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DEVELOPMENT OF A MEASURE OF THE PROCESS OF INFORMED DECISION-MAKING ABOUT PRENATAL GENETIC SCREENING IN EXPECTANT WOMEN

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by

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Dedication

I dedicate this dissertation to my mother, Balwinder Kaur. She made me the woman I am today. In all the world there is no other, who can take the place of my dear mother.

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Development of a Measure of the Process of Informed Decision-Making about Prenatal Genetic Screening in Expectant Women

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Existing literature shows low levels of informed decision-making regarding prenatal genetic screening (IDM-PGS) in expectant women. In an attempt to increase autonomy and promote more ethical healthcare, this study aims to develop an instrument to measure of an expectant woman's informed decision-making regarding prenatal genetic screening. The instrument was developed based on review of the literature. Thorough psychometric testing including content validity analysis, cognitive interviewing, and readability analysis, as well as exploratory administration for criterion-related validity, construct validity, factor analysis and reliability was performed. The population of interest is women who are pregnant or may become pregnant, between the ages of 18 and 34, with no known genetic predispositions. A sample of eight women was recruited for the cognitive interviews, 433 for the exploratory administration and 111 participants in the two-week retest. Results show the instrument is a valid and reliable measure of IDM-PGS. Content validity was achieved after two rounds of expert review and feedback. Cognitive interviews showed high understanding of items in the instrument. Readability analysis resulted in a high grade level, but was justified in the highly technological nature of the information. Criterion-related validity showed a statistically significant ability for the instrument to predict participant action based on results from the IDM-PGS. Construct validity was validated by exploratory factor

analysis and known group analysis. Factor analysis resulted in factor loading in line with the developed conceptual model. Known group analysis showed individuals with medical training were significantly more likely to measure high on the IDM-PGS. Reliability was confirmed. The highly valid and reliable nature of this instrument shows its general applicability to various settings. Thus, healthcare providers can apply this instrument in clinical settings to measure the IDM-PGS in expectant women. The instrument is adaptable and should be adapted in diverse populations. In addition to future implementation and study, the results of this study indicate policy implications as well. Policy level changes and implementation of this instrument could increase IDM-PGS for all expectant couples.

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Chapter One: Introduction

Approximately one-quarter of prenatal, perinatal, and immediate post-natal infant deaths result from a genetic condition or birth defect (Human Genetic Disease, 2010). In the United States, 6.8 out of every 1000 live births result in infant death (World Bank, 2011). This factor translates to more than 27 thousand infant deaths per year in America (American Pregnancy Association, 2011; WHO, 2013a). Congenital malformations are the number one cause of infant death in the United States (CDC, 2007; Lee et al., 2001). With technological advancements in the field of genetics, modern medicine can screen an expectant woman during pregnancy to determine her unborn fetus' risk of having a genetic anomaly (de Jong et al., 2011; Lippman, 1991). Prenatal genetic screening (PGS) is a technological advancement with the potential to decrease infant morbidity and mortality by identifying harmful and, at times, life-threatening conditions for early intervention, such as in utero surgery or abortion (Gortmaker & Wise, 1997; March of Dimes, 2008; Noble, 2003; Watson et al., 2006; WHO, 2013b).

Expectant women make numerous decisions prenatally (Chiang, 2006; Goldberg, 2009; Santalahti, 1998; van den Berg et al., 2008). All medical decisions, including the decision for or against prenatal genetic screening, should be well-informed (Bekker et al., 1999; Goldberg, 2009; Leo, 1999; Moulton & King, 2010; Stacey et al., 2012). Unfortunately, ensuring informed decision-making in health care is limited and implemented inconsistently (Declercq, Sakala, Corry, & Applebaum, 2006; Goldberg, 2009; Jung, Wensing, & Grol, 1997; Levy, 1999b; Marteau, Dormandy, & Michie, 2001). In the prenatal period, expectant mothers make many decisions that impact the life and health of their fetus. An expectant mother's choices have a direct impact on health

through medical decisions, and a more indirect impact to health such as selection of the child's future pediatrician, diet or lifestyle choices. All of these decisions impact a fetus in the course of the expectant mother's pregnancy and well into the fetus' life. Research shows 51% of expectant women make truly informed decisions about PGS (Van den Berg et al., 2006). Informed decision-making about prenatal genetic screening (IDM-PGS) is defined as an expectant woman choosing a specific course of action based on herself as the decision maker, the decisional context, moral judgments, emotional responses, cognitive appraisals, framing issues and availability of resources. This definition is adapted from various definitions reviewed in the literature of IDM and PGS as independent concepts.

Ensuring decisions are made with adequate information and are consistent with an expectant woman's values and beliefs, are essential to ethical health care with positive psychological outcomes (Garcia et al., 2008; Marteau & Dormandy, 2001; Potter et al., 2008; Santalahti, et al., 2008). The concept of shared decision-making can promote informed decision-making in expectant women while maintaining a patient's autonomy (Friedberg et al., 2013; Griffith & Tengnah, 2013; Jaques et al., 2004; Lipkin, 2013; Moulton & King, 2010). An expectant mother should have the option to make decisions collectively with a healthcare professional, spouse and/or family, allowing a dispersion of the burden and greater insight into a highly technical and impactful decision (Dugas, 2012; Segal & Shahar, 2009; Vanstone et al., 2012). Though not synonymous, informed choice is often considered a preliminary step to informed decision-making (IDM) (Hibbard & Peters, 2003; Jorgensen et al., 2009; Woolf et al., 2005). Informed choice is defined as a choice based on relevant knowledge, yet is consistent with one's values and is implemented in behavior (Barnett, Ogden & Daniells, 2008; Marteau et al., 2001; Robertson, Dixon & Le Grande, 2008).

STATEMENT OF THE PROBLEM

Informed decision-making is an important aspect of PGS, as in any decision-based healthcare intervention (Chiang et al., 2006; Dugas et al., 2012; Guillemin & Gillam, 2006; Potter et al., 2008; Santalahti, 1998; Schoonen et al., 2011; van den Berg et al., 2008; Vlemmix et al., 2012). Decisions made with inadequate information often do not align with the decision-makers' values and beliefs, thus violating autonomy and leading to poorer psychological outcomes (Garcia et al., 2008; Marteau & Dormandy, 2001; Potter et al., 2008; Santalahti, et al., 2008). Further research exploring how well-informed expectant women are or are not prior to PGS decisions is needed to support the development of information-promoting interventions, including decision aids; however, this exploration cannot be done without a valid instrument to measure this phenomenon.

By increasing understanding of the prenatal genetic informed decision-making of expectant women, health care providers and researchers will be able to identify, develop, and evaluate programs and interventions to support mothers through the prenatal process.

SIGNIFICANCE

Worldwide, ten million children under the age of die five each year; four million within the first month of life and two million within the first 24 hours (Mason, 2008). Twenty-seven thousand infants die in the United States before the age of one (WHO, 2013a). The United Nations ranks the United States of America (US) 33rd in infant mortality rate, and the Central Intelligence Agency (CIA) World Factbook places the US at 50th (CIA, 2013a). These numbers reflect poorer infant mortality rates in the US than countries such as Canada, Cuba, South Korea, Israel, Portugal and Japan, all of whom had higher infant mortality rates 50 years ago. The lack of improvement in rank is not a result of lack of technological advancement, as the US leads most of the world in medical advancements. Genetic conditions and birth defects contribute to 23% of prenatal,

perinatal or immediate post-natal infant deaths (Human Genetic Disease, 2010; Nussbaum, McInnes, & Willard, 2007). Three to six percent of infants have a birth defect, and half of these have a direct genetic link (Arnold & Self, 2012; Human Genetic Disease, 2010). Prenatal genetic screening, though initially developed to promote maternal health, has rapidly transitioned to an equal focus on promoting fetal health by identifying potentially life-threatening conditions during pregnancy (Gortmaker & Wise, 1997; March of Dimes, 2008; Noble, 2003; Rappaport, 2008; Watson et al., 2006; WHO, 2013b).

Genetic assessment technology has advanced such that many congenital malformations and other genetic anomalies can be screened for and diagnosed in the prenatal period (de Jong et al., 2011; Lippman, 1991). The first step towards prenatal genetic diagnosis is prenatal genetic screening, such as maternal serum screens: a relatively non-invasive and cost-effective blood draw. PGS determines the risk for the genetic anomalies: Down syndrome, neural tube defects, and/or trisomy 18. The results of the prenatal screen are sensitive to gestational age and maternal age and thus are independent for each pregnancy. If the results of the genetic screens are positive and the expectant woman wishes to have more information, she must agree to more invasive, risky, and expensive testing for a diagnostic, or conclusive, result.

The American College of Obstetrics and Gynecologists (2007) recommends that all women be provided with reproductive health screenings, including PGS. Subsequently, the impact of PGS is widespread and healthcare providers are charged with ensuring that decision-making is well-informed. Having a valid measure of informed decision-making to ensure a decision is informed will facilitate a healthcare provider's role in prenatal genetic screening.

METHODOLOGICAL FRAMEWORK

The methodological framework for this study is Classical Test Theory (CTT). Classical test theory is one of the oldest psychometric methodologies, with origins as far back as the early 20th century (Kline, 2005; de Klerk, 2008). CTT allows researchers to account for error in measurement, both systematic and unsystematic influences (de Klerk, 2008).

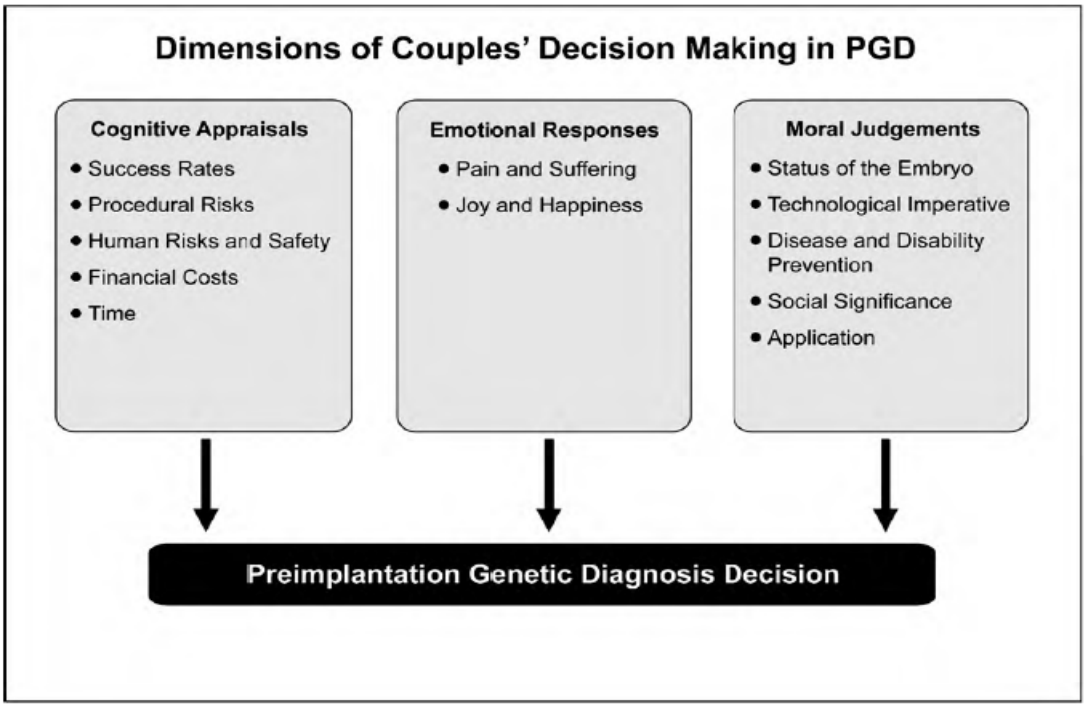
Though many forms of CTT exist, the basic premise of CTT revolves around a basic formula. This formula is a measurement of an observed score as calculated from the actual true score plus the measurement error. Calculating a true score would be the mean score obtained if the test was implemented an infinite number of times (Kline, 2005). Provided the impossibility of calculating the mean of an infinite measure, CTT dictates the addition of error as a means to account for the difference in observed scores and true scores (Kline, 2005; de Klerk, 2008). This error can be assessed in a number of ways. In this research, the test-retest method provides feedback regarding variance in multiple administrations of this instrument. Correlation of results of multiple administrations should reduce measurement error. A large sample size also reduces measurement error, as larger sample sizes lead to increasingly reliable findings (Kline, 2005).

CONTEXTUAL FRAMEWORK

Hershberger and Pierce (2010) developed a framework for decision-making in pre-implantation genetic diagnosis called *Dimensions of couples' decision-making in pre-implantation genetic diagnosis*. Their framework provides a context for this study and is shown in Figure 1 below. Despite Hershberger and Pierce's framework being based in the pre-implantation period (before the prenatal period), many of the key concepts they outline in the model regarding IDM in pre-implantation diagnosis play an important role in PGS. Major constructs of Hershberger and Pierce's model include cognitive appraisal,

emotional responses and moral judgments. In this framework, cognitive appraisal represents a couples' interpretation of success rates, procedures and short- and long-term health risks. Hershberger and Pierce further show how emotional response to pain, suffering, joy and happiness can have a great impact on decision-making. Finally, the third concept of moral judgment explores how a couple's evaluation of the embryo status, technological imperative, disability and disease prevention, social significance and application from a moral perspective can significantly impact the decision they make. These three constructs are shown collectively to lead to a pre-implantation diagnosis decision. The key concepts within these three constructs are important to the decision-making process as empirically shown in Hershberger and Pierce's research for pre-implantation genetic diagnosis; however, they can provide context to decision-making about PGS as well. Thus, these key concepts and constructs contextually guided the choice of items to include in this instrument development. The Hershberger and Pierce figure indicates that the three constructs (cognitive appraisals, emotional responses, and moral judgment) lead directly to a decision. In this study, the IDM-PGS represents an intermediate step not illustrated in the model – the process by which an informed decision is made.

Figure 1. Dimensions of Couples' Decision-making in Pre-implantation Genetic Diagnosis



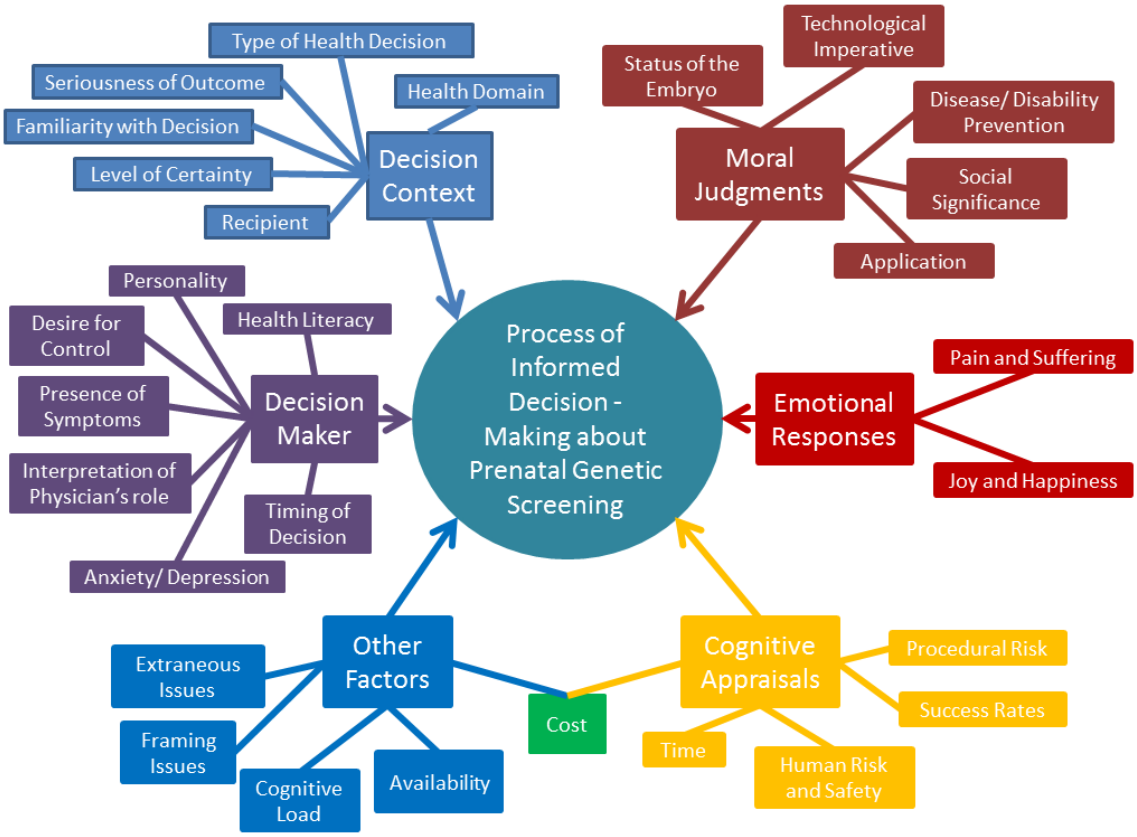
Note: Figure 1 reprinted from *Patient Education & Counseling*, 81 /1, Hershberger, P. E., & Pierce, P. F., "Conceptualizing couples' decision making in PGD: emerging cognitive, emotional, and moral dimensions", Pages 53-62, Copyright (2010), with permission from Elsevier (see Appendix C).

An annotated bibliography and systematic review published in 1999 by Bekker et al. defined decision-making as a complex concept. This study found multiple factors including decision context, the decision maker and a variety of other influences impacted informed decision making. Decision context variables were outlined to include the type of health decision, seriousness of the outcome, familiarity with the decision, level of certainty of the decisional outcome, the health domain in which the decision best fits and the recipient of the outcome of the decision (Bekker et al., 1999; Charise et al., 2011; Weiner, et al., 2010; Weiner, et al., 2013). The decision maker as an individual was shown to be influenced by their individual desire to be involved in a decision as based on

their personality, desire for control, presence of symptoms of illness, anxiety or depression, when the decision is being made, their interpretation of the physician's role, and their health literacy (Bekker et al., 1999). The various other factors include framing issues, cognitive load, extraneous issues, cost and availability of the health service or intervention as key aspects of informed decision-making.

For the purposes of this study, the Hershberger and Pierce model has been adapted in conjunction with the concepts outlined in Bekker et al.'s (1999) systematic review of informed decision making to a model, see Figure 2 below, which links the hybrid concepts to the informed decision-making about prenatal genetic screening process.

Figure 2. Adapted Model of the Process of Informed Decision-Making about Prenatal Genetic Screening



PURPOSE, AIMS AND RESEARCH QUESTIONS

The purpose of this study is to develop and test an instrument to measure informed decision-making in prenatal genetic screening for use with expectant women. The specific aims of this study are to answer the following research questions:

1. How valid is the proposed measure (informed decision-making about prenatal genetic screening) for measuring informed decision-making about prenatal genetic screening?
    1. What is the content validity index for the proposed measure of IDM-PGS?

2. What is the criterion-related validity for the proposed measure of IDM-PGS?
3. What is the construct validity for the proposed measure of IDM-PGS?

2. How reliable is the proposed measure (informed decision-making about prenatal genetic screening) for measuring informed decision-making about prenatal genetic screening?
    1. What is the Cronbach's alpha for the proposed measure of IDM-PGS?
    2. What is the two-week test-retest reliability for the proposed measure of IDM-PGS
    3. What is the item-by-item readability for the proposed measure of IDM-PGS?

Study design for the development of the IDM-PGS instrument includes psychometric evaluation and an exploratory study. Development of the IDM-PGS instrument followed a process common to instrument development with a progression of logical steps. Initially, the construct of interest, IDM-PGS, was defined from a thorough review of the literature. Based on this definition, a review of current literature and expert feedback, an array of items were formulated. Items were reviewed and filtered using construct, criterion-related, and content validity calculations. Finally, study testing for reliability through implementation of the study was conducted (LoBiondo-Wood and Haber, 1998; Polit & Beck, 2008).

DEFINITIONS:

Informed decision-making

Informed decision-making is defined as an individual of sound and reasonable mind correctly using information regarding potential advantages and disadvantages of

every potential course of action relating to the decision at hand to make that decision in accordance with their own beliefs and values (Bekker et al., 1999).

Prenatal genetic screening

Prenatal genetic screening is defined as a maternal serum screen for genetic disease in the prenatal period. This screen includes both triple and quad screens, measuring alpha-fetoprotein (AFP), human chorionic gonadotropin (hCG), unconjugated estriol (uE3) and inhibin A to determine risk of fetal aneuploidy, such as Down syndrome, or neural tube defects such as, spina bifida (Dugoff, et al., 2005; Mikail, 2008; Waletzky & Gupta, 2011).

Expectant women

Expectant women are defined as women who are pregnant at the time a decision is made about prenatal genetic screening.

Reliability

A measure is considered reliable if the results it calculates are consistent and dependable over multiple measurements (Polit & Beck, 2008; Powers & Knapp, 2006).

Validity

A measure of quality defined as an instrument that is accurately able to measure the construct it claims to measure (Polit & Beck, 2008; Powers & Knapp, 2006).

Categorical Variable

A variable that is intrinsically non-numerical and discrete in value, either nominal or ordinal (Gunst & Manson, 1980; Polit & Beck, 2008).

Continuous Variable

A variable that has an infinite range of potential values, either interval or ratio (Polit & Beck, 2008).

Latent Variable

The underlying construct an instrument is attempting to measure, often abstract and directly immeasurable (Polit & Beck, 2008).

Manifest Variable

The variable that is observable, measurable and representative of the latent variable, or the construct of interest (Polit & Beck, 2008).

ASSUMPTIONS:

The assumptions of this study are acknowledged as:

1. Participants answer the questionnaire and demographic information honestly.
2. Participants have access to and are literate about computers for an online questionnaire.
3. The participant who states they are responding to the online questionnaire is in fact the individual responding.
4. The participant who responds to online questionnaire does so in a private setting without influence of others.
5. All the data are valuable.

LIMITATIONS:

The limitations of this study are acknowledged as:

1. A convenience sample from the population of interest was recruited; thus, results cannot be generalized to all women.
2. All data are self-reported and are subject to participant bias.
3. Internet-based research requires the participant to have access to the internet.

CHAPTER SUMMARY

This chapter states the statement of the problem, significance, methodological framework, contextual framework, purpose, aims and research questions, definitions, assumptions and limitations of this study. Chapter Two provides a literature review on the variables of interest.

Chapter Two: Literature Review

In this chapter, extant literature about prenatal genetic screening technology, general informed decision-making, shared decision-making, and informed decision-making about prenatal genetic screening is reviewed and critiqued.

MODERN PRENATAL GENETIC SCREENING TECHNOLOGY

From the time Mendel manipulated pea plants to Watson and Crick's discovery of the double helix to the more recent completion of the Human Genome Project (HGP), the science of genetics has grown and continues to grow exponentially. The HGP was an international research collaboration through which all human genes were mapped out to outline their structure, organization and function. Through the work of the HGP, the enormity of the human genome was understood and more readily appreciated. The more than 20,000 genes discovered and mapped opened the door to a complex and, to date, largely misunderstood system in the human body. Over more than a decade of work, researchers have discovered that a vast majority of the work is yet left to be completed. Though mapping the human genome led to uncovering tremendous amounts of information, it also led to thousands of questions that now have a vehicle by which they can be answered (NHGRI, 2012).

The foundation laid by these early researchers has led to advancements in genetic science that allow the use of this technology in the assessments and diagnostics of modern medicine. Genetic assessment technology has advanced such that many congenital malformations and other genetic anomalies can be screened for and diagnosed prenatally. Routine prenatal care promotes healthy fetal development while monitoring for potential complications (British Medical Journal, 2013; Health Resources and Services Administration, 2013; March of Dimes, 2008).

PRENATAL GENETIC SCREENING

The preliminary step towards a prenatal genetic diagnosis is prenatal genetic screening (PGS). PGS is a minimally invasive and cost-effective blood test drawn from a pregnant woman. The risks associated with this medical procedure are those associated with any venous puncture or needle stick. These include pain, infiltration, phlebitis, thrombosis, hematoma, venous spasm, vessel collapse, cellulitis, infection, hypersensitivity, sepsis, emboli, shock and death (Josephson & Keegan, 1999). Venous access is a common health intervention and though risks do exist, the risk of serious complication is rare in venipuncture (Cheever, 2007).

The two types of PGS are maternal serum screening and quadruple screening. The maternal serum screen measures maternal levels of the biomarkers alpha-fetoprotein (AFP), human chorionic gonadotropin, and unconjugated estriol. These biomarkers are measured in relation to a normal pregnancy to estimate the fetus' risk of having Down syndrome (trisomy 21), neural tube defects (such as spina bifida, anencephaly, Smith Lemli Optiz syndrome, and abdominal wall defects), and Edwards syndrome (trisomy 18) (Lashley, 2005; Mikail, 2008; Waletzky & Gupta, 2011). Trisomy refers to a genetic condition in which there are three copies of a particular chromosome rather than the normal two copies (Cummings, 2011).

Levels of maternal serum AFP that are higher than those of a normal pregnancy are linked with a higher risk of open neural tube defect. When levels of AFP are decreased, relative to decreased levels of human chorionic gonadotropin and unconjugated estriol, the fetus is at a greater risk for having Trisomy 18. Finally, if levels of AFP and unconjugated estriol are decreased while levels of the Human Chorionic Gonadotropin and Inhibin-A are increased, the fetus is deemed to be at a greater risk for Down Syndrome (Arnold & Self, 2011; Waletzky & Gupta, 2011).

The maternal serum screen remains the most common screening tool due to its low cost and convenience (Arnold & Self, 2011; Ciarleglio, Bennett, Williamson, Mandelll & Marks, 2003; De Crespigny & Chervenak, 2006; Waletzky & Gupta, 2011). The quadruple screen adds the hormone inhibin-A, a biomarker that helps to increase the accuracy of the screen results (Mikail, 2008; Waletzky & Gupta, 2011). However, despite this additional biomarker, neither the maternal serum screen nor the quadruple genetic screens detect 100% of the genetic diseases they are designed to detect. This inexactitude refers to the predictive value of a genetic test, which is the proportion of positive screens that are truly positive to all positive screens (Abramsky & Chapple, 1994). This unequal proportion defines the concept of a false positive, an unaffected pregnancy that measures at or above the cutoff level for the marker as a positive screen would measure (Haddow et al., 1998). Thus, a false positive result shows the screening results to reflect an affected fetus when, in fact, the fetus is not affected. Before the introduction of the quadruple screen, maternal serum screenings detected about 65% of fetuses with Down syndrome. The addition of the fourth biomarker has allowed for a substantial improvement, detecting 80% of Down syndrome fetuses. These screenings also allow for detection of 90% of open neural tube defects and between 60% to70% of fetuses with trisomy 18 (Slack et al., 2006). However, there is a 5% risk of false positive results (Arnold & Self, 2011; Slack et al., 2006).

The results of PGS can be impacted by gestational age and maternal age. Expected blood levels of the biomarkers measured in these serum screens fluctuate over the course of a pregnancy and are ideally evaluated between weeks 15 and 18 of gestation. Thus, precise gestational dating is vital to ensure accurate interpretation of results (American Pregnancy Association, 2006). If the results of the genetic screens are positive for genetic risk and the expectant woman wishes to have more information, she

must agree to more invasive and expensive testing for a diagnostic result. A positive screen is not diagnostic of a genetic condition, but rather a proportion relative to a 'predetermined cut-off' of values that would be healthy in a normal pregnancy (Cartier et al., 2012). Results of screens outside the range of the predetermined cut-off are deemed a positive risk and may potentially be affected by the condition of interest, if the results of the screen are within the range of the predetermined cut-off, the fetus is most likely unaffected by the condition of interest. Of note, neither the positive nor negative results are conclusive as the risk of false positives and false negatives leaves room for error and deception of the true results. Examples of these pre-determined cut-offs in the prenatal screening period can be found in Figure 3, reprinted with permission.

Figure 3. Current Genetic Screening Options and Performance Sensitivity

Current available screening options and their screening performance*

Screening option	Markers	Trimester	Term risk cut-off	DR, %	FPR, %	OAPR
Options that meet the minimum standard						
FTS[1,2]	NT, free β-hCG, PAPP-A, MA	First	1 in 325	83	5.0	1:27
Quad screening[3]	AFP, uE3, free β-hCG, inhibin A, MA	Second	1 in 385	77	5.2	1:50
IPS[1,2]	NT, PAPP-A, AFP, uE3, free β-hCG/ total hCG, inhibin A, MA	First and second	1 in 200	87	1.9	1:10
IPS without inhibin A[1]	NT, PAPP-A, AFP, uE3, total hCG, MA	First and second	1 in 200	88	3.0	1:20
Serum IPS[1,2]	PAPP-A, AFP, uE3, free β-hCG/ total hCG, inhibin A	First and second	1 in 200	85	4.4	1:26
Options that do not meet the minimum standard						
Maternal age[4]	MA	First and second	1 in 385	44	16	1:218
Triple screening[4]	AFP, uE3, total hCG, MA	Second	1 in 385	71	7.2	1:59

*Some centres in Canada may offer variation on IPS (sequential screening or contingent screening) with cut-offs set that achieve at least the minimum standard.

DR: detection rate; FPR: false positive rate; OAPR: odds of an affected pregnancy result; FTS: first trimester combined screening; NT: nuchal translucency; hCG: human chorionic gonadotropin; PAPP-A: pregnancy associated plasma protein A; MA: maternal age; IPS: integrated prenatal screening; AFP: alpha fetaprotein; uE3: unconjugated estriol.

[1] Wald NJ, Rodeck C, Hackshaw AK, Walters J, Chitty L, Mackinson AM. First and second trimester antenatal screening for Down's syndrome: the results of the Serum, Urine and Ultrasound Screening Study (SURUSS). J Med Screen 2003;10:56–104.

[2] Malone FD, Canick JA, Ball RH, Nyberg DA, Comstock CH, Bukowski R, et al. First- and Second-Trimester Evaluation of Risk (FASTER) Research Consortium. First-trimester or second-trimester screening, or both, for Down's syndrome. N Engl J Med 2005;353:2001–11.

[3] Wald NJ, Kennard A, Hackshaw A, McGuire A. Antenatal screening for Down's syndrome. J Med Screen 1997;4:181–246.

[4] Summers AM, Farrell SA, Huang T, Meier C, Wyatt PR. Maternal serum screening in Ontario using the triple marker test. J Med Screen 2003;10:107–11.

Note: From "Counseling considerations for prenatal genetic screening" (p. 491) by Cartier, L., Murphy-Kaulbeck, L., Wilson, R. D., Audibert, F., Brock, J. A., Carroll, J., . . . Pastuck, M. in J Obstet Gynaecol Can (2012). Permission to reprint has been provided courtesy of the Society of Obstetricians and Gynaecologists of Canada (see Appendix D).

PRENATAL GENETIC TESTING

Prenatal genetic testing (PGT) differs from PGS in that the results of PGT are diagnostic. PGT is usually done subsequent to PGS when the screen is positive. Women who are of advanced maternal age (35 years of age or older), have a genetic risk in their family history, are medically recommended, or screen positive for genetic risk should be offered the more invasive diagnostic PGT. PGT includes amniocentesis, chorionic villi sampling, or fetal blood sampling. PGT diagnoses the presence of a genetic disorder or anomaly within the fetus at greater than 99% accuracy. The aforementioned PGTs require puncturing the amniotic sac and collecting a fluid, tissue or blood sample, respectively, from within the womb and thus pose a greater risk of harm to the woman and fetus than

the simple blood draw for PGS (Alfirevic, et al. 2003; American Pregnancy Association, 2006; Cartier & Murphy-Kaulbeck, 2012).

The state of California was a leader in the standardization of prenatal genetic screening through the California Prenatal Screening Program. This program requires clinicians to offer PGS services to all women. Women are expected to be educated about the varying options provided in the program by their prenatal health care provider and may choose to refuse all genetic assessment technology at that time. The California Department of Public Health (CDPH) has outlined in great detail for providers and patients what they deem as the best practices to reduce misinformation and miscommunication (CDPH, 2010; Mikail, 2008). Proponents of this innovative program tote its cost-effectiveness to the patient, universal compliance, and promotion of patient autonomy (Cunningham & Tompkinison, 1999; Flessel & Lorey, 2011). However, critics cite the lack of individuality leading to excess unnecessary cost and the "cold, computerized letter from the state" disclosing results (p. 100) as reasons to terminate the program (Priver, 1987 & Lasnover, 1987). Whether the system devised by the CDPH is adequate for the remainder of the nation is a task to be tackled by those national agencies addressing public health issues.

The American College of Obstetricians and Gynecologists (ACOG) recently published guidelines recommending all women, regardless of age, be offered Down syndrome screening (ACOG, 2004; 2007). This ACOG recommendation implies that prenatal genetic screening should be offered to all expectant women who receive prenatal care, but not all expectant women must be offered the more invasive, genetic testing. If an expectant woman vocalizes a lack of interest in the results of any prenatal genetic assessment, some health care providers may choose to withhold these offers in an attempt to increase the quality of care provided. Expectant women may have a number of reasons

to be opposed to receiving prenatal genetic screening or be in favor of it. Though, certainly not a requirement, an expectant woman may choose to disclose her concerns. A health care provider should not be obligated to determine why an individual feels one way or another but rather to ensure that individual's feelings and subsequent decision-making are based on a strong foundation of knowledge and education about the decision being made. Whether fear of a negative result, lack of interest, opposition to abortion, lack of financial resources or fear of being discriminated against are the reason, it is vital that health care professionals provide expectant women with adequate information to make a fully informed decision (Cartier, et al., 2012).

INFORMED DECISION-MAKING

The 1914 Supreme Court Ruling on *Schloendorff v Society of New York Hospital* marked the beginning of informed consent litigation and precedence with the statement that "every human being of adult years and sound mind has a right to determine what shall be done with his own body" (p. 129). The ethical implications of pre-war Nazi Germany that led to the development of the Nuremberg Code in 1947 highlighted the requirement of informed decision-making and consent issues on an international scale. The Nuremberg code defined the term *voluntary consent* by ensuring a decision-making individual has adequate knowledge about the subject matter, understanding of the issue and the legal capacity to make a competent and informed decision. Though the Nuremberg Code addressed issues surrounding biomedical research, more recent discussions purport the importance of informed decision-making in clinical practice (Manson & O'Neill, 2007). Despite the innumerable advantages of ensuring clinical decisions are informed, there are some limitations to this approach from a medical perspective.

Bekker et al. (1999) published a systematic review defining decision-making as a complex concept, influenced by multiple factors. Every decision is impacted by its decision context (Bekker et al., 1999). Contextual variables include the type of health decision, seriousness of the outcome, familiarity with the decision, level of certainty of the decisional outcome, the health domain in which the decision best fits and the recipient of the outcome of the decision (Bekker et al., 1999; Charise et al., 2011; Weiner, et al., 2010; Weiner, et al., 2013). The decision maker and the degree to which that individual wishes to be involved in the decision also impacts informed decision-making. An individual's desire to be involved in a decision is based on many factors, including their personality, desire for control, presence of symptoms of illness, anxiety or depression, when the decision is being made, their interpretation of the physician's role, and their health literacy. A number of other influences can impact decision-making as well. Framing issues, cognitive load and extraneous issues are key aspects of informed decision-making.

The method by which the decision-related information is presented to the decision maker can greatly impact which decision is made by changing the decision makers' understanding about making an informed decision. Slight contextual changes, either positive or negative, can impact a decision-maker's view of the issues that he or she must consider in making the decision (Bekker et al., 1999; Charise et al., 2011; Tversky & Kahneman, 1981; Weiner, et al., 2010; Weiner, et al., 2013). Excess cognitive load results from too much information being given to an individual to interpret in the amount of time provided (Vogel-Walcutt, et al., 2011; Whitney et al., 2008). Cognitive load refers to the amount of information an individual is cognitively processing. Increasing the amount of information to process with no increased cognitive ability leads to increased cognitive load. Excess cognitive load has been shown to have harmful effects on the

ability of an individual to make informed decisions (Croskerry, 2002; Vogel-Walcutt, et al., 2011; Whitney et al., 2008). Exceeding saturation for an individual's ability to understand and interpret information leads to inappropriate decision-making based on misunderstandings and inadequate interpretations. Frequently, individuals fall back on heuristics to deal with this excess cognitive load, which though beneficial to help ease cognitive load, usually leads to a suboptimal understanding of the issue and thus a decision that is not well-informed (Bekker et al., 1999; Whitney et al., 2008).

The cognitive load involved in medical decision-making can be reduced while maintaining an individual's autonomy by keeping the decision well-informed through the use of decisional aids. The Agency for Healthcare Research and Quality (AHRQ) define decisional aids as tools to help patients prioritize their own values and beliefs when discussing treatment options with their healthcare provider. A decisional aid promotes informed choice based on an individual's values and beliefs by exercises that clarify the values impacted by varying choices. Decision aids have been widely used in the perinatal time period with significantly improved decision-making outcomes (Bekker et al., 2004; Dugas, 2012; Frost et al., 2009; Glazier et al., 1997; Hunter et al., 2005; Kuppermann et al., 2009; Montgomery et al., 2007; Nassar et al., 2007; Shorten et al., 2005; Stewart et al., 2003). Kuppermann et al. (2009) measured the effect of an interactive prenatal genetic testing decision tool on outcomes of informed decision-making and found the use of the tool led to greater informed decision-making. Communication between a healthcare provider and patient can be positively affected through the use of decision aids (Stacey et al., 2011). The improvement in communication and reduction of cognitive load facilitates informed decision-making.

Finally, extraneous issues vary within situations and must be accounted for individually. Although Bekker et al. (1999) did not specify which factors play into every

decision-making process, as they are highly unique and individual based on each situation, the authors did provide an example with factors such as cost and availability of the health service or intervention. The out-of-pocket cost associated with an optional health service or intervention is often the deciding factor for the average expectant mother. These optional healthcare decisions are weighed in a cost-benefit analysis by patients who are limited to any extent financially and thus factor into a decision-making process (Shiell, Au, Jonhston, & Tough, 2010). Examples of the availability of the health service or intervention include how readily available transportation is, the amount of time the service or intervention takes versus how much time an individual has available, or if there is a waiting period before the service or intervention becomes available. Extraneous factors are unique to each situation of medical decision-making and a healthcare provider must play a role in each situation to minimize limitations in autonomy as a result of extraneous factors. Educating health care professionals about the role these factors play in medical decision-making is fundamental in achieving informed decision-making.

ETHICAL CONSIDERATIONS

As previously stated, the highly technical nature of the science involved with modern medicine makes considering all options based on relevant knowledge difficult for many individuals. Thus, the health care professional must take responsibility and ensure the patient is thoroughly informed prior to the decision-making process. A health care provider is responsible for ensuring that an individual is agreeing to an intervention. The agreement must be based on understanding the information essential to making that decision, avoiding influences that would promote a specific decision and thus consent to an intervention, and consenting by giving permission as opposed to an automatic consent with a decision to act otherwise. These factors are key in securing informed consent is

being achieved (Faden and Beauchamp, 1986). In the seminal opinion written by Judge Spottswood W. Robinson, III in 1972 in the case *Canterbury v. Spence*, "suits charging failure by a physician to adequately disclose the risks and alternatives of proposed treatment are not innovations in American law. They date back a good half century" (p. 4). Thus, healthcare professionals have been held both ethically and legally liable for ensuring informed decision-making for many years.

Health care professionals have many ethical responsibilities to their clients. The four ethical principles defined by Beauchamp and Childress (1979) have become foundational to medical ethics. These principles are beneficence, non-maleficence, justice and, of key importance to this study, autonomy.

Beneficence

Beneficence guides healthcare professionals to promote the patient's best interest and well being (Beauchamp & Childress, 1979; Chabon, 2011; Clarke, 1993; Lo, 2000; Noble-Adams, 1996). A health care professional is charged with caring for a patient such to lead to the best possible outcome for that patient. Unfortunately, many health care treatments and procedures entail a degree of harm. Most frequently this is a physical harm, resulting in pain, injury, or trauma. However, the risk of psychological, emotional, societal or economic harm must also be factored by a health care professional. All health care decisions require a balance of non-maleficence and beneficence to tip the benefit to risk ratio in the positive direction for a patient (Gillon, 1994). With its introduction as a moral principle by Beauchamp and Childress (1979), beneficence should be approached as a health care professional's duty to a patient to promote good, and prevent and remove as much harm as possible (Chabon, 2011; Clarke, 1993; Lo, 2000; Noble-Adams, 1996).

Non-Maleficence

Many early ethicists did not distinguish the principle of non-maleficence from beneficence. Similar to, but more narrowly focused than beneficence, non-maleficence focuses on avoiding harm. Health care professionals are diligently taught to prioritize avoiding and preventing harm in their care. Florence Nightingale introduced the concept of *Primum Non Nocere*, Latin for 'above all, do no harm', to the nursing profession (Clarke, 1993; Smith, 2005). Distinction is made between intentional and unintentional harm. Often unintentional harm is unavoidable, such as in medical treatments that may cause harm to a patient in the process of promoting healing (Beauchamp & Childress, 1979; Lo, 2000). Surgery, radiation/ chemotherapy, and injections all inflict varying degrees of physical harm through pain, risk for infection, and even death. However, as earlier outlined in beneficence, often harm is necessary to do good and benevolently treat a patient. A more clearly immoral harm is intentional harm. Intentional harm is never acceptable in a health care professional's care except in the rarest circumstances. An example of this circumstance may be self-defense or protecting another from harm (Beauchamp & Childress, 1979). Despite these specific circumstances, health care professionals are charged with avoiding and preventing harm when caring for patients to the best of their ability.

Justice

The ethical principle of justice refers to distribution of resources. In health care, there are a limited number of personnel, financial and healthcare resources. The principle of distributive justice requires these scarce health care resources be equally distributed to those in need. Health care providers are charged with the responsibility of treating all patients equally, both in benefit and in burden. For example, a nurse must equally distribute her or his time to those patients whose health care require dedication of equal

time. Through the equal distribution of her/his time, the benefit of the nurse's care and the burden of the lack of availability due to caring for other patients is spread to each of the patients in a fair and just manner. This distribution equality is required despite an individual's social or economic stature (Beauchamp & Childress, 1979; Gillon, 1994).

Autonomy

Finally, the ethical principle of autonomy promotes individuals to make their own health decisions based on their own values and beliefs. Implementing an autonomous environment in health care allows patients to make health care decisions that, if plausible, comply with their moral principles. The qualifier of plausibility is necessary in accordance with the ethical principle of justice mentioned above. Though autonomy should always be highly regarded in patient care, if the health care resources are not available it is reasonable that the course of action an individual would want to follow may be outside the means available for that individual's care (Atkins, 2000; Beauchamp & Childress, 1979; Chervenak & McCullough, 2011). The ethical principle of autonomy charges a health care professional to remove his or her own biases from the care of a patient and prioritize the patient's beliefs and values. Health care professionals must be cognizant not only of removing their own bias but promoting the moral principles of the patient. Autonomy promotes the client as the primary decision maker, as opposed to the traditional paternalistic health care provider as primary or sole decision maker.

PATIENT-FOCUSED DECISION-MAKING

As medical decision-making transitions from a dogmatic physician-centered approach to a primarily patient focused decision-making process, patients are not only encouraged, but rather required, to be active participants in their health care. Often patient participation is promoted with little or no patient preparation for the complexity,

technicality and gravity of the decisions they are participating in making (Rao, 2007). Due to the lack of preparation, patients often forgo by choice or by lack of understanding key information provided by health care professionals. Similarly, patients may react fearfully to information a health care professional feels is routine from lack of understanding as well (Manson & O'Neill, 2007). Information a health care professional feels is routine due to increased medical knowledge and exposure, often patients have rarely encountered the information and the situation they are being faced with in these medical decision-making scenarios. For example, an average American family has 2.06 children (CIA, 2013b). Thus, an average American woman will only encounter the medical information pertinent to childbirth two times in her life, hardly allowing enough exposure to push past novice skills. A health care professional must never assume a patient adequately understands information required in a medical decision, regardless if they feel the information is routine.

The terms informed consent and informed decision-making are often used interchangeably. However, these terms, in fact, define two distinct but interrelated concepts. Informed consent refers to the communication of the information to a patient that leads to making a medical decision in an informed and voluntary manner (ACOG, 2009; Beauchamp, 2011; McCoy, 2008). Informed decision-making is a precursory thought process in individuals before giving informed consent, and informed consent cannot be achieved without an informed decision-making process. When a decision is made with relevant information about the advantages and disadvantages of each course of action and is considered in accordance with beliefs, it is called an informed decision (Bekker, 2003). Whether a medical intervention is received or refused requires an active decision-making process. Ideally, the decision-making process is well informed, which thus leads to informed consent. Due to the inconsistencies within and between literatures

discussing informed consent, for purposes of this paper, the focus remained on informed decision-making.

Unfortunately, true informed decision-making is frequently not ideally achievable, some even argue informed decision-making is often unachievable (Lo, 2000; Manson & O' Neill, 2007). Inadequately informed decisions can result from multiple variables in a decision-making situation, such as inability to comprehend the medical information. However, health care professionals must engage themselves fully towards promotion of autonomy and removing paternalism. Though not irrational, it is unethical for a health care professional to assume a patient cannot fully comprehend medical information well enough to make an informed medical decision. Informed consent is more than solely a legal requirement, it is an ethical requirement all health care professionals must work to achieve for every patient (Lo, 2000; Manson & O' Neill, 2007).

Informed Decision-Making about Prenatal Genetic Screening

Ensuring well-informed decisions is of particular ethical and regulatory importance when a significant degree of personal, identifiable information about an individual is provided, such as in the case of prenatal genetics. Knowledge of an individual's genetic information is very different from other kinds of knowledge about an individual (Mason & O'Neill, 2007). Genetic information can dictate susceptibility to negative health outcomes secondary to genetically inherited disease, and may lead to genetic discrimination. The two-part Genetic Information Nondiscrimination Act (GINA) of 2009 is aimed directly at protecting individuals from facing genetic discrimination from employers and insurance carriers. Despite being a positive step towards preventing genetic discrimination, even GINA has its limitations and cannot protect individuals in

every circumstance. Employers with less than 15 employees, benefits provided by the Veterans Health Administration or the Indian Health Service, and life, disability and long-term care insurance are not covered by GINA (Genetics Home Reference, 2013). Thus, genetic information should be dealt with distinctly in terms of societal and individual rights and responsibilities when compared to other health information. This difference is referred to as genetic exceptionalism, a concept stemming from the *Draft Genetic Privacy Act,* which was produced by the ethical, legal and social issues (ELSI) program of the Human Genome Project. The *Draft Genetic Privacy Act* purports genetic information, due to its propensity to lead to discrimination, requires special protections that are unique to other personal information.

Decision-making is a multifactorial process in health care, influenced by the context in which a decision is being made, the decision maker, information framing issues, cognitive load on the decision maker and extraneous factors affecting availability and obtainability (Bekker et al., 1999). These factors in their most general form translate across various health care settings, including prenatal genetic screening. The decisional context in a prenatal genetic screening decision has specific features. The perspective of the decision maker impacts this context of a decision. The seriousness of the outcome depends on the values and beliefs of the expectant woman. For instance, a result that has a devastating impact on some expectant women could potentially have a minimal impact on another. An example of this is seen when a pregnancy is found to have a high risk of Down syndrome; an expectant woman who has strong beliefs against abortion could interpret these results as destiny, accept them and carry the baby to term. On the other hand, an expectant woman who would be willing to contemplate abortion may perceive a fetus with an increased risk of Down syndrome as reason to terminate her pregnancy and thus a devastating loss. Similarly, whether or not an expectant woman plans to pursue

further testing could also dictate the seriousness of the outcome. If an expectant woman does wish to pursue the more invasive prenatal genetic testing, the outcome of the screening has the potential of having a much more serious impact on a fetus, as per the risks defined earlier, than if the expectant woman would not plan to pursue prenatal genetic testing. Context is also impacted by an expectant woman's familiarity with prenatal genetic screening decisions. Depending on how many pregnancies requiring prenatal genetic screening this expectant woman has had in the past, her level of comfort in making that decision can vary. An expectant woman who has made an informed decision about prenatal genetic screening in the past is more likely to make an informed decision in the future.

Having past experience with informed decision-making regarding prenatal genetic screening decreases the impact of extraneous factors on an individual's decision-making. For example, as earlier discussed, the effects of framing of health information would not impact an expectant woman who has made an informed decision about prenatal genetic screening in the past. However, in situations of first time decision-makers, the health care professional's framing of the risks and benefits of prenatal genetic screening can alter the decision made. Variations in tone, inflection, or other displays of personal bias can be perceived by a decision-maker and inappropriately affect the decision (Bekker et al., 1999; Charise et al., 2011; Tversky & Kahneman, 1981; Weiner, et al., 2010; Weiner, et al., 2013). Similarly, past experience with making a prenatal genetic screening decision may reduce cognitive load in a future prenatal genetic screening decision. However, as the technology continuously changes, health care professionals must continue to educate their patients, who can have any varied length of time between their pregnancies, and inform them of changes in the technology and science of prenatal genetic screening and the potential impact on a prenatal genetic screening decision.

Many extraneous issues are situation specific. One such example of a situation specific extraneous issue to consider is that of increased knowledge leading to delayed maternal emotional bonding to the fetus. Some believe that the delay in maternal bonding is a positive process and can lead to a healthier transition if a woman is going to abort or miscarry. Others believe that this delay is a negative process in that women do not protect their fetuses in a healthy way and may practice riskier health behaviors, such as consuming alcohol (Rowe et al. 2009).

Though the expectant woman's decision regarding prenatal genetic screening is hers to make, the recipients of this decisional context also include her partner, the unborn fetus and, due to the genetic component, more distant relatives with genetic ties, such as the expectant couples' parents and siblings. Regardless of the involvement of potential recipients of health information in prenatal genetic screening, a health care professional is responsible to ensure that whatever decision regarding prenatal genetic screening is made, it is informed regardless of being an individual or shared decision.

SHARED DECISION-MAKING

The decision maker, in the realm of prenatal genetic screening, can be limited solely to the expectant woman or can be expanded to include her partner and/or health care provider if shared decision-making is being considered. Shared decision-making allows clinicians, patients and influential individuals, such as family members, to have open communication about the risks of a medical decision, benefits of a medical decision, and other potential options of a medical decision. This communication not only discusses factors involved in the decision but also reveals the decisional pathway preferred by all of the deciding parties to allow for a decision to be made jointly (Dugas et al., 2012; Griffith & Tengnah, 2013; Lipkin; 2013; Moulton & King, 2010).

Though familial involvement in shared decision-making can be positive and beneficial by providing knowledge and promoting a families beliefs and values, potential negatives can arise as well. Familial roles could lead to a more dominant individual monopolizing and persuading a patient towards a decision that does not align with the patient's values and beliefs (Epstein & Gramling, 2013; Ho, 2008). Dominating roles can result from personality characteristics but also from cultural role differences in familial dynamics. Some cultures have strong matriarchal or patriarchal roles in families that are charged with making key decisions, which could lead to vetoing of the patient's decision (Goldbas, 2013; Ho, 2008). Age, gender roles and attitude towards illness vary culturally as well. A number of cultures believe in not disclosing a diagnosis to the elderly, females, the terminally ill or those patients with a low likelihood of survival (Back & Huak, 2005; Ho, 2008). Many cultures have male dominated decision-making roles, thus in the PGS scenario, the expectant woman's values and beliefs may be considered secondary to her husband's (Back & Huak, 2005; Ho, 2008). An excessively persuasive parent or spouse can remove patient autonomy during shared decision-making. Healthcare professionals involved in shared decision-making must facilitate a positive familial role while ensuring the patients values and beliefs are manifested within the decision that is made.

A healthcare provider has an active role in shared decision-making to ensure the process maintains the highest level of autonomy for the patient. Even shared decision-making requires that patients be educated on the risks, benefits and other potential intervention options for the medical decision to ensure that the decision is made in alignment with the patient's values and beliefs (Dugas et al., 2012, p. 1968). The addition of participation from the healthcare provider in a decision-making process adds the benefit of a health care provider's medical knowledge to the decision-making. A healthcare provider must be cognizant of the potential downfall to a patient's autonomy

when adding a recommendation of a course of action (Frongillo et al., 2013). In situations involving highly technical material, and thus a greater involvement from a healthcare provider, healthcare providers should be trained to filter their own beliefs and values out of a decision process involving shared decision-making to provide the patient with the most autonomy possible.

Shared decision-making when properly implemented has the potential to enhance individual autonomy while minimizing paternalism (Griffith & Tengnah, 2013). Implementing shared decision-making models universally, however, can be drastic and short sighted.

PREVIOUSLY PUBLISHED INSTRUMENTS REGARDING SIMILAR CONCEPTS

Few instruments exist in the currently available literature that measure concepts similar or identical to informed decision-making about prenatal genetic screening. Michie et al. (2002) developed a Multi-dimensional Measure of Informed Consent (MMIC). The MMIC measures pregnant women for informed choices based on assessing knowledge, attitudes and behavior regarding prenatal genetic screening (Marteau et al, 2001). This is scale most closely reflects the concept being measured in this study. Both the MMIC and the IDM-PGS explore prenatal genetic screening literacy, and an expectant woman's values and beliefs. However, the MMIC is distinct in its measure of the decided action an expectant woman chooses in prenatal genetic screening, while the measure of IDM-PGS explores the process involved in the decision making as a means of ensuring decisions made will be informed. The IDM-PGS aims to obtain information regarding the informed status of the expectant woman prior to her making a decision regarding prenatal genetic screening, a more upstream approach. Though the MMIC has been widely implemented in various settings (Potter et al., 2008; Gourounti & Sandall, 2011), the

distinction between the IDM-PGS requires the development of a new measure to assess the population prior to implementation of the prenatal genetic screen.

Table 1. Previously Published Instruments Regarding Similar Concepts

Author	Year	Title	Topic	Participants	Cronbach's Alpha (α)
Michie et al.	2002	The multi-dimensional measure of informed choice: a validation study	Measure if pregnant women have a positive attitude towards, have relevant knowledge about and undergo a screening test for Down Syndrome.	225 pregnant women	Knowledge scale = 0.68 Attitude scale = 0.78
Van den Berg et al.	2008	Understanding Pregnant Women's Decision Making Concerning Prenatal Screening	Measure: perceived risk, perceived severity, attitude toward termination of pregnancy, response efficacy, attitude towards having a prenatal screen, subjective norm, anxiety, and intention to test	1,666 pregnant women	Range from 0.59-0.88 in each scale
O'Connor et al.	1997	Decisional Conflict Scale Ottawa Civic Hospital	Measure "uncertainty in making a choice", "modifiable factors contributing to the uncertainty" and "perceived effective decision making"	Range from 28-360 in each group measured	Range from 0.78-0.89 in each group measured

Van den Berg and colleagues (2008) also explore expectant women's decision making regarding prenatal genetic screening. However, as opposed to developing a uniform overall scale for their measurements, Van den Berg et al. (2008) administered eight distinct scales measuring the concepts: perceived risk, perceived severity, attitude toward termination of pregnancy, response efficacy, attitude towards having a prenatal screen, subjective norm, anxiety, and intention to test. The constructs measured individually encompass important aspects encompassed in the IDM-PGS as well,

however, by combining each of these separate scales into a single measure implementation and consensus of results is simplified.

Various measures of informed decision-making and informed consent exist across a multitude of disciplines. Dr. Annette O'Connor developed a decisional conflict scale, often known as the Ottawa Decisional Conflict Scale, which measures uncertainty in making a decision, perceived effective decision making, and modifiable factors such as lacking information, unclear values, and inadequate social support that enhance decision maker uncertainty in making a choice (O'Connor, 1997). This 16-item General Decisional Conflict Scale provides useful insight into scale items that determine confidence in decision making and certainty of choice. This scale and others represent more general or distinctly specific measures of informed consent and informed decision-making which, while maintaining key constructs that apply to the IDM-PGS, do not provide the level of specificity needed to address the need for the development of the IDM-PGS.

CONCLUSION OF LITERATURE REVIEW

Deciding whether or not to receive PGS requires a thorough consideration of medical harm versus benefit, personal beliefs and values, familial input, and societal norms and pressures (Henneman et al., 2008). Every expectant woman brings a unique mix of biological risk factors to each of her pregnancies. Not only does the woman's genetic disease history and family history impact the risk for fetal genetic disease, but also, advancing maternal age can increase risk from one pregnancy to the next. The balance of these variables dictates the information a health care provider will give to an expectant woman and the physician's medical recommendation for or against PGS. Unfortunately, many PGS decisions are made based solely on the gist of information

health care professionals provide (Reyna, 2008). In other words, expectant women often are not thoroughly informed on all aspects of receiving PGS and are only given information deemed important by the provider.

As with infant mortality statistics, many other countries have progressed further in prenatal genetic decision-making than the United States. In the Netherlands, for example, expectant women are offered prenatal genetic information only if they indicate an interest. In other words, if they refuse further information or counseling, they will not be inundated with unnecessary information of no concern to them (Schoonen et al., 2012). The International Society of Nurses in Genetics has published a position statement regarding informed decision-making and consent, and the American Nurses Association, in conjunction with the International Society of Nurses in Genetics, has published the scope and standards of genetics and genomics nursing (ISONG/ ANA, 2007; ISONG, 2011). Both these guidelines delineate a nurse's role in genetics and genomics. However, there are no universally-accepted guidelines for discussing genetic risk information in the United States (Ethegary & Perrier, 2007). This lack of universality often leads to health care professionals asserting their own beliefs and values on medical recommendations they make to their clients. However, an individual's values and beliefs may not correlate with a physician's recommendations (Reyna, 2008; Van den Berg et al. 2005). This difference in beliefs highlights the importance of being well-informed to make autonomous decisions about PGS and contributes to the significant difference that exists between PGS intention and actual screening behavior (McDaniel, 2005).

As previously discussed, Bekker et al.'s (1999) systematic review defines decision-making as a complex concept. Decisions are impacted by various constructs including: decisional context (Bekker et al., 1999) including the type of health decision, seriousness of the outcome, familiarity with the decision, level of certainty of the

decisional outcome, the health domain of the decision and the decisional outcome recipient; the decision maker, their personality, desire for control, presence of symptoms of illness, anxiety or depression, when the decision is being made, their interpretation of the physician's role, and their health literacy; framing issues; cognitive load; and, extraneous issues. Hershberger and Pierce (2010) developed a framework for decision-making in pre-implantation genetic diagnosis that outlines major constructs including: cognitive appraisal, the interpretation of success rates, procedures and short- and long-term health risks; emotional responses to pain, suffering, joy and happiness; and, moral judgments in which evaluation of the embryo status, technological imperative, disability and disease prevention, social significance and application from a moral perspective impact decision-making. Both the models outlined by Bekker et al. (1999) and Hershberger and Pierce (2010) are distinctly useful in their respective focus areas; however, to address this unique area of study, the synthesis and adaptation of these two models provides for a comprehensive model for contextual guidance for the development, analysis and implementation of the measure of informed decision making about prenatal genetic screening.

CHAPTER SUMMARY

The development of an instrument to measure IDM-PGS can be used to conduct an exploratory administration of the instrument that allows for results to be generalized to the population. Once results are generalized, current gaps and deficiencies in IDM-PGS may be identified. These results promote the advent of interventions and development of clinical decision pathways. Clinical decision pathways assisting expectant women in making PGS decisions and improved instructional information for the health care provider will, ideally, improve an expectant woman's IDM-PGS.

Chapter Three: Methods

This chapter describes the methods used in this instrument development and the exploratory administration of the instrument. Research design, method, sample and procedure are described in detail. Data analyses occurred using the statistical analysis software SPSS (Statistical Package for the Social Sciences) –Version 21. Owing to the lack of existing measures of IDM-PGS, development of an instrument is essential to advance the science. Psychometric testing was conducted to provide evidence of adequate instrument development for further use in research and practice.

DESIGN

Study design for the development of the IDM-PGS instrument included psychometric evaluation and an exploratory study guided by classical test theory (DeMars, 2010; Reckase, 2009; Steinberg, 2001). Development of the IDM-PGS instrument followed a process common to instrument development with a progression of logical steps. Initially, the construct of interest, IDM-PGS, was defined from a thorough review of the literature. Based on this definition, a review of current literature and expert feedback on an array of items were formulated. Cognitive interviews were conducted with a sample from the population of interest. Items were reviewed and filtered using construct, criterion-related and content validity calculations. Finally, testing for reliability through implementation of the exploratory administration of the instrument was conducted (LoBiondo-Wood and Haber, 1998; Polit & Beck, 2008).

The research questions addressed in this exploratory administration of the instrument are:

1. How valid is the proposed measure (informed decision-making about prenatal genetic screening) for measuring informed decision-making about prenatal genetic screening?
    1. What is the content validity index for the proposed measure of IDM-PGS?
    2. What is the criterion-related validity for the proposed measure of IDM-PGS?
    3. What is the construct validity for the proposed measure of IDM-PGS?
2. How reliable is the proposed measure (informed decision-making about prenatal genetic screening) for measuring informed decision-making about prenatal genetic screening?
    1. What is the Cronbach's alpha for the proposed measure of IDM-PGS?
    2. What is the two-week test-retest reliability for the proposed measure of IDM-PGS?
    3. What is the item-by-item readability for the proposed measure of IDM-PGS?

IDM-PGS DEFINITION

Based on the results of the literature review, the construct of informed decision-making about prenatal genetic screening is defined as an expectant woman choosing a specific course of action based on herself as the decision maker, the decisional context,

moral judgments, emotional responses, cognitive appraisals, framing issues and availability of resources.

ITEM FORMULATION

Data for item formulation were compiled from numerous sources to construct a multidimensional instrument for IDM-PGS. The adapted contextual framework provided a primary medium for item development (Bekker et al., 1999; Hershberger & Pierce, 2010). Key concepts and major constructs from the models, such as health literacy, seriousness of the outcome, cognitive load, procedural risks, emotional responses and social significance, were used as contextual bases for items. Additionally, a 20-item list of essential information for healthcare professionals to provide to patients pursuing genetic testing (see Figure 4, reprinted with permission) from *Essentials of Clinical Genetics in Nursing Practice* (Lashley, 2007, p. 84) provided key factors worth consideration in the decision-making process and guided further item development.

Figure 4. Information for the Client Considering Genetic Testing

- The reason that testing is appropriate for this person or family.
- What is being tested for.
- What estimation of risk and for surveillance can be done without genetic testing.
- What the procedure being considered entails, including description, cost, length of time, and where it is to be done.
- What can and cannot be tested. If relevant, this should include the information that some mutations will be looked for and detected, other rare ones might not be, and that negative results refer only to whatever was being tested and not to every genetic disorder. If one is testing for cystic fibrosis, the most common mutations in the population group will be looked for, but not ever vary rare mutation will be tested for. Usually within the context of an affected family, however, it a specific mutation has already been identified in a blood relative, it will be specifically looked for when another family member is undergoing genetic testing.
- What both positive and negative results mean, including that negative results do not necessarily translate to a zero risk and that a positive test may result in fear and anxiety, whereas negative results can also have emotional and relationship impact.
- The accuracy, validity, and reliability of the test including the likelihood of false-negative or false-positive results and the suitability of this test for the information the client is seeking.
- The possibility that testing will not yield additional risk information.
- The length of time between the procedure and when the results are obtained.
- How the results will be communicated to the client.
- What will be analyzed.
- Whether the actual test result will be revealed. For example, in Huntington disease, in some cases there may be some correlation between the number of CAG repeats and the predicted age of onset but there is a gray area, so some centers do not disclose the actual number although such disclosure is generally recommended.
- What happens to the sample used for testing—who owns it, what uses are possible.
- A discussion of the possible risks of life and health insurance coverage or employment discrimination after testing results are done, although there may be benefits such as if a person is free of a certain mutation, better insurance rates or coverage might result.

Figure 4. (Continued)

- The level of confidentiality of results and what this means (who can know or find out the results).
- Risks of psychological distress and negative impact on not only the individual but also the family including stigmatization and altered self-image.
- Risk of passing on the mutation in the disorder being tested for to offspring and the meaning of the risks.
- What disclosure the client might consider for other family members and those he or she will tell (if anyone) about the test results; what obligation the health provider might feel to inform other family members.
- Provision for referral for periodic surveillance, further testing, lifestyle changes, or treatment after testing if needed.
- What these mean in the context of both positive and negative tests. As in other genetic testing, a negative test can have several meanings: that the individual is truly free of the disease, that the result is false negative due to laboratory error, or that the person possesses alternate alleles other than what could be or what was tested for.

Note: From "Essentials of clinical genetics in nursing practice" (p. 84) by Lashley, F. Permission to reprint this table has been provided courtesy of Springer Publishing Company, Inc. (see Appendix E).

Although Lashley's (2007) list is defined specifically towards pre-implantation genetic testing, similar concepts were appropriately applied to the pursuit of prenatal genetic screening information as well. Lashley expresses that the items she describes provide the basic information an individual needs before making a decision about whether or not to pursue genetic testing, and thus genetic screening. Lashley developed her list based on a review of the literature and her expert opinion as a nurse geneticist, making this list a useful source for item formulation. For example, this list provided information for the development of items addressing interpretation of results, lifestyle changes, and insurance discrimination issues.

Key concepts in the review of the literature generated additional items not addressed in Lashley's (2007) list or the adapted contextual framework. These additions included items about other treatment options, and diagnosis versus risk. The review also

validated the importance of the items developed based on the adapted contextual framework and Lashley's list.

A large conceptual map was created using the in-depth review of the literature, the items discussed in Lashley's list, the findings of Bekker et al.'s (1999) review of the literature and the Hershberger and Piece model (2010). All items were mapped and key concepts were found to contextually guide the definition of IDM-PGS outlined above and the contextual model. Items that did not conform to the constructs outlined in the definition or in the contextual model were removed.

ITEM-BY-ITEM READABILITY

Instrument readability is often considered an important assessment of validity. In the early 20th century, research on vocabulary in achievement tests found that if the vocabulary of questionnaires was too difficult, the questionnaire became a measure of reading ability as opposed to a measure of the content as initially intended (Chall, 1958). In an effort to minimize the conversion of this instrument for IDM-PGS into a health literacy assessment or a reading ability assessment, item readability was kept as low as possible. The readability for each item of the IDM-PGS was calculated using the SMOG readability calculator (Kouame, 2010) after revision based on recommendations from the expert panelists and before implementing the instrument for exploratory analyses. Readability calculators, such as the SMOG, use a complex formula based on the number of syllables in a word and sentence length to calculate an items anticipated level of understanding and correlates that number to the reading grade of an individual (McLaughlin, 1969). The University of Nottingham created an online calculator for the SMOG calculations that was used to measure the readability of the items of this instrument (The University of Nottingham, n.d.).

CONTENT VALIDITY.

Once the instrument was created, psychometric evaluation began. The first psychometric evaluation in this instrument development process measured content validity. Content validity refers to the adequacy of item sampling (De Vellis, 2012). A content validity index (CVI) reflects the appropriateness of the items in the instrument in assessing the construct of interest (Polit, Beck & Owen, 2007). Both individual item (I-CVI) and overall scale CVI (S-CVI) calculations were used to determine the relevance of each item and the overall scale to the construct of interest.

The CVIs were calculated by assembling a panel of experts by word-of-mouth recommendations from contacts of the principal researcher. The members of this panel included three doctorally-prepared nurses researching various aspects of genetics and genomics, including two focused specifically on maternal-child genetics and genomics; one masters-prepared nurse with many years of experience in maternal-child nursing and faculty experience in genetics; and two retired obstetric-gynecologist physicians. To calculate the CVIs, expert panelists were instructed to respond "for each of the items on the scale, please indicate how relevant you think it is for the construct of informed decision making in prenatal genetic screening." Additionally, a definition of IDM-PGS was provided to the expert panelists as "An expectant woman choosing a specific course of action based on her understanding of the purpose of receiving PGS, the procedure of PGS, an estimation of the risks involved with PGS, interpretation of the results of PGS and other potential options or implications of PGS." In order to ensure consistency in response from expert panelists, definitions of each response were provided. The responses were defined as 4 represented a response of "highly relevant", 3 represented "quite relevant". 2 was defined as "somewhat relevant" and response 1 was defined as "not relevant". Each participant responded to the relevance of each item in the survey.

Often, CVI collection is an iterative process requiring multiple rounds of data collection to improve relevance by addition, removal or clarification of items. Each individual item was scaled for I-CVI. Then the collective responses provided scores for calculating the S-CVI. The CVIs help the researcher determine whether or not IDM-PGS is sufficiently measured by the items developed. Expert panelists' suggestions about the instrument were considered and implemented if appropriate. Polit, Beck and Owen (2007) suggest two options for calculating CVI. The S-CVI universal approach requires all content experts to rate each item favorably, thus a minimum S-CVI of 0.70. The second approach is the S-CVI average in which I-CVI is calculated and then averaged, results of all the content experts should yield a value around 0.90. For purposes of this study, the S-CVI universal approach was taken and thus a minimum of 0.70 was set for adequate relevance of the instrument.

COGNITIVE INTERVIEWS

Prior to exploratory implementation of this instrument, cognitive interviews were done to assess if individuals understood the instrument items and to improve the design of the instrument (Knafl et al., 2007). Verbal probing occurred concurrently with item administration in these audio-recorded cognitive interviews. The interviewer conducted a think-aloud interviewing exercise to initiate the cognitive interview process. This initial exercise probe asked the interviewee to complete a minor task, in this situation count the number of windows in their house, and in the process think-aloud everything they are thinking of or picturing. Once this task was completed the interviewer would read each item to the interviewee and the interviewee would then repeated back the question in their own words as an assessment of the ability of the question to ask what it was developed to

ask. Further discussion promoting explanation was prompted by the interviewer per the interviewer's discretion (Willis, 1999).

Sample

The population of interest for the audio-recorded cognitive interviews included:

- Expectant women
- 18 – 34 years of age
- No known genetic predispositions or risk factors (advanced maternal age – greater than 35, family history of genetic risk, etc.)

Exclusion criteria were established to narrow the population to those of interest in this exploratory administration. Adolescents were excluded owing to excess complications, such as legal consent, and those factors that exist in decision-making with expectant women who are not of legal age. For example, limited autonomy for pregnant teenagers include situations such as many states not allowing a teenager access to an abortion without parental permission. These limitations create changes in informed decision-making and thus impact how adolescents would have IDM-PGS.

Sample size

Cognitive interviewers minimize the importance of sample size as a quantitative value and rather stress the importance of gathering a variety of diverse individuals within sample inclusion criteria (Willis, 1999). The audio-recorded cognitive interviews were planned on a minimum of four and up to twelve participants depending on when saturation of findings were obtained.

Data collection

All participant research was conducted per approval from the Institutional Review Board (IRB) at the University of Texas at Austin. Snowball sampling, flyers and social

media were used to collect a convenience sample of women who met the inclusion criteria. Participants were asked to nominate one or two potential participants, flyers were posted around The University of Texas at Austin campus inviting women who met inclusion criteria to contact the investigator if interested in the exploratory administration, classes and organizations with a dominant female population were approached with permission from course faculty, and social media such as Facebook, Twitter and blogs were also used for recruitment. Consent cover letters were provided to all participants and verbal consent were provided prior to the interviews.

The audio-recorded cognitive interviews lasted approximately thirty minutes each. Privacy and confidentiality of the audio-recorded cognitive interview participants were maintained by conducting the audio-recorded interviews in a private area with which the participants were familiar and comfortable. The recordings were maintained under password-protection until transcription of the interviews and study completion, these recordings have now been destroyed. Additionally since the audio-recorded interviews collected no names or personal identifiable information, and consent forms were not obtained with names and signatures, anonymity of participants was maintained, further advancing privacy and confidentiality.

Data Analysis

Cognitive interview results represent a more argumentative and applied interpretation of items in the instrument than an evaluation from the instrument developer alone (Willis, 1999). The data collected should enhance a general discussion about the understandability of the items and is immeasurable on a quantitative scale (Willis, 1999). Transcripts of cognitive interviews were collected and analyzed based on a review of participant understanding and interpretation of each item in a systematic method. Results

of the item-by-item review of participant responses allowed for suggestions towards modification and improvement of instrument items for increased understanding (Knafl et al., 2007; Miles & Huberman, 1994). Modifications to item wording, presentation and order were made based on systematic review of the results of cognitive interviews.

EXPLORATORY ADMINISTRATION OF THE INSTRUMENT

After compilation and preliminary testing of the developed instrument for IDM-PGS, exploratory administration of the developed IDM-PGS instrument was done. This exploratory administration was conducted with women who fit within the inclusion criteria outlined below.

Sample

The population of interest was narrowed to include two distinct groups of women who fit the inclusion criteria:

- Women
- 18 – 34 years of age
- No known genetic predispositions or risk factors (advanced maternal age – greater than 35, family history of genetic risk, etc.)

Similar to the cognitive interviews' exclusion criteria, adolescents were excluded due to excess complications, such as legal consent, that exist in decision-making with individuals that are not of legal age. The same example applies in that limited autonomy occurs for pregnant teenagers in a situation such as states not allowing a teenager access to an abortion without parental permission.

To distinguish the two known groups of women, the principal researcher collected demographic data from the women recruited to specify how to group their results. The first group was from the general population, and was assumed to be naïve in terms of

prenatal genetic health information. The second group was recruited from women who fit the above inclusion criteria but had received some training or education regarding prenatal genetic health information. This second group included women such as those with prior pregnancies who received prenatal care, nursing students, nurses, or physicians. The demographic data sheet (Appendix B) included questions specifically addressing parity, medical health-related training of the participant and occupational information.

Sample Size

According to Norman and Streiner (1994), a minimum sample size of five participants per item, with at least 100 subjects total, is adequate to ensure construct validity in instrument development. Marteau et al. (2001; Michie et al., 2002) developed a twelve-item measure of informed choice with a sample size of 66 pregnant women; a ratio of 5.5 participants per item. Instrument development is often conducted based on what is considered by many researchers a standard 10 participants to one item ratio (Nunnally, 1978). For purposes of this instrument development study a seven participant to one item ratio was used. Thus, with 37 items, a minimum of 259 participants were needed to be recruited to complete the exploratory administration, which is the psychometric evaluation of the survey. However, in order to ensure sufficient and usable data from 259 participants was retrieved, the population was over-sampled by 15%. Thus, a minimum 298 participants' data were collected and analyzed. Of note, Classical Test Theory notes that larger sample sizes tend to represent more reliable results and thus reduce the measurement error leading to an observed score that more closely represents the true score for a latent variable (Kline, 2005).

Data Collection

All participant research was conducted per approval from the Institutional Review Board (IRB) at The University of Texas at Austin. Flyers and social media were used to collect data from a convenience sample of women who met the inclusion criteria. Qualtrics provided an additional platform for administration of the instrument. A unique URL was created for this particular study. Flyers were posted around The University of Texas at Austin campus with the primary investigator's email address and the link to the survey, inviting women who met inclusion criteria to contact the investigator if interested in the exploratory administration. Classes and organizations with a predominantly female population were approached with permission from course faculty. Recruitment also occurred by providing the primary investigator's email address and link to the survey on social media such as Facebook, Twitter and blogs. Additionally, Qualtrics Panels were used to recruit participants. Qualtrics provides access to members of the general population through their network of respondents. Acceptance and completion of the survey was voluntary. After indicating an interest in the exploratory administration, participants received a thorough explanation of risks and benefits of participation in the exploratory administration in an electronic consent page. The participants were provided the option to bypass contacting the primary investigator as all recruitment material was included in the survey link. The survey was anonymous with minimal risk. Completion of the survey takes between fifteen to thirty minutes. The participants responded to the survey on a secure password-protected website. Qualtrics maintains IP addresses as a means to avoid multiple entries for a single respondent. Respondents not collected through Qualtrics were given the option to enter an email address for a two-week follow-up for retest. Two weeks after completion of the instrument, the participant was emailed

asking for their involvement to complete the instrument retest. Qualtrics panel respondents were contacted for the two week follow-up retest through Qualtrics.

CRITERION–RELATED VALIDITY.

Criterion-related validity is a measure of a test's relevance and usefulness by describing how well the results of a measurement fare in a real world outcome. In measurement of IDM-PGS, criterion-related validity was assessed through the administration of two items developed to explore the predictive validity of a survey respondents planned outcome. This predictive validity assessed if study participants feel they would have IDM-PGS if the situation they were presented with in this survey was their present situation. These two items represent a prediction of the course of action the individual would follow. A high correlation of the results of the other items in the IDM-PGS instrument with these two items shows the criterion-related validity of the instrument. However, of note, assessment of criterion-related validity in this IDM-PGS instrument is limited in that the attitude of the participant towards their likely course of action is a prediction based on their perception of reality and not necessarily the reality itself (Field, 2013).

CONSTRUCT VALIDITY.

To thoroughly evaluate the construct validity of the developed instrument, the known-groups method, and exploratory factor analysis were employed. Hattie and Cooksey (1984) explain that instrument scores must differ across groups that are known to be different in a key characteristic related to the phenomenon of concern. For the development of the IDM-PGS instrument, the sample described below was categorized into separate groups with expectedly distinct characteristics that should contribute to a significant variation in IDM-PGS. The groups explored in this study included women

who have had a prior pregnancy versus those who have never been pregnant, women who were pregnant at the time of the instrument was administered versus women who were not pregnant at the time the instrument was administered and women who have had formal medical training versus women who have had no medical training. Ideally by exploration of the differences in these three groups, construct validity can be further emphasized as these groups should represent a difference in levels of IDM-PGS. Scores on the IDM-PGS are expected to differ in the group of women who have been pregnant before and the group of women who are pregnant currently through the assumption that prenatal care has been or is currently being received and this knowledge regarding prenatal genetic screening would be increased versus the groups of women who have never been pregnant and women who are not currently pregnant, respectively. Similarly, scores for the group who have received medical training is assumed to be higher than those who have not been trained medically, as medical training should contain some sort of prenatal genetic health-related coursework, such as a nursing student, nurse or physician. Theoretically, individuals with prior knowledge about prenatal genetics, through pregnancy or medical training, should have a higher level of literacy surrounding prenatal genetics and thus have a higher level of IDM-PGS. The known-group differences were first analyzed by bivariate correlations between the values of the overall mean of the items with the demographic characteristics defining the groups of interest (i.e. past pregnancy, currently pregnant and medical training). Further analyses of group differences, using independent samples *t*-tests for each of the three demographic known groups of interest, were also performed.

An exploratory factor analysis was performed to provide further evidence of construct validity after completion of the exploratory administration of the instrument. The primary functions of this factor analysis were to highlight the structure of the latent

variables underlying the items in the instrument, and focus the data to make the results more manageable by reducing multicollinearity (De Vellis, 2012; Field, 2013). Parsimonious measurement of the latent variable should ideally occur with factor analysis resulting in the fewest number of explanatory constructs represented by the most common variance (Field, 2013). Items should load with similar factors as outlined by the definition of IDM-PGS and the adapted contextual framework. Results of the exploratory administration were factor analyzed and the principal researcher will use these results to improve the instrument for future study.

RELIABILITY

Results of the exploratory administration of this instrument provide data for reliability measurements. Reliability measures the consistency, predictability and dependability of an instrument's performance (De Vellis, 2012; Polit & Beck, 2008). To assess the reliability of this instrument, the principal researcher used Cronbach's alpha and test-retest reliability measures.

Cronbach's Alpha

The Cronbach's alpha coefficient for internal consistency was used to measure how consistently multiple items combined to measure a single concept. For many years researchers used split-half reliability and odd-even reliability measures to provide a numerical reference to the internal consistency of an instrument, but in 1951 Cronbach developed a statistical measure that averages all possible split-half reliabilities to give a single score representing intercorrelation amongst the items of the instrument (Cronbach, 1951; De Vellis, 2012). An ideal Cronbach's alpha that approaches or surpasses 0.80 was considered adequate reliability for the instrument to be used in further research

(Cronbach, 1951). Nevertheless, lower values of Cronbach's alpha may be more realistic and were accepted due to the highly psychological nature of this instrument.

In order to ensure adequate measurement of the Cronbach's alpha, the researcher conducted multiple measures. The first was an overall measure of Cronbach's alpha, due to the high item count, adequate reliability was expected. However, based on the results of factor analysis, the formula for Cronbach's alpha was applied to each factor found, thereby testing the reliability of every subscale that may potentially exist. Similarly, item-total statistics were also measured to ascertain the effect of each individual item on the overall and subscale reliability. Ideally each item should maintain a Cronbach's alpha near the overall Cronbach's alpha or the subscale Cronbach's alpha. Items whose individual removal causes a large improvement in Cronbach's alpha may need to be removed from the instrument (Cronbach, 1951; Field, 2013).

Test-Retest Reliability.

Another measure of reliability that was used in this exploratory administration was the test-retest. Test-retest reliability provides a measure of temporal stability, or the consistency of scores over time from one temporally unique situation to the next (De Vellis, 2012; Polit & Beck, 2008). To measure test-retest reliability the instrument was administered on two separate occasions and the scores correlated. A strong correlation suggests adequate test-retest reliability of the instrument. The test-retest reliability also represents a good indicator for measurement error as defined by Classical Test Theory. Highly correlated test-retest results indicate that measurement error is likely minimal (Kline, 2005).

For the IDM-PGS, the option for participants to opt for a retest occurred during the first administration of the instrument. At the end of the instrument, participants were

provided the option to enter their email address and were contacted two weeks after completion of the survey for a follow-up survey. The scores of the two administrations were correlated and the Pearson's correlation coefficient of the comparison was analyzed for a score approaching +1.0 (DiIorio, 2006). In order to correlate the results of a particular participant's IDM-PGS test with their retest results, a unique, anonymous identification code was created through the Qualtrics panels. For individuals whose data were collected outside of Qualtrics, unidentifiable information was collected to create a unique alphanumeric identifier for each respondent. This unique identifier included questions such as first letter of the respondents' birth month, number of siblings, and birth year. This unique identifier was created using coded values for first letter of birth month, number of siblings, birth year and gender to maintain anonymity (Lyons et al., 2009; Meray et al., 2007). The same unique identifier questions were asked during the two-week retest, and identically coded, thereby allowing the researcher to correlate the data for test-retest reliability calculations.

CHAPTER SUMMARY

This chapter provided an overview of the methods that were used in this exploratory administration of the IDM-PGS instrument. Details of the design and exploratory administration of the instrument were described. Planned data analyses were included.

Chapter Four: Results

This chapter presents the results of the instrument development and the exploratory administration of the Informed Decision-Making regarding Prenatal Genetic Screening (IDM-PGS) instrument.

INSTRUMENT DEVELOPMENT

The adapted conceptual framework, Lashley's (2007) list of essential information to consider for genetic testing and numerous literature sources led to the development of an initial set of over 100 items for potential inclusion in the instrument. These items were conceptually mapped to ensure they addressed all concepts pertinent to the definition of IDM-PGS. The items were further reviewed by a group of colleagues who were either doctoral students or research faculty at the School of Nursing. In this group setting, items were collaboratively added, removed and reworded for clarity and parsimony. After this review, 85 items remained for inclusion in this instrument.

These 85 items were then further evaluated for content validity through expert panelists. Detailed explanation of items saved versus those removed for inclusion occurs later in this chapter. Upon completion of content validity analysis, 37 items remained in the final instrument that was used for exploratory administration.

Item-by-Item Readability

The overall SMOG readability for this instrument was initially calculated at 18.1. Item-by-item readability scores are shown below in Table 2. The average of the item-by-item readability was 17.9, indicating a reading level of an individual who has done some graduate school (McLaughlin, 1969).

Table 2. Item-by-Item SMOG Readability Score

Item	SMOG Readability Score
1	19.6
2	19.6
3	18
4	18
5	18
6	18
7	18
8	22.1
9	20.9
10	19.6
11	18
12	16.2
13	18
14	16.2
15	18
16	16.2
17	16.2
18	18
19	16.2
20	16.2
21	22.1
22	16.2
23	18
24	18
25	18
26	19.6
27	18
28	18
29	18
30	16.2
31	16.2
32	18
33	22.1
34	13.8
35	13.8
36	16.2
37	18

When the term “prenatal genetic screen” or “prenatal genetic screening” is reduced to “PGS” for evaluation purposes, the overall instrument SMOG readability for this instrument was re-calculated at 15.4. The average of the item-by-item SMOG readability was then re-calculated at 14.5. A readability of 14.5 denotes individuals with some college education should be able to comprehend the instrument.

Content Validity.

The initial set of 85 items was sent to three expert panelists for review. The members of this initial panel included three doctorally-prepared nurses researching various aspects of genetics and genomics, including two focused specifically on maternal-child genetics and genomics, and the other with a primary focus on informed decision making. The results were returned and analyzed resulting in a S-CVI of 0.497. Numerous I-CVI’s fell well below adequate levels, items scoring lower than a 0.25 were removed and the CVI was recalculated at 0.65.

Based on recommendations of the expert panelists, a number of items were added prior to resending for CVI analysis. The items were resent to the previous three expert panelists and three new experts including one masters-prepared nurse with many years of experience in maternal-child nursing and teaching experience in genetics, and two retired obstetric-gynecologic physicians.

The results from the second round returned a S-CVI of 0.53. One of the expert panelists from the initial set did not respond to the second request and thus the results from those items that were unchanged from the initial request were carried over to represent her responses. Another of the initial expert respondents returned the second round request for CVI by responding with all zeros for content validity. Thus the responses from these two panelists were removed from the content validity index

measurements. Finally, items with I-CVI scores lower than 0.33 were removed and the final S-CVI for the remaining 37 items was 0.78, thus acceptable for content validity of this instrument.

Cognitive Interviews

Prior to completion of the content validity analysis, when the instrument still included 48 items, cognitive interviews were conducted. Eight cognitive interviews of expectant women between the ages of 24 to 34, with an average age of 30, were conducted by the primary investigator. These eight participants included five Caucasians, one Black, one Hispanic and one Asian Indian woman. All participants were pregnant at the time of the interviews. The average gestational age was 29.1 weeks, ranging from 17 to 39.5 weeks. Five participants were primiparas and the remaining three were expecting their second child.

The results of the cognitive interviews were very consistent in that most interviewees were able to interpret and accurately verbalize back the intended meaning of each item. Of the eight participants, two participants tended to repeat back the items of the instrument verbatim as opposed to in their own words as requested, despite numerous prompts from the interviewer. No single item was flagged by any two or more participants as misinterpreted. Eight items were flagged by only one participant as an incorrect interpretation. All eight of the incorrect responses related to an inaccurate interpretation of the level of diagnosis versus risk assessment being measured in the IDM-PGS instrument. When the item was reread and the distinguishing term was emphasized on two of the incorrect responses, both participants self-corrected to the correct response.

After completing the content validity index analyses and the cognitive interviews, the final 37-item instrument was administered to an exploratory group of women.

Criterion-related Validity

The criterion-related validity was calculated independently for the initial administration and the two-week retest administration. Since items 36 and 37 were aimed at measuring criterion-related validity, the results of these items were combined at each time point respectively to measure predictive validity. In the exploratory administration situations the analysis resulted in highly significant ($p < 0.01$), but a relatively small correlation, which was only at the initial administration (see Table 3). The correlation between the re-test administration and the predictive question was not statistically significant.

Table 3. Criterion-Related Validity Correlation

		TotalPred_1	TotalPred_2
TotalTime1	Pearson Correlation	.215**	
	Sig. (2-tailed)	.000	
	N	279	
TotalTime2	Pearson Correlation		.160
	Sig. (2-tailed)		.094
	N		111

**. Correlation is significant at the 0.01 level (2-tailed). TotalTime1: Mean of all results in the initial exploratory administration. TotalTime2: Mean of all results in the two-week retest time point. TotalPred_1: Mean of the results of the two predictive variables in the initial exploratory administration. TotalPred_2: Mean of the results of the two predictive variables in the two-week retest time point.

Construct Validity

Construct validity was calculated using the known-groups method and exploratory factor analysis. The results of the known-groups method was calculated by a

bivariate correlation between the values of the overall mean of the items with the demographic characteristics defining the groups of interest. The overall mean included all of the items except two items that were added as a measure of predictive or criterion-related validity at the initial administration time. The demographic characteristics of interest included medical training and pregnancy, both currently and in the past. The results of this analysis were all small correlations with only the demographic group of participants who had medical training as a significant correlation (see Table 4). Of note, the demographic group of women who had been pregnant in the past fell just outside of significance in this situation with $p = 0.051$. As can be seen in Table 4, all three demographic known groups of interest show a negative correlation which is the expected outcome based on item coding, as these items are reverse coded (Field, 2013).

Table 4. Known Groups Correlation Initial Administration

		TotalTime1	Mdtrn	PstPrg	NwPrg
TotalTime1	Pearson Correlation	1	-.146*	-.116	-.009
	Sig. (2-tailed)		.014	.051	.878
	N	284	284	284	284

*. Correlation is significant at the 0.05 level (2-tailed). TotalTime1: Mean of all results in the initial exploratory administration. Mdtrn: Item measuring medical training. PstPre: Item measuring if participants had a previous pregnancy. NwPrg: Item measuring if a participant was currently pregnant.

At the two-week retest time point, none of the correlations with medical training, previous pregnancy or current pregnancy were significantly related (see Table 5).

Table 5. Known Groups Correlation Two-Week Retest Administration

		TotalTime2	Mdtrn_2	PstPrg_2	NwPrg_2
TotalTime2	Pearson Correlation	1	-.113	-.138	.152
	Sig. (2-tailed)		.239	.147	.111
	N	111	111	111	111

TotalTime2: Mean of all results in the two-week retest time point. Mdtrn_2: Item measuring medical training at the two-week retest time point. PstPre: Item measuring if participants had a previous pregnancy at the two-week retest time point. NwPrg: Item measuring if a participant was currently pregnant at the two-week retest time point.

Group differences were further analyzed using independent samples *t*-tests for each of the three demographic known groups of interest: medically trained versus not medically trained; pregnant before versus never pregnant; pregnant currently versus not currently pregnant. Results showed a significant ($p < 0.05$) difference in means only in the independent samples *t*-test analysis for the demographic group of past medical training versus those without medical training at the initial exploratory administration time point (see Table 6 and Table 7). The Levene's test showed significance, so the t-test for "Equal variances not assumed" was used and in the medically trained versus not medically trained group, the group difference in mean was significant at $p = 0.037$.

Table 6. Levene's Test for Medically Trained Versus Not Medically Trained Individuals

		Levene's Test for Equality of Variances		t-test for Equality of Means	
		F	Sig.	t	df
TotalTime1	Equal variances assumed	6.896	.009	2.476	282
	Equal variances not assumed			2.121	78.300

Table 7. T-Test for Medically Trained Versus Not Medically Trained Individuals

		t-test for Equality of Means		
		Sig. (2-tailed)	Mean Difference	Std. Error Difference
TotalTime1	Equal variances assumed	.014	.18058	.07293
	Equal variances not assumed	.037	.18058	.08514

Results of the independent samples *t*-test did not show a significant difference between the group of women who had been pregnant in the past versus those who had never been pregnant (see Table 8 and Table 9).

Table 8. Levene's Test for Pregnant Before Versus Never Pregnant

		Levene's Test for Equality of Variances		t-test for Equality of Means	
		F	Sig.	t	df
TotalTime1	Equal variances assumed	3.280	.071	1.959	282
	Equal variances not assumed			1.980	278.512

Table 9. T-Test for Pregnant Before Versus Never Pregnant

		t-test for Equality of Means		
		Sig. (2-tailed)	Mean Difference	Std. Error Difference
TotalTime1	Equal variances assumed	.051	.11727	.05985
	Equal variances not assumed	.049	.11727	.05924

Similarly, results of the independent samples *t*-test did not show a significant difference between the group of women who were pregnant at the time of the instrument

implementation versus those who were not pregnant at the time of instrument implementation (see Table 10 and Table 11). The Levene's test did not show significance, so the t-test for "Equal variances assumed" was used and in the previously pregnant versus never pregnant group, the group difference in means approached significance at $p = 0.051$. Though not significant at the $p < 0.05$ level, a $p = 0.051$ is very close to significance, and as if often used with instrument developments, a less stringent cutoff for *p*, such as $p < 0.10$ would consider a $p = 0.051$ significant. In the currently pregnant versus not currently pregnant group comparison, the Levene's test did not show significance, so the t-test for "Equal variances assumed" was used; the group difference in mean was not significant at $p = 0.878$. This *p*-value is well outside range of any level of significance used in *t*-test analyses.

Table 10. Levene's Test for Currently Pregnant Versus Not Currently Pregnant

		Levene's Test for Equality of Variances		t-test for Equality of Means	
		F	Sig.	t	df
TotalTime1	Equal variances assumed	.359	.549	.154	282
	Equal variances not assumed			.162	92.343

Table 11. T-Test for Currently Pregnant Versus Not Currently Pregnant

		t-test for Equality of Means		
		Sig. (2-tailed)	Mean Difference	Std. Error Difference
TotalTime1	Equal variances assumed	.878	.01158	.07513
	Equal variances not assumed	.871	.01158	.07135

Next a factor analysis was conducted on the results of the two independent administrations. For the initial administration of the instrument, the last initial Eigenvalues that remained at 1.0 or above were at component number 10, which suggests that the 35 items in this instrument intended to analyze IDM-PGS loaded on 10 factors (see Table 12). An Eigenvalue refers to the variance in items of an instrument that is explained by the principal component or factor (Pett, Lackey, & Sullivan, 2003).

Table 12. Eigen Values for Initial Exploratory Administration

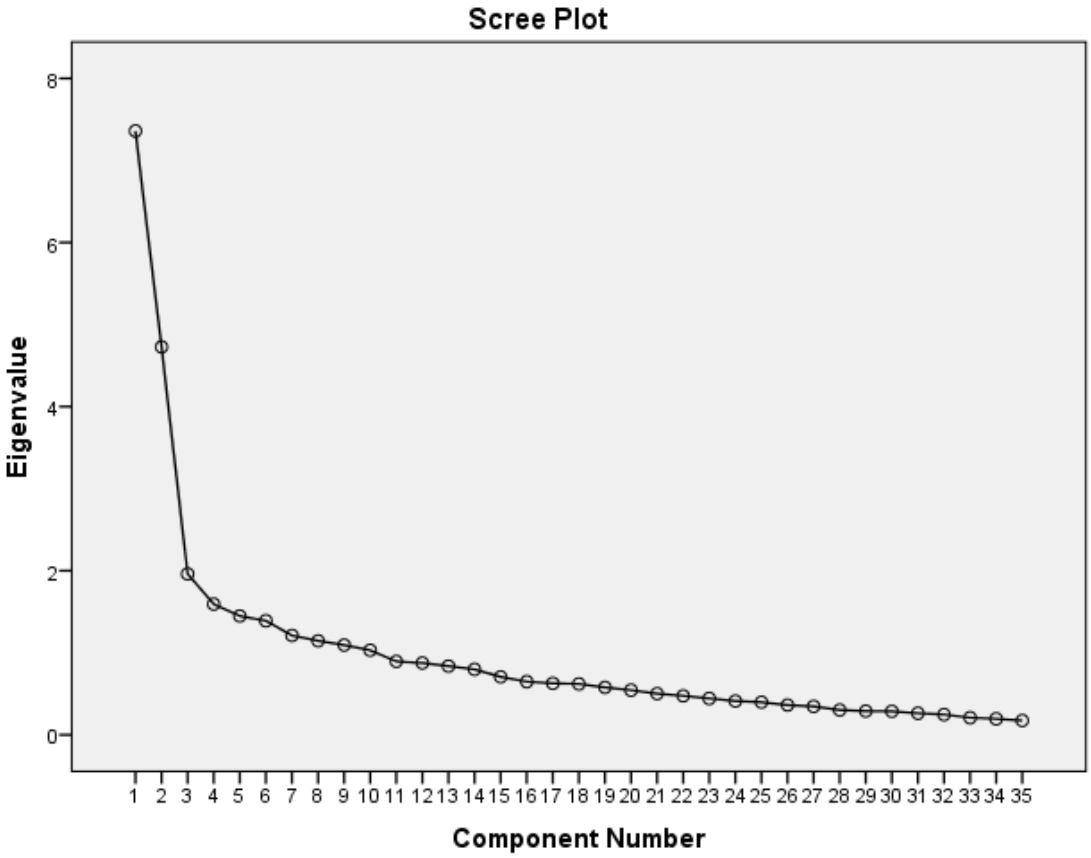
Component	Initial Eigenvalues			Extraction Sums of Squared Loadings			Rotation Sums of Squared Loadings
	Total	% of Variance	Cumulative %	Total	% of Variance	Cumulative %	Total
1	7.359	21.025	21.025	7.359	21.025	21.025	6.063
2	4.728	13.507	34.533	4.728	13.507	34.533	4.181
3	1.961	5.603	40.136	1.961	5.603	40.136	2.055
4	1.592	4.550	44.686	1.592	4.550	44.686	1.784
5	1.449	4.139	48.825	1.449	4.139	48.825	1.762
6	1.390	3.972	52.797	1.390	3.972	52.797	1.632
7	1.211	3.459	56.255	1.211	3.459	56.255	1.561
8	1.146	3.274	59.529	1.146	3.274	59.529	1.402
9	1.095	3.127	62.657	1.095	3.127	62.657	1.311
10	1.030	2.944	65.601	1.030	2.944	65.601	1.210
11	.895	2.557	68.157				
12	.875	2.501	70.659				
13	.838	2.394	73.052				
14	.798	2.279	75.332				
15	.706	2.019	77.350				
16	.648	1.852	79.202				

Table 12. (continued)

17	.628	1.794	80.996
18	.621	1.774	82.769
19	.579	1.653	84.423
20	.544	1.556	85.978
21	.502	1.435	87.413
22	.476	1.359	88.772
23	.443	1.266	90.038
24	.412	1.176	91.214
25	.398	1.138	92.352
26	.363	1.037	93.389
27	.347	.991	94.381
28	.302	.864	95.244
29	.289	.826	96.070
30	.287	.820	96.890
31	.263	.753	97.643
32	.247	.706	98.349
33	.208	.595	98.944
34	.195	.558	99.502
35	.174	.498	100.000

Pett and colleagues (2003) suggest extracting factors until the addition of a further extracted factor would account for less than 5% of the explained variance. By this method the results produced three components of the ten initial components overall with 21.025%, 13.507% and 5.603% of variance explained by these first three components respectively. The fourth component would explain only 4.550% of the variance. Similarly, the results of the scree plot of Eigenvalues of the initial instrument administration (see Figure 5) indicated three or four major components.

Figure 5. Scree Plot of Initial Administration of Instrument.



The factor loading on the component matrix analysis showed that though 10 factors loaded on the instrument, only 6 factors loaded with the highest correlations as can be seen on Table 13, Table 14 and Table 15. However, this table was not analyzed further as a rotated factor analysis was performed.

Table 13. Component Matrix

	1	2	3	4	5	6	7	8	9	10
Q1	-.261	.533	.352	.304	.289	.148	.161	-.137	.008	.070
Q2	.036	.284	.580	.403	.363	.109	.076	-.067	-.219	-.014
Q3	.769	.069	-.091	.052	-.088	-.189	-.100	.140	-.068	.115
Q4	.644	.006	-.040	-.025	.000	-.094	-.147	.053	.164	.074
Q5	-.301	.542	.165	.220	.064	-.084	-.113	.133	.201	.264
Q6	-.508	.002	-.158	-.053	.385	.143	-.386	-.023	.208	.073
Q7	.350	.034	.434	-.169	-.199	-.298	.267	.010	-.371	.028
Q8	.009	.566	-.535	.191	-.034	.037	.093	.109	.048	-.047
Q9	.474	-.024	-.249	.277	.164	.141	.175	.148	-.198	-.109
Q10	-.194	.501	-.175	.034	.209	.171	.309	.248	-.118	.066
Q11	.614	.063	.137	.082	.142	.155	-.176	-.053	.224	-.075
Q12	.783	.058	-.086	-.092	-.076	-.094	-.009	-.061	.057	.009
Q13	.646	-.034	-.150	-.098	-.043	.022	.005	-.068	.106	-.028
Q14	.141	.650	-.263	.202	-.012	-.070	-.149	-.191	-.127	-.112
Q15	-.333	.355	.414	-.039	.111	-.361	.060	.346	.067	.105
Q16	.537	.253	.223	.038	-.172	-.313	-.259	-.029	-.142	-.130
Q17	-.004	.768	-.013	-.106	-.157	-.243	-.239	.033	.058	.057
Q18	.616	.130	-.125	.228	.124	.101	-.203	-.356	-.266	-.098
Q19	.674	.249	.016	.114	.171	.028	-.211	-.161	-.189	-.075
Q20	.457	-.392	-.159	.091	.354	.020	.189	.019	.119	.305
Q21	.723	.062	-.189	.094	.173	.003	.228	.094	.065	.135
Q22	.509	-.142	.249	.015	.117	.073	-.281	.211	.286	.041
Q23	.446	.033	.349	-.219	.029	.170	.262	-.109	.274	-.419
Q24	.004	.629	-.075	-.075	-.101	.041	.303	-.073	.244	-.340
Q25	-.085	.588	.065	-.003	.029	.052	-.051	.251	-.075	-.185
Q26	.463	-.078	.056	.135	-.041	-.214	.363	-.300	.361	.063
Q27	.337	-.176	.143	-.125	.036	.395	-.150	.475	-.026	-.385
Q28	.123	.748	-.149	-.108	-.134	.109	.057	.042	.112	.145
Q29	.678	.312	.080	-.018	-.047	.063	-.008	.162	.147	.159
Q30	.685	.252	.042	-.135	-.008	-.082	.026	.128	-.034	.081
Q31	-.118	.495	.050	-.228	-.411	.490	-.006	-.117	-.068	.215
Q32	.324	-.034	.327	-.003	-.318	.549	-.009	-.115	-.063	.356
Q33	.274	-.332	-.232	.323	-.250	.053	.141	.362	-.187	.043
Q34	.472	-.094	.041	-.547	.354	.017	.045	.034	-.199	.165
Q35	-.063	.398	-.208	-.636	.402	-.011	.061	-.107	-.184	.030

Table 14. Component Matrix Factor Loading with Item Descriptions

#	Short Description of Item	1	2	3	4	5	6	7	8	9	10
Q3	Fetus might have genetic disease	.769									
Q4	Fetus DNA for risk of genetic disease	.644									
Q6	Would wait until birth	-.508									
Q9	Ultrasound less accurate	.474									
Q11	Blood test for maternal protein markers	.614									
Q12	Down Syndrome	.783									
Q13	Spina Bifida	.646									
Q16	Results might be wrong	.537									
Q18	Positive means risk for genetic disease	.616									
Q19	Positive means might be at risk for genetic disease	.674									
Q20	Trust results	.457									
Q21	More information than regular prenatal checks	.723									
Q22	Checks maternal protein markers	.509									
Q23	Results protected, cannot deny baby insurance	.446									
Q26	Results available to anyone with permission	.463									
Q29	May have emotional effect	.678									
Q30	Lifestyle may change	.685									
Q1	Diagnose fetal genetic disease		.533								
Q5	Fetal DNA free of genetic disease		.542								
Q8	Ultrasound equally accurate		.566								
Q10	Ultrasound for thickness of fetal neck		.501								
Q14	Positive means no risk of genetic disease		.650								

Table 14 (continued)

Q17	Results always right	.768				
Q24	Problem is pre-existing condition and uninsurable	.629				
Q25	Results available to anyone	.588				
Q28	No emotional effect	.748				
Q31	No risks	.495				
Q2	Cannot diagnose fetal genetic disease		.580			
Q7	Can wait until birth		.434			
Q15	Positive means specific problem		.414			
Q34	Have treatment options			-.547		
Q35	No treatment options			-.636		
Q32	Risk of pain, bleeding, infection and death				.549	
Q27	Results only to partner and participant					.475
Q33	No options for equally accurate medical information					.362

Table 15. List of Factor Loadings from Component Matrix

1	2	3	4	5	6	7	8	9	10
Q3	Q1	Q2	Q34		Q32		Q27		
Q4	Q5	Q7	Q35				Q33		
Q6	Q8	Q15							
Q9	Q10								
Q11	Q14								
Q12	Q17								
Q13	Q24								
Q16	Q25								
Q18	Q28								
Q19	Q31								
Q20									
Q21									
Q22									
Q23									
Q26									
Q29									
Q30									

Rotated analysis is considered the gold standard for factor analysis. The factor analyses component matrix was Varimax or Orthogonally rotated for increased interpretability and usefulness of results (Nunnally & Bernstein, 1994). The results of this rotated factor analysis resulted in 10 factors as well (see Table 16, Table 17 and Table 18), with the specific items loading very similar to the un-rotated 10 factors. Items that loaded on components 3, 5, 6 & 7 all loaded on the dimension evaluating decision context. Component 8 loaded items evaluating cognitive appraisals. Components 9 and 10 loaded their items on the other factors identified by constructs of the adapted model. Components 1, 2 and 4 were divided amongst various dimensions of interest. The subscales and groups of factors were named as per Table 19.

Table 16. Rotated Component Matrix

	1	2	3	4	5	6	7	8	9	10
Q1	-.180	.319	.737	-.045	-.087	-.036	.005	.099	.098	-.127
Q2	.032	-.003	.861	.017	.139	.108	-.037	.024	.039	.072
Q3	.737	.062	-.131	.150	.274	.124	-.026	-.036	-.138	.004
Q4	.679	-.025	-.121	.021	.044	.059	-.009	-.018	.020	-.008
Q5	-.036	.438	.410	-.203	-.174	-.319	-.129	.036	-.208	-.128
Q6	-.275	-.008	.084	-.224	-.664	-.059	.148	-.108	-.193	.060
Q7	.184	-.107	.114	-.047	.770	-.052	.130	.053	.044	-.022
Q8	.032	.732	-.085	.266	-.168	.101	-.046	-.077	-.058	-.088
Q9	.310	.050	.040	.555	.061	.279	-.018	-.096	.019	.121
Q10	-.188	.527	.240	.335	-.069	-.153	.233	.033	-.027	.044
Q11	.641	-.053	.138	-.017	-.104	.180	-.048	.063	.214	.110
Q12	.709	.035	-.188	.082	.213	.193	.066	.027	.129	-.065
Q13	.569	-.020	-.229	.124	.071	.189	.063	.047	.173	-.032
Q14	.145	.651	.124	-.074	.003	.392	-.020	-.073	-.095	-.108
Q15	-.122	.227	.387	-.250	.179	-.567	.042	-.206	-.120	.034
Q16	.525	.145	.066	-.279	.423	.212	-.105	-.103	-.062	.086
Q17	.173	.709	.073	-.407	.090	-.080	.049	-.002	-.159	-.058
Q18	.471	.047	.123	.088	.065	.696	.029	.027	-.029	-.034
Q19	.604	.119	.191	.018	.122	.474	.103	-.009	-.015	.070
Q20	.434	-.404	-.014	.461	-.156	-.019	.145	-.062	-.008	-.226
Q21	.654	.061	-.008	.452	.071	.074	.109	-.026	.098	-.123
Q22	.632	-.246	.077	-.072	-.118	-.112	-.095	.028	.055	.237
Q23	.326	-.073	.078	-.058	.162	.027	.074	.060	.740	.166
Q24	-.021	.662	.043	-.046	.043	-.018	.018	-.016	.493	-.052
Q25	-.024	.559	.233	-.083	.051	-.070	.054	-.030	.001	.283
Q26	.413	-.120	-.008	.116	.136	-.005	-.143	-.054	.398	-.495
Q27	.266	-.141	-.063	.156	-.013	-.007	-.011	.059	.195	.757
Q28	.204	.730	.041	-.042	-.013	-.077	.129	.259	.029	-.092
Q29	.717	.226	.060	.093	.125	-.059	.025	.195	.071	.040
Q30	.656	.176	-.009	.085	.281	.020	.169	.061	.039	.046
Q31	-.135	.447	-.021	-.159	.024	.013	.085	.731	.018	.038
Q32	.248	-.177	.108	.041	.106	.066	-.087	.792	.025	.050
Q33	.134	-.136	-.237	.545	.186	.016	-.335	.012	-.196	.145
Q34	.405	-.228	-.060	.076	.142	.006	.687	.047	.011	.092
Q35	-.064	.320	-.033	-.111	-.057	.036	.833	-.054	.027	-.018

Table 17. Rotated Component Matrix Factor Loading with Item Descriptions

#	Short Description of Item	1	2	3	4	5	6	7	8	9	10
Q3	Fetus might have genetic disease	.737									
Q4	Fetus DNA for risk of genetic disease	.679									
Q11	Blood test for maternal protein markers	.641									
Q12	Down Syndrome	.709									
Q13	Spina Bifida	.569									
Q16	Results might be wrong	.525									
Q19	Positive means might be at risk for genetic disease	.604									
Q21	More information than regular prenatal checks	.654									
Q22	Checks maternal protein markers	.632									
Q29	May have emotional effect	.717									
Q30	Lifestyle may change	.656									
Q5	Fetal DNA free of genetic disease		.438								
Q8	Ultrasound equally accurate		.732								
Q10	Ultrasound for thickness of fetal neck		.527								
Q14	Positive means no risk of genetic disease		.651								
Q17	Results always right		.709								
Q24	Problem is pre-existing condition and uninsurable		.662								
Q25	Results available to anyone		.559								
Q28	No emotional effect		.730								
Q1	Diagnose fetal genetic disease			.737							
Q2	Cannot diagnose fetal genetic disease			.861							

Table 17 (continued)

Q9	Ultrasound less accurate	.555					
Q20	Trust results	.461					
Q33	No options for equally accurate medical information	.545					
Q6	Would wait until birth		-.664				
Q7	Can wait until birth		.770				
Q15	Positive means specific problem			-.567			
Q18	Positive means risk for genetic disease			.696			
Q34	Have treatment options				.687		
Q35	No treatment options				.833		
Q31	No risks					.731	
Q32	Risk of pain, bleeding, infection and death					.792	
Q23	Results protected, cannot deny baby insurance						.740
Q26	Results available to anyone with permission						-.495
Q27	Results only to partner and participant						.757

Table 18. List of Factor Loadings from Rotated Component Matrix

1	2	3	4	5	6	7	8	9	10
Q3	Q5	Q1	Q9	Q6	Q15	Q34	Q31	Q23	Q26
Q4	Q8	Q2	Q20	Q7	Q18	Q35	Q32		Q27
Q11	Q10		Q33						
Q12	Q14								
Q13	Q17								
Q16	Q24								
Q19	Q25								
Q21	Q28								
Q22									
Q29									
Q30									

Table 19. Factor Subscales Names

Factor	Subscale Name/ Description
1	Reason to have screen
2	General knowledge of screening
3	Diagnosis of genetic disease
4	Accuracy of screening
5	Option to wait
6	Meaning of positive results
7	Treatment options
8	Risk
9	Legal protections of fetus
10	Availability of results

EXPLORATORY ADMINISTRATION

Recruitment for a preliminary exploratory administration of the instrument was initially conducted through snowball sampling, flyers and social media. After three weeks

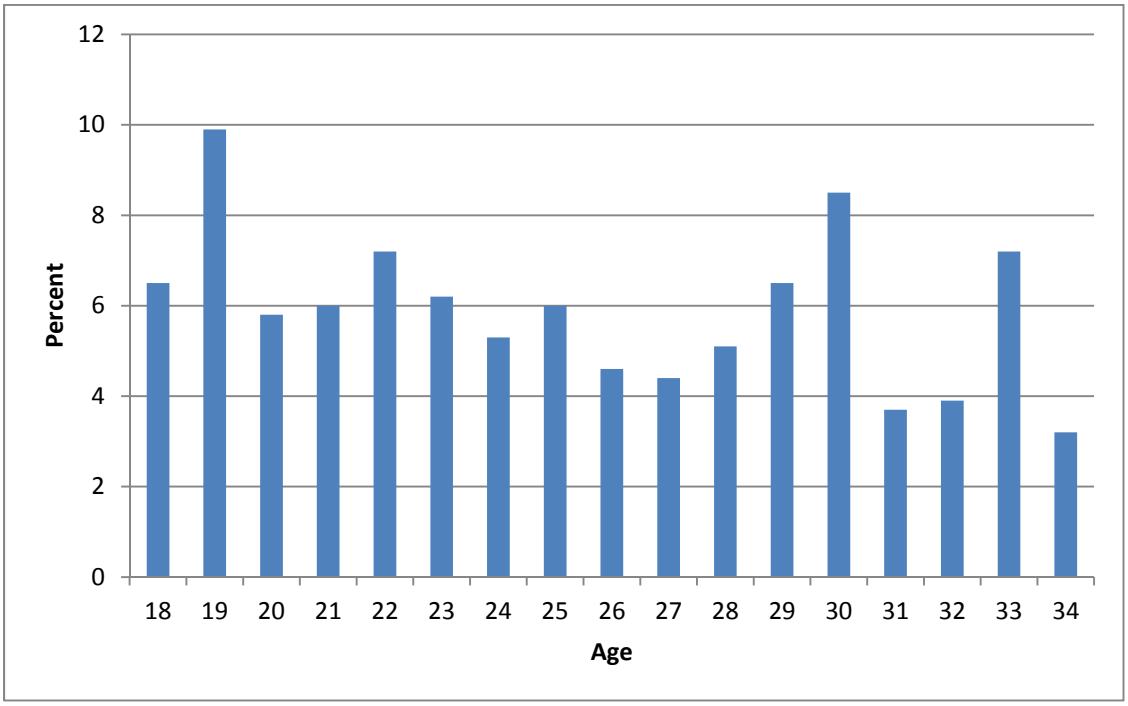
of recruitment in this manner, only 27 participants were enrolled. Qualtrics' panels were then employed to further expand participant recruitment. Qualtrics provides access to a large network of potential respondents through their network. These respondents can be accessed based on specific inclusion criteria of the study. Upon final completion of the initial administration, 433 valid participant responses were collected. The two-week retest resulted in 111 valid participant responses being collected.

Sociodemographic characteristics

Initial Exploratory Administration

The initial exploratory administration of the instrument resulted in 433 female participants who met the inclusion criteria for participation in this instrument development administration. The ages of the women in the sample were rather equally dispersed amongst the age range of interest (18 - 34) with a mean age of 26.36, and a standard deviation of 4.93 (see Figure 6).

Figure 6. Age Frequency Bar Chart



A majority (59.8%) of women in this sample were single, with 37.2% married women, 2.3% women were divorced or separated and 0.7% were widows (see Table 20). This sample was also predominately heterosexual at 74.6% with only 9.0% stating they were bisexual and 3.7% stating they were lesbian (see Table 21). Results of the sexual orientation should correlate with those of the partner's gender, as one's sexual orientation would determine their partner's gender. However, in the results of this sample, the partner's gender was calculated at 66.3% male and 21.0% female, both with 12.7% missing (see Table 22), which did not correlate with the 74.6% heterosexual orientation result.

Table 20. Marital Status of Initial Exploratory Administration Group

		Frequency	Percent	Valid Percent	Cumulative Percent
Valid	Single	259	59.8	59.8	59.8
	Married	161	37.2	37.2	97.0
	Divorced/ Separated	10	2.3	2.3	99.3
	Widows	3	.7	.7	100.0
	Total	433	100.0	100.0	

Table 21. Sexual Orientation of Initial Exploratory Administration Group

		Frequency	Percent	Valid Percent	Cumulative Percent
Valid	Heterosexual	323	74.6	85.4	85.4
	Homosexual	16	3.7	4.2	89.7
	Bisexual	39	9.0	10.3	100.0
	Total	378	87.3	100.0	
Missing	System	55	12.7		
Total		433	100.0		

Table 22. Partner Gender of Initial Exploratory Administration Group

		Frequency	Percent	Valid Percent	Cumulative Percent
Valid	Male	287	66.3	75.9	75.9
	Female	91	21.0	24.1	100.0
	Total	378	87.3	100.0	
Missing	System	55	12.7		
Total		433	100.0		

Non-Hispanic, White women were the largest racial group in this administration at 38.8%, black women made up 10.2% and Asians and Hispanic women each made up

9.5%, Hawaiian/ Pacific Islanders, Native/ American Indian and the other group made up less than 1% each, and 30.3% of participants did not respond to this demographic question (see Table 23).

Table 23. Race of Initial Exploratory Administration Group

		Frequency	Percent	Valid Percent	Cumulative Percent
Valid	Non-Hispanic, White	168	38.8	55.6	55.6
	Hispanic	41	9.5	13.6	69.2
	Black	44	10.2	14.6	83.8
	Asian	41	9.5	13.6	97.4
	Hawaiian/ Pacific Islander	2	.5	.7	98.0
	American Indian/ Alaskan Native	2	.5	.7	98.7
	Other	4	.9	1.3	100.0
	Total	302	69.7	100.0	
Missing	System	131	30.3		
Total		433	100.0		

A majority of the women in this sample had with some college or higher education: 25.6% had some college education, 20.6% were college graduates and 8.1% were or had been in graduate school. Only 3.0% had not completed high school and 12.5% had only completed high school (see Table 24). Within this group, 54.5% of women had no formal medical health-related training and 15.2% were trained medically (see Table 25). The employment status demographic showed this group was predominantly employed at 29.3%. Within this sample, 16.4% of women were students, 15% were stay-at-home partners, 8.3% were unemployed and 0.7% were in the military (see Table 26). All three of these demographic questions were also not answered by 30.3% of participants.

Table 24. Education History of Initial Exploratory Administration Group

		Frequency	Percent	Valid Percent	Cumulative Percent
Valid	Some High School	13	3.0	4.3	4.3
	High School Diploma	54	12.5	17.9	22.2
	Some College	111	25.6	36.8	58.9
	College Graduate	89	20.6	29.5	88.4
	Graduate School	35	8.1	11.6	100.0
	Total	302	69.7	100.0	
Missing	System	131	30.3		
Total		433	100.0		

Table 25. Medical Training of Initial Exploratory Administration Group

		Frequency	Percent	Valid Percent	Cumulative Percent
Valid	Trained	66	15.2	21.9	21.9
	Not Trained	236	54.5	78.1	100.0
	Total	302	69.7	100.0	
Missing	System	131	30.3		
Total		433	100.0		

Table 26. Employment Status of Initial Exploratory Administration Group

		Frequency	Percent	Valid Percent	Cumulative Percent
Valid	Employed	127	29.3	42.1	42.1
	Unemployed	36	8.3	11.9	54.0
	Student	71	16.4	23.5	77.5
	Military	3	.7	1.0	78.5
	Stay at Home Partner	65	15.0	21.5	100.0
	Total	302	69.7	100.0	
Missing	System	131	30.3		
Total		433	100.0		

Thirty-three percent of the women participating in this initial instrument development administration stated they were spiritual and religious. Twenty-one and a half percent stated they were spiritual and non-religious and 15.2% denied being spiritual or religious (see Table 27). This demographic question also was not answered by 30.3% of participants.

Table 27. Spirituality of Initial Exploratory Administration Group

		Frequency	Percent	Valid Percent	Cumulative Percent
Valid	Spiritual, Non-Religious	93	21.5	30.8	30.8
	Spiritual, Religious	143	33.0	47.4	78.1
	Non-Spiritual, Non-Religious	66	15.2	21.9	100.0
	Total	302	69.7	100.0	
Missing	System	131	30.3		
Total		433	100.0		

A majority of participants in this administration had no children (40.6%). The percentage of women with children was inversely related to the number of children.

Those women with only one child constituted 13.2% of participants, 10.4% had two, 2.5% had three, 2.3% had four and less than one percent (0.7%) had five or more children (see Table 28). Synchronously, 37.4% of the women had never been pregnant and 32.3% had been pregnant before (see Table 29). Of women participating in this administration, 14.1% were pregnant at the time that the results of this study were collected and 55.7% confirmed not being pregnant at the time of completion of this instrument (see Table 30). Again, 30.3% of participant did not respond to these demographic questions.

Table 28. Number of Children of Initial Exploratory Administration Group

		Frequency	Percent	Valid Percent	Cumulative Percent
	0	176	40.6	58.3	58.3
	1	57	13.2	18.9	77.2
	2	45	10.4	14.9	92.1
Valid	3	11	2.5	3.6	95.7
	4	10	2.3	3.3	99.0
	5+	3	.7	1.0	100.0
	Total	302	69.7	100.0	
Missing	System	131	30.3		
Total		433	100.0		

Table 29. Past Pregnancy History of Initial Exploratory Administration Group

		Frequency	Percent	Valid Percent	Cumulative Percent
	Pregnant Before	140	32.3	46.4	46.4
Valid	Never Pregnant	162	37.4	53.6	100.0
	Total	302	69.7	100.0	
Missing	System	131	30.3		
Total		433	100.0		

Table 30. Current Pregnancy Status of Initial Exploratory Administration Group

		Frequency	Percent	Valid Percent	Cumulative Percent
Valid	Pregnant Now	61	14.1	20.2	20.2
	Not Pregnant Now	241	55.7	79.8	100.0
	Total	302	69.7	100.0	
Missing	System	131	30.3		
Total		433	100.0		

Two-Week Retest Administration

The two-week retest administration of the instrument resulted in responses from 111 of the initial 433 female participants. The results of the sociodemographic characteristics of the retest group should ideally reflect a smaller but true ratio of the results described above. As expected, the ages of the women in the two-week retest sample were similarly equally dispersed over the range of interest with a mean of 26.55 and a standard deviation of 4.99. Since a large percentage (74.4%) of the results represents individuals who completed the initial exploratory administration and not the retest, those results are blank in data output. Thus, in data analysis only valid percentage will be considered in discussion of the results. The majority (59.5%) of women in the two-week retest sample were single and 40.5% were married. No divorced, separated, or widowed women participated in this second administration of the instrument (see Table 31). This sample was also predominately heterosexual at 98.2% with the remaining 0.2% stating they were bisexual (see Table 31). The sexual orientation in this iteration of instrument administration did correlate with the results of the partner's gender calculated at 100% male (see Table 32 and Table 33).

Table 31. Marital Status of Two-Week Retest Administration Group

		Frequency	Percent	Valid Percent	Cumulative Percent
	Single	66	15.2	59.5	59.5
Valid	Married	45	10.4	40.5	100.0
	Total	111	25.6	100.0	
Missing	System	322	74.4		
Total		433	100.0		

Table 32. Sexual Orientation of Two-Week Retest Administration Group

		Frequency	Percent	Valid Percent	Cumulative Percent
	Heterosexual	109	25.2	98.2	98.2
Valid	Bisexual	2	.5	1.8	100.0
	Total	111	25.6	100.0	
Missing	System	322	74.4		
Total		433	100.0		

Table 33. Partner Gender of Two-Week Retest Administration Group

		Frequency	Percent	Valid Percent	Cumulative Percent
Valid	Male	111	25.6	100.0	100.0
Missing	System	322	74.4		
Total		433	100.0		

Non-Hispanic, White women were the largest racial group in this administration as well at 56.8%, Hispanic women comprised 14.4% of the retest sample, Asian women represented 13.5%, black women made up 10.8% and Hawaiian/ Pacific Islanders were

less than one percent of the sample. 3.6 percent of women participating in the two-week retest marked their race as ‘other’ (see Table 34).

Table 34. Race of Two-Week Retest Administration Group

		Frequency	Percent	Valid Percent	Cumulative Percent
Valid	Non-Hispanic, White	63	14.5	56.8	56.8
	Hispanic	16	3.7	14.4	71.2
	Black or African American	12	2.8	10.8	82.0
	Asian	15	3.5	13.5	95.5
	Hawaiian/ Pacific Islander	1	.2	.9	96.4
	American Indian/ Alaskan Native	4	.9	3.6	100.0
	Total	111	25.6	100.0	
Missing	System	322	74.4		
Total		433	100.0		

Similar to the initial exploratory administration, a substantial majority of the women in the two-week retest sample had at least some college or higher. Both some college education and college graduate evenly made up 72.0% of the sample. 9.0% were or had been in graduate school. Only 4.5% had not completed high school and 14.4% had only completed high school (see Table 35). Of the women participating in the two-week retest, 83.8% had no formal medical health-related training and 16.2% were trained medically (see Table 36). The employment status demographic showed this group was predominantly employed at 40.5%. Just under a quarter (23.4%) of women in this sample were students, 22.5% were stay at home partners, 13.5% were unemployed and none were in the military (see Table 37).

Table 35. Educational History of Two-Week Retest Administration Group

		Frequency	Percent	Valid Percent	Cumulative Percent
Valid	Some High School	5	1.2	4.5	4.5
	High School Diploma	16	3.7	14.4	18.9
	Some College	40	9.2	36.0	55.0
	College Graduate	40	9.2	36.0	91.0
	Graduate School	10	2.3	9.0	100.0
	Total	111	25.6	100.0	
Missing	System	322	74.4		
Total		433	100.0		

Table 36. Medical Training of Two-Week Retest Administration Group

		Frequency	Percent	Valid Percent	Cumulative Percent
Valid	Trained	18	4.2	16.2	16.2
	Not Trained	93	21.5	83.8	100.0
	Total	111	25.6	100.0	
Missing	System	322	74.4		
Total		433	100.0		

Table 37. Employment Status of Two-Week Retest Administration Group

		Frequency	Percent	Valid Percent	Cumulative Percent
Valid	Employed	45	10.4	40.5	40.5
	Unemployed	15	3.5	13.5	54.1
	Student	26	6.0	23.4	77.5
	Stay at Home Partner	25	5.8	22.5	100.0
	Total	111	25.6	100.0	
Missing	System	322	74.4		
Total		433	100.0		

Over half (52.3 %) of the women participating in this initial instrument development administration stated they were spiritual and religious. Only 27.9% stated they were spiritual and non-religious and 19.8% denied being spiritual or religious (see Table 38).

Table 38. Spirituality of Two-Week Retest Administration Group

		Frequency	Percent	Valid Percent	Cumulative Percent
Valid	Spiritual, Non-Religious	31	7.2	27.9	27.9
	Spiritual, Religious	58	13.4	52.3	80.2
	Non-Spiritual, Non-Religious	22	5.1	19.8	100.0
	Total	111	25.6	100.0	
Missing	System	322	74.4		
Total		433	100.0		

As was the case in the initial exploratory administration of this instrument, a majority of participants in this administration had no children (52.3%). The percentage of women with children was inversely related to the number of children. Nearly one in five (19.8%) of participants had one child, 18.0% had two, 4.5% had three, 3.6% had four and 1.8% had five or more children (see Table 39). Similarly, 55.0% of the women had never been pregnant and 45.0% had been pregnant before (see Table 40). At the time the results of this study were collected, 13.5% of women were pregnant and 86.5% confirmed not being pregnant at the time of completion of this instrument (see Table 41).

Table 39. Number of Children of Two-Week Retest Administration Group

		Frequency	Percent	Valid Percent	Cumulative Percent
	0	58	13.4	52.3	52.3
	1	22	5.1	19.8	72.1
	2	20	4.6	18.0	90.1
Valid	3	5	1.2	4.5	94.6
	4	4	.9	3.6	98.2
	5+	2	.5	1.8	100.0
	Total	111	25.6	100.0	
Missing	System	322	74.4		
Total		433	100.0		

Table 40. Past Pregnancy History of Two-Week Retest Administration Group

		Frequency	Percent	Valid Percent	Cumulative Percent
	Pregnant Before	61	14.1	55.0	55.0
Valid	Never Pregnant	50	11.5	45.0	100.0
	Total	111	25.6	100.0	
Missing	System	322	74.4		
Total		433	100.0		

Table 41. Current Pregnancy Status of Two-Week Retest Administration Group

		Frequency	Percent	Valid Percent	Cumulative Percent
	Pregnant Now	15	3.5	13.5	13.5
Valid	Not Pregnant Now	96	22.2	86.5	100.0
	Total	111	25.6	100.0	
Missing	System	322	74.4		
Total		433	100.0		

RELIABILITY

Cronbach's Alpha

In the initial exploratory administration of this instrument, the internal reliability was calculated with a Cronbach's Alpha of 0.785. Though this is just slightly shy of the 0.80 goal for adequate reliability (Cronbach, 1951), results indicate that removal of a single item, Item 6, alone would increase the reliability to 0.806 (see Table 42). Of note, as per the above statistical calculations as well, only 35 items are evaluated in the reliability calculations as the two predictive validity items were not included.

Table 42. Item-Total Statistics of the Initial Exploratory Administration of this Instrument

	Scale Mean if Item Deleted	Scale Variance if Item Deleted	Corrected Item-Total Correlation	Cronbach's Alpha if Item Deleted
Q1	164.5520	299.838	.128	.787
Q2	164.2832	296.038	.220	.782
Q3	162.2652	288.692	.524	.771
Q4	162.5520	292.155	.388	.775
Q5	163.8925	302.945	.095	.788
Q6	165.0824	328.932	-.345	.806
Q7	163.1577	297.076	.209	.783
Q8	162.9964	295.737	.267	.780
Q9	163.2939	295.928	.291	.779
Q10	163.6416	300.310	.142	.786
Q11	162.9319	287.711	.458	.772
Q12	162.4444	286.823	.522	.770
Q13	162.6523	293.357	.373	.776
Q14	162.8208	288.284	.395	.774
Q15	164.1935	309.581	-.010	.792
Q16	162.8710	289.552	.459	.773
Q17	162.7957	291.753	.377	.775

Table 42 (continued)

Q18	162.9068	289.322	.451	.773
Q19	162.6129	284.231	.601	.767
Q20	163.2688	306.075	.087	.786
Q21	162.5986	289.306	.538	.771
Q22	163.2796	297.123	.265	.780
Q23	163.0143	295.820	.317	.778
Q24	163.3513	293.783	.335	.777
Q25	163.2545	293.140	.250	.781
Q26	163.2688	297.327	.211	.782
Q27	163.3118	301.726	.128	.786
Q28	162.4122	284.488	.469	.771
Q29	162.3620	281.009	.641	.765
Q30	162.0932	284.409	.579	.768
Q31	163.3477	301.386	.152	.785
Q32	163.6201	299.503	.210	.782
Q33	163.8029	311.691	-.043	.791
Q34	163.2222	296.094	.265	.780
Q35	163.1147	301.850	.154	.784

The two-week retest administration of this instrument resulted in a Cronbach's Alpha of 0.763. Removal of item 6 reflected the largest increase in internal reliability in the retest administration as well (see Table 43) to an increase to $\alpha = 0.787$. This approaches the preferred level of 0.80 in this retest administration (Cronbach, 1951).

Table 43. Item-Total Statistics of the Two-Week Retest Administration of this Instrument

	Scale Mean if Item Deleted	Scale Variance if Item Deleted	Corrected Item-Total Correlation	Cronbach's Alpha if Item Deleted
Q1_2	163.8702	276.175	.087	.768
Q2_2	163.4656	278.112	.070	.768
Q3_2	161.6718	259.099	.651	.743
Q4_2	161.8855	271.518	.268	.758

Table 43 (continued)

Q5_2	162.9618	269.791	.239	.759
Q6_2	164.3053	298.829	-.286	.787
Q7_2	162.3740	276.374	.132	.764
Q8_2	162.7863	264.646	.340	.754
Q9_2	162.7176	277.297	.127	.764
Q10_2	163.2824	279.266	.049	.769
Q11_2	162.2137	267.646	.398	.753
Q12_2	161.8473	265.884	.461	.750
Q13_2	162.0763	264.286	.436	.750
Q14_2	162.3664	264.634	.326	.754
Q15_2	163.4275	280.924	.025	.770
Q16_2	162.0840	264.416	.458	.750
Q17_2	162.0992	264.536	.373	.752
Q18_2	162.1450	266.202	.417	.752
Q19_2	162.0611	262.581	.486	.748
Q20_2	162.7176	283.204	-.004	.769
Q21_2	162.0076	267.977	.449	.752
Q22_2	162.5115	273.683	.216	.760
Q23_2	162.3130	265.617	.405	.752
Q24_2	162.6718	267.176	.332	.754
Q25_2	162.5344	263.835	.301	.756
Q26_2	162.5802	274.769	.144	.764
Q27_2	162.3893	270.947	.217	.760
Q28_2	161.8855	259.010	.441	.748
Q29_2	161.7481	258.682	.517	.745
Q30_2	161.5649	258.802	.544	.745
Q31_2	162.6031	273.057	.219	.760
Q32_2	162.8168	270.058	.257	.758
Q33_2	163.1832	287.612	-.103	.773
Q34_2	162.4198	275.153	.176	.761
Q35_2	162.5344	268.512	.283	.757

Two-Week Test-Retest Reliability

The two-week retest reliability was calculated by calculating a Pearson's correlation of the total means of the results at the initial administration and the results at the two-week retest administration. The results indicate a large correlation of $r = 0.821$ that is statistically significant ($p < 0.01$) (see Table 44).

Table 44. Two-Week Test-Retest correlation

		TotalTime1	TotalTime2
TotalTime1	Pearson Correlation	1	.821**
	Sig. (2-tailed)		.000
	N	284	111
TotalTime2	Pearson Correlation	.821**	1
	Sig. (2-tailed)	.000	
	N	111	111

**. Correlation is significant at the 0.01 level (2-tailed).

CHAPTER SUMMARY

This chapter presented the results of the instrument development and the exploratory administration of this instrument, including sociodemographic characteristics of the samples. The implications of these results are discussed in the next chapter.

Chapter Five: Discussion

An instrument measuring the process of informed decision making about prenatal genetic screening was developed as the focus of this study. Through development and implementation of this instrument in the clinical setting, health care providers should be able to ensure expectant women are making informed decisions regarding receiving or refusing prenatal genetic screening, thus, promoting autonomy and healthcare decisions more in line with each individual's beliefs and values. A discussion of the results of the instrument development and the exploratory administration of this instrument are presented in this chapter. Results are first discussed in light of the conceptual framework for the study. This discussion is followed by a more detailed interpretation of the findings, conclusions from the instrument development process, policy implications, and suggestions for future research.

RESULTS OF STATISTICAL ANALYSIS

The IDM-PGS instrument is a general measure of how well informed an expectant woman is regarding making decisions in prenatal genetic screening. The instrument was developed based on review of the literature. Thorough psychometric testing including content validity analysis, cognitive interviewing, and readability analysis, as well as exploratory administration for criterion-related validity, construct validity, factor analysis and reliability was performed.

Item Formulation

Item formulation for the IDM-PGS was based on a variety of sources in pertinent literature and expert feedback. Though initial item formulation was highly inclusive, limitations to item inclusion in the final instrument were made after content validity

analyses and cognitive interviews. These limitations resulted in a loss of inclusion of certain constructs of the adapted model for IDM-PGS**.** Though the six major factors of decision context, decision maker, moral judgments, emotional responses, cognitive appraisals and other factors were all still addressed within the remaining items, a number of the sub-constructs were no longer measured. Constructs such as a decision maker's desire for control, and the moral judgment of the technological imperative were no longer measured as a result of content validity and cognitive interviewing. By removing these items, the instrument was no longer able to measure aspects such as personality characteristics of the decision maker, or specific indicators of social significance of the results. However, the statistical results of this instrument development suggest these changes are not substantial, and thus this instrument still adequately measures IDM-PGS.

Situation-based variations in the context of measuring IDM-PGS must be considered. A number of aspects of the model cannot be measured with universal items that would adequately assess the construct across diverse groups (Bekker et al., 1999; Charise et al., 2011; Tversky & Kahneman, 1981; Weiner, et al., 2010; Weiner, et al., 2013). For example, cost associated with conducting a prenatal genetic screen can vary from situation to situation based on an individual's insurance or lack thereof, governmental policies or funding, et cetera. Thus, a universal item or set of items to measure cost associated with screening would be shortsighted. The responses to these items have the potential of varying greatly based on the context of the situation of the respondent, measuring the level of informed decision-making related to the issue of cost would be impossible.

Similarly, availability of the resources required in prenatal genetic screening, such as obstetricians or laboratories, varies in diverse settings. In some regions or countries, access to prenatal genetic screening may be available to all citizens at no cost and at

every physician's office; however, in other locales, it may be the case that individuals must have insurance and access to specific physicians to access prenatal care and thus prenatal genetic screening. The varied scenarios require distinct items to measure this access; no universal item can measure the variation in the locations described above. Even constructs such as time involved in conducting the prenatal screen can affect an individual's informed decision-making and thus may need to be considered in measurement of IDM-PGS; however, because time involved in measurement can vary greatly between settings, and even within individual situations within a single setting, no single item measuring time can be universally included in this instrument. Thus, modifications to items, or the addition or removal of items may become necessary for adequate application of the IDM-PGS instrument in diverse settings. As the adapted model indicates, modifications to the instrument may need to encompass items that are unique to the situation, such as cost of the screening, availability, or time involved in getting the screening.

Cultural modifications may also need to occur. For example, items assessing social significance of results of prenatal genetic screening may need to be added as the social impact of results can differ across diverse cultural backgrounds. Though some cultures may minimize the significance of results, other cultures may find results negative or even go so far as to stigmatize the fetus or expectant woman. Thus, modifications to the instrument to assess for cultural or societal norms should be considered for unique cultural scenarios. Other cultural modifications include accounting for shared decision-making characteristics in various cultures. If an individual is from a culture that relies heavily on shared decision-making, the IDM-PGS instrument may need to be administered amongst all parties involved in the decision-making process to ensure that every individual who is entitled to contribute to the decision-making process is

adequately informed (Dugas et al., 2012; Griffith & Tengnah, 2013; Lipkin; 2013; Moulton & King, 2010). Thus, the shared-decision is based on the thoughts and conclusions of adequately-informed individuals.

A healthcare provider should also be responsible for assessing characteristics of the individual with whom the instrument is being administered. As delineated in the adapted model, various factors can affect the decision maker, such as health literacy, desire for control, propensity towards anxiety or individual interpretation of the health care provider's role. If the health care provider determines the situation requires a more thorough assessment of an individual prior to or simultaneous with the administration of the IDM-PGS instrument, assessments and modifications may need to occur to add items to assess these factors.

Item-by-Item Readability

With complex technological advancements in science, common measures for readability, despite being the current gold standard in item development, are found to be limited in addressing complex topics. As such, terms like 'prenatal genetic screening' cannot be short-handed by verbiage with fewer syllables or words. In situations with complex or technical terminology, simplification of terms may actually remove autonomy and reduce informed decision-making through misrepresentation of information. These situations call on the application of linguistic models and constructs to enhance understanding of complex terminology (Zarcadoolas, 2011). For situations such as the IDM-PGS instrument development, the SMOG readability levels were very high, denoting individuals requiring graduate level education for adequate understanding. However, this is arguably linked to the highly technical language of concepts involved. The cognitive interviews were conducted with a majority of individuals who did not have

graduate education yet, based on the results of the cognitive interviews, understanding of the instrument was still present. The technique of engaging the reader to most obviously note the most important message or most salient point in each item in simple syntax and vocabulary was used. Thus, despite a decrease in the SMOG readability when the term 'prenatal genetic screening' was changed to 'PGS' and 'informed decision making' was changed to 'IDM', these key terms were kept in the items. In an effort to reduce SMOG readability levels the salient point of the item, or the question that was being asked, was put in more simple language.

Content validity.

Content validity analyses often require multiple iterations through expert panelists. In order to achieve adequate content validity per the universal approach, the S-CVI scores should be 0.70 or higher (Polit, Beck & Owen, 2007). In this research study adequate content validity for the IDM-PGS instrument required two rounds of analysis with the expert panel and the subsequent removal of 48 items. Thus, upon completion of the content validity analyses, the revisions of the IDM-PGS instrument resulted in a valid final instrument that includes 37 items with a content validity index that was adequate at 0.78. As Polit, Beck and Owen (2007) state, many instrument developers do not discuss how and if content validity was analyzed including the two instrument development papers discussed in chapter two (Michie et al., 2002; Van den Berg et al., 2008). However, Schulman-Green et al. (2012) and Weber et al. (2011) both discuss their content validity analysis. Schulman-Green et al. (2012) found, on a 13-item scale measuring knowledge of care options including curative, palliative and hospice care, a S-CVI of 0.869 on the first round of expert questions. Their second round was sent to different experts and returned a CVI of 0.852. Both values were well over an acceptable

cutoff for content validity analysis. Weber et al. (2011) had a staggering 0.99 content validity on a 64-item scale measuring parenting intention, decision-making, and expectations, also clearly an adequate value. Though the results of the IDM-PGS scale did not render a CVI as high as other similar scales on information about decisions published in the literature to date such as the Knowledge of Care Options instrument (Schulman-Green et al., 2012) and the Parenting Paradigms Scale (Weber et al., 2011), the CVI of 0.78 is still considered adequate using the universal approach to measure CVI. The results of scales revolving around well-researched areas of the literature, such as parenting and knowledge of care, can explicably have substantially higher CVI values as a result of longstanding research, universal guidelines and more widely accepted practice.

Cognitive Interviews.

Cognitive interviews were conducted to measure if individuals in the population of interest were able to adequately understand the instrument items in their intention and to improve the design of the instrument (Knafl et al., 2007). Participants of the cognitive interviews tended towards an accurate understanding of the intended meaning of the items in the IDM-PGS instrument. However, of the eight items that were inaccurately interpreted, only one participant misinterpreted any one given item. No consistency across participants in misinterpretation was seen during the cognitive interviews. These results imply that overall the individual items of this instrument are able to assess accurately the construct they are aimed at assessing. The inaccurate interpretations represent a misunderstanding of the intent of the items to assess diagnosis versus risk assessment. Additionally, of these eight incorrect interpretations, two instances were clarified by rereading the item that was initially interpreted incorrectly with an emphasis on the terms in the item that are meant to distinguish between the diagnostic or risk

assessment. When asked again with emphasis, both participants responded with an accurate interpretation. The high degree of accurate interpretation also supports the earlier discussion of the readability. Though readability is high, the accurate interpretation during cognitive interviews represents acceptable readability in this instrument.

Based on findings from the cognitive interviews, a few adaptations were made prior to the exploratory administration of the IDM-PGS. The exploratory administration was conducted using an online questionnaire platform and not an interview-based administration, thus key terms in each item that assist the reader in correctly interpreting the intent of the questions towards assessing diagnosis versus risk assessment were highlighted by bolding and underlining. The goal of this added emphasis was to reduce the misinterpretation of the item by highlighting key distinguishing terms. The cognitive interview process further enhanced the instrument's ability to measure IDM-PGS. Ensuring the instrument is adequately interpreted amongst the population of interest strengthens the ethical considerations made in developing the instrument. By increasing adequate interpretation, the autonomy of the decision-making process is enhanced (Atkins, 2000; Beauchamp & Childress, 1979; Chervenak & McCullough, 2011). The decisions are subsequently more patient-focused as opposed to health care provider-focused allowing the decision to be informed based on the values and beliefs of the patient (ACOG, 2009; Beauchamp, 2011; Bekker, 2003; McCoy, 2008).

Sociodemographic Characteristics.

In the initial exploratory administration of this instrument, data from 433 participants were collected, which was well over the 298 participants deemed via power analysis as required for adequate instrument development for a 37-item scale. With 111

participants, there was adequate response to the two-week retest for reliability. In both samples, a well-dispersed age range allowed for a representative sample of the general population. Similarly, the samples were predominantly heterosexual, though homosexual and bisexual women were included. Race, education, employment status, spirituality, pregnancy status and number of children of expectant women in these samples also mirrored what would be expected in the general population making the results more translatable across broader, potentially nationwide, samples.

Criterion–related validity.

Criterion-related validity measures relevance and usefulness by describing how well the results of a measurement are applicable in a real world outcome. In the IDM-PGS instrument, the administration of two items developed to explore the predictive validity of a survey respondents planned outcome measured criterion-related validity (Field, 2013). The initial exploratory administration resulted in a highly significant but low strength correlation in the predictive values and total time. Despite predictive validity being used in previously published instruments regarding similar concepts (Marteau et al., 2011), Field (2013) suggests that assessing predictive validity is often impractical as objective criteria measuring a predicted action may not exist. However, results from the administration of the IDM-PGS indicate the measure used in this study may predict participant behavior. Such an interpretation would be enhanced in further study with additional specific objective criteria.

Construct validity.

The known-groups method and exploratory factor analysis were used to measure construct validity of the IDM-PGS in this study. Bivariate correlation between the values of the overall mean of the items with the demographic characteristics defining the groups

of interest was used to calculate the known-groups method. The groups include medically trained individuals versus those who are not medically trained, individuals who are currently pregnant and those who are not pregnant, and women who have been pregnant in the past and those who have never been pregnant. Of the three known-group comparisons measured to calculate construct validity, only one difference between groups was statistically significant. Decision making can be impacted by various contextual variables including an individual's familiarity with the decision, their health literacy, cognitive load, etc. (Bekker et al., 1999; Charise et al., 2011; Tversky & Kahneman, 1981; Weiner, et al., 2010; Weiner, et al., 2013). Those who were medically trained versus lay-persons were found to have significantly higher IDM-PGS with a *p*-value at 0.014. Assuming individuals with medical training have higher levels of informed decision making regarding any medical decision for which they would have received training is not implausible. In an ideal situation, more medical training would lead to higher levels of IDM-PGS. The negative correlations with each of the other groups represents the item coding (these items were reverse-coded) and thus negative correlations actually represent an increase in IDM-PGS for those with medical training.

However, of note, participants who claimed to be trained medically identified their training with descriptions that this researcher feels are not representative of the goal of this demographic quality. For example, a number of participants described formal health training as having been trained in Cardiopulmonary Resuscitation (CPR). Though not an inaccurate description of having formal medical training, CPR training does not encompass training related to genetic screening or testing as was the intent in that demographic characteristic. Thus, the impact of medical training on IDM-PGS should be reanalyzed in the future with a more specific definition or example of what is meant by "formal medical training" such as specific courses taken in colleges and universities. In

these future studies of construct validity, re-wording the item used to measure if a participant is adequately medically trained regarding genetic screening or testing is vital to creating a distinction among groups and thus support construct validity.

Of the three known-groups none had a statistically significant correlation in the two-week retest administration of the IDM-PGS. The lack of significance is not an unexpected result based on the substantially smaller sample size likely resulting in a loss of power and thus a lower likelihood of finding significance (Field, 2013).

No significant increase in IDM-PGS was found within the groups of women who had been pregnant before or within the group of women who were pregnant at the time of completion of the measure. The group of women who had been pregnant in the past versus those who had never been pregnant narrowly missed a significant correlation with IDM-PGS with a *p*-value of 0.051. Though not significant at $p < 0.01$ or $p < 0.05$, instrument development researchers have been known to use more lenient levels of significance and at times use a level of significance set at $p < 0.10$, in which situation this value would in fact be significant. This result suggests the idea that individuals who have been pregnant before may have multiple children, may have had prior experience with prenatal genetic screening multiple times and thus have a greater knowledge of this process. A possible explanation of these results also includes individuals who are currently pregnant may not have been far enough along in their pregnancies to have considered or potentially even heard of prenatal genetic screening. These findings are further supported by a previous study suggesting familiarity with a decision being an influencing variable on decision-making (Bekker et al., 1999) as women with multiple previous pregnancies can be assumed to have more familiarity with the decision.

These group differences support construct validity. Testing known group differences is to test the hypothetical or assumed contrast between groups of interest. In

this situation, one would hypothesize individuals with medical training, those who have had children previously and those who are pregnant would have more knowledge and thus make more informed PGS decisions than those without. The results confirmed the hypothesis for individuals with medical training having higher levels of IDM-PGS. Though the results could not be labeled as significant for those individuals who have been pregnant in the past, a $p = 0.051$ approaches significance and thus provides evidence that there likely is a difference in the groups based on that criterion.

Exploratory factor analysis.

Data from the exploratory administration of the IDM-PGS of 433 women were factor analyzed and this analysis resulted in the 37 items loading on 10 factors. In this IDM-PGS instrument development items' loading on 10 factors does not match the adapted conceptual model as six factors would be ideal to parallel the six factors as delineated in the conceptual framework; however, when further analyzed, the 10 factors could be grouped in such a way that the items loaded similarly to the conceptual framework's six factors.

The items that loaded on factors 1 and 2 represented the largest portion of the items on the IDM-PGS instrument. Nineteen items fell within this first and second factor. Since these items did not align with any one specific context on the conceptual framework, the items in these factors would have to be divided amongst the various components of the adapted model and could not mirror the conceptual model in the intended way. In these overly inclusive factors, the variations within each item linking it to the factor that it was created in an attempt to measure was likely too subtle for the factor analysis software to distinguish differences and thus these factors should not be taken as a unique factors (De Vellis, 2012).

Items that loaded on factors or components 3, 4, 5 and 7 were intended to measure decision context. These four components represent nine of the items, a relatively large proportion for one contextual group. Decision context covers a variety of facets that the instrument developer, expert panelists and cognitive interviewees agreed were very important so the heavy weight on this factor is acceptable, or justified. These eight items when grouped could be considered a sub-scale within the IDM-PGS.

Factor 6 represented two items that evaluated the meaning of positive results. This factor seems to align with the contextual model under the moral judgment group since a measure of positive results can play into the moral judgment involved with the process of making an informed decision.

Both of the items that loaded on factor 8 were aimed specifically at measuring cognitive appraisals by measuring risk. Cognitive appraisals are a key factor in the IDM-PGS process that a decision maker would use. Though two items alone does not constitute enough to provide for a robust subscale measurement, these two items possibly combined with some of the items in the remaining factors could potentially constitute such a scale. This warrants further study.

Factors 9 and 10 loaded with items that were intended to measure the “other factors” concept within the adapted model. Three items loaded on this factor measuring subscales named "legal protection of fetus" and "availability of results". This factor encompasses a wide range of topics as it covers extraneous factors, framing issues, cognitive load, availability and cost. Thus, a three-item subscale of this factor would not provide a feasible measurement of a specific construct.

The scree plot analysis also did not recover similar results as the factor analysis on the exploratory implementation of the IDM-PGS. The scree plot showed between three and four factors, substantially fewer than the 10 found by the factor analysis. The

scree plot results were further interpreted from the conceptual model and are thus not likely as accurate of an analysis as the factor analysis above. Regardless, a Varimax or Orthogonally rotated factor analysis remains the gold standard for factor analysis, so results of the scree plot are of lesser importance than the factor analysis. Further study to reduce the ambiguity found in the subscale analyses of this factor analysis would include conducting an item-by-item correlation. The goal would be to eliminate the vagueness and ambiguity of why the items may have loaded the way they did. The items may also need to go back to content experts for further validation.

Reliability

The reliability of this instrument was seen in the high Cronbach's alpha (α = 0.785) calculated at the initial exploratory administration of instrument implementation. Removal of a single item would elevate the Cronbach's alpha to the suggested cutoff of 0.80. This item represents the measure of if an individual would wait until birth to get genetic screening. Though the reason removing this item improves reliability is not clear. Even the two-week retest administration resulted in a reliability of α = 0.765. Though not necessarily low reliability, the results of the two-week retest administration data having a lower Cronbach's alpha is not an unexpected finding due to the substantially lower number of participants in the retest sample. The high Cronbach's alpha in this study represents good internal consistency and thus an independently reliable instrument when used with this sample on a single administration.

Additionally, when comparing the initial administration with the two-week test-retest reliability, this resulted in a highly significant and strong correlation in the two iterations of this instrument implementation. These results indicate a good correlation of

the reliability between the test and the retest. Thus, this result represents low measurement error, which indicates good internal consistency of the instrument (Field, 2013).

ETHICAL IMPLICATIONS

Given the calculated psychometric characteristics of the IDM-PGS, using the instrument developed in this study will allow health care providers to treat prospective parents more ethically by ensuring the decision made about prenatal genetic screening is in line with the individuals' values and beliefs. As Faden and Beauchamp (1986) described, numerous factors affect informed consent. Such factors include autonomy, competence, coercion, manipulation and persuasion. The developed instrument explores various factors, including decision context, decision maker, moral judgments, emotional responses, cognitive appraisals and other factors to determine if an individual has adequate IDM-PGS. Through the application of this instrument in clinical settings, healthcare providers will act accordingly with the four ethical pillars of beneficence, non-maleficence, justice and autonomy (Beauchamp & Childress, 1979). Using the IDM-PGS instrument maintains beneficence by continuing to promote the patient's best interest and thus enhance good while removing as much harm as possible (Chabin, 2011; Clarke, 1993; Lo, 2000; Noble-Adams, 1996).

Similarly, using this instrument allows for non-maleficence by continuing to avoid harm (Clarke, 1993; Lo, 2000; Smith, 2005). Higher levels of IDM-PGS facilitate healthcare providers in guiding patients towards decisions most in accordance with their patient's personal values and beliefs. By acting in accordance with one's values and beliefs, a patient is less likely to inflict emotional harm unto themselves.

Though not a specific issue due to abundance of resources for prenatal genetic screening in the United States, justice is also promoted by implementing the IDM-PGS in areas where limited resources exist. The IDM-PGS instrument could enhance justice by avoiding frivolous use of resources for individuals who would prefer not to have PGS or subsequent further testing (Beauchamp & Childress, 1979; Gillon, 1994). Further study could be done to determine that the IDM-PGS can be easily used by anyone from any race, socioeconomic level, or educational level.

A clear enhancement of autonomy exists by the use of the IDM-PGS instrument as implementation of this instrument is aimed at ensuring informed decisions are made. The IDM-PGS was developed in an attempt to ensure decisions were made in accordance with the decision-maker's values and beliefs in regards to prenatal genetic screening. Thus, implementation of this instrument enhances prioritization of the patients' values and beliefs and thus their autonomy (Atkins, 2000; Beauchamp & Childress, 1979; Chervenak & McCollough, 2011).

Implementation of the IDM-PGS promotes each ethical pillar. Healthcare providers are held to a high ethical standard. Enhancing beneficence, non-maleficence, justice and autonomy in every healthcare situation is necessary for healthcare providers to diligently care for patients. The IDM-PGS promotes ethical healthcare and should thus be implemented widely in clinical settings. Similar to the four ethical pillars Beauchamp and Childress (1979) presented, the Belmont report (Department of Health, Education and Welfare, 1979) discusses three ethical principles. These include respect for persons, beneficence and justice. Beneficence and justice overlap almost identically to Beauchamp and Childress' (1979) ethical principles. However, the ethical principle of respect for persons contains features of autonomy and adds the concept of protecting individuals with diminished autonomy. In situations of shared decision making or situations in which

autonomy is diminished in emergent circumstances, healthcare providers must maintain the ethical principle of respect for persons.

Ethical issues arose in discussions with content validity experts related to whether or not the IDM-PGS should cover more controversial topics, such as miscarriage and abortion as a result of invasiveness of further testing or selectively due to results of genetic analyses. Though prenatal genetic screening itself has very low risk, implications of results of this screening can cause an expectant woman to pursue further more invasive testing and intervention. This further testing itself carries risk as well as may diagnose genetic disease that some expectant women may deem as reason to abort. Both of these potential outcomes are important issues to consider in pursing genetic analysis of any kind. However, for purposes of this research related to prenatal genetic screening, the researcher - with feedback from expert panelists - concluded that an item discussing potential further testing and intervention should be kept vague. Expert panelists suggested not explicitly mentioning the risk of miscarriage or the potential abortion as an intervention. From an ethical perspective, not mentioning these outcomes should not impact the results of the IDM-PGS as an item measuring understanding of further exploration and potential intervention exists.

A further complication of ethics involved in prenatal genetic screening is the consideration of the fetus as a unique and separate individual that the expectant woman. Fortunately the decisions involved in PGS are mostly low risk and low consequence decisions. The decisions of greater ethical concern are those made subsequent to PGS, such as prenatal genetic testing, amniocentesis, chorionic villus sampling, fetal blood sampling and even abortion. However, since deciding on pursing PGS is a precursory decision to the more invasive testing, healthcare providers must stress the importance of universally ethical decision-making. The greatest ethical considerations occur when

conflict arises between maternal rights and fetal rights. At times ethical pillars such as beneficence and autonomy contradict one another. For example, in situations such as alcohol consumption during pregnancy, the most beneficent action would be for the expectant woman not to drink and thus cause the least harm to the fetus. However, the expectant mother, by the ethical principle of autonomy, is entitled to act at her own discretion and may choose to drink when she is pregnant. Arguably, a situation in which alcohol consumption during pregnancy is occurring, the greatest good and least harm would occur by prohibiting alcohol consumption by the expectant mother. However in situations such as prenatal genetic screening and testing the option for the greatest good is not quite as clear. The most ethically inline action varies in individuals with diverse values and beliefs. An expectant woman who would intervene and treat her fetus for a genetic anomaly may benefit most from risky, invasive genetic testing. On the contrary, individuals with a more fatalistic or religiously-driven outlook may have the most ethical experience with no intervention and allowing 'destiny' or 'God's will' to happen.

Moore (2000) and Lowery (2004) suggest healthcare providers are responsible for promoting the most ethical care by promoting discussions with their patients, providing education and being aware of laws and community practices. Though Lowery (2004) focuses on cancer genetics, overlying concepts regarding impact of the law on genetic testing decisions remain universal. All genetic screening and testing results can lead to discrimination but state and federal laws such as the Genetic Information Non-Discrimination Act of 2003 (S. 1053) aim to prohibit employment and insurance discrimination. As with genomic science in general, this aspect of genomic science is rapidly expanding. Further research will be required to determine ethical processes and outcomes. Instruments such as the IDM-PGS can be used in clinical practice to help keep

ethical issues such as these in the forefront of healthcare providers' care, especially as more applications of genetics and genomics become available.

POLICY IMPLICATIONS

A key point of consideration is policy-level implementation for screening-based assessments such as this instrument. Policy-level changes in healthcare are necessary for population-wide improvements. As genetic technology proliferates within the healthcare field, many population-wide policies and procedures are being recommended, and at times even implemented, through both governmental and scientific organizations. The California Prenatal Screening Program (CPSP) implemented through the California Department of Health is one such population-wide program (CDPH, 2010). The CPSP requires prenatal genetic screening services be offered to all women by health care providers. This program has outlined for providers best practice for avoiding miscommunication and misinformation of genetic assessment technology (CDPH, 2010; Mikail, 2008). Further research into the potential effectiveness of this program, if implemented nationally, should be conducted before the actual implementation occurs. However, the potential to mirror the California program nationally would allow a smooth transition for increased prenatal screening population-wide.

In conjunction with the implementation of a nation-wide prenatal screening program, promoting autonomy and enhancing informed decision-making should be implemented population-wide as well. Though small-scale efforts to ensure informed decision-making occurs regarding prenatal genetic screening are necessary within programs or individual states, a more global scale policy-level change allows larger scale impact and improvements. Thus, if federal-level policy changes are implemented for

ensuring informed-decision making, populations of expectant women will be more likely to make well-informed decisions in-line with their values and beliefs.

Additionally, provided that this instrument has a focus on prenatal genetic screening - an issue affecting all individuals who are or who may become pregnant and their families - the potential impact of this study is very widespread. The potential to touch nearly every individual in the country adds gravity to the impact factor of this research and thus must be considered by policymakers as a means by which to improve healthcare autonomy and subsequently outcomes. It is the suggestion of this researcher that this instrument be added to basic prenatal genetic procedures as a means to ensure that all expectant women have adequate informed decision making about prenatal genetic screening so that their decisions can be well-informed and in-line with their values and beliefs.

RECOMMENDATIONS AND IMPLICATIONS FOR FUTURE RESEARCH

The results of this study show that the IDM-PGS has adequate reliability and validity to be utilized for application in real world scenarios with similar populations of expectant women. For application in real life situations, this instrument should be administered by health care providers to patients considering prenatal genetic screening. The administration of this instrument would allow health care providers to ensure expectant women have been adequately informed prior to making a decision for or against proceeding with prenatal genetic screenings. If the results of the implementation of this instrument in a clinical setting reflect an individual with low IDM-PGS, a healthcare provider can intervene and provide more thorough teaching about prenatal genetic screening to enhance informed decision-making.

Though this study was not administered in a clinical setting for purposes of this research, future study in a clinical setting is recommended. In this study, retrieval of a large sample size quickly, to allow this research to be completed prior to major advancements in the rapidly changing technology was necessary. Various online and clinical resources and an open discussion with the expectant woman can improve IDM-PGS. Further application of the IDM-PGS in research with expectant women in diverse clinical settings will also contribute to the psychometric soundness and stability of the instrument. These further studies may also include nursing interventions in which clinicians provide information and decision tools to assist expectant women and their partners in making informed decisions.

As discussed earlier, modifications to this instrument may be required to account for cultural, societal, geographical, or other variations in settings that can impact IDM-PGS (Bekker et al., 1999; Charise et al., 2011; Tversky & Kahneman, 1981; Weiner, et al., 2010; Weiner, et al., 2013). A specific area of future study would be to explore the religiosity piece further. Religion plays a large role in decision-making in many healthcare settings, especially in prenatal situations and genetic information seeking. Further research exploring the impact of religion on decision-making can help guide care and clinical application. As can be expected with any modifications to an established instrument, psychometric properties would have to be reassessed to ensure adequate reliability and validity in the modified instrument with the specific sample.

In addition to application in clinical settings, the results of this study open a plethora of implications for future study. Though expectant women remain the primary audience for this instrument, in an effort to promote patient-focused decision-making (ACOG, 2009; Beauchamp, 2011; McCoy, 2008), in culturally diverse settings, the IDM-PGS instrument can still be administered. In those cultures that tend towards group or

patriarchal decision-making, this same instrument could be adapted for administration to the decision maker, or makers, involved in prenatal genetic screening decision-making to assess IDM-PGS (Epstein & Gramling, 2013; Ho, 2008).

The data received through these two administrations of this instrument can also be further analyzed to provide greater information not addressed by the limited number of research questions in this research study. Due to the various demographic characteristics being assessed, the data can be analyzed for variations in results based on these demographic qualities. For example, further assessment of differences in sexuality, race, education, employment status, spirituality, pregnancy status and number of children as they relate to IDM-PGS can provide great insight to guide future research. If results reflect differences in groups, further research exploring where these differences arise could be necessitated.

The IDM-PGS instrument is highly specific towards the goal of measuring informed decision-making in those expectant women being offered or interested in pursuing prenatal genetic screening. However, with minor adjustments this instrument can very easily be applied to prenatal genetic testing, such as amniocentesis or chorionic villus sampling. Even as our genetic technologies advance, the more minimally invasive prenatal genetic diagnosis can also be assessed with minor modifications to this instrument.

Limitations

As previously discussed, the limitations of this study are acknowledged as:

1. A convenience sample from the population of interest was recruited; thus, results cannot be generalized to all women. Despite a diverse group of participants, the nature of a convenience sample indicates

lack of generalizability. In this situation, the Qualtrics' panel recruitments techniques may have set up limitations to target audiences and thus reduced selection from the population as a whole.

2. All data are self-reported and are subject to participant bias. Participant bias can arise in various situations. For example, despite cognitive interviewing to enhance construct validity, the potential for misinterpretation remains.
3. Internet-based research requires the participant to have access to the Internet. This limitation feeds into the first limitation in that the measure does not reflect the entire population as a result of elimination of those potential participants without access to the Internet. Thus, generalizability of the instrument is limited.

Despite these limitations, the findings from this study provide evidence that this tool can be used in research and clinical settings as a measure of informed decision-making regarding prenatal genetic screening. The psychometric evaluation of this study indicates that this developed instrument has adequate reliability and validity and is thus an appropriate measure of IDM-PGS.

CONCLUSIONS

In an effort to increase the informed decision-making of expectant women considering prenatal genetic screening, an instrument measuring the process of informed decision-making about prenatal genetic screening was developed in this study. Psychometric evaluation and implementation of this instrument show adequate validity and reliability for use in additional studies and in clinical practice. Thus, these results allow for the broader implementation of this instrument to measure IDM-PGS in

expectant women in clinical settings thereby potentially enhancing autonomy and improving health outcomes that are in line with the decision maker's values and beliefs. The potential wide-spread implementation of this instrument in clinics as well as research settings, and potentially even policy-level changes will allow this instrument to have a much larger population-wide impact.

APPENDIX A – EXCERPT FROM INFORMED DECISION-MAKING IN PRENATAL GENETIC SCREENING QUESTIONNAIRE

Informed Decision-making in Prenatal Genetic Screening Questionnaire

Instructions for participants:
Answer the questions by selecting the choice that corresponds to your response.

For purposes of this exercise, assume you are a pregnant woman who is being offered prenatal genetic screening. *Prenatal genetic screening* is a **non-diagnostic blood test** on a pregnant woman to check the risk that the unborn baby will have a genetic disease such as an aneuploidy, like Down syndrome, or neural tube defects, like spina bifida.

1	This prenatal genetic screen is to **diagnose** if my unborn baby has a genetic problem.
	Strongly Disagree 7 6 5 4 3 2 1 Strongly Agree

2	Prenatal genetic screening **cannot diagnose** if my unborn baby has a genetic problem.
	Strongly Disagree 7 6 5 4 3 2 1 Strongly Agree

3	Prenatal genetic screening is to tell me if my unborn baby **might** have a genetic problem.
	Strongly Disagree 7 6 5 4 3 2 1 Strongly Agree

4	This prenatal genetic screen checks my unborn baby's DNA for the **risk** of a genetic problem.
	Strongly Disagree 7 6 5 4 3 2 1 Strongly Agree

APPENDIX B – DEMOGRAPHIC DATA SHEET

D1 Gender:
- ❍ Male (1)
- ❍ Female (2)

D2 Age:
- ❍ 18 (1)
- ❍ 19 (2)
- ❍ 20 (3)
- ❍ 21 (4)
- ❍ 22 (5)
- ❍ 23 (6)
- ❍ 24 (7)
- ❍ 25 (8)
- ❍ 26 (9)
- ❍ 27 (10)
- ❍ 28 (11)
- ❍ 29 (12)
- ❍ 30 (13)
- ❍ 31 (14)
- ❍ 32 (15)
- ❍ 33 (16)
- ❍ 34 (17)

D3 Marital Status:
- ❍ Single (1)
- ❍ Married (2)
- ❍ Divorced/ Separated (3)
- ❍ Widowed (4)

D4 Partner's gender:
- ❍ Male (1)
- ❍ Female (2)

D5 Race
- ❍ Non-Hispanic, White (1)
- ❍ Hispanic (2)
- ❍ Black or African American (3)
- ❍ Asian (4)
- ❍ Hawaiian/ Pacific Islander (5)
- ❍ American Indian/ Alaskan Native (6)
- ❍ Other (7)

D6 Ethnicity:

D7 Employment Status:
- ❍ Employed (1)
- ❍ Unemployed (2)
- ❍ Student (3)
- ❍ Military (4)
- ❍ Stay at home partner (5)

D8 If applicable, occupation:

D9 Spirituality:
- ❍ Spiritual, non-religious (1)
- ❍ Spiritual, religious (2)
- ❍ Non-spiritual, non-religious (3)

D10 If applicable, religion:

D11 Education
- ❍ Some high school (1)
- ❍ High school diploma (2)
- ❍ Some college (3)
- ❍ College graduate (4)
- ❍ Graduate school (5)

D12 Have you had any formal medical health-related training (i.e. nursing student, nurse, or physician)?
- ❍ Yes (1)
- ❍ No (2)

D13 If yes, please describe your formal medical health-related training:

D14 How many children do you have?
- ❍ 0 (1)
- ❍ 1 (2)
- ❍ 2 (3)
- ❍ 3 (4)
- ❍ 4 (5)
- ❍ 5+ (6)

D15 Have you ever been pregnant before?
- ❍ Yes (1)
- ❍ No (2)

D16 Are you pregnant now?
- ❍ Yes (1)
- ❍ No (2)

D17 If yes, how far along are you in your pregnancy?

D18 How many siblings do you have?
- ❍ 0 (1)
- ❍ 1 (2)
- ❍ 2 (3)
- ❍ 3 (4)
- ❍ 4 (5)
- ❍ 5+ (6)

D19 What is the first letter of your birth month?
- ❍ J (1)
- ❍ F (2)
- ❍ M (3)
- ❍ A (4)
- ❍ S (5)
- ❍ O (6)
- ❍ N (7)
- ❍ D (8)

D20 How tall are you?

Feet (1)

Inches (2)

D21 Do you or does anyone in your family have any genetic condition?
- ❍ Yes (9)
- ❍ No (10)

D22 If applicable, what genetic condition do they have?

APPENDIX C – PERMISSION TO REPRINT FIGURE 1

APPENDIX D – PERMISSION TO REPRINT FIGURE 3

APPENDIX E – PERMISSION TO REPRINT FIGURE 4

References

Abramsky, L., & Chapple, J. (1994). *Prenatal diagnosis: the human side*: Chapman & Hall.

ACOG committee on practice bulletins (2007). ACOG Practice Bulletin No. 77: Screening for fetal chromosomal abnormalities. *Obstetric Gynecology, 109* (1), 217-227.

ACOG Committee Opinion No. 296: First trimester screening for fetal aneuploidy. (2004) *Obstetrics & Gynecology, 101*, 215-217.

ACOG Committee Opinion No. 439: Informed consent. (2009). *Obstetrics & Gynecology, 114* (2 Pt 1), 401-408. doi: 10.1097/AOG.0b013e3181b48f7f

AHRQ. (n. d.). Patient Decision Aids. *US Department of Health & Human Services.* Retrieved from http://effectivehealthcare.ahrq.gov/index.cfm/tools-and-resources/patient-decision-aids/

Alfirevic, Z., Mujezinovic, F. & Sundberg, K. (2003). Amniocentesis and chorionic villus sampling for prenatal diagnosis. *Cochrane Database of Systematic Reviews* (3), CD003252. doi: 10.1002/14651858.CD003252

American College of Obstetricians and Gynecologists. (2007). *Guidelines for Perinatal Care* (6th ed.). Washington, DC: American Academy of Pediatrics.

American College of Obstetricians and Gynecologists (2007). Screening for fetal chromosomal abnormalities. *Obstetrics & Gynecology* (109), 217-227.

American Pregnancy Association. (2006). *Prenatal Testing.* Retrieved from http://www.americanpregnancy.org/prenataltesting/

American Pregnancy Association. (2011). *Pregnancy Statistics.* Retrieved from http://www.americanpregnancy.org/main/statistics.html

Anderson, G. (1998). Creating moral space in prenatal genetic services. *Qualitative Health Research, 8*, 168-187.

Anderson, G. (1999). Nondirectiveness in prenatal genetics: Patients read between the lines. *Nursing Ethics, 6*, 126-136.

Atkins, K. (2000) Autonomy and the subjective character of experience. *Journal of Applied Philosophy, 17* (1), 71-79.

Arnold, K. M. R., & Self, S. B. (2012). Genetic screening and counseling: Family medicine obstetrics. *Primary Care: Clinics in Office Practice, 39*, 55-70. doi: 10.1016/j.pop.2011.11.010

Back, M. F., & Huak, C. Y. (2005). Family centered decision-making and non-disclosure of diagnosis in a South East Asian oncology practice. Psychooncology, 14(12), 1052-1059. doi: 10.1002/pon.918

Bane, A., Brown, L., Carter, J., Cote, C., Crider, K., de la Forest, S., ... (1998). Life and death decisions: America's changing attitudes towards genetic engineering, genetic testing and abortion, 1972-98. *International Social Work, 46*, 209-219.

Barnett, J., Ogden, J., & Daniells, E. (2008). The value of choice: a qualitative study. *British Journal of General Practice, 58*(554), 609-613. doi: 10.3399/bjgp08X330717

Beauchamp, T. L., & Childress, J. F. (1979). *Principles of biomedical ethics*. New York: Oxford University Press.

Beauchamp, T. L. (2011). Informed consent: its history, meaning, and present challenges. *Cambridge Quarterly of Healthcare Ethics, 20*(4), 515-523. doi: 10.1017/s0963180111000259

Bekker, H., Thornton, J. G., Airey, C. M., Connelly, J. B., Hewison, J., Robinson, M. B., . . . Pearman, A. D. (1999). Informed decision-making: an annotated bibliography and systematic review. *Health Technology Assess, 3*(1), 1-156.

Bekker, H. L. (2003). Genetic screening: facilitating informed choices. *Nature encyclopedia of the Human Genome,* 926- 930.

Bekker, H. L., Hewison, J., & Thornton, J. G. (2004). Applying decision analysis to facilitate informed decision-making about prenatal diagnosis for Down syndrome: a randomised controlled trial. *Prenatal Diagnosis, 24,* 265e275.

British Medical Journal. (2013). Routine Antenatal Care. *BMJ Evidence Centre.* Retrieved from http://bestpractice.bmj.com/best-practice/monograph/493.html

Browner, C. H., Preloran, M., & Press, N. A. (1996). The effects of ethnicity, education and an informational video on pregnant women's knowledge and decisions about a prenatal diagnostic screening test. *Patient Education & Counseling, 27,* 135-146.

California Department of Public Health. (2010). *Welcome to the California Prenatal Screening Program (PNS).*Retrieved from http://www.cdph.ca.gov/programs/pns/Pages/default.aspx

Cartier, L., Murphy-Kaulbeck, L., Wilson, R. D., Audibert, F., Brock, J. A., Carroll, J., . . . Pastuck, M. (2012). Counselling considerations for prenatal genetic screening. *Journal of Obstetrics and Gynaecology Canada, 34*(5), 489-493.

Central Intelligence Agency (2013a). Infant Mortality Rate. *The World Factbook.* Retrieved from https://www.cia.gov/library//publications/the-world-factbook/rankorder/2091rank.html

Central Intelligence Agency (2013b). Total Fertility Rate. *The World Factbook.* Retrieved from https://www.cia.gov/library/publications/the-world-factbook/fields/2127.html

Centers for Disease Control and Prevention. (2007). National Vital Statistics Report. *U.S. Department of Health and Human Services, 57* (8), 1-20.

Chabon, S., Morris, J., & Lemoncello, R. (2011). Ethical deliberation: a foundation for evidence-based practice. *Seminars in Speech and Language, 32*(4), 298-308. doi: 10.1055/s-0031-1292755

Chall, J. S. (1958). *Readability: an appraisal of research and application*: Ohio State University.

Charise, A., Witteman, H., Whyte, S., Sutton, E. J., Bender, J. L., Massimi, M., . . . Elf, M. (2011). Questioning context: a set of interdisciplinary questions for investigating contextual factors affecting health decision-making. *Health Expectations, 14*(2), 115-132. doi: 10.1111/j.1369-7625.2010.00618.x

Cheever, K. (2007). *IV Therapy Demystified : A Self-Teaching Guide: A Self-Teaching Guide*: Mcgraw-hill.

Chervenak, F. A., & McCullough, L. B. (2011). Ethical issues in perinatal genetics. *Seminars in Fetal & Neonatal Medicine, 16*(2), 70-73. doi: 10.1016/j.siny.2010.10.004

Chiang, H. H., Chao, Y. M., & Yuh, Y. S. (2006). Informed choice of pregnant women in prenatal screening tests for Down's syndrome. *Journal of Medical Ethics, 32*(5), 273-277. doi: 10.1136/jme.2005.012385

Ciarleglio, L. J., Bennett, R. L., Williamson, J., Mandelll, J. B., & Marks, J. H. (2003). Genetic counseling throughout the life cycle. *The Journal of Clinical Investigation, 112,* 1280-1286.

Clarke, J. B. (1993). Ethical issues in health education. *British Journal of Nursing, 2*(10), 533-538.

Croskerry, P. (2002). Achieving quality in clinical decision making: cognitive strategies and detection of bias. Academic Emergency Medicine, 9(11), 1184-1204.

Cummings, M.R. (2011). *Human heredity: Principles and issues* (9th ed.). Belmont, CA: Brooks/Cole.

Cunningham, G. C., & Tompkinison, D. G. (1999). Cost and effectiveness of the California triple marker prenatal screening program. *Genetics in Medicine, 1*(5), 199-206. doi: 10.1097/00125817-199907000-00006

De Crespigny, L., & Chervenak, F. A. (2006). *Prenatal tests: The facts.* New York: Oxford University Press.

de Jong, A., Dondorp, W. J., Frints, S. G. M., de Die-Smulders, C. E. M., & de Wert, G. M. W. R. (2011). Advances in prenatal screening: the ethical dimension. [10.1038/nrg3036]. *Nature Reviews Genetics, 12*(9), 657-663.

de Klerk, G. Classical test theory (CTT). In M. Born, C.D. Foxcroft & R. Butter (Eds.), Online Readings in Testing and Assessment, International Test Commission, http://www.intestcom.org/Publications/ORTA.php

Declercq, E. R., Sakala, C., Corry, M. P., & Applebaum, S. (2007). Listening to Mothers II: Report of the Second National U.S. Survey of Women's Childbearing Experiences: Conducted January-February 2006 for Childbirth Connection by

Harris Interactive(R) in partnership with Lamaze International. *Journal of Perinatal Education, 16*(4), 15-17. doi: 10.1624/105812407x244778

DeMars, C. (2010). *Item Response Theory*: Oxford University Press, USA.

Department of Health, Education and Welfare. (1979). *The Belmont report: Ethical principles and guidelines for the protection of human subjects of research.* Washington, DC: OPRR Reports.

DiIorio, C. K. (2006). *Measurement in Health Behavior: Methods for Research and Evaluation*: Wiley.

Dugas, M., Shorten, A., Dube, E., Wassef, M., Bujold, E., & Chaillet, N. (2012). Decision aid tools to support women's decision-making in pregnancy and birth: a systematic review and meta-analysis. *Social Science & Medicine, 74*(12), 1968-1978. doi: 10.1016/j.socscimed.2012.01.041

Epstein, R. M., & Gramling, R. E. (2013). What is shared in shared decision-making? Complex decisions when the evidence is unclear. *Medical Care Research and Review, 70*(1 Suppl), 94S-112S. doi: 10.1177/1077558712459216

Etchegary, H., & Perrier, C. (2007). Information processing in the context of genetic risk: Implications for genetic-risk communication. *Journal of Genetic Counseling, 16*(4), 419-425.

Field, A. (2013). Discovering Statistics using IBM SPSS Statistics: SAGE Publications.

Flessel, M. C., & Lorey, F. W. (2011). The California Prenatal Screening Program: "options and choices" not "coercion and eugenics". *Genetics in Medicine, 13*(8), 711-713. doi: 10.1097/GIM.0b013e3182272e25

Friedberg, M. W., Van Busum, K., Wexler, R., Bowen, M., & Schneider, E. C. (2013). A Demonstration Of Shared Decision-making In Primary Care Highlights Barriers

To Adoption And Potential Remedies. *Health Affairs, 32*(2), 268-275. doi: 10.1377/hlthaff.2012.1084

Frongillo, M., Feibelmann, S., Belkora, J., Lee, C., & Sepucha, K. (2013). Is there shared decision-making when the provider makes a recommendation? *Patient Education & Counseling, 90*(1), 69-73. doi: 10.1016/j.pec.2012.08.016

Frost, J., Shaw, A., Montgomery, A., & Murphy, D. (2009). Women's views on the use of decision aids for decision delivery following a previous caesarean section: qualitative interview study. *BJOG: An International Journal of Obstetrics & Gynaecology*, 116, 896e905.

Galbraith, J. I., Moustaki, I., Bartholomew, D. J., & Steele, F. (2002). *The Analysis and Interpretation of Multivariate Data for Social Scientists*: Taylor & Francis.

Garcia, E., Timmermans, D. R., & van Leeuwen, E. (2008). The impact of ethical beliefs on decisions about prenatal screening tests: searching for justification. *Social Science & Medicine, 66*(3), 753-764. doi: 10.1016/j.socscimed.2007.10.010

Genetics Home Reference. (2013). What is genetic discrimination? *US National Library of Medicine*. Retrieved August 28, 2013, from http://ghr.nlm.nih.gov/handbook/testing/discrimination

Gillon, R. (1994). Medical ethics: four principles plus attention to scope. *British Medical Journal, 309*(6948), 184-188.

Glazier, R., Goel, V., Holzapfel, S., Summers, A., Pugh, P., & Yeung, M. (1997). Written patient information about triple-marker screening: a randomized controlled trial. *Obstetrics & Gynecology*, 90, 769e774.

Goldbas, A. (2013). Whose body is it anyway? [Article]. *International Journal of Childbirth Education, 28*(2), 79.

Goldberg, H. (2009). Informed decision-making in maternity care. *Journal of Perinatal Education, 18*(1), 32-40. doi: 10.1624/105812409x396219

Gortmaker, S. L., & Wise, P. H. (1997). The first injustice: socioeconomic disparities, health services technology, and infant mortality. *Annual Review of Sociology, 23*, 147-170. doi: 10.1146/annurev.soc.23.1.147

Gourounti, K., & Sandall, J. (2011). The validation and translation of Multidimensional Measure of Informed Choice in Greek. Midwifery, 27(2), 170-173. doi: 10.1016/j.midw.2009.04.006

Griffith, R. & Tengnah, C. (2013). Shared decision-making: Nurses must respect autonomy over paternalism. *British Journal of Community Nursing, 18* (6), 303-306.

Guillemin, M., & Gillam, L. (2006). Attitudes to genetic testing for deafness: the importance of informed choice. *Journal of Genetic Counseling, 15*, 51-59.

Gunst, R. F., & Mason, R. L. (1980). *Regression Analysis and its Application: A Data-Oriented Approach*: Taylor & Francis.

Haddow, J. E., Palomaki, G. E., Knight, G. J., Williams, J., Miller, W. A., & Johnson, A. (1998). Screening of Maternal Serum for Fetal Down's Syndrome in the First Trimester. *New England Journal of Medicine, 338*(14), 955-962. doi: doi:10.1056/NEJM199804023381404

Hattie, J. & Cooksey, R. W. (1984). Procedures for assessing the validities of tests using the “known-groups” method. *Applied Psychological Measurement, 8* (3), 295-305.

Ho, A. (2008). Relational autonomy or undue pressure? Family's role in medical decision-making. *Scandinavian Journal of Caring Science, 22*(1), 128-135. doi: 10.1111/j.1471-6712.2007.00561.x

Health Resources and Services Administration. (n.d.). Prenatal services: Did you know… *US Department of Health & Human Services.* Retrieved from http://www.mchb.hrsa.gov/programs/womeninfants/prenatal.html

Henneman, L., Marteau, T. M., & Timmermans, D. R. M. (2008). Clinical geneticists and genetic counselors' views on communication of genetic risks: A qualitative study. *Patient Education and Counseling, 73*, 42-49.

Hershberger, P. E. & Pierce, P. F. (2010). Conceptualizing couples' decision-making in PGD: Emerging cognitive, emotional and moral dimensions. *Patient Education and Counseling, 81*, 53-62.

Hibbard, J. H., & Peters, E. (2003). Supporting informed consumer health care decisions: data presentation approaches that facilitate the use of information in choice. *Annual Review of Public Health, 24*, 413-433. doi: 10.1146/annurev.publhealth.24.100901.141005

Human Genetic Disease. (2010). In *Encyclopædia Britannica*. Retrieved April 28, 2010, from Encyclopædia Britannica Online: http://www.britannica.com/EBchecked/topic/228874/human-genetic-disease

Hunter, A. G. W., Cappelli, M., Humphreys, L., Allanson, J. E., Chiu, T. T., Peeters, C., et al. (2005). A randomized trial comparing alternative approaches to prenatal diagnosis counseling in advanced maternal age patients. *Clinical Genetics, 67*, 303e313.

ISONG/ANA (2007). *Genetics and Genomics Nursing: Scope and Standards of Practice*. Washington DC: American Nurses Association.

ISONG (2011). Informed decision-making and consent: The role of nursing. *International Society of Nurses in Genetics*. Retrieved from: http://www.isong.org/ISONG_position_statements.php

Josephson, D. L., & Keegan, L. (1999). *Intravenous infusion therapy for nurses: principles & practice*: Delmar.

Jorgensen, K. J., Brodersen, J., Hartling, O. J., Nielsen, M., & Gotzsche, P. C. (2009). Informed choice requires information about both benefits and harms. *Journal of Medical Ethics*, *35*(4), 268-269. doi: 10.1136/jme.2008.027961

Jung, H. P., Wensing, M., & Grol, R. (1997). What makes a good general practitioner: do patients and doctors have different views? *British Journal of General Practice, 47*(425), 805-809.

Kline, T. (2005). Psychological Testing: A Practical Approach to Design and Evaluation: SAGE Publications.

Knafl, K., Deatrick, J., Gallo, A., Holcombe, G., Bakitas, M., Dixon, J., & Grey, M. (2007). The analysis and interpretation of cognitive interviews for instrument development. Res Nurs Health, 30(2), 224-234. doi: 10.1002/nur.20195

Kouame, J. B. (2010). Using readability tests to improve the accuracy of evaluation documents intended for low-literate participants. *Journal of Multidisciplinary Evaluation, 6* (14), 132-139.

Kuppermann, M., Norton, M. E., Gates, E., Gregorich, S. E., Learman, L. A., Nakagawa, S., et al. (Jan 2009). Computerized prenatal genetic testing decision assisting tool: a randomized controlled trial. *Obstetrics & Gynecology, 113*(1), 53e63.

Lashley, F. R. (2005). *Clinical genetics in nursing practice* (3rd ed.). New York, NY: Springer Publishing Company, LLC.

Lashley, F. R. (2006). *Essentials of clinical genetics in nursing practice*. New York, NY: Springer Publishing Company, LLC.

Lasnover, A. L. (1987). Criticism of the California alpha fetoprotein screening program. *Western Journal of Medicine, 146* (1), 100.

Lee, K., Khoshnood, B., Chen, L., Wall, S. N., Cromie, W. J., & Mittendorf, R. L. (2001). Infant mortality from congenital malformations in the United States, 1970-1997. *Obstetrics & Gynecology, 98*(4), 620-627.

Leo, R. J. (1999). Competency and the Capacity to Make Treatment Decisions: A Primer for Primary Care Physicians. *Primary Care Companion to The Journal of Clinical Psychiatry, 1*(5), 131-141.

Levy, V. (2006). Protective steering: a grounded theory study of the processes by which midwives facilitate informed choices during pregnancy. 1998. *Journal of Advanced Nursing, 53*(1), 114-122; discussion 122-114. doi: 10.1111/j.1365-2648.2006.03688.x

Lipkin, M. (2013). Shared decision-making. *JAMA Internal Medicine, 173*(13), 1204-1205. doi: 10.1001/jamainternmed.2013.6248

Lippman, A. (1991). Prenatal genetic testing and screening: constructing needs and reinforcing inequities. *American Journal of Law & Medicine, 17*(1-2), 15-50.

Lo, B. (2000). *Resolving Ethical Dilemmas: A Guide for Clinicians*: Lippincott Williams & Wilkins.

LoBiondo-Wood, G., & Haber, J. (1998). *Nursing research: Methods, critical appraisal, and utilization*. St. Louis, MO: C. V. Mosby.

Loftus, E. F., & Fries, J. F. (2008). The potential perils of informed consent. *Mcgill Journal Of Medicine: MJM: An International Forum For The Advancement Of Medical Sciences By Students, 11*(2), 217-218.

Lowery, K. M. (2004). Legal and ethical issues in cancer genetics nursing. *Seminars in Oncology Nursing, 20* (3), 203-208.

Lumley, M. A., Zamerowski, S. T., Jackson, L., Dukes, K. & Sullivan, L. (2006). Pyscosocial correlates of pregnant women's attitudes toward prenatal maternal serum screening and invasive diagnostic testing: Beyond traditional risk status. *Genetic Testing, 10*, 131-138.

Lyons, R. A., Jones, K. H., John, G., Brooks, C. J., Verplancke, J. P., Ford, D. V., . . . Leake, K. (2009). The SAIL databank: linking multiple health and social care datasets. BMC Med Inform Decis Mak, 9, 3. doi: 10.1186/1472-6947-9-3

Manson, N. C., & O'Neill, O. (2007). *Rethinking informed consent in bioethics.* Cambridge, UK: University Press.

March of Dimes. (2008, May). *Pregnancy & Newborn: Health Education Center - Prenatal Care.* Retrieved from http://www.marchofdimes.com/pnhec/159_513.asp

Marteau, T. M. (1995). Towards informed decision-making about prenatal testing: A review. *Prenatal Diagnosis, 15*, 1215-1226.

Marteau, T. M. & Dormandy, E. (2001). Facilitating informed choice in prenatal testing: How well are we doing? *American Journal of Medical Genetics, 106*, 185-190.

Marteau, T. M., Dormandy, E., & Michie, S. (2001). A measure of informed choice. *Health Expectations, 4*, 99-108.

Mason, B. (2008) *UNICEF report: Infant mortality rates still high.* Retrieved from http://www.wsws.org/articles/2008/jan2008/mort-j31.shtml

McCoy, M. (2008). Autonomy, consent, and medical paternalism: legal issues in medical intervention. *Journal of Alternative and Complementary Medicine, 14*(6), 785-792. doi: 10.1089/acm.2007.0803

McDaniel, S. H. (2005). The psychotherapy of genetics. *Family Process, 44*(1), 25-44.

McLaughlin, G. H. (1969). SMOG Grading-a New Readability Formula. *Journal of Reading, 12*(8), 639-646. doi: 10.2307/40011226

Meray, N., Reitsma, J. B., Ravelli, A. C., & Bonsel, G. J. (2007). Probabilistic record linkage is a valid and transparent tool to combine databases without a patient identification number. J Clin Epidemiol, 60(9), 883-891. doi: 10.1016/j.jclinepi.2006.11.021

Michie, S., Dormandy, E., & Marteau, T. M. (2002). The multi-dimensional measure of informed choice: a validation study. Patient Education And Counseling, 48(1), 87-91. doi: http://dx.doi.org/10.1016/S0738-3991(02)00089-7

Mikail, C. N. (2008). *Public health genomics: The essentials.* San Fransisco, CA: Josey-Bass.

Miles, M., & Huberman, A. (1994). Qualitative data analysis (2nd. ed). Thousand Oaks, CA: Sage.

Montgomery, A. A., Emmett, C. L., Fahey, T., Jones, C., Ricketts, I., Patel, R. R., et al. (Jun 23 2007). Two decision aids for mode of delivery among women with previous caesarean section: randomised controlled trial. *British Medical Journal, 334*(7607), 1305.

Moore, M. L. (2000). Ethical issues for nurses providing perinatal care in community settings. J Perinat Neonatal Nurs, 14(2), 25-35.

Moulton, B., & King, J. S. (2010). Aligning ethics with medical decision-making: the quest for informed patient choice. *The Journal of Law, Medicine & Ethics, 38*(1), 85-97. doi: 10.1111/j.1748-720X.2010.00469.x

Moyer, A., Brown, B., Gates, E., Daniels, M., Brown, H. D., & Kuppermann, M. (1999). Decisions about prenatal testing for chromosomal disorders: Perceptions of a diverse group of pregnant women. *Journal of Women's Health & Gender-Based Medicine, 8*, 521-531.

Nassar, N., Roberts, C., Raynes-Greenow, C., Barratt, A., & Peat, B. (2007). Evaluation of a decision aid for women with breech presentation at term: a randomized controlled trial. *BJOG: An International Journal of Obstetrics & Gynaecology*, 114, 325e333.

National Human Genome Research Institute (NHGRI). (2012). An overview of the human genome project. Retrieved from http://www.genome.gov/12011238

Noble, L. (2003). Developments in neonatal technology continue to improve infant outcomes. *Pediatric Annals, 32*(9), 595-603.

Noble-Adams, R. (1999). Ethics and nursing research. 1: Development, theories and principles. *British Journal of Nursing, 8*(13), 888-892.

Norman, G.R., & Streiner, D.L. (1994). *Biostatistics: The bare essentials*. St. Louis, MO: Mosby.

Nunnally, J. C. (1978). *Psychometric Theory* (2nd ed.). New York, NY: McGraw-Hill.

Nunnally, J. C., & Bernstein, I. H. (1994). *Psychometric Theory* (3rd ed.) New York, NY: McGraw-Hill.

O'Connor, A. M. (1997). Decisional Conflict Scale Ottawa Civic Hospital (2nd ed.).

Pett, M. A., Lackey, N. R., & Sullivan, J. J. (2003). *Making sense of factor analysis: The use of factor analysis for instrument development in health care research*. Thousand Oaks, CA: Sage.

Polit, D. F., & Beck, C. T. (2008). *Nursing Research: Generating and Assessing Evidence for Nursing Practice*: Wolters Kluwer Health/Lippincott Williams & Wilkins.

Polit, D. F., Beck, C. T., & Owen, S. V. (2007). Is the CVI an acceptable indicator of content validity? Appraisal and recommendations. *Research in Nursing & Health, 30*, 459-467.

Potter, B. K., O'Reilly, N., Etchegary, H., Howley, H., Graham, I. D., Walker, M., . . . Wilson, B. J. (2008). Exploring informed choice in the context of prenatal testing: findings from a qualitative study. *Health Expect, 11*(4), 355-365. doi: 10.1111/j.1369-7625.2008.00493.x

Powers, B. A., & Knapp, T. R. (2006). *Dictionary of Nursing Theory and Research*: 3rd Edition: Springer Publishing Company.

Priver, D. M. (1987). Criticism of the California alpha fetoprotein screening program. *Western Journal of Medicine, 146* (1), 100.

Rao, G. (2007). *Rational medical decision-making.* The McGraw-Hill Companies

Rappaport, V. J. (2008). Prenatal diagnosis and genetic screening--integration into prenatal care. *Obstetrics & Gynecology Clinics of North America, 35*(3), 435-458.

Reckase, M. (2009). *Multidimensional Item Response Theory*: Springer.

Reise, S. P., & Waller, N. G. (2009). Item response theory and clinical measurement. *Annual Review of Clinical Psychology*, 5, 27-48. doi: 10.1146/annurev.clinpsy.032408.153553

Reyna, V. F. (2008). A theory of medical decision-making and health: Fuzzy trace theory. *Medical decision-making, 28,* 850-865.

Robertson, R., Dixon, A., & Le Grand, J. (2008). Patient choice in general practice: the implications of patient satisfaction surveys. *Journal of Health Services Research & Policy, 13*(2), 67-72. doi: 10.1258/jhsrp.2007.007055

Santalahti, P., Hemminki, E., Latikka, A. M., & Ryynanen, M. (1998). Women's decision-making in prenatal screening. *Social Science & Medicine, 46*(8), 1067-1076.

Schloendorff v Society of New York Hospital, 211 N.Y. 125; 105 N.E. 92; N.Y. LEXIS 1028 (1914).

Schoonen, M., Wildschut, H., Essink-Bot, M., Peters, I., Steegers, E. & de Koning, H. (2012) The provision of information and informed decision-making on prenatal screening for Down syndrome: A questionnaire- and register-based survey in a non-selected population. *Patient Education and Counseling, 87,* 351-359.

Schulman-Green, D., Ercolano, E., Jeon, S., & Dixon, J. (2012). Validation of the knowledge of care options instrument to measure knowledge of curative, palliative, and hospice care. J Palliat Med, 15(10), 1091-1099. doi: 10.1089/jpm.2011.0514

Segal, I., & Shahar, Y. (2009). A distributed system for support and explanation of shared decision-making in the prenatal testing domain. *Journal of Biomedical Informatics, 42*(2), 272-286. doi: 10.1016/j.jbi.2008.09.004

Shiell, A., Au, F., Jonhston, D., & Tough, S. (2010). Eliciting individual preferences for health care: a case study of perinatal care. *Health Expectations, 13*(1), 4-12. doi: 10.1111/j.1369-7625.2009.00551.x

Shorten, A., Shorten, B., Keogh, J., West, S., & Morris, J. (Dec 2005). Making choices for childbirth: a randomized controlled trial of a decision-aid for informed birth after cesarean. *Birth, 32*(4), 252e261.

Shultz, K. S. & Whitney, D. J. (2005) *Measurement theory in action.* Thousand Oaks, CA: Sage Publications, Inc.

Slack, C., Lurix, K., Lewis, S. & Lichten, L. (2006). Prenatal genetics: The evolution and future directions of screening and diagnosis. *Journal of Perinatal and Neonatal Nursing, 20* (1), 93-97.

Smith, C. M. (2005). Origin and uses of primum non nocere--above all, do no harm! *Journal of Clinical Pharmacology, 45*(4), 371-377. doi: 10.1177/0091270004273680

Stacey, D., Bennett, C. L., Barry, M. J., Col, N. F., Eden, K. B., Holmes-Rovner, M., . . . Thomson, R. (2011). Decision aids for people facing health treatment or screening decisions. *Cochrane Database of Systematic Review* (10), CD001431. doi: 10.1002/14651858.CD001431.pub3

Steinberg, L. S. 2001. The consequences of pairing questions: context effect in personality measurement. *Journal of Personality and Social Psychology,* 81, 332–42

Stewart, A., Sodhi, V., Harper, N., & Yentis, S. M. (2003). Assessment of the effect upon maternal knowledge of an information leaflet about pain relief in labor. *Anaesthesia*, 58, 1003e1022.

The University of Nottingham. (n.d.). *Education Development: Innovation Technologies.* Retrieved from: http://www.niace.org.uk/misc/SMOG-calculator/smogcalc.php

Tversky, A., & Kahneman, D. (1981). The Framing of Decisions and the Psychology of Choice. *Science, 211*(4481), 453-458. doi: 10.2307/1685855

Toutenburg, H. & Everitt, B. S. (1985). Introduction to Latent Variable Models. Chapman and Hall, London 1984. 107 pp., £ 9.50. *Biometrical Journal, 27*(6), 706-706. doi: 10.1002/bimj.4710270617

United Nations, *World Population Prospects: 2006 revision,* Table A 18-19 (2006)

Van den Berg, M., Timmermans, D. R. M., Kate, L. P. T., Van Vugt, J. M. G. & Van der Wal, G. (2006). Informed decision-making in the context of prenatal screening. *Patient Education and Counseling, 63,* 110-117.

Van den Berg, M., Timmermans, D. R., Knol, D. L., van Eijk, J. T., de Smit, D. J., van Vugt, J. M., & van der Wal, G. (2008). Understanding pregnant women's decision-making concerning prenatal screening. *Health Psychology, 27*(4), 430-437. doi: 10.1037/0278-6133.27.4.430

Vanstone, M., Kinsella, E. A., & Nisker, J. (2012). Information-sharing to promote informed choice in prenatal screening in the spirit of the SOGC clinical practice guideline: a proposal for an alternative model. *Journal of Obstetrics & Gynaecology Can, 34*(3), 269-275.

Vogel-Walcutt, J. J., Gebrim, J. B., Bowers, C., Carper, T. M., & Nicholson, D. (2011). Cognitive load theory vs. constructivist approaches: which best leads to efficient, deep learning? *Journal of Computer Assisted Learning, 27*(2), 133-145. doi: 10.1111/j.1365-2729.2010.00381.x

Waletzky, J. & Gupta, M. (2011). Prenatal quad screen: Technical brief. *Cleveland Clinic Laboratories*. Retrieved from: http://clevelandcliniclabs.com/portals/66/PDF/TechBriefs/TB_PrenatalQuadScreen.pdf

Watson MS, Mann MY, Lloyd-Puryear MA, et al. (2006) Newborn screening: Toward a uniform screening panel and system [executive summary]. *Pediatrics, 117*(5 Pt. 2):S296-307

Weber, S., Hill, B. J., Ren, D., & Beatty, R. L. (2011). Measurement of Parenting Intention, Decision-Making, and Expectations. Perspectives in Psychiatric Care, 47(1), 35-47. doi: 10.1111/j.1744-6163.2010.00262.x

Weiner, S. J., Schwartz, A., Sharma, G., Binns-Calvey, A., Ashley, N., Kelly, B., . . . Harris, I. (2013). Patient-centered decision-making and health care outcomes: an observational study. *Annals of Internal Medicine, 158*(8), 573-579. doi: 10.7326/0003-4819-158-8-201304160-00001

Weiner, S. J., Schwartz, A., Weaver, F., Goldberg, J., Yudkowsky, R., Sharma, G., . . . Abrams, R. I. (2010). Contextual errors and failures in individualizing patient care: a multicenter study. *Annals of Internal Medicine, 153*(2), 69-75. doi: 10.1059/0003-4819-153-2-201007200-00002

Whitney, P., Rinehart, C. A., & Hinson, J. M. (2008). Framing effects under cognitive load: the role of working memory in risky decisions. *Psychonomic Bulletin & Review, 15*(6), 1179-1184. doi: 10.3758/pbr.15.6.1179

Willis G. Cognitive interviewing. A 'how to' guide. Research Triangle Park, NC: Research Triangle Institute, 1999.

Woolf, S. H., Chan, E. C., Harris, R., Sheridan, S. L., Braddock, C. H., 3rd, Kaplan, R. M., . . . Tunis, S. (2005). Promoting informed choice: transforming health care to dispense knowledge for decision-making. *Annals of Internal Medicine, 143*(4), 293-300.

World Bank. (2011). *World Development Indicators.* Retrieved from http://data.worldbank.org/data-catalog/world-development-indicators?cid=GPD_WDI

World Health Organiazation (WHO). (2013a). MDG 4: Child health: Infant mortality by country. *Global Health Observatory Data Repository.* Retrieved from: http://apps.who.int/gho/data/node.main.526?lang=en

World Health Organization (WHO). (2013b). Congenital Anomalies. *Media Centre.* Retrieved from: http://www.who.int/mediacentre/factsheets/fs370/en/

Zarcadoolas, C., Pleasant, A., & Greer, D. S. (2003). Elaborating a definition of health literacy: A commentary. *Journal of Health Communication, 8,* 119-120.

Zarcadoolas, C., Pleasant, A., & Greer, D. S. (2006). *Advancing health literacy: A framework for understanding and action*. San Fransisco, CA: John Wiley & Sons, Inc.

Zarcadoolas, C. (2011). The simplicity complex: exploring simplified health messages in a complex world. *Health Promotion International, 26*(3), 338-350. doi: 10.1093/heapro/daq075

Vita

Mandeep Kaur was born in New Delhi, India and immigrated to the United States at a very young age. She was raised with one older sister in Austin, Texas. She had the honor of graduating with a Bachelor of Science in Nursing from the University of Texas at Austin in December of 2007. Her nursing practice has been diverse with clinical experience in labor and delivery, corporate nursing in a semiconductor company and legal nurse consulting in device and pharmaceutical mass tort litigation. She has had the opportunity to work in various areas of academia through teaching assistant work in hospital clinicals, skills lab and didactic courses.

Her interest in research throughout her educational career sparked her interest in pursuing a Doctor of Philosophy at the University of Texas at Austin beginning in 2008. Her work with expectant mothers in the labor and delivery guided her to pursue research regarding informed decision-making about prenatal genetic screening.

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