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A Feasibility Analysis of a Pilot Study Comparing Prenatal Genetic Service Delivery Outcomes Using the Self-Determination Theory

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A Feasibility Analysis of a Pilot Study Comparing
Prenatal Genetic Service Delivery Outcomes
Using the Self-Determination Theory

by

Lindsey N. Victoria

A thesis submitted in partial fulfillment
of the requirements for the degree of
Masters of Science
with a concentration in Genetic Counseling
Department of Global Health
College of Public Health
University of South Florida

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ABSTRACT

Genetic counselors, along with the National Society of Genetic Counselors, desire evidence-based research and data assessing the value of genetic counseling in genetic service delivery. This pilot study was designed to gather data about genetic counseling outcomes as well as analyze the feasibility of a study looking at new genetic outcome measures in the prenatal setting. Implementation of the methods used for data collection were evaluated by analysis of the appropriateness, acceptability, feasibility, fidelity, and adoption of the research protocol at three sites. We found that there is a hierarchy between implementation outcomes and it may be necessary to satisfy one implementation outcome before the next one can be achieved. We also found that patient engagement is a key component to evaluating the success of methods used for data collection. These findings may be useful to individuals designing future research studies used to measure genetic counseling outcomes.
INTRODUCTION

Several genetic counselors in conjunction with the National Society of Genetic Counselors have called for more attention on demonstrating the value of genetic counseling and the important role genetic counselors play in genetic service delivery. As evidence-based medicine is the gold-standard for justifying clinical practice strategies it is essential to develop research studies to measure genetic counseling outcomes. Genetic counseling outcomes have been analyzed in the past; however, this has not been done using simple, short measures and methods that could be easily implemented in prenatal clinics. Self-determination theory has been used as a framework to guide development of studies to evaluate other healthcare professions and may be a suitable framework for genetic counseling as well. As the goal is widespread adoption of methods to evaluate genetic counseling, it is also important to analyze the implementation of a study to identify aspects that can be improved, automated, or simplified. This article reviews several areas that genetic counselors can draw upon when developing research protocols to evaluate genetic counseling outcomes and measuring the efficiency of implementation of those protocols. This research also discusses progress on a multi-site research study to evaluate implementation of methods to collect data as part of a feasibility study looking at new genetic outcome measures in prenatal genetic counseling settings.
**Evidence-Based Medicine**

Evidence-based medicine involves health care professionals making decisions on how to care for their patients using the most recent evidence gathered from research obtained from the medical community (Sackett, Rosenberg, Gray, Haynes, & Richardson, 1996). Evidence-based medicine, which originated in mid-19th century Paris, has been widely accepted as the gold standard for determining ideal methods for patient care and improving medical practices since the 1990’s (Sackett et al., 1996). At this time, there was an influx of researchers and providers using evidence based practices due to increased social after the Evidence-Based Medicine Working Group identified evidence-based medicine as the new paradigm for medical practice (Evidence-Based Medicine Working Group, 1992; Pope, 2003; Sackett et al., 1996). The Evidence-Based Medicine Working Group saw evidence-based medicine as an essential tool for health care providers when navigating the increasing amount of uncertainties in medicine due to the rapid increases in literature and technology, concerns for medical costs, and scrutiny about patient outcomes in health care (Evidence-Based Medicine Working Group, 1992).

Evidence-based medicine has not been without criticism as the movement has brought about an abundance of evidence, some with little clinical utility, which is defined as how useful the newly gained research is for clinical practice (Greenhalgh, Howick, & Maskrey, 2014). It is important to remember that at its root, evidence-based medicine is about providing the best clinical care for patients and research endeavors should be designed with the needs of the patient in mind. Research findings should be presented in ways that are easy for the audience, which may include both clinicians and patients, to understand (Greenhalgh et al., 2014). It is essential to ensure the right questions are being asked during research and that the study will have clinical utility.
Genetic Counseling and the Reciprocal Engagement Model

Genetic counseling is defined according to the National Society of Genetic Counselors as “the process of helping people understand and adapt to the medical psychological and familial implications of genetic contributions to disease” (Resta et al., 2006). Genetic counselors receive a Master’s degree from an accredited genetic counseling training program, where they learn the communication skills, genetic knowledge, critical thinking skills, counseling skills, and psychosocial assessment skills needed to provide optimum patient care (Fine, Baker, Fiddler, & ABGC Consensus Development Consortium, 1996). The profession continues to evolve and increase in complexity as it takes on a more prominent role in health care and gathers an increased presence in the media and public health (Veach, Bartels, & Leroy, 2007). Prenatal genetic counseling is becoming especially challenging due to an increase in potentially diagnosable disorders and availability of more testing options (Minkoff & Berkowitz, 2014).

Evidence-based medicine was identified as a tool to navigate the increasing amount of literature and available technology in medicine in the 1990’s and could be applied as an equally useful tool to navigate the increasing amount of genetic information available (Evidence-Based Medicine Working Group, 1992).

Evidence-based outcomes research for genetic counseling services is limited, particularly in prenatal settings, and it is essential to design protocols and validate measures that can be used to evaluate services and guide practice as it evolves and grows (Redlinger-Grosse et al., 2016). The importance of identifying the goals of genetic counseling and creating a model for practice lead to a consensus conference that created The Reciprocal-Engagement model, which identifies 5 tenets of genetic counseling and 17 goals to achieve these tenets in practice (Veach et al.,
The first tenet is that information is key, which highlights the importance of knowledge for both the counselor and the patient (Veach et al., 2007). The second tenet is that the relationship is integral to genetic counseling; without a strong relationship between the counselor and the patient, shared decision making is less likely to occur (Veach et al., 2007). The quality of a patient’s relationship with their provider has been shown to directly affect the health and outcomes for patient (Beach, Keruly, & Moore, 2006). Improving communication and the relationship between the patient and provider has been shown to increase the efficiency of shared-decision making (Adams & Drake, 2006). The third tenet is that patient autonomy must be supported throughout the session so that the patient is empowered to make decision (Veach et al., 2007). The fourth tenet is that patients are resilient and the counselor should recognize the patient’s ability to adapt to situations (Veach et al., 2007). The fifth tenet is that the patient’s emotions make a difference and the counselor should remain aware of the patient’s emotions throughout the session to better understand their feelings and values (Veach et al., 2007). These tenets highlight the importance of being patient-centered, empowering the patient, focusing on the relationship between the patient and provider, and addressing the patient’s emotions (Veach et al., 2007).

The 17 goals identified by the REM were further simplified to the following factors: understanding and appreciation between the counselor and patient, support and guidance provided by the counselor, facilitating patient decision-making, and patient-centered education (Hartmann, Veach, MacFarlane, & LeRoy, 2015; Redlinger-Grosse et al., 2016). Important factors associated with genetic counseling were identified, but they did not develop methods or specific measures that could be used to evaluate these outcomes in clinical practice (Hartmann et al., 2015; Redlinger-Grosse et al., 2016; Veach et al., 2007). Previous publications have
identified challenges in evaluating genetic counseling outcomes emphasizing the lack of reliable and valid measures, uncertainty about the patient needs based on advances in technology, and uncertainty about how to measure outcomes across genetic counseling specialties (Redlinger-Grosse et al., 2016). Pilot studies and feasibility studies are important to test new outcome measures, identify challenges, and create solutions to increase success of future research studies.

**Self-Determination Theory**

Self-determination theory states that high levels of autonomy, relatedness, and competence will lead to enhanced self-motivation (Ryan & Deci, 2000). Patients who are self-motivated and engaged in their health care have better care experiences resulting in improved health outcomes (Hibbard & Greene, 2013). Autonomy is defined as the ability for an individual to make decisions according to their own free will. The importance of autonomy is noted in self-determination theory as well as the third tenet of the reciprocal engagement model. Relatedness, defined as the need for connectedness with others, is a critical aspect of self-determination theory as well as the second tenet of the reciprocal engagement model. Competence is defined in SDT as the feeling of being capable of completing a task; however, to be truly competent using a more standard definition of the word, it also requires knowledge. Competence therefore encompasses both the first REM tenet, “knowledge is key”, and the third, which advises counselors to empower patient to make decisions. Overlap between the REM tenets and self-determination theory is notable given that self-determination theory has been successfully applied to evaluate outcomes in a variety of settings including healthcare, education, workplace, and physical activity (Ng et al., 2012).
**Shared-Decision Making**

Shared-decision making is a service delivery model that involves the patient and the genetic counselor working together to make informed decisions about testing and screening options. When a genetic counselor successfully uses shared-decision making they provide the patient with accurate information, while supporting the decision making process for the patient (Elwyn et al., 2012). Shared-decision making has been shown to improve satisfaction, reduce decisional conflict, and increase patient knowledge and informed choice (Durand et al., 2014; Shay & Lafata, 2015). Despite the evidence supporting the use of shared-decision making health care providers are not affectively using in in their clinics citing lack of time, too much information to cover, and doubts that patients have the competence to participate in a session (Stevenson, Barry, Britten, Barber, & Bradley, 2000). In 2001 the Institute of Medicine highlighted the benefits of shared-decision making and recommended redesigning health care to incorporate the model; however, there are still few health care providers consistently using shared-decision making in practice (Couët et al., 2015).

**Implementation Framework**

Implementation science arose in response to a recognized need to evaluate success and failures in implementing evidence-based programs as well as the need to understand the effects of making changes in procedures, processes, or programs. There are numerous factors that influence whether practice changes or implementation of new processes will be successful and understanding these is essential for evaluating genetic counseling outcome studies (E. Proctor et al., 2011; Redlinger-Grosse et al., 2016). A research study or evaluation process may fail because the measures utilized were inefficient to assess the outcome or because the study was
implemented poorly (E. Proctor et al., 2011). Therefore, it is important to not only analyze the measures and data that will be collected, but also the procedures used to obtain the data within a clinical setting given that the priority must be to maintain high quality patient care with limited disruption to usual service delivery.

A framework to evaluate implementation of this research study was modeled after previously published frameworks and taxonomy (E. K. Proctor et al., 2009; E. Proctor et al., 2011). According to this framework implementation outcomes must be met before service outcomes and client/patient outcomes can be achieved. Implementation outcomes are described in detail in a later section and include: acceptability, adoption, appropriateness, costs, feasibility, fidelity, penetration, and sustainability (E. Proctor et al., 2011). Service outcomes in this framework are defined using the Institute of Medicine’s standards of care, which requires healthcare delivery to be safe, effective, patient-centered, timely, efficient, and equitable (Institute of Medicine (US) Committee on Quality of Health Care in America, 2001; E. Proctor et al., 2011).

Genetic counseling services have been found to be more time-consuming and labor intensive than other medical services (Sukenik-Halevy, Ludman, Ben-Shachar, & Raas-Rothschild, 2016). For these reasons, it is especially important to consider clinic flow and efficiency when developing a research protocol to study genetic counseling outcomes. It is important when performing research not to disrupt the delivery of services. The implementation process of a research study is an essential, and often over-looked, component of the research process. An objective of this pilot study is to refine methods used to evaluate genetic counseling services and improve feasibility of future studies on a larger scale. Identifying barriers to implementing research protocols to evaluate genetic counseling outcomes and solutions to these
challenges could increase uptake of studies to evaluate genetic counseling outcomes in the future.
STUDY GOALS AND OBJECTIVES

The objectives of this study are: 1) To refine measures and methods used to evaluate genetic counseling services in order to assess and improve feasibility of future studies on a large scale and 2) To measure patient knowledge and recall, perception of autonomy support, self-competence, relatedness, and decision making in the prenatal setting.

Limited research has measured genetic counseling outcomes (Redlinger-Grosse et al., 2016). This study serves to fill that gap by creating simple, brief measures based on Self-Determination Theory that could be used to evaluate outcomes in the future. The study will help refine measures used to assess prenatal outcomes, evaluate feasibility of similar studies on a larger scale, and serve as a key step in helping achieve the long-term goal of improving patient outcomes. We seek to identify which implementation outcomes are most important for successful implementation in order to guide development of future studies.
METHODS

This multi-site implementation study involves data collection at three prenatal sites within three university settings in the South-Eastern United States. Institutional Review Board (IRB) approval was obtained from institution #1 and #2 and is in the process of being obtained from institution #3. Methods for recruitment and survey distribution vary between sites due to differences in clinic flow and requests made by the IRB at each site. Designated and approved study team members at each site view the patient schedule and charts weekly to identify potential participants, who are 18 years or older, able to read and speak English, capable of consenting to participate, and scheduled to be seen at one of the participating centers to discuss risks for fetal chromosome conditions. To preserve anonymity, patient-generated identification codes were used to match each participant’s pre-visit questionnaire to the post-visit questionnaire at all sites.

Recruitment and Survey Distribution

Institution #1

A research team member approaches each patient who qualifies for the study, provides them with information about the study, answers their questions and then obtains signed consent if they choose to participate in the study. Pre-visit surveys are distributed by study team members in clinic prior to the session using a tablet purchased for this study. Upon completion of the pre-visit survey, participants click on a link to a different website (unlinked to their survey responses), where they provide their e-mail address, which is used to send a link to the post-visit
survey. Participants are also provided a sheet of paper with the link and a QR code that can be used to access the post-visit questionnaire. The participants are then instructed to complete the post-visit questionnaire on their own person smart phone, tablet, or computer.

**Institution #2**

Pre-visit questionnaires are distributed using a messenger service available through the electronic medical record approximately 7 days prior to their visit. Written documentation of consent was waived by the IRB. Participants review the consent form and e-mail the researchers if they have any questions or concerns. Those who are interested in participating are instructed to complete the pre-visit questionnaire at home on their computer, tablet, or smart phone prior to attending the visit. When they complete the pre-visit questionnaire, they provide their e-mail address on a separate website, which is used to send them a link to the post-visit questionnaire. Participants at site 3 complete all aspects of the research study outside of clinic.

**Institution #3**

Methods for recruitment are still being developed in this clinic because it is in the process of obtaining IRB approval. The submitted proposal uses methods similar to the methods used at Institution #1. However, a waiver of written informed consent has been requested at this site due to the minimal risk nature of the study, which is assessing only patient opinions, attitudes, and recall about their visit and will not collect protected health information as part of the survey.

**Implementation**

Throughout the planning and implementation process e-mails and field notes were collected by the primary investigator documenting implementation challenges, discussions
surrounding solutions, and changes that were implemented. These field notes and e-mails were then reviewed and discussed among the researchers.
MEASURES

Patient/Client Experiences, Values, Attitudes, and Decisions

Demographic, non-identifying information about participants including the participant’s age, ethnicity, and education level are collected from pre-visit surveys. Both pre- and post- visit surveys collect data evaluating patient knowledge, decision making competence, patient values related to screening and testing for fetal chromosome changes, and empowerment to make a decision about genetic testing. Post-visit surveys also assess the extent to which the patient felt autonomy support from their provider and their perceptions of relatedness to their healthcare provider.

Autonomy support is measured using the health care climate control questionnaire (G. C. Williams & Deci, 2001). It consists of 6 Likert scale questions that assess how well the patient feels the provider supported their autonomy. Minor language modifications were made to the validated questions on this scale in order to fit the research study better. These modifications can be seen in Table 1.

Perceived patient competence to make a decision about genetic testing is being measured in the pre-visit and post-visit surveys using a newly created scale. Validated scales used to measure perceived competence in previous studies were analyzed and determined to be inappropriate for a study analyzing outcomes that did not include interventions (G. C. Williams & Deci, 1996; Geoffrey C. Williams, Freedman, & Deci, 1998). Therefore, the researchers used
those questions to develop 4 new questions shown in Table 2 that are rated using a 5-points Likert scale.

**Table 1.** Autonomy support questions created from a modified version of The Health Care Climate Control Questionnaire

<table>
<thead>
<tr>
<th>Original vs. Modified</th>
<th>Autonomy Support Questions</th>
</tr>
</thead>
<tbody>
<tr>
<td>Original</td>
<td>I feel that my physician has provided me choices and options.</td>
</tr>
<tr>
<td>Modified</td>
<td>I feel that my healthcare provider gave me choices and options about genetic testing.</td>
</tr>
<tr>
<td>Original</td>
<td>I feel understood by my physician.</td>
</tr>
<tr>
<td>Modified</td>
<td>I feel understood by my healthcare provider.</td>
</tr>
<tr>
<td>Original</td>
<td>My physician conveys confidence in my ability to make changes.</td>
</tr>
<tr>
<td>Modified</td>
<td>This provider is confident in my ability to make decisions.</td>
</tr>
<tr>
<td>Original</td>
<td>My physician encourages me to ask questions.</td>
</tr>
<tr>
<td>Modified</td>
<td>My provider encouraged me to ask questions.</td>
</tr>
<tr>
<td>Original</td>
<td>My physician listens to how I would like to do things.</td>
</tr>
<tr>
<td>Modified</td>
<td>My provider listened to how I would like to do things.</td>
</tr>
<tr>
<td>Original</td>
<td>My physician tries to understand how I see things before suggesting a new way to do things.</td>
</tr>
<tr>
<td>Modified</td>
<td>My provider tries to understand how I see things before making suggestions.</td>
</tr>
</tbody>
</table>

**Table 2: Perceived competence questions using a newly created scale**

<table>
<thead>
<tr>
<th>Perceived Competence Questions</th>
</tr>
</thead>
<tbody>
<tr>
<td>I feel I am able to make a decision about prenatal screening and testing.</td>
</tr>
<tr>
<td>I feel confident I can ask questions if I don’t understand my choices.</td>
</tr>
<tr>
<td>I am capable of understanding the information enough to make a decision.</td>
</tr>
<tr>
<td>I have the ability to figure out what choice is best for me.</td>
</tr>
</tbody>
</table>

Relatedness is measured in the post-visit survey using a modified version of the validated Patient-Doctor Depth or Relationship Measure (Ridd, Lewis, Peters, & Salisbury, 2011). The scale was modified from 8 questions to 4 to decrease the amount of time the questionnaire would take patients to complete. Questions were chosen based on which seemed most appropriate for
this study. The word doctor was changed to provider to better fit this study. The questions use a Likert scale and are shown in Table 3.

**Table 3: Patient-Provider relatedness questions modified from the Patient-Doctor Depth-of-Relationship Scale**

<table>
<thead>
<tr>
<th>Original vs. Modified</th>
<th>Patient-Provider Relatedness Questions</th>
</tr>
</thead>
<tbody>
<tr>
<td>Original</td>
<td>I feel totally relaxed with this doctor.</td>
</tr>
<tr>
<td>Modified</td>
<td>I feel totally relaxed with this provider.</td>
</tr>
<tr>
<td>Original</td>
<td>This doctor takes me seriously.</td>
</tr>
<tr>
<td>Modified</td>
<td>This provider takes me seriously.</td>
</tr>
<tr>
<td>Original</td>
<td>This doctor really cares for me.</td>
</tr>
<tr>
<td>Modified</td>
<td>This provider really cares for me.</td>
</tr>
<tr>
<td>Original</td>
<td>This doctor really knows how I feel about things.</td>
</tr>
<tr>
<td>Modified</td>
<td>This provider really knows how I feel about things.</td>
</tr>
<tr>
<td>Unused Question</td>
<td>I know this doctor very well.</td>
</tr>
<tr>
<td>Unused Question</td>
<td>This doctor knows me as a person.</td>
</tr>
<tr>
<td>Unused Question</td>
<td>I know what to expect with this doctor.</td>
</tr>
<tr>
<td>Unused Question</td>
<td>This doctor accepts me the way I am.</td>
</tr>
</tbody>
</table>

Knowledge about genetic testing options and chromosome conditions is assessed via 8 questions that were designed by the study team to assess the type of knowledge needed to make an informed decision. This includes knowledge about the cause of chromosome conditions, clinical characteristics of chromosome conditions, what conditions can be found using the testing/screening methods, and risks and limitations of testing. These questions were developed by a team of 3 genetic counselors with input from 3 other genetic counselors and an MD, who specializes in maternal fetal medicine. These questions asked the patient to select yes, no, or unsure for each question. Questions can be found in Table 4.

The SURE scale from the Ottawa Decision Support Framework is used in the pre- and post-visit surveys to measure the patient’s empowerment to make a decision (Légaré et al., 2010). In this case empowerment to make a decision is being defined as the opposite of
decisional conflict and reflects the patients’ feeling that they know what decision is best for them, they have the information and support they need to make a decision, they understand the benefits and risks, and they understand which benefits and risk are most important to them. Questions were slightly modified to improve clarity for this study and match the formatting with other questions. These modifications can be seen in Table 5.

**Table 4: Knowledge questions using a newly created scale**

<table>
<thead>
<tr>
<th>Knowledge Questions</th>
</tr>
</thead>
<tbody>
<tr>
<td>A chromosome condition is caused by a change in the amount of genetic information that a person has.</td>
</tr>
<tr>
<td>There is a cure for some chromosome conditions if we find out about it early.</td>
</tr>
<tr>
<td>Chromosome conditions are most often inherited from the parents.</td>
</tr>
<tr>
<td>Chromosome conditions can cause health problems or learning problems.</td>
</tr>
<tr>
<td>A screening test done during pregnancy can tell us for sure if a baby has a genetic condition.</td>
</tr>
<tr>
<td>A diagnostic test can sometimes cause a problem with the pregnancy, but most women have no problems after the test.</td>
</tr>
<tr>
<td>Screening tests sometimes cause harm to an unborn baby.</td>
</tr>
<tr>
<td>A baby could still have a genetic condition even if diagnostic testing is normal.</td>
</tr>
</tbody>
</table>

**Table 5: Empowerment to make a decision questions using the SURE scale**

<table>
<thead>
<tr>
<th>Original vs. Modified</th>
<th>SURE Scale Questions</th>
</tr>
</thead>
<tbody>
<tr>
<td>Original</td>
<td>Do you know the benefits and risk of each option?</td>
</tr>
<tr>
<td>Modified</td>
<td>I know the benefits and risks of the genetic testing options.</td>
</tr>
<tr>
<td>Original</td>
<td>Are you clear about which benefits and risks matter most to you?</td>
</tr>
<tr>
<td>Modified</td>
<td>I am clear about which benefits and risks matter most to me.</td>
</tr>
<tr>
<td>Original</td>
<td>Do you have enough support and advice to make a choice?</td>
</tr>
<tr>
<td>Modified</td>
<td>I have enough support and advice to make a choice.</td>
</tr>
<tr>
<td>Original</td>
<td>Do you feel SURE about the best choice for you?</td>
</tr>
<tr>
<td>Modified</td>
<td>I feel sure about the best choice for me.</td>
</tr>
</tbody>
</table>

Additional questions are included on both surveys to assess the extent to which the patient values the genetic information that can be gained from prenatal testing and how useful and important the testing is to them. This will then be compared to determine if their values
match their reported decision about having genetic testing. Two four-part values questions were creating using a modified version of the multi-dimensional measure of informed choice (MMIC) attitude scale (Marteau, Dormandy, & Michie, 2001). These questions asked patients their opinions about genetic screening and diagnostic tests using semantic differential scales. Words were altered on the MMIC scale to increase clarity and create questions that would be easier for someone with lower literacy to understand. An additional 4 Likert-type values questions were newly created for this study to ascertain how it compared to the MMIC questions. Values questions are shown in Table 6.

**Table 6: Value questions using a modified version of the MMIC semantic differential scale and a newly created scale**

<table>
<thead>
<tr>
<th>Scale</th>
<th>Original vs. Modified</th>
<th>Values Questions</th>
</tr>
</thead>
<tbody>
<tr>
<td>Modified MMIC semantic</td>
<td>Original</td>
<td>Bad thing/ Good thing</td>
</tr>
<tr>
<td>differential Scale</td>
<td>Modified</td>
<td>Bad choice/ Good choice</td>
</tr>
<tr>
<td></td>
<td>Original</td>
<td>Unimportant/ Important</td>
</tr>
<tr>
<td></td>
<td>Modified</td>
<td>Unimportant/ Important</td>
</tr>
<tr>
<td></td>
<td>Original</td>
<td>Harmful/ Beneficial</td>
</tr>
<tr>
<td></td>
<td>Modified</td>
<td>Harmful/ Helpful</td>
</tr>
<tr>
<td></td>
<td>Original</td>
<td>Unpleasant/ Pleasant</td>
</tr>
<tr>
<td></td>
<td>Modified</td>
<td>Going to make me worry more/ Going to make me worry less</td>
</tr>
<tr>
<td>Newly created scale</td>
<td>I only want to know about health problems for my unborn baby if there is something that can be done to fix the health problems before birth. I am willing to take a small chance that there could be a problem because of a test, if it means I will find out if my unborn baby has a chromosome condition. Having genetic testing will make me worry more than if I don't have genetic testing. I want to know as much as I can about the health of my unborn baby.</td>
<td></td>
</tr>
</tbody>
</table>
Respondents are asked to provide a written response of why they made the decision they did, what they found helpful during the visit, and what they think would be more helpful in the future. These questions will provide qualitative information about the patient’s experience.

**Implementation Outcomes**

The following implementation outcomes were described in a previous study as essential for measuring the implementation success of a study: acceptability, adoption, appropriateness, costs, feasibility, fidelity, penetration, and sustainability (E. Proctor et al., 2011). Outcomes have been altered slightly to fit the current study, which consists of collecting data through pre-visit and post-visit surveys as described in more detail in the following section. We also determined it was important to analyze patient engagement for studies involving patient participation for research.

A prior study defined the level of analysis for each implementation outcomes, such as the individual provider, individual consumer, or organization. Here we will identify the level of analysis as described in the previous paper as well as identify the specific individual or group that conducted the analysis or made the decision about the implementation outcome for this study. Analysis methods are listed in Table 7 and described in more detail in the following paragraphs.
Table 7: Level of analysis and evaluation/analysis methods for each implementation outcome

<table>
<thead>
<tr>
<th>Implementation outcome</th>
<th>Definition</th>
<th>Level of analysis (Individual or group conducting the analysis)</th>
<th>Evaluation/ analysis method</th>
</tr>
</thead>
<tbody>
<tr>
<td>Appropriateness</td>
<td>Usefulness and practicality of the research study</td>
<td>Individual providers (Research team)</td>
<td>Analysis of field notes and e-mails from researchers</td>
</tr>
<tr>
<td></td>
<td></td>
<td>Organization (Institutional Review Boards)</td>
<td>Approval of research study and protocol</td>
</tr>
<tr>
<td></td>
<td></td>
<td>Organization (Other relevant organizations [i.e. NSGC])</td>
<td>Literature review and approval by other organizations</td>
</tr>
<tr>
<td>Acceptability</td>
<td>Perception that the study is satisfactory</td>
<td>Individual provider (Research team)</td>
<td>Analysis of e-mails and provider involvement in relevant research activities (i.e. phone conferences, recruitment)</td>
</tr>
<tr>
<td></td>
<td></td>
<td>Individual consumer (Potential participants)</td>
<td>Proportion of participants approached for recruitment at site 1 vs. number who participated at site 1</td>
</tr>
<tr>
<td></td>
<td></td>
<td>Organization (Institutional Review Boards)</td>
<td>Approval of research study and protocol</td>
</tr>
<tr>
<td>Feasibility</td>
<td>Ease at which the study can be implemented</td>
<td>Individual provider (Research team)</td>
<td>Analysis of field notes and e-mails from researchers</td>
</tr>
<tr>
<td>Fidelity</td>
<td>Degree to which the study is implemented as described in the original research protocol</td>
<td>Individual provider (Research team)</td>
<td>Analysis of field notes and e-mails from researchers</td>
</tr>
<tr>
<td></td>
<td></td>
<td>Organization (Institutional Review Boards)</td>
<td>Analysis of IRB notes and requested protocol changes</td>
</tr>
<tr>
<td>Adoption</td>
<td>Uptake of the Study</td>
<td>Individual provider (Research team)</td>
<td>Number of providers and sites that are recruiting</td>
</tr>
<tr>
<td>Ongoing Patient Engagement</td>
<td>Percentage of all eligible participants that complete the full study</td>
<td>Research study</td>
<td>Proportion of eligible patients at site 1 and site 2 who complete the pre-visit survey and proportion who complete both surveys</td>
</tr>
</tbody>
</table>
Acceptability is defined as the perception that the study is satisfactory. Acceptability must be satisfied by research team members and other organizations before other important implementation outcomes can be met and the study can be implemented successfully. The research team, who will be involved in the project, must accept the study, otherwise there will be no one to successfully collect the data. This was evaluated here by analyzing e-mails and provider involvement and satisfaction with the study. Potential participants will not be interested in participation if they do not find the study acceptable. Participant acceptability was analyzed by calculating the number of participants at Site 1 who agreed to participate after a study team member informed them of the study. Site 2 was not used because there are many variables such as not seeing the message, time-constraints, or viewing the message as spam that may prevent participation outside of acceptability. The protocol must be acceptable by the institutional review boards at each site, prior to patient involvement and data collection, which can be measured by the acquisition of successful IRB approval at the research sites.

Adoption is the uptake of a research study or the willingness for the research team to participate in the study and occurs when the research team successfully begins implementing a study in their clinic. Adoption was measured by the number of study team members at the various sites who were willing to adopt the study in their clinics and begin recruitment and data collection.

Appropriateness is the usefulness or practicality of the research. This is essential before the study can begin and is important for development of the study protocol and research ideas. The study must be deemed appropriate by members of the research team because if they do not see any clinical utility to the research, they will not be interested in adopting the study in their clinic. Appropriateness was evaluated by analyzing e-mails and provider involvement in the
development of the study. The IRB at each site must also view the study as appropriate, identifying the benefits as worth the potential risks to patients. It is also important for relevant outside organizations to find the study appropriate, otherwise there will be no potential for future uptake or publications of the findings. The relevance was evaluated using a literature review to determine if the study could potentially fill any research gaps and through approval from other organizations through e-mails or grants.

Feasibility is the ease at which the study can be implemented by researchers. The feasibility of a study is identified by the research team members involved in implementing this study. Feasibility of the study was analyzed by reviewing field notes and e-mails from researchers about the ease of the study and challenges that were identified during data collection and recruitment.

Fidelity is the degree to which the study is implemented as described in the original research protocol. Fidelity was measured by analyzing e-mails and field notes about the implementation process from members of the research team. It was also measured by analyzing notes from IRB approvals and changes requested by the IRB to the research protocol.

Implementation cost is defined as the cost of running a research study and is used to analyze the cost-benefit ratio of a study. There were no significant costs associated with this study. Therefore, we did not think implementation cost to be an essential component for measuring success of this study.

Penetration is institutional spread of a study, or the level at which other institutions uptake the procedures or methods of a study. Sustainability is the level at which a study can continue or be useful long-term. The end goal for this study is that the methods and measures are used in other research endeavors at other institutions. Penetration and sustainability will be
important for future studies. However, penetration and sustainability were not identified as essential components for the implementation of this pilot study and were not analyzed as part of this study.

We found that patient involvement in the research was also an important measurement to analyze success of the implementation of a study. Patient engagement was measured by calculating the proportions of all eligible patients who completed the pre-visit survey and the proportion who completed both the pre- and post-visit surveys.

**Service System**

Although service outcomes were not measured, these were considered when developing the study protocol. Service system measures are defined using the Institute of Medicine’s standards of care, which requires clinical care to be safe, effective, patient-centered, timely, efficient, and equitable (Institute of Medicine (US) Committee on Quality of Health Care in America, 2001; E. Proctor et al., 2011). Consideration of these outcomes explains why we determined that we would not approach people who were clearly emotional because we wanted our research approach to be patient-centered. We also determined that we would not include anyone who could not decide whether to participate quickly because it is important to maintain efficiency of the clinic.
DATA ANALYSIS

Patient/Client Experiences, Values, Attitudes, and Decisions

Demographic information will be analyzed using a chi-square test for categorical variable and ANOVA for continuous variables. Survey data will be analyzed using SPSS. A paired T-test will be run to compare whether there is a difference in pre-visit and post-visit measures of knowledge, competence, and empowerment to make a decision. ANOVA and linear mixed models will be used to compare measures. Participants responses to the written portion of the questionnaire will be analyzed for word and phrase repetitions to determine if any ideas or concepts were commonly mentioned by participants.

Implementation Outcomes

Upon review and discussion of the field notes, e-mails, and institutional review board applications themes and data were pulled out and categorized according to the implementation outcome. Data categorization was reviewed by two of the researchers until consensus was reached in terms of presenting key data and themes for relevant implementation outcome categories. Analysis of participation was measured by analyzing the participation rate. The participation rate is defined as the percentage of people eligible that completed various parts of the study including recruitment, the pre-visit questionnaire, and the post-visit questionnaire.
RESULTS

Patient/ Client Experiences, Values, Attitudes, and Decisions

Preliminary demographic information from the completed pre-visit surveys is summarized in Table 8. The average age of participants was 32 years. Most participants identified as white. All participants had completed at least 11-12th grade, received a GED, or completed equivalent vocational school or higher.

Table 8: Demographic information for participants at site #1 and site #2

<table>
<thead>
<tr>
<th>Variable</th>
<th>Response Categories</th>
<th>n=6</th>
</tr>
</thead>
<tbody>
<tr>
<td>Age, mean (SD)</td>
<td>32 (4.65)</td>
<td></td>
</tr>
<tr>
<td>Ethnicity</td>
<td></td>
<td></td>
</tr>
<tr>
<td>White</td>
<td>3 (50%)</td>
<td></td>
</tr>
<tr>
<td>Black or African American</td>
<td>1 (16.7%)</td>
<td></td>
</tr>
<tr>
<td>Hispanic, Latina, or Spanish</td>
<td>2 (33.3%)</td>
<td></td>
</tr>
<tr>
<td>American Indian or Alaska Native</td>
<td>0 (0%)</td>
<td></td>
</tr>
<tr>
<td>Native Hawaiian or Other pacific Islander</td>
<td>0 (0%)</td>
<td></td>
</tr>
<tr>
<td>Asian</td>
<td>0 (0%)</td>
<td></td>
</tr>
<tr>
<td>Middle Easern, Arab, or North African</td>
<td>0 (0%)</td>
<td></td>
</tr>
<tr>
<td>Prefer not to answer</td>
<td>0 (0%)</td>
<td></td>
</tr>
<tr>
<td>Some other race, ethnicity, or origin</td>
<td>0 (0%)</td>
<td></td>
</tr>
<tr>
<td>Education Level</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Did not complete 6th grade</td>
<td>0 (0%)</td>
<td></td>
</tr>
<tr>
<td>6th-8th grade</td>
<td>0 (0%)</td>
<td></td>
</tr>
<tr>
<td>9th-10th grade</td>
<td>0 (0%)</td>
<td></td>
</tr>
<tr>
<td>11-12th grade, GED, or equivalent vocational school</td>
<td>2 (33.3%)</td>
<td></td>
</tr>
<tr>
<td>Some college</td>
<td>1 (16.7%)</td>
<td></td>
</tr>
<tr>
<td>Graduated college</td>
<td>2 (33.3%)</td>
<td></td>
</tr>
<tr>
<td>Completed postgraduate degree</td>
<td>1 (16.7%)</td>
<td></td>
</tr>
</tbody>
</table>
Data collection will continue at all sites until 30 participants are recruited at each site for a total number of at least 90 participants at all sites. The protocols were altered at collection sites based on information from this feasibility analysis. Protocol alterations and justifications for them will be detailed in this manuscript.

**Implementation Outcomes**

During our evaluation of the implementation of our feasibility study, we found that satisfying certain implementation outcomes were important before other implementation outcomes could be satisfied. We also found that cost, penetration, and sustainability were not relevant implementation outcomes when helping describe success of this and similar studies and analysis of these outcomes was not performed on this study. Patient engagement was not included in the previously reported implementation outcomes and was added for this study.

A summary of the implementation results and relevant themes identified can be found in Table 9. A summary of the challenges and solutions for future studies is summarized in Table 10. Results for each implementation outcomes are described in further detail in the following section.
Table 9: Results and themes identified at each level of analysis for each implementation outcome

<table>
<thead>
<tr>
<th>Implementation Outcome</th>
<th>Individual or group involved in analysis</th>
<th>Results and relevant themes identified</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Appropriateness</strong></td>
<td>Research team</td>
<td>Researchers expressed desire for publication of research noting the importance and relevance of the study</td>
</tr>
<tr>
<td></td>
<td>Institutional Review Boards</td>
<td>Institutional review boards at Site #1 and Site #2 approved study; Site #3 is in the process of approval</td>
</tr>
<tr>
<td></td>
<td>Organizations (i.e. NSGC)</td>
<td>Little research has been done to measure genetic counseling outcomes and develop measures and protocols to analyze outcomes; large research gap deems research significantly appropriate; NSGC Prenatal SIG provided $200 grant for research</td>
</tr>
<tr>
<td><strong>Acceptability</strong></td>
<td>Research team</td>
<td>Met via phone conference and e-mail to discuss study protocol and timeline; actively involved in recruitment of patients; helped create, edit, or approve final survey measures</td>
</tr>
<tr>
<td></td>
<td>Potential participants</td>
<td>100% of patients approached in-person for recruitment at Site #1 agreed to participate in the study</td>
</tr>
<tr>
<td></td>
<td>Institutional Review Boards</td>
<td>Institutional review boards at site #1 and site #2 approved the study, but at site #1 they required signed patient consent, while site #2 considered it exempt given no protected health information was collected with the study data, the ultimate goal was quality improvement of clinical services, and study risks were low; site #3 is in the process of approval</td>
</tr>
<tr>
<td><strong>Feasibility</strong></td>
<td>Research team</td>
<td>Acknowledgement of time constraints in clinic; concerns raised about lack of time for providers to approach patients at site #2; concerns about work load of IRB application; Emotional distress noted among 3 patients at site #1 limiting the number of patients eligible for recruitment</td>
</tr>
<tr>
<td><strong>Fidelity</strong></td>
<td>Research team</td>
<td>Concerns about lack of post-visit participation and need to alter protocol to increase recruitment by approaching them to complete the post-visit survey before they leave; request by research team at site #2 to send survey link before clinic visit to improve feasibility</td>
</tr>
<tr>
<td></td>
<td>Institutional Review Boards</td>
<td>Consent changes required: Request to acquire signed consent at Site #1</td>
</tr>
<tr>
<td><strong>Adoption</strong></td>
<td>Research team</td>
<td>All sites submitted IRB approval; Site #1 and Site #2 began collecting data after approval but site #3 is still awaiting approval</td>
</tr>
<tr>
<td><strong>Patient Engagement</strong></td>
<td>Research study</td>
<td>To date 0% of participants at Site #1 and Site #2 have completed both the pre-visit and post-visit surveys.</td>
</tr>
</tbody>
</table>
Table 10: Challenges encountered for each implementation outcome and solutions to addressing each challenge

<table>
<thead>
<tr>
<th>Implementation Outcome</th>
<th>Challenges</th>
<th>Solutions</th>
</tr>
</thead>
<tbody>
<tr>
<td>Appropriateness</td>
<td>Little research has been done to measure genetic counseling outcomes and develop measures and protocols to analyze outcomes, which creates a large research gap, but makes it more difficult to define what is appropriate</td>
<td>Analyze research that has been used to evaluate health care measures used in other settings and tweak for genetic counseling</td>
</tr>
<tr>
<td>Acceptability</td>
<td>Multi-site study involves many personnel; varying opinions about protocol development; different needs depending on site</td>
<td>Designate primary person to make final calls about protocol and act as a liaison between sites; host meetings involving all personnel in study to ensure satisfaction between all parties; remain open-minded to ideas and the possibility of altering strategies</td>
</tr>
<tr>
<td>Feasibility</td>
<td>Time constraints in clinic; emotional distress of patients limiting number of patients; eligible for recruitment; lack of clarity in researchers’ roles</td>
<td>Develop a protocol with time constraints in mind; communicate with clinics to determine what will work for that site; consider the fact that all patients may not be recruited due to time constraints or emotional distress; incorporate delays in data collection into timeline; communicate efficiently with research team members to ensure roles are clear</td>
</tr>
<tr>
<td>Fidelity</td>
<td>Protocol alterations requested at each site by institutional review board; necessity for more protocol alterations due to lack of patient engagement</td>
<td>Begin communication with IRB personnel early in protocol development; consider the fact that protocols may differ between sites; consider the fact that protocol may need to be altered if it is not working successfully and this requires IRB amendments</td>
</tr>
<tr>
<td>Adoption</td>
<td>Clinic is often limited on time and under-staffed; patient care is prioritized over research endeavors</td>
<td>Ensure researchers involved in study are dedicated to completing study; incorporate delays in data collection into timeline of research project; consider alternative strategies for distributing surveys that may increase feasibility (e.g messaging services)</td>
</tr>
<tr>
<td>Patient Engagement</td>
<td>Patients were not motivated to complete surveys outside of clinic</td>
<td>Consider altering protocol to ensure patients complete both the pre-visit and post-visit surveys in clinic</td>
</tr>
</tbody>
</table>
Appropriateness

Researchers expressed a strong desire for publication of the research, noting the importance and relevance of the study. Institutional review boards at site #1 and site #2 approved the research study through required modifications and site #3 is in the process of approval. A literature review identified that there is a significant gap in research studies that measure genetic counseling outcomes, especially in the prenatal setting. Although some previously validated measures of patient changes could be used (such as the genetic counseling outcomes scale), these are lengthy and thus it is more appropriate and acceptable to have brief measures, which was the impetus for those we are piloting. Furthermore the 28-item genetic counseling outcomes scale does not measure multiple constructs within a single theory. Self-determination theory is simple and had been successfully applied to many other fields, both in and out of health-care. Questions were modified slightly from previously validated measures in order to apply to the study. Measures have also not been applied in this context and it is critical to test them and determine their validity and reliability within the setting they intend to be used for larger studies. Testing the appropriateness of measures is critical because the measures need to be sensitive to change and should capture key aspects related to the REM goals of genetic counseling. The appropriateness of this study was recognized by the National Society of Genetic Counselors Prenatal Special Interest Group when they awarded the study $200 to be applied to the research endeavors.

Acceptability

Given that recruitment success would rely on clinician involvement, we made the clinicians at each site full partners in the study and involved them in all aspects of study design
so as to help increase the likelihood that it would be acceptable to them. Acceptability was noted by the research team’s willingness to participate in many different aspects of the study. Team members communicated via phone conferences and e-mail on multiple occasions throughout the project to discuss study protocol development, recruitment endeavors, and the timeline for the research project. Team members provided feedback and approval of the final survey measures and research methods prior to submitting for IRB approval at the research sites. Two of the sites wrote and signed letters of support for the study. Two individuals completed IRB Human Subjects training in order to participate on the study team at site #1. Two researchers communicate weekly via e-mail to discuss eligible participants approached for recruitment at site #1 and 100% of participants approached for recruitment at site #1 agreed to participate in the study, suggesting that the study is deemed acceptable by patients. Two researchers coordinate weekly via e-mail to distribute the pre-visit surveys to eligible participants at site #2.

Institutional review boards at Site #1 and Site #2 approved the study and Site #3 is in the process of seeing IRB approval.

One of the largest challenges with a multi-site study is the number of personnel involved in development of the protocol. When there are multiple people involved in the process, there is more room for disagreements and difficulty coordinating meetings to discuss aspects of the study to ensure all research team members agree about various aspects of the study. Individuals of the study teams may have varying opinions about protocol development and each site may have different needs in order to successfully recruit patients.

Designating a primary person to make final calls about the protocol and act as a liaison between sites can improve communication between study team members and sites. Regular meetings between all personnel in the study would help to ensure satisfaction between all parties,
but we found it difficult to schedule and thus one person communicated at various times to all the different sites. It is important to remain open-minded to ideas and the possibility of altering strategies. What is acceptable to one site or to one study team member, may not be acceptable to another. Therefore, maintaining good communication is an essential component for ensuring a study is acceptable.

**Feasibility**

The major challenge with feasibility is the time constraints placed on a genetic counseling clinic and the required participation of clinicians in recruitment. Researchers expressed many concerns about the feasibility of the study during protocol development. Researchers were concerned with the time constraints in clinic because it is important to prioritize quality patient care and they worried there would not be enough time to recruit patient and allow them to complete the questionnaire without affecting patient care.

Due to the limitations on time it is more difficult to allow patients the time needed to complete both the pre-visit and post-visit surveys in clinic. These challenges were addressed by developing a research protocol that considered the importance of managing time well and avoiding slowing down clinic. Every clinic has a different structure and flow, so it was important to communicate with each clinic in order to determine what would work best for their location, which was used to guide protocol development.

Researchers were worried about the additional workload the research study would put on them including recruitment endeavors and the IRB approval process and expressed a lack of clarity about their role in the study. Once recruitment began, study team members voiced concerns about the limited number of eligible patients due to some patient’s emotional distress.
prior to a visit. It is inevitable that some patients will be ineligible for the study based on emotional distress or limited time will prevent patients from being recruited for the study; therefore, the timeline for the project should consider these potential and likely delays in data collection.

**Fidelity**

Site #1 and site #2 expressed concerns about the lack of post-visit participation and the need to alter the protocol in order to increase recruitment of participants. At site #1 participants were agreeing to participate and completing the pre-visit survey, but they would not complete the post-visit survey after their session. At site #2 there was a low level of participation using the messenger service to recruit patients. Additional protocol alterations were requested at each site by their respective institutional review boards which made it difficult to design a study with high fidelity. Site #2 allowed the study to go through as exempt, while site #1 required collection of documentation of informed consent, despite our desire to waive documentation of informed consent given the low-risk nature of the study.

Protocol design for a study assessing genetic counseling outcomes is already difficult due to time constraints in clinic and the emphasis on patient care. Design is further complicated by unclear guidelines and extreme variability in the requirements of the institutional review boards. While it is only a temporary solution, it can help to begin communication with the Institutional Review Board personnel early in protocol development. Meeting with these individuals early on allows for the protocol to be developed with their input from the beginning, preventing the need to go back and alter aspects of the protocol during the development process. It is also important
to recognize when the protocol is not working and adjust the methods to improve implementation. These changes are discussed further in the section titled ‘patient engagement’.

**Adoption**

All 3 research sites submitted for IRB approval and site #1 and site #2 began data collection once approval was obtained. Sites expressed that they were willing to participate in the study as long as it did not interfere with patient care or clinic flow. In order to increase the likelihood of adoption, it is important to consider all of the other implementation outcomes and be open to alternative strategies for data collection. The methodology and survey distributions methods at site #1 would not have worked at site #2 due to the limited amount of time available between patients. Therefore, an alternative methodology, which included using messaging services to distribute surveys was developed. When discussing adoption of the study with various sites, it is essential to ensure clinicians are dedicated to completing the study and collecting data, which is why we included the clinicians as research partners.

**Patient Engagement**

As shown in Figure 1, 5 eligible participants have been identified at site #1 and identified to participate in the study. 4 participated only in the pre-visit survey. 1 participated in the post-visit survey but did not complete it. Therefore, there were 0 participants that completed both the pre-visit and post-visit questionnaires at site #1.
As shown in Figure 2, 35 eligible participants have been identified at site #2 and were sent an invitation to participate in the research study. 33 did not participate in any part of the study. 1 participant only completed part of the pre-visit survey and did not complete the demographic information. 1 participant completed the pre-visit survey but did not complete the post-visit survey. Therefore, there were 0 participants at site #2 that completed both the pre-visit and post-visit questionnaires.
Site #3 is still in the process of obtaining IRB approval and data collection is expected to begin in the near future.

After preliminary data collection began, it became clear that the methods used were not collecting data quick enough for the study to be efficient. Many patients at Site #1 are ineligible for the study because they do not meet the inclusion criteria; usually because they are receiving genetic counseling for an indication unrelated to discussing fetal chromosome conditions and more rarely due to being too distraught to be approached about research. Furthermore, at site #1 participants were not completing the post-visit survey. Site #2 had an extensive number of eligible patients on the weekly schedule; however, most of these patients were not completing the survey after being reached out to by messenger service. It was also impossible to message all patients because not everyone had the messenger service activated. Therefore, protocol alterations were discussed in order to improve patient response rates. At site #1, it was decided that patients would be asked to complete the post-visit survey on site as well. Post-visit surveys
would be distributed by a study team member not involved in the genetic counseling session, allowing clinic flow to continue while also ensuring the data is collected. At site #2 study team members discussed recruiting some patients in person, instead of solely using the messenger service. Feasibility of the study was emphasized during protocol development, but it is additionally important to consider patient engagement. It may be necessary to increase provider engagement in order to improve patient engagement even at the expense of making things more feasible through automation.
DISCUSSION

Protocol development for a research study involved in collecting data about patient outcomes is complicated. There are multiple levels that must be considered before data collection can begin in order to improve the chance of success for a study. For example, the researcher should contemplate how they are going to implement the study and consider implementation outcomes such as appropriateness, acceptability, feasibility, fidelity, adoption, and patient engagement. Further complicating studies conducted in a clinical setting is the need to prioritize quality patient care. This means the study should neither interfere with nor reduce the safety, effectiveness, patient-centeredness, timeliness, efficiency, and equitability of clinical care. Patient reported outcomes measured in this study include: knowledge, competence, autonomy-support, relatedness, values, and empowerment to make a decision. Figure 3 summarizes these outcomes in the context of implementation outcomes and quality patient care outcomes.

![Figure 3: Summarization of significant research outcomes, implementation outcomes, and quality patient care outcomes identified](image-url)
During the implementation evaluation of our feasibility study, we further refined and modified Proctor’s model of implementation outcomes to create an updated and more specific model that can be used for future studies in genetic counseling outcomes (Figure 4). One key finding from our evaluation is that implementation outcomes may actually have a hierarchical ordering such that the ability to achieve one outcome may be dependent on whether another outcome is achieved. Furthermore, some implementation outcomes may be more important than others for the successful implementation of genetic counseling outcomes studies. For example, study team members prioritized appropriateness and feasibility in addition to providing quality patient care during protocol development and implementation. Before researchers accepted the protocol, they required information about how it would be feasible in clinic and confirmation that the measurements and methods were appropriate and would be beneficial to the academic community. Once these needs were met and the researcher’s questions were satisfied they agreed to accept and ultimately adopt the protocol into clinic.

As shown in our results, meeting previous implementation needs does not guarantee patient engagement and the successful collection of data relevant to the study. Therefore, we found that patient engagement was also an important outcome to consider when designing a research study and have included it in our model. During our study this required reevaluation of the protocol to increase the levels of patient engagement. If it is essential to alter the protocol at any point in the process due to the inability to satisfy one of the implementation outcomes, then it is valuable to reconsider the implementation outcomes and attempt to satisfy all implementation outcomes before similar studies can have wide-spread adoption, penetration, and sustainability. It would not be efficient to implement a study using previous methods or
measures if those methods and measures were not found to work at any of the small number of sites that participate in the feasibility study. This highlights the value of small scale pilot studies before ramping up to include more sites as this gives us the opportunity to identify what works under various conditions and work out all the changes and modification before recruiting more sites and applying to more IRBs.

**Figure 4:** A model showing the sequence and interaction of implementation outcomes identified in this study

We found in our evaluation that, while researchers may initially view feasibility as the most important aspect of protocol development, this does not always ensure the study will be successful. While our study was easy to implement at site #2 and required low levels of maintenance for researchers, it did not inspire a high level of patient engagement. Whereas the protocol developed at site #2 is more difficult to implement in clinic and requires a higher level of provider engagement but may ultimately lead to a higher level of patient engagement if patients are asked to complete the post-visit questionnaire in clinic by a third party (not one of the clinicians. At Site #2 the goal of using a messenger service was to increase feasibility and ease at which the study could be implemented, but utilization of the messenger service has resulted in a low rate of patient engagement. While it may lower feasibility and require more effort from researchers, it may be beneficial to complete surveys in clinic to improve the level of
patient engagement. We found there to be an inverse relationship between feasibility and patient engagement and a direct relationship between provider/researcher engagement and patient engagement during this analysis study (Figure X). Further study and research is required to evaluate and quantify the relationship between these constructs in different studies, but this is the first to our knowledge to report on an apparent hierarchy and relationships between constructs.

**Figure 5:** A visual representation of the inverse relationship between feasibility and patient engagement and the direct relationship between provider engagement and patient engagement.
CONCLUSION

Surveys involving pre-visit and post-visit data collection in a prenatal clinical setting are ambitious, and it is essential to consider all implementation outcomes and the importance of quality patient care before we can successfully collection patient outcomes data (even though that may be the ultimate goal for clinical research studies). Satisfaction of researchers, potential participants, regulatory organizations (i.e. IRB), and other relevant organizations may be necessary for successful implementation of research or other changes in clinical procedures. Protocol development and research implementation are complicated by the need to balance implementation outcomes, but such considerations may be needed for research and other clinical changes to be successful. It may be necessary to balance tension that may occur from the apparent interdependency of certain implementation outcomes if achieving one leads to a reduction in another implementation outcome and it may not be possible to maximize all implementation outcomes. Patient engagement may be challenging, though it may improve by designing a protocol that involves patients completing both the pre-visit and post-visit surveys while they are still in clinic. Because modifying a protocol takes time, it would be valuable to apply what we have learned in future studies so that researchers can weigh the pros and cons of various approaches in their own particular setting. Research methods for the study analyzed in this paper were shown to be unsatisfactory and protocol alterations may be needed to obtain enough patient reported data to validate the newly created and modified measures (that are based
on Self-Determination Theory) because this is the first time they are being used in a prenatal genetic counseling setting.
REFERENCES


Appendix A: Pre-Visit Survey

Researchers at the University of South Florida, Wake Forest University School of Medicine, and Vanderbilt University Medical Center are trying to improve our prenatal services by conducting a research study. We hope you will help us by answering this short survey. It will take you 5-10 minutes. If you do choose to fill out the survey we will also ask you to do a second short survey after your visit. Your answers will be kept private. We will not ask anything that could be used to identify you. You may choose not to take this survey. We will give you the best care we can whether or not you take this survey.

Please read and sign the consent form first. If you have questions, please ask the person who gave you this survey.

1. The following questions will be used to match the pre-visit survey to the post-visit survey without recording any identifying information about yourself. Please answer these questions to the best of your knowledge to ensure accurate matching of the surveys.
   a. What YEAR were you born?
   b. What are the FIRST TWO LETTERS of your MOTHER’S first name?
      (MOTHER means the person you call mother and could be your birth or adoptive mother.)
   c. What are the FIRST TWO LETTERS of your FATHER’S first name?
      (FATHER means the person you call father and could be your birth or adoptive father.)
   d. How many BROTHERS do you have (Include full and half-brothers)?
   e. How many SISTERS do you have (Include full and half-sisters)?
   f. What is the FIRST letter of your MIDDLE name (If none, write X)?

2. Why are you being seen for today’s visit? (check all that apply)
   □ Something was found on the ultrasound that may be of concern
   □ To find out the sex of my unborn baby
   □ The results of my bloodwork showed a higher chance for my baby to have a genetic condition or birth defect
   □ I have a family history of a birth defect or genetic condition
   □ My doctor asked me to come
   □ I will be age 35 or older at the time of delivery
   □ Other (please explain here)

3. Sometimes, a genetic test can help find out more about the health of your unborn baby. What role do you want to have in choosing whether or not to have a genetic test during your pregnancy? (Choose the one option that fits you best)
☐ Decide all by myself
☐ Have my healthcare provider help me make a decision
☐ Have a partner, family member, or friend help me make a decision
☐ Have my healthcare provider decide what is best for me
☐ Have someone else decide what is best for me
☐ Have my healthcare provider and a partner, family member, or friend help me make a decision

4. **Please answer the following questions about genetic testing during your pregnancy. A few examples of genetic testing include noninvasive prenatal screening (cell free DNA testing), amniocentesis, and quad screen.**

<table>
<thead>
<tr>
<th>I get to decide if I want to have genetic testing.</th>
<th>Yes ☐</th>
<th>No ☐</th>
<th>Unsure ☐</th>
</tr>
</thead>
<tbody>
<tr>
<td>I feel like my healthcare provider will pressure me to have genetic testing.</td>
<td>Yes ☐</td>
<td>No ☐</td>
<td>Unsure ☐</td>
</tr>
<tr>
<td>I have a choice between different types of genetic tests.</td>
<td>Yes ☐</td>
<td>No ☐</td>
<td>Unsure ☐</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>I know the benefits and risks of the genetic testing options.</th>
<th>Yes ☐</th>
<th>No ☐</th>
<th>Unsure ☐</th>
</tr>
</thead>
<tbody>
<tr>
<td>I am clear about which benefits and risks matter most to me.</td>
<td>Yes ☐</td>
<td>No ☐</td>
<td>Unsure ☐</td>
</tr>
<tr>
<td>I have enough support and advice to make a choice.</td>
<td>Yes ☐</td>
<td>No ☐</td>
<td>Unsure ☐</td>
</tr>
<tr>
<td>I feel sure about the best choice for me.</td>
<td>Yes ☐</td>
<td>No ☐</td>
<td>Unsure ☐</td>
</tr>
<tr>
<td>I only want to know about health problems for my unborn baby if there is something that can be done to fix the health problem before birth.</td>
<td>Yes ☐</td>
<td>No ☐</td>
<td>Unsure ☐</td>
</tr>
<tr>
<td>I am willing to take a small chance that there could be a problem because of the test if it means I will find out if my unborn baby has a chromosome condition.</td>
<td>Yes ☐</td>
<td>No ☐</td>
<td>Unsure ☐</td>
</tr>
<tr>
<td>Having genetic testing will make me worry more than if I don’t have genetic testing.</td>
<td>Yes ☐</td>
<td>No ☐</td>
<td>Unsure ☐</td>
</tr>
<tr>
<td>I want to know as much as I can about the health of my unborn baby.</td>
<td>Yes ☐</td>
<td>No ☐</td>
<td>Unsure ☐</td>
</tr>
</tbody>
</table>

5. **Please check yes, no, or unsure next to each statement below. We do not expect you to know the answers to these questions. In fact, many people don’t know this information. If you don’t know please check “unsure”.

| A chromosome condition is caused by a change in the amount of genetic information that a person has. | Yes ☐ | No ☐ | Unsure ☐ |
There is a cure for some chromosome conditions if we find out about it early. | Yes ☐ | No ☐ | Unsure ☐ |
Chromosome conditions are most often inherited from the parents. | Yes ☐ | No ☐ | Unsure ☐ |
Chromosome conditions can cause health problems or learning problems. | Yes ☐ | No ☐ | Unsure ☐ |
A screening test done during pregnancy can tell us for sure if a baby has a genetic condition. | Yes ☐ | No ☐ | Unsure ☐ |
A diagnostic test can sometimes cause a problem with the pregnancy, but most women have no problems after the test. | Yes ☐ | No ☐ | Unsure ☐ |
Screening tests sometimes cause harm to an unborn baby. | Yes ☐ | No ☐ | Unsure ☐ |
A baby could still have a genetic condition even if diagnostic testing is normal. | Yes ☐ | No ☐ | Unsure ☐ |

6. For each of the statements below, please choose the answer that shows how you feel today.

<table>
<thead>
<tr>
<th>I feel I am able to make a decision about prenatal screening and testing.</th>
<th>Strongly disagree</th>
<th>Disagree</th>
<th>Neutral</th>
<th>Agree</th>
<th>Strongly agree</th>
</tr>
</thead>
<tbody>
<tr>
<td>I feel confident I can ask questions if I don’t understand my choices.</td>
<td>Strongly disagree</td>
<td>Disagree</td>
<td>Neutral</td>
<td>Agree</td>
<td>Strongly agree</td>
</tr>
<tr>
<td>I am capable of understanding the information enough to make a decision.</td>
<td>Strongly disagree</td>
<td>Disagree</td>
<td>Neutral</td>
<td>Agree</td>
<td>Strongly agree</td>
</tr>
<tr>
<td>I have the ability to figure out what choice is best for me.</td>
<td>Strongly disagree</td>
<td>Disagree</td>
<td>Neutral</td>
<td>Agree</td>
<td>Strongly agree</td>
</tr>
</tbody>
</table>

7. For each statement below, please circle the number that shows how you feel about having a diagnostic genetic test during your pregnancy. Diagnostic genetic tests include amniocentesis and chorionic villus sampling.

**Having a diagnostic genetic test during my pregnancy is…**

A bad choice for me

| 1 | 2 | 3 | 4 | 5 | 6 | 7 |

A good choice for me

| 1 | 2 | 3 | 4 | 5 | 6 | 7 |

Unimportant to me

| 1 | 2 | 3 | 4 | 5 | 6 | 7 |

Important to me

| 1 | 2 | 3 | 4 | 5 | 6 | 7 |

Harmful

Helpful
8. For each statement below, please circle the number that shows how you feel about having a genetic screening test during your pregnancy. Some examples of genetic screening tests include noninvasive prenatal screening (cell free DNA test), quad screen, and first trimester screen.

**Having a genetic screening test during my pregnancy is...**

<table>
<thead>
<tr>
<th>A bad choice for me</th>
<th>A good choice for me</th>
</tr>
</thead>
<tbody>
<tr>
<td>1 2 3 4 5 6 7</td>
<td>1 2 3 4 5 6 7</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Unimportant to me</th>
<th>Important to me</th>
</tr>
</thead>
<tbody>
<tr>
<td>1 2 3 4 5 6 7</td>
<td>1 2 3 4 5 6 7</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Harmful</th>
<th>Helpful</th>
</tr>
</thead>
<tbody>
<tr>
<td>1 2 3 4 5 6 7</td>
<td>1 2 3 4 5 6 7</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Going to make me worry more</th>
<th>Going to make me worry less</th>
</tr>
</thead>
<tbody>
<tr>
<td>1 2 3 4 5 6 7</td>
<td>1 2 3 4 5 6 7</td>
</tr>
</tbody>
</table>

9. How old are you currently? ___________ years

10. Which of these categories describe you?

a. White
b. Hispanic, Latina, or Spanish
c. Black or African American
d. Native Hawaiian or Other Pacific Islander
e. Middle Eastern, Arab, or North African
f. American Indian or Alaska Native
g. Asian
h. I prefer not to answer
i. Some other race, ethnicity or origin (please list below)

11. What is the last grade or level of school you have completed?

a. Did not complete 6th grade
b. 6th-8th grade
c. 9th-10th grade
d. 11th-12th grade, GED, or equivalent vocational school
e. Some college
f. Graduated college
g. Completed postgraduate degree

12. Please click the following link which will direct you to a separate survey, which will be used to provide a link and reminder e-mail to complete the post-visit survey as well as a copy of the informed consent document. Collecting your e-mail address in a separate
survey prevents it from being connected to your survey responses. This allows your responses to remain anonymous.

LINK
Appendix B: E-Mail Collection Survey

E-mail addresses will be used to provide a link to the post-visit survey and a copy of the informed consent document. E-mail addresses will not be connected to survey data. They will be deleted within a month after the study is completed. If you withdraw from the study, we will delete your e-mail address.

1. What is your e-mail address that you would like to provide for further information related to the study?
Appendix C: Post-Visit Survey

Researchers at the Vanderbilt University Medical Center, University of South Florida, and Wake Forest University School of Medicine are trying to improve our prenatal services. We hope you will continue to help us by answering this short survey about your visit today. It will take you 5-10 minutes.

1. The following questions will be used to match the pre-visit survey to the post-visit survey without recording any identifying information about yourself. Please answer these questions to the best of your knowledge to ensure accurate matching of the surveys.
   a. What YEAR were you born?
   b. What are the FIRST TWO LETTERS of your MOTHER’S first name? (MOTHER means the person you call mother and could be your birth or adoptive mother.)
   c. What are the FIRST TWO LETTERS of your FATHER’S first name? (FATHER means the person you call father and could be your birth or adoptive father.)
   d. How many BROTHERS do you have (Include full and half-brothers)?
   e. How many SISTERS do you have (Include full and half-sisters)?
   f. What is the FIRST letter of your MIDDLE name (If none, write X)?

2. Which genetic counselor did you see for your genetic counseling session to discuss risk from chromosomes conditions (i.e. Down syndrome)?
   - Martha Dudek
   - I shared in the decision with a partner, family member or friend
   - I had my healthcare provider decide what is best
   - I had my healthcare provider decide what is best

3. At your prenatal visit, you may have made a choice about having a genetic test. If so, what role did you play when making that choice? (Choose one)
   - I decided myself after hearing information
   - I shared in the decision with my healthcare provider
   - I shared in the decision with a partner, family member or friend
   - I had my healthcare provider decide what is best
   - I had someone else (other than my healthcare provider) decide what is best
   - I have not discussed genetic testing
   - I have not made a decision about genetic testing

4. How satisfied were you with your role in this decision?
   - Very unsatisfied
   - Very satisfied
   - 1
   - 2
   - 3
   - 4
   - 5
   - 6
   - 7

5. Please answer the following questions about genetic testing during your pregnancy.

   - I know the benefits and risks of the genetic testing options. Yes □  No □  Unsure □
<table>
<thead>
<tr>
<th>Question</th>
<th>Yes □</th>
<th>No □</th>
<th>Unsure □</th>
</tr>
</thead>
<tbody>
<tr>
<td>I got to decide if I wanted to have genetic testing.</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>I feel like the healthcare provider I met pressured me to have genetic testing.</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>I am clear about which benefits and risks matter most to me.</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>I have enough support and advice to make a choice.</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>I feel sure about the best choice for me.</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>I only want to know about health problems for my unborn baby if there is something that can be done to fix the health problem before birth.</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>I am willing to take a small chance that there could be a problem because of the test if it means I can know for certain if my unborn baby has a chromosome condition.</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Having genetic testing will make me worry more than if I don't have genetic testing.</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>I want to know as much as I can about the health of my unborn baby.</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>I had a choice between different types of genetic tests.</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>I got to decide if I wanted to have genetic testing.</td>
<td></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

6. **We want to find out how we can do better at giving you information. During your visit we may not have talked about everything on the list below. We do not expect you to know all the answers. Please check yes, no, or unsure next to each item. If you don’t know for certain please check “unsure”.

<table>
<thead>
<tr>
<th>Fact</th>
<th>Yes □</th>
<th>No □</th>
<th>Unsure □</th>
</tr>
</thead>
<tbody>
<tr>
<td>A chromosome condition is caused by a change in the amount of genetic information that a person has.</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>There is a cure for some chromosome conditions if we find out about it early.</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Chromosome conditions are most often inherited from the parents.</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Chromosome conditions can cause health problems or learning problems.</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>A screening test done during pregnancy can tell us for sure if a baby has a genetic condition.</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>A diagnostic test can sometimes cause a problem with the pregnancy, but most women have no problems after the test.</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Screening tests can sometimes cause harm to an unborn baby.</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>A baby could still have a genetic condition even if diagnostic testing is normal.</td>
<td></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>
7. For each statement below, please circle the number that shows how you feel about having a diagnostic genetic test during your pregnancy. **Diagnostic genetic tests** include amniocentesis and chorionic villus sampling. **Having a diagnostic genetic test during my pregnancy is…**

A bad choice for me | A good choice for me
---|---
1 | 2 | 3 | 4 | 5 | 6 | 7

Unimportant to me | Important to me
---|---
1 | 2 | 3 | 4 | 5 | 6 | 7

Harmful | Helpful
---|---
1 | 2 | 3 | 4 | 5 | 6 | 7

Going to make me worry more | Going to make me worry less
---|---
1 | 2 | 3 | 4 | 5 | 6 | 7

8. For each statement below, please circle the number that shows how you feel about having a genetic screening test during your pregnancy. Some examples of **genetic screening tests** include noninvasive prenatal screening (cell free DNA test), quad screen, and first trimester screen. **Having a genetic screening test during my pregnancy is…**

A bad choice for me | A good choice for me
---|---
1 | 2 | 3 | 4 | 5 | 6 | 7

Unimportant to me | Important to me
---|---
1 | 2 | 3 | 4 | 5 | 6 | 7

Harmful | Helpful
---|---
1 | 2 | 3 | 4 | 5 | 6 | 7

Going to make me worry more | Going to make me worry less
---|---
1 | 2 | 3 | 4 | 5 | 6 | 7

9. The healthcare provider you met with may or may not have talked about a lot of information. Please tell us if you remember each of the following being discussed during your visit.

<table>
<thead>
<tr>
<th>I was told the specific chance that my unborn baby could have a chromosome condition.</th>
<th>Yes ☐</th>
<th>No ☐</th>
<th>Unsure ☐</th>
</tr>
</thead>
<tbody>
<tr>
<td>We talked about what the genetic tests would or would not tell me.</td>
<td>Yes ☐</td>
<td>No ☐</td>
<td>Unsure ☐</td>
</tr>
<tr>
<td>I was told that I might get an unexpected test result.</td>
<td>Yes ☐</td>
<td>No ☐</td>
<td>Unsure ☐</td>
</tr>
<tr>
<td>We talked about how much money the genetic test would cost.</td>
<td>Yes ☐</td>
<td>No ☐</td>
<td>Unsure ☐</td>
</tr>
</tbody>
</table>
10. For each of the statements below, please choose the answer that shows how you feel about your visit.

<table>
<thead>
<tr>
<th>Statement</th>
<th>Strongly disagree</th>
<th>Disagree</th>
<th>Neutral</th>
<th>Agree</th>
<th>Strongly agree</th>
</tr>
</thead>
<tbody>
<tr>
<td>I feel that my healthcare provider gave me choices and options about genetic testing.</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>I feel I am able to make a decision about prenatal screening and testing.</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>I feel totally relaxed with this provider.</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>I feel understood by my healthcare provider.</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>I feel confident I can ask questions if I don’t understand my choices.</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>This provider takes me seriously.</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>I am capable of understanding the information enough to make a decision.</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>This provider really cares for me.</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>My provider is confident in my ability to make decisions.</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>This provider really knows how I feel about things.</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>My provider encouraged me to ask questions.</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>My provider listened to how I would like to do things.</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>My provider tried to understand how I see things before making suggestions.</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>I have the ability to figure out what choice is best for me.</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

11. Did you have any genetic testing? (Choose one)
☐ I am not sure or I don’t remember
☐ I am still deciding if I want testing
☐ No
☐ Yes
If yes, what test did you have? -

12. Choosing whether or not to have genetic testing is often a personal decision. Testing is not always needed or a person may decide that one test is better for them than another. We want to understand how people choose from the different options. Please tell us why you made the choice you did about genetic testing.
13. Part a: What about your visit did you find most helpful?
___________________________________________________________________________
___________________________________________________________________________
___________________________________________________________________________
___________________________________________________________________________
___________________________________________________________________________

Part b: What could we do to be even more helpful in the future?
___________________________________________________________________________
___________________________________________________________________________
___________________________________________________________________________
___________________________________________________________________________
___________________________________________________________________________
Appendix D: Informed Consent Document

Informed Consent to Participate in Research Involving Minimal Risk and Authorization to Collect, Use and Share Your Health Information

Pro # 00036617

You are being asked to take part in a research study. Research studies include only people who choose to take part. This document is called an informed consent form. Please read this information carefully and take your time making your decision. Ask the researcher or study staff to discuss this consent form with you, please ask him/her to explain any words or information you do not clearly understand. The nature of the study, risks, inconveniences, discomforts, and other important information about the study are listed below.

We are asking you to take part in a research study called:

A Pilot Study Comparing Prenatal Genetic Counseling Patient Reported Outcomes Using the Self-Determination Theory

The person who is in charge of this research study is Dr. Deborah Cragun. This person is called the Principal Investigator. However, other research staff may be involved and can act on behalf of the person in charge.

The research will be conducted at University of South Florida Health, Wake Forest University School of Medicine, and Vanderbilt University Medical Center.

Purpose of the study

The purpose of this study is to compare service delivery models at prenatal centers. This will be done using pre-visit and post-visit surveys that will gather information from you about your prenatal visit. This information will be used to improve prenatal genetic services in the future.

Why are you being asked to take part?

We are asking you to take part in this research study because you are receiving prenatal services at a participating center and you may discuss chances of having a baby with a chromosome condition. It is important for us to ask you and other women receiving prenatal genetic services these questions so that we can improve these services in the future.
Study Procedures:
If you take part in this study, you will be asked to:

1. Complete a 5-10 minute survey before the genetic counseling visit on a tablet provided to you or your smart phone. This survey will ask you:
   a. What you already know about prenatal genetic screening and testing
   b. Your personal values and attitudes related to genetic counseling, screening, and testing
2. Provide an email address. This will be kept separate from you survey data and used only to:
   a. Send a link and reminder e-mail to complete the post-visit survey
   b. Send an additional copy of the informed consent document
3. Complete a 5-10 minute survey after the genetic counseling visit using your own personal tablet, smart phone, or computer. This will ask about your:
   a. Knowledge of prenatal genetic screening and testing after the session
   b. Personal values related to prenatal genetic counseling, screening, and testing
   c. Opinions about your prenatal visit.

Total Number of Participants
About 30-35 individuals will take part in this study at USF. A total of 90-100 individuals will participate in the study at all sites.

Alternatives / Voluntary Participation / Withdrawal
You do not have to participate in this research study.

You should only take part in this study if you want to volunteer. You should not feel that there is any pressure to take part in the study. There will be no penalty or loss of benefits you are entitled to receive if you do not take part in this study. The care you receive during your visit will be the same whether or not you participate.

Benefits
The potential benefits of participating in this research study include:

1. Some participants may find satisfaction in contributing to research that is expected to help develop strategies to improve prenatal genetic services. However, other participants may find no benefits to participating.

Risks or Discomfort
This research is considered to be minimal risk. That means the risks associated with this study are the same as what you face every day. There are no known additional risks to those who take part in this study.

We will keep your e-mail address in a separate database from your survey responses. We will do everything we can to keep your e-mail information safe and will only use it for the purposes described for this study. However, we cannot guarantee absolute confidentiality.

Compensation
You will receive no payment or other compensation for taking part in this study.
Costs
It will not cost you anything to take part in the study.

Privacy and Confidentiality
The surveys do not ask for any personal information about yourself. We will ask for your email address, but this will be kept separate from your survey data. We will do our best to keep all your records private and confidential. We cannot guarantee absolute confidentiality. Certain people may need to see your study records. These individuals include:

- The research team, including the Principal Investigator, study coordinator, and research staff at USF.
- The research team members at Vanderbilt and Wake Forest. Information shared with Vanderbilt and Wake Forest will not contain any identifiers as the data will be anonymous.
- Certain government and university people who need to know more about the study, and individuals who provide oversight to ensure that we are doing the study in the right way.
- Any agency of the federal, state, or local government that regulates this research.
- The USF Institutional Review Board (IRB) and related staff who have oversight responsibilities for this study, including staff in USF Research Integrity and Compliance.

We may publish what we learn from this study. If we do, we will not include your name. We will not publish anything that would let people know who you are. It is possible, although unlikely, that unauthorized individuals could gain access to your responses. Confidentiality will be maintained to the degree permitted by the technology used. No guarantees can be made regarding the interception of data sent via the internet. However, your participation in this online survey involves risks similar to a person’s everyday use of the Internet. If you complete and submit an anonymous survey and later request your data be withdrawn this may or may not be possible as the researcher may be unable to extract anonymous data from the database.

You can get the answers to your questions, concerns, or complaints
If you have any questions, concerns or complaints about this study, or experience an unanticipated problem, e-mail Dr. Deborah Cragun at dcragun@health.usf.edu.

If you have questions about your rights as a participant in this study, or have complaints, concerns or issues you want to discuss with someone outside the research, call the USF IRB at (813) 974-5638 or contact by email at RSCH-IRB@usf.edu.

Authorization to Use and Disclose Protected Health Information (HIPAA Language)

The federal privacy regulations of the Health Insurance Portability & Accountability Act (HIPAA) protect your identifiable health information. The only potentially identifying
information that we are asking you for is your e-mail. By giving us your e-mail, you are permitting the University of South Florida to use this information for research purposes. You are also allowing us to share your e-mail with individuals or organizations other than USF who are also involved in the research and listed below.

The following groups of people may also be able to see your health information and may use that information to conduct this research:

- The medical staff and researchers, who are part of this research study, will review your information to determine if you meet the criteria to participate in this study.
- The USF Institutional Review Board (IRB), Vanderbilt University Institutional Review Board, Wake Forest University Institutional Review Board and its related staff who have oversight responsibilities for this study, including staff in USF Research Integrity and Compliance and the USF Health Office of Clinical Research.
- Data Safety Monitoring Boards or others who monitor the data and safety of the study;

Anyone listed above may use consultants in this research study, and may share your information with them. If you have questions about who they are, you should ask the study team. Individuals who receive your health information for this research study may not be required by the HIPAA Privacy Rule to protect it and may share your information with others without your permission. They can only do so if permitted by law. If your information is shared, it may no longer be protected by the HIPAA Privacy Rule.

By signing this form, you are giving your permission to use and/or share your information as described in this document. As part of this research, USF may collect, use, and share the following information:

- Your e-mail address

You can refuse to sign this form. If you do not sign this form you will not be able to take part in this research study. However, your care outside of this study and benefits will not change. Your authorization to use your health information will not expire unless you revoke (withdraw) it in writing. You can revoke this form at any time by sending a letter clearly stating that you wish to withdraw your authorization to use your health information in the research. If you revoke your permission:

- You will no longer be a participant in this research study;
- We will stop collecting new information about you and will not e-mail you a link to the follow up survey;
- We will use the information collected prior to the revocation of your authorization. This information may already have been used or shared with others; and
- Staff may need to follow-up with you if there is a medical reason to do so.

To revoke this form, please write to:

Principal Investigator: Deborah Cragun
For IRB Study # Pro00036617
Interdisciplinary Research Building
3720 Spectrum Blvd, Suite 304
Tampa, FL 33612

While we are conducting the research study, we cannot let you see or copy the research information we have about you. After the research is completed, you have a right to see the information about you, as allowed by USF policies. You will receive a signed copy of this form.
Consent to Take Part in this Research Study
And Authorization to Collect, Use and Share Your Health Information for Research

I freely give my consent to take part in this study and authorize that my health information as agreed above, be collected/disclosed in this study. I understand that by signing this form I am agreeing to take part in the research. I have received a copy of this form to take with me.

_______________________________________________________________  ________________________
Signature of Person Taking Part in Study                    Date

_______________________________________________________________
Printed Name of Person Taking Part in Study

Statement of Person Obtaining Informed Consent
I have carefully explained to the person taking part in the study what he or she can expect from their participation. I confirm that this research subject speaks the language that was used to explain this research and is receiving an informed consent form in their primary language. This research subject has provided legally effective informed consent.

_______________________________________________________________  ________________________
Signature of Person obtaining Informed Consent                    Date

_______________________________________________________________
Printed Name of Person Obtaining Informed Consent
Appendix E: IRB Approval Confirmation

January 3, 2019

Deborah Cragun, PhD
Global Health
3720 Spectrum Blvd
Suite 304
Tampa, FL 33612

RE: Expedited Approval for Initial Review
IRB#: Pro0036617
Title: A Pilot Study Comparing Prenatal Genetic Counseling Patient Reported Outcomes Using the Self-Determination Theory

Study Approval Period: 1/3/2019 to 1/3/2020

Dear Dr. Cragun:

On 1/3/2019, the Institutional Review Board (IRB) reviewed and APPROVED the above application and all documents contained within, including those outlined below.

Approved Item(s):
Protocol Document(s):
Prenatal Study Protocol, Version 1, 11-25-18

Consent/Assent Document(s)*:

*Please use only the official IRB stamped informed consent/assent document(s) found under the "Attachments" tab. Please note, these consent/assent documents are valid until the consent document is amended and approved.

It was the determination of the IRB that your study qualified for expedited review which includes activities that (1) present no more than minimal risk to human subjects, and (2) involve only procedures listed in one or more of the categories outlined below. The IRB may review
research through the expedited review procedure authorized by 45CFR46.110. The research proposed in this study is categorized under the following expedited review category:

(5) Research involving materials (data, documents, records, or specimens) that have been collected, or will be collected solely for nonresearch purposes (such as medical treatment or diagnosis).

(7) Research on individual or group characteristics or behavior (including, but not limited to, research on perception, cognition, motivation, identity, language, communication, cultural beliefs or practices, and social behavior) or research employing survey, interview, oral history, focus group, program evaluation, human factors evaluation, or quality assurance methodologies.

Your study qualifies for a waiver of the requirements for the process of informed consent as outlined in the federal regulations at 45CFR46.116(d) which states that an IRB may approve a consent procedure which does not include, or which alters, some or all of the elements of informed consent, or waive the requirements to obtain informed consent provided the IRB finds and documents that (1) the research involves no more than minimal risk to the subjects; (2) the waiver or alteration will not adversely affect the rights and welfare of the subjects; (3) the research could not practicably be carried out without the waiver or alteration; and (4) whenever appropriate, the subjects will be provided with additional pertinent information after participation. (Patient schedules and medical records)

Your study qualifies for a waiver of the requirement for signed authorization as outlined in the HIPAA Privacy Rule regulations at 45CFR164.512(i) which states that an IRB may approve a waiver or alteration of the authorization requirement provided that the following criteria are met (1) the PHI use or disclosure involves no more than a minimal risk to the privacy of individuals; (2) the research could not practicably be conducted without the requested waiver or alteration; and (3) the research could not practicably be conducted without access to and use of the PHI. A partial waiver of HIPAA Authorization is granted for recruitment/screening only; signed Authorization will be obtained as part of the informed consent process. Pursuant to this partial waiver, the study team is allowed to review PHI from the USF OB/GYN’s weekly patient schedule and medical records (EPIC) of adult pregnant patients scheduled to receive genetic counseling services at USF to confirm eligibility as outlined in the protocol.

45 CFR 46, Subpart B This research involving pregnant women and/or fetuses was approved under 45 CFR 46.204

As the principal investigator of this study, it is your responsibility to conduct this study in accordance with IRB policies and procedures and as approved by the IRB. Any changes to the approved research must be submitted to the IRB for review and approval via an amendment. Additionally, all unanticipated problems must be reported to the USF IRB within five (5) business days.

We appreciate your dedication to the ethical conduct of human subject research at the University of South Florida and your continued commitment to human research protections. If you have
any questions regarding this matter, please call 813-974-5638.

Sincerely,

Kristen Salomon, Ph.D., Chairperson
USF Institutional Review Board
ABOUT THE AUTHOR

Lindsey Nicole Victoria grew up in Frankfort, Illinois, which is a suburb outside of Chicago. She was raised with her younger brother and sister and moved to Florida with her family when she was twenty-one. She graduated from Florida Gulf Coast University with her Bachelor of Science in Biology degree in 2014. While in school, she decided to pursue a career in genetic counseling. She will be graduating from University of South Florida with her Master of Science in Public Health with a concentration in Genetic Counseling degree in May 2019. She experienced a variety of genetic counseling specialties while in school and has been most drawn to prenatal. When she graduates, she plans to work as a prenatal genetic counselor.