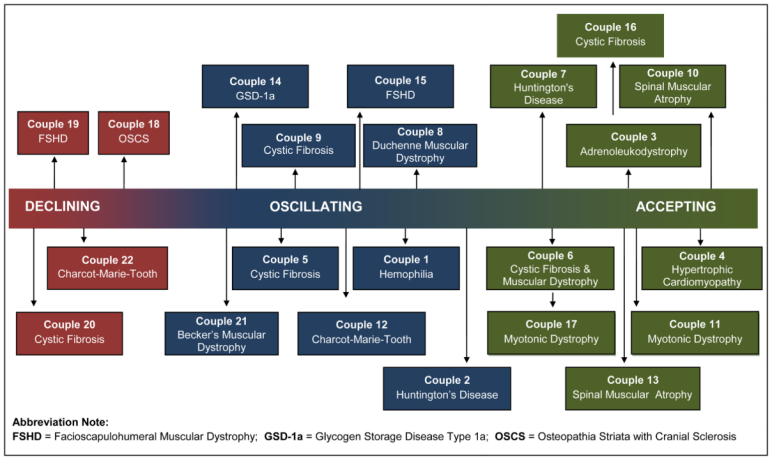

Figure 2.
Spectrum of Decision Types and Genetic Disorders by Couple

Table 1

Interview Guide: Primary Research Question and Sample of Probes

Primary Question

- Please think aloud about your decision experience surrounding PGD. Be as detailed or take as much time as you need to express your experience at this time.

Probes

- How did you learn about PGD and how was your partner involved in this learning process?
- How is your understanding about PGD similar and also different from that of your partner?
- Can you describe what it is like to be faced with this decision?
- Who or what was helpful to you in making this decision?
- How long has it taken you and your partner to reach a decision about PGD?
- What “words of wisdom” would you give to other couples who are in the midst of deciding upon the use of PGD?
- Is there any part of your experience in making the decision about PGD use with your partner that was not discussed that you feel is important and you would like to share at this time?

Table 2

Reproductive History of the Sample

Couple	Reproductive History
1	Two naturally conceived pregnancies that resulted in first trimester miscarriages.
2	Intra uterine insemination conceived son, age 2 years. Subsequently completed four additional intra uterine insemination cycles without establishing pregnancy.
3	Naturally conceived son, age 3 years, diagnosed with adrenoleukodystrophy.
4	No prior pregnancies or reproductive history.
5	Two naturally conceived pregnancies, both terminated after diagnosis of cystic fibrosis via chorionic villus sampling testing.
6	Intra uterine insemination conceived daughter, age 10 months, with cystic fibrosis.
7	Underwent two PGD cycles without achieving pregnancy.
8	No prior pregnancies or reproductive history.
9	Underwent two PGD cycles without achieving pregnancy. Have since adopted a son, age 2 years.
10	Naturally conceived son diagnosed with spinal muscular atrophy that survived for 2 months.
11	IVF conceived son, age 1 year, diagnosed with myotonic dystrophy.
12	No prior pregnancies or reproductive history.
13	Naturally conceived son died of spinal muscular atrophy just before reaching his first birthday.
14	Natural conception followed by miscarriage shortly after undergoing chorionic villus sampling testing.
15	PGD conceived daughter, age 1 year, who is without facioscapulohumeral muscular dystrophy.
16	Naturally conceived son, age 5 years, who is without cystic fibrosis.
17	IVF conceived daughter, age 1 year, diagnosed with myotonic dystrophy.
18	Naturally conceived son, stillborn, affected with osteopathia striata with cranial sclerosis.
19	No prior pregnancies or reproductive history.
20	Two naturally conceived children. One child diagnosed with cystic fibrosis at about 6 months of age and one child identified as cystic fibrosis carrier.
21	No prior pregnancies or reproductive history.
22	No prior pregnancies or reproductive history.