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Amish Perspectives of the Genetic Counseling Process

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Dedication

I would like to dedicate this study to the Amish, who have graciously taught us so much.

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Abstract

The Amish are a population with a high concentration of genetic disorders who have informed our understanding of several genetic conditions. This culturally unique group has special need for genetic services. While clinics have been established to care for Amish individuals, such as the Community Health Clinic in Indiana, little research has been done on Amish perspectives of these services, specifically genetic counseling. Amish individuals who received genetic counseling from the Community Health Clinic were sent recruitment letters and a questionnaire via mail. The questionnaire consisted of demographic questions, a 7-item adapted Genetic Counseling Satisfaction Scale (GCSS), and open-response questions. Thirty-three individuals completed the questionnaire. The majority of participants were aware they were receiving genetic counseling (81.8%), and most participants received genetic counseling from a genetic counselor (54.5%) versus a geneticist (39.4%). The mean satisfaction scores for each 5-point Likert-scale question showed that overall, participants were satisfied with their experience with genetic counseling, with mean scores ranging from 4.58 to 4.77. Descriptive and univariate statistics showed some statistically significant differences in satisfaction when comparing males versus females, referral type, and whether the participant saw a genetic counselor or a geneticist. Open-ended responses also showed that participants were satisfied with their genetic counseling. These responses centered around three themes: rapport-building aspects of the session, cultural appropriateness of the session, and the impact of the

session on medical decision-making. In this first study to explore Amish perceptions of a genetics clinic tailored to their particular way of life, we found that the Amish served by the Community Health Clinic felt respected, that their care was culturally sensitive, and they were satisfied with the genetic services they received.

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Chapter 1: Background

1.1 Genetic counseling and culture

Genetic counseling is a process inherently tied to the culture of both the counselor and the counselee. The term culture can be defined as, "shared values, goals, expectations, beliefs, perceptions, and behaviors" among a group of people who share similar origins and styles of living (Randall-David, 1989). Common topics addressed in a genetic counseling session include reproductive decision making, health, and disability. Beliefs, practices and values surrounding these issues are strongly tied to a person's world view (Weil, 2001). Culture is an integral component of the genetic counseling process and a significant aspect in providing successful and effective care (Weil, 2000). The American Board of Genetic Counseling (ABGC) practice-based competencies include expectations about cultural competency, and the ability to tailor sessions to meet the needs of culturally different patients ("ABGC - Genetic Counseling Standards & Best Practices | ABGC," 2015.). Cultural competency can be defined as "the ability to provide skilled treatment to members of diverging cultural backgrounds through the use and knowledge of differing cultures, and self-awareness of one's attitudes, beliefs, behaviors, and biases linked to culture and cultural differences" (Leroy, Veach & Bartels, 2010, p. 227).

The Genetic Counseling Competence Toolkit serves as a resource to improve cultural competency in the profession. The "culturally humble" genetic counselor is one who realizes that each cross-cultural interaction can be viewed as a learning opportunity

and that lessons learned should be shared. Genetic counselors should be open and willing to try new approaches and ask their patients to share their stories and experiences with them (Warren, 2011).

Many studies exist in the literature that focus on specific cultural groups and how genetic services are perceived and delivered (Awwad, Veach, Bartels, & LeRoy, 2008; Baars, van Dulmen, Velthuizen, van Riel, & Ausems, 2017; Barlow-Stewart et al., 2006; Kinney, Gammon, Coxworth, Simonsen, & Arce-Laretta, 2010; Kowal, Gallacher, Macciocca, & Sahhar, 2015; Mittman, Bowie, & Maman, 2007; Thompson et al., 2015; Tsai et al., 2017). While it is impossible to summarize any particular culture, and people of a certain culture do not necessarily all share the same views and behaviors, it is still important to explore different cultural responses to genetic services. Several common themes emerge among the current literature on culture and genetic services.

A commonality among many non-Western cultures is community or family-based decision making. Individualism is a major component of Western medicine, whereas many cultures have a strong sense of community that extends to health care (Weil, 2000). Southeast and East Asian women living in the U.S. that were interviewed with regard to prenatal genetic counseling said that pregnancy decisions would be made as a family rather than as an individual (Tsai et al., 2017). Similarly, Chinese-Australians interviewed regarding genetics services expressed that senior family members should be involved as they typically play a role in decision-making for the family (Barlow-Stewart et al., 2006). In Orthodox Jewish tradition, the Rabbi provides mediation between modern health technology and ancient laws, and is therefore involved in health care utilization among this community (Mittman et al., 2007). Genetic counselors with experience

working with Indigenous Australians said that engaging family members in decision-making dialogue can be important as many Indigenous people involve community elders in their decisions (Kowal et al., 2015). These studies emphasize the need for inclusion of family and/or community members in decision making among certain cultures.

In cross-cultural genetic counseling, establishing rapport plays an even more essential role than in a typical session (Weil, 2001). Cultural needs described by genetic counselors who have worked with Indigenous Australians include spending extra time on rapport. This can likely be attributed to the population's distrustful attitude towards health care services stemming from their previous negative experiences with government policies (Kowal et al., 2015). Turkish and Moroccan participation in genetic counseling in the Netherlands was also found to be affected by previous negative experience with health care services (Baars et al., 2017). It can prove helpful to ask about what the patient understands about the indication for their appointment as this allows them to tell their story. Additionally, if any distrust is voiced by a patient the counselor could respond in a non-defensive manner to help open conversation even further (Weil, 2001).

Communication issues can create barriers to genetic counseling. In a study focused on the Latino community and amniocentesis refusal, Mexican-origin women did not understand that the protein discussed in relation to prenatal screening is not affected by the protein in their diets. For example, one woman who screened positive for a chromosome abnormality said she would simply eat more protein. Additionally, the non-directive nature of genetic counseling was a source of miscommunication for some women. One participant said that she did not want the amniocentesis because everyone was calm and reassuring (Browner, Mabel Preloran, Casado, Bass, & Walker, 2003).

Similar communication issues were found in a study regarding Latinos' attitudes towards cancer genetic counseling. Participants suggested providing clear definitions of "gene" and "mutation," and to limit unnecessary information such as detailed information on genetics (Kinney et al., 2010). Participants of a another study on Latina perceptions of prenatal genetic counseling suggested providing information regarding what genetic counseling is and what to expect beforehand as well as providing written or print resources during the session as ways to improve communication issues (Thompson et al., 2015).

Certain beliefs among cultures can also affect genetic counseling. For example, Latinos' interviewed regarding cancer genetic counseling brought up cultural taboos about cancer. In their culture, cancer can be viewed as contagious and is often seen as a death sentence. There is also shame associated with cancer, thus it is often not discussed (Kinney et al., 2010). Chinese-Australians also mentioned that cancer is a taboo topic and that illness is often not discussed in their culture (Barlow-Stewart et al., 2006). In Turkey and Morocco, cancer is also not spoken of and is seen as a death sentence (Baars et al., 2017). These cultural beliefs can prevent patients from obtaining genetic counseling and are important to be aware of when taking a family history as individuals may not be aware of cancer in their families.

There are differing beliefs among different cultures regarding having a child with disabilities. Southeast and East Asian individuals interviewed about prenatal genetic counseling described shame that would be associated with the family if they had a child with a disability, and that community members might think that the child is punishment for a wrongdoing in a past life (Tsai et al., 2017). Native Palestinians responding to

hypothetical prenatal situations expressed that they would not want to marry someone with a family history of intellectual disability (Awwad et al., 2008). Latinas interviewed about prenatal genetic counseling did express the desire for a healthy baby, however, they did not feel the need for prenatal diagnosis, citing the desire to leave it up to God (Thompson et al., 2015). Weil explains that the dominant U.S. culture perceives nature as something that can be understood and potentially changed by humans. Other cultures have a more accepting perspective of nature and thus are more accepting of those with disabilities and take issue with some of the testing options offered in genetic counseling sessions such as prenatal diagnosis (2000).

In many of the studies previously discussed, participants expressed that they saw benefits to genetic counseling. Latinos felt that cancer genetic counseling could open the doors to earlier treatment, provide information for family members, and encourage more frequent screening. Members of this community felt that cost would be a barrier to pursuing genetic counseling, however, as many Latinos are low-income and uninsured (Kinney et al., 2010). Turkish and Moroccan individuals in the Netherlands noted similar benefits to genetic counseling, such as knowing more about personal and family risks (Baars et al., 2017). In the prenatal setting, Latinas felt that learning the risk for a chromosome abnormality in their pregnancy was a helpful part of the session and made them feel reassured (Thompson et al., 2015).

Exploring and understanding these cultural beliefs is an important part of a genetic counselor's role. In order to best serve individuals of different cultures, genetic counselors need to know about the beliefs and values these cultures share as this promotes cultural competency and provides patients with the most appropriate care.

1.2 The Amish

One group of people with a unique culture, the Amish, have contributed a great deal to our understanding of genetics (Strauss & Puffenberger, 2009). The Amish are members of a Christian church formed out of the Anabaptist movement in South Germany and Switzerland in 1525 (Cates, 2014). The Anabaptists believed in adult baptism and that an individual should be able to make an informed choice about their faith as opposed to being born into one. As the Anabaptists were not Catholic or traditional Protestant, they faced persecution for their beliefs in the 16th-century. An early leader of the Anabaptist movement was Menno Simons and those who still follow his teachings are known as Mennonites (Francomano, 1996). Under the leadership of Jakob Ammann, the Amish separated from the Mennonites in 1693 because they felt they needed to be more isolated from those who did not follow their belief system (Kowal et al., 2015). Ammann advised a "sharper separation from the world and more severe shunning of unrepentant members." Because of the persecution they faced in Europe, the Amish accepted William Penn's offer of religious tolerance in Pennsylvania in the 1740's, and their last congregation in Europe died out in 1936 (Cates, 2014). Several other groups have roots in the Anabaptist movement. Today, Amish, Old Order and Conservative Mennonites, Old Order Brethren, and Hutterites are collectively referred to as "Plain" people, owing to their plain dress and simple way of life (Strauss & Puffenberger, 2009).

Today, there are 330,270 Amish individuals in North America, with 63% living in the states of Ohio (Holmes and Wayne Counties), Pennsylvania (Lancaster and Mifflin Counties), and Indiana (Elkhart and Lagrange Counties). ("Amish Population Profile,

2018," 2018; Francomano, 1996). Approximately 40 "orders" of Amish exist, each varying in terms of their degree of separation from the world, technology usage, and adherence to tradition, and over 475 settlements that span 30 states including Ontario (Cates, 2014). Among the 40 Amish "orders" there are Old Order Amish, Swartezentruber Amish, New Order Amish and Beachy Amish. The Old Older Amish is the largest group in the United States. The Swartzentruber Amish are more conservative than the Old Order Amish and are unlikely to utilize modern health care, while the New Order Amish are perceived as less conservative. The Beachy Amish are the least conservative and typically are accepting of modern medicine (Francomano, 1996). Amish group range from "low" groups of Amish communities, seen as more traditional, to "higher" groups that have more interaction with the world. The openness a group of Amish might feel toward those of the outside world can depend greatly on whether they belong to a more traditional or more progressive group (Cates, 2014).

Amish culture is deeply intertwined with religion. Amish communities are divided into "church districts" that are typically based on the population of an area, and the congregation is known as the Gmay. A Gmay usually consists of 75 to 150 individuals. Four to five "ministers" lead each congregation, including a bishop, a deacon, and several preachers. The bishop is the leader of the congregation and is responsible for baptisms, weddings, and similar rituals. The deacon is responsible for financial aspects of the Amish community, including health care and fire or storm damage (Cates, 2014). The clergy is elected by the community. In this patriarchal society, senior bishops hold authority among the clergy. The bishop's authority is granted by God and he is responsible for the physical and spiritual health of his congregation. If the

bishop perceives an outside service to be in conflict with their religious views, he can prohibit use of this service (Miller-Fellows, Adams, Korbin, & Greksa, 2018). At home, the husbands and fathers hold authority, however mothers and fathers typically make decisions together (Francomano, 1996).

The core beliefs of the Amish center around living simply and separately from the rest of the world. The Amish typically avoid modernization and physical connections to the outside ("English") world. To accomplish this, they prefer no or limited use of electricity, wearing plain dress, and using horse and buggy for transportation purposes, and pacifism. The practices by which each congregation of Amish people live by is taught and upheld through *Ordnung*, which is an oral tradition that advises on how to live. This includes rules about clothes, technology, higher education and divorce, among other things. *Ordnung* details might be slightly different from community to community (Cates, 2014). Amish culture is rooted in ordinary, day-to-day life practices, and *Ordnung* provides instructions for these practices to help accomplish the goal of peaceful, supportive community (Francomano, 1996).

The pervasiveness of religion in Amish culture extends to matters of health. Any illness is seen as a reflection of God's will. Additionally, Amish do not see illness through the lens of symptoms, but rather as an inability to perform their typical duties such as farming or housekeeping. Therefore, Amish individuals may not present to medical care until an illness is severe. Modern medicine is not prohibited in Amish communities, but typically home remedies and folk medicine are preferred. Health care professionals working with the Amish should therefore be accepting of their use of alternative medicine, if it does not cause harm. The Amish may choose not to use medical

interventions, and typically weigh the degree to which an intervention would disrupt the family or community. An additional factor in Amish utilization of health care is their religious proscription against the use of insurance. The Amish believe that commercial insurance takes away from the concept of leaning on their own community for support (Miller-Fellows et al., 2018) This means that medical interventions can be costly. The Amish do, however, have a system in place in which the community pools finances to be used in case a member of the community needs medical care (Francomano, 1996).

Family is also an important aspect of Amish culture. Amish adults are rarely unmarried, and it is important to note that marriages are not arranged. The Amish do intermarry within their cultural group as they believe in staying separate from the outside world (Francomano, 1996). On average, Amish couples have five to eight children (Cates, 2014). The Amish keep meticulous genealogical records to honor their families and provide stability and foundation for where the current generation fits in. When entering church, the Amish walk in according to age, so an Amish individual will walk behind and in front of the same people their entire life (Francomano, 1996).

Amish education is guided by their culture as well. Typically, Amish schools consist of one room where children of all ages learn in English. Children are usually taught by an unmarried woman who has recently completed her education. Subjects such as reading, writing, arithmetic and world geography are taught until 8th grade. Once a child has completed 8th grade, education moves into the home where girls learn about homemaking and boys study farming. In order to satisfy state requirements, children keep a journal of what they have learned until age 16 that a teacher will review. At home, a form of German called “Pennsylvania Dutch” is typically spoken and learned from

parents, and at church a form of High German, distinct from Pennsylvania Dutch, is used. Therefore, many Amish individuals are trilingual (Francomano, 1996).

In a health care situation, there are several things to keep in mind to have a culturally competent interaction with an Amish individual. Amish individuals do not typically use formal titles and prefer the use of first names. Therefore, rapport can be built using a professional title and then first name, such as "Nurse Bill." Additionally, Amish children are often referred to as their father's name followed by their first name, for example "Eli's Adam." This can be helpful in distinguishing individuals who share the same name, especially since there are only a handful of last names in these communities. Health care professionals should also keep in mind that English is usually a second language for Amish people, so they may not communicate as well but this should not be conflated with a lack of intelligence or literacy. However, biological and medical principles are not an area of focus in Amish schooling, thus professionals should take the time to clearly define words and use visual aids when explaining concepts. Finally, photography is a taboo subject in Amish communities. Some Amish families will allow the use of photography if they feel it will only be used for medical purposes, but others refuse to allow it (Francomano, 1996).

With these aspects of Amish culture in mind, health services in these communities must be provided by those who have a good understanding of the Amish and their beliefs. When services are provided, the experience that Amish individuals have with this service spreads throughout the community quickly. Therefore, it is important to provide services carefully and thoughtfully, and potentially include family members and the bishop in these conversations (Miller-Fellows et al., 2018).

1.3 The Amish and healthcare

Cultural aspects of the Amish affect the uptake in medical services, including genetic services. One clear example is the use of newborn screening among the Amish. Newborn screening is difficult to accomplish in the Wisconsin Plain population, which includes both Old Order Amish and Mennonite communities, for a variety of reasons (Kuhl et al., 2017). The main barrier is the lack of medical follow-up due to lack of medical insurance, the difficulty of getting to a medical facility, and the idea that a doctor does not need to be seen unless a child is seriously ill (Kuhl et al., 2017). In order to gain a better understanding of newborn screening in the Wisconsin Plain population, Sieren et al. (2015) surveyed about their utilization of newborn screening and their attitudes towards it. In Wisconsin, around 2% of births occur outside of hospitals, and the majority of those occur among Plain communities. The study found that 40% of households screened all their children, and of those who responded to the survey, most thought that the screening was important. Of note, one of the main reasons that individuals did not have their children screened was due to lack of awareness about newborn screening.

In an attempt to help alleviate the issue of the lack of newborn screening among the Amish, the Wisconsin Plain population was provided with carrier testing kits to be distributed via midwives and at community meetings (Kuhl et al., 2017). In order to build trust with the community, outreach activities with an educational component were performed that included community elders at community meetings. While the carrier testing kits did not identify anyone that had not already been diagnosed, the study was successful at gaining trust and providing information about the importance of newborn screening to this community through their outreach portion of the methods. These

findings speak to the idea that health care services must be provided in a culturally sensitive fashion.

Another study assessed cancer screening practices among the Amish in Ohio Appalachia (Katz et. al, 2011). Like the newborn screening survey, the study found that cancer screening among Amish was significantly lower compared to non-Amish individuals in the same area. It was found that Amish individuals did not think screening was necessary as they did not believe they were likely to get cancer. The authors believe this may be due to the hesitation the Amish have in utilizing preventative medicine.

The Amish are also known for having low immunization rates. A study done on the Amish population in Holmes County, Ohio aimed to discover the reason for these low rates (Wenger, McManus, Bower, & Langkamp, 2011). The immunization rate in this population at the time of the study was 45% as compared to national rate of 85%. The study found that the Amish weighed the possibility of adverse effects of the vaccines more heavily than other factors such as financial burden, accessibility or religious issues when deciding whether to vaccinate their children. Interestingly, many participants in this study allowed their children to have some vaccines but not all of them.

A special concern within Amish communities is health literacy. Formal education stops in 8th grade in Amish communities and continues at home with a focus on learning how to farm and run households (Francomono, 1996). Those who have a limited education usually have lower health literacy, and health literacy is likely a predictor of how well individuals understand and utilize genetic information (Lea, Kaphingst, Bowen, Lipkus, & Hadley, 2011). Health literacy also has implications for how individuals utilize health services and how it impacts health behaviors. In order to assess health

literacy in Amish communities in Ohio Appalachia, Amish and non-Amish individuals completed interviews and were given the rapid estimate of adult literacy in medicine (REALM) instrument to compare the two groups (Katz, Ferketich, Paskett, & Bloomfield, 2013). The REALM scores of the Amish individuals were significantly lower than non-Amish, indicating limited health literacy. The authors suggest that culture is a contributing factor to the limited health literacy, citing the unique educational practices among the Amish, and the limited exposure to media and technology.

1.4 The Amish and medical genetics

In 1962, Victor McKusick learned of the Amish population's high incidence of achondroplasia. He recognized that two recessive conditions, Ellis-van Creveld syndrome and cartilage-hair hypoplasia, were present in the population and were incorrectly diagnosed as achondroplasia (Strauss & Puffenberger, 2009). This sparked the beginning of many genetic studies on the Amish population. McKusick (1964) realized that this population was an excellent source to study genetic disease due to their geographical isolation, high rate of consanguinity and recessive conditions, large family sizes, and excellent genealogy records. However, these initial years of studying the Amish did not have a focus on providing clinical services to the Amish, and many research subjects (affected with genetic conditions) passed away due to the lack of services (Strauss & Puffenberger, 2009).

In 1989, Dr. D. Holmes and Caroline Morton opened the Clinic for Special Children in Lancaster County, Pennsylvania. This clinic serves the Amish and Mennonite populations of children affected by genetic conditions and operates on the idea that research and clinical care go hand in hand. The Clinic for Special Children has given the

Amish a logistically feasible place to receive care. The Clinic has a history of incorporating advanced biochemical and genetic laboratory techniques into their care. Dr. Puffenburger, a molecular geneticist, was hired in 1998 to help advance the Clinic's molecular techniques. Dr. Strauss, the Medical Director, plays a large role in the research and clinical aspects at the Clinic for Special Children. The Clinic serves over 1,000 patients with over 264 known genetic conditions. Many specialty services are also made available to patients as part of the Clinic's vision to be a medical home for the individuals they treat ("History & Mission," 2016).

The Clinic for Special Children is funded through annual auctions, donations, collaborative relationships, and fees paid by patients ("Our Impact," 2016). Four annual quilt auctions are organized by members of the Plain community that contribute to one third of the budget. The Clinic has always worked to keep costs minimal for their patient population through efforts such as utilization of publicly available molecular information and reducing hospitalizations and laboratory costs. For example, in 2010, the $1.5 million dollar operating budget saved the community an estimated $20 million in medical costs. Keeping costs minimal is important for their patient population, as 95% are uninsured (Strauss, Puffenberger, & Morton, 2012).

In 2008, The Community Health Clinic (CHC) was established in Topeka, Indiana. Its inception was based on the need for a more logistically reasonable place for children affected by genetic disorders to receive care ("Our Story," n.d.). Since it opened, Amish individuals have been receiving genetic counseling from both physicians and genetic counselors. The Community Health Clinic is like the Clinic for Special Children as many health care services are provided to Amish individuals in one place, including

newborn screening services, speech therapy, audiology and nutrition services. They also maintain low costs for patients and rely on donations and fundraisers such as auctions to provide for their patients. Additionally, for some patients, the CHC makes house visits to increase compliance.

The CHC has also helped with other aspects of care. Within the community that CHC serves, a healthcare sharing ministry has been established called the Plain Church Group Ministry (PCGM). Amish deacons and members of this group negotiate with hospitals to obtain discounts for their community needs. For example, PCGM has discounted rates for formula that children with metabolic conditions need. They can get formula for wholesale price and then a 2% administrative fee is charged by the PCGM to cover their services. In order for PCGM to be formalized, Amish individuals must carry a card that shows they are members of PCGM. Many hospitals and medical groups, including CHC, send bills directly to PCGM. If an Amish family asks for help or the bill exceeds $3500, PCGM will contact the deacon to help with cost sharing. When the family receives the bill, they pay however much they can afford and then the rest is covered by a fund that PCGM has access to. If an Amish individual feels strongly that they do not want to bill through PCGM then they can pay a self-pay price (R. Evans, personal communication, April 8, 2019).

Another aspect that makes the CHC unique is the length of time of the appointments. A new patient appointment is scheduled to be 2-3 hours. This initial appointment may or may not include genetic counseling, and sometimes patients come back for genetic counseling. Ultimately, the physician at the clinic spends a great deal of time with patients (R. Evans, personal communication, April 8, 2019).

As for testing, CHC does whole exome sequencing through the CSC on a research basis, so the turnaround time can be lengthy. Other testing is done through standard labs such as Invitae that have reasonable turnaround times. (R. Evans, personal communication, April 8, 2019).

While the established clinics provide much needed genetics care to Amish individuals, no studies to date detail patient perceptions of the genetic counseling process or their experiences with genetic counseling. One study, however, sought to examine the Amish population's general knowledge of genetic disorders and attitudes towards medical care, including genetic counseling by interviewing 17 Amish families were interviewed in Lancaster County, Pennsylvania (Brensinger & Laxova, 1995). Only three of 12 families with a child with a genetic disorder knew about genetic counseling services, and only two were able to provide an explanation of genetic counseling. The majority of those interviewed, however, would have considered genetic counseling and wanted to know more about their child's condition.

1.5 Genetic conditions in the Amish

Many genetic conditions affect the Amish and Mennonite populations. Two conditions that were first treated in the Amish and Mennonite populations are maple syrup urine disease (MSUD) and glutaric acidemia Type 1 (GA1). MSUD is an autosomal recessive disorder in which the body cannot process leucine, isoleucine, and valine. The byproducts of these amino acids build up in the body causing a sweet odor in the urine. Poor feeding, abnormal movements, delayed development are hallmark symptoms, and if untreated, it can cause seizures and death. The general population

incidence of this condition is 1 in 185,000, while the Old Order Mennonite population has an estimated incidence of 1 in 380 (Strauss, Puffenberger, & Morton, 1993).

GA1 is also a recessive condition that affects the body's ability to process amino acids. Build-up of the amino acids lysine, hydroxylysine and tryptophan can cause damage to the basal ganglia and result in issues with movement as well as intellectual disability. The general population incidence of this condition is estimated to be 1 in 30,000 to 1 in 40,000, while the Amish population incidence is 1 in 300 (Strauss, Puffenberger, Robinson, & Morton, 2003, p. 1).

Table 1 provides a selected list of many other genetic conditions that affect the Amish population, adapted from Strauss and Puffenberger's "Genetics, Medicine, and the Plain People," (2009) and Francomano's, "Amish Culture" (1996).

Exome sequencing has also recently been used in Plain populations to help identify pathogenic variants for conditions. The combination of SNP microarrays and gene sequencing have been used to map loci in individuals affected with genetic conditions. At the Clinic for Special Children, loci had been mapped for 28 genetic disorders found within the Amish and Mennonites since 2004, but for 11 of these conditions no causative genes were found. While genetic mapping is relatively easy and cost-efficient, the homozygous blocks typically contained hundreds of genes. A collaboration between the Clinic for Special Children and the Broad Institute using phenotype data, autozygosity mapping and exome sequencing resulted in the identification of pathogenic variants for seven disorders (Henderson & Anbar, 2009)

Table 1.1. Selected autosomal recessive genetic conditions more common among the Amish

Disorder	Gene	Symptoms	Incidence	Citation
3-methyl-crotonyl-glycinuria	*MCCC2*	Hypoglycemia, hyperammonemia, lactic academia, tachypnea, vomiting		(Gibson, Bennett, Naylor, & Morton, 1998)
Amish microcephaly	*SLC25A19*	Microcephaly, underdeveloped brain, micrognathia, hepatomegaly, life expectancy 6 months	1 in 500 in Old Order Amish	(Biesecker, 1993)
Cartilage-hair hypoplasia	*RMRP*	Short stature, skeletal abnormalities, hypotrichosis, immune deficiency	1 in 1300 in Older Order Amish	(Mäkitie & Vakkilainen, 1993)
Cortical dysplasia and focal epilepsy	*CNTNAP2*	Gross motor delay, hypotonia, intellectual disability, seizures, autistic behavior		(Strauss et al., 2006)
Cystic fibrosis	*CFTR*	Respiratory system damage, digestive system issues		(Henderson & Anbar, 2009)
Ellis-van Creveld syndrome	*EVC*	Short stature, particularly short forearms and lower legs, polydactyly, dental abnormalities, heart defects		(McKusick, 2000)
Galactosemia	*GALT*	Feeding difficulties, lethargy, failure to thrive, jaundice, liver damage, cataracts		(McKusick, Hostetler, & Egeland, 1964)
Glutaric aciduria, type 3	*C7orf10*	Failure to thrive, diarrhea, vomiting, many asymptomatic		(Sherman et al., 2008)
McKusick-Kaufman syndrome	*MKKS*	Polydactyly, heart defects, genital abnormalities (hydrometrocolpos in females, hypospadias in males)	1 in 10,000 in Old Order Amish	(Slavotinek, 1993)
Nemaline rod myopathy	*TNNI*	Myopathy throughout body, swallowing difficulties, scoliosis, contractures, typically fatal in childhood	1 in 500	(North & Ryan, 1993)
Phenylketonuria	*PAH*	Intellectual disability, seizures, autism-like features	1 in 1000 in Plain Community	(Schwoerer, Drilias, Kuhl, Mochal, & Baker, 2018)
Propionic acidemia	*PCCB*	Vomiting, lethargy, hypotonia, failure to thrive		(Kidd, Wolf, Hsia, & Kidd, 1980)
Sitosterolemia	*ABCG8*	Tendon xanthomas, premature coronary artery disease, atherosclortic disease, elevated levels of plant sterols		(Lee, Lu, & Patel, 2001)
Troyer syndrome	*SPART*	Spasticity of leg muscles, paraplegia, muscle wasting in hands and feet, short stature, developmental delay		(Patel, Harlalka, & Crosby, 1993)

Through the study, a mutation in the *SLC6A3* gene was found to cause infantile Parkinsonism-dystonia syndrome, a progressive movement disorder characterized by difficulty walking, eating, and talking. Psychomotor retardation, epilepsy and craniofacial dysmorphism (PMRED) is a condition marked by psychomotor delay, dysmorphic features, focal or generalized seizures, and heart issues such as aortic stenosis. A mutation in the *SNIP1* gene was found to be the cause of this condition. A mutation in the *CRADD* gene was found to cause nonsyndromic mental retardation. For many of the pathogenic variants, in vitro studies provided further evidence of pathogenicity. This study shows the clear potential for exome sequencing to elucidate pathogenic variants for conditions among the Amish in the future. (Puffenberger et al., 2012).

Given that new technology is being used to study genetic conditions in the Amish, it is important to consider the health literacy of this population and to deliver patient sensitive services. One way to ensure that the Amish population is receiving appropriate genetic services is to assess their satisfaction with genetic counseling they have received. Since only one study to date has focused on Amish perspectives, it seems pertinent to revisit this topic especially given the advances in technology.

1.6 Patient satisfaction with genetic counseling

Patient satisfaction is an important measure to consider for the continuous improvement of genetic counseling services. Previous studies have focused on counseling in the prenatal and cancer settings and have shown that satisfaction with genetic counseling is high (Bleiker et al., 1997; Bober, Hoke, Duda, & Tung, 2007; Kaduri, Zlotogora, & Peretz, 1998.; Nordin, Lidén, Hansson, Rosenquist, & Berglund, 2002; Tercyak, Johnson, Roberts, & Cruz, 2001). Several studies found that patients were

satisfied with the counselor's ability to listen and thoroughly answer questions, and overall genetic counseling was found to be helpful (Bober et al., 2007; Tercyak et al., 2001; Veach, Truesdell, LeRoy, & Bartels, 1999.) Areas of dissatisfaction included patient perception that genetics professionals were not communicating information with other health care professionals and that the primary health care provider did not seem to be involved (Bleiker et al., 1997).

More recently, patient satisfaction has been assessed with regard to alternative delivery models for genetic counseling. Satisfaction was found to be high for group counseling in both the cardiology and prenatal settings (Otten, Birnie, Ranchor, van Tintelen, & van Langen, 2015; Cloutier, et al., 2017). Patient satisfaction with telegenetics (live videoconferencing) services has also been assessed (Buchanan et al., 2015). While patients were less likely to present for appointments, satisfaction was high among those who did attend. Additionally, no difference in satisfaction was noted between those who received in-person genetic counseling versus those who received telegenetics services.

A number of measures have been developed to measure patient satisfaction. These measures typically include qualitative and quantitative items. Many of these are long and take time to complete. The Genetic Counseling Satisfaction Scale (GCSS), a six-item Likert scale measure, was developed as a way to assess patient satisfaction in a simple, quick manner (DeMarco, Peshkin, Mars, & Tercyak, 2004). First used in the prenatal genetics setting (Tercyak et al., 2001), the measure was also found to be reliable in the cancer setting (DeMarco et al., 2004). The GCSS has also been used in the cardiology setting (Otten et al., 2015) and to assess satisfaction with telegenetics

(Buchanan et al., 2015). The GCSS assesses patient perceptions regarding rapport, information provided, and counseling provided (DeMarco et al., 2004).

One of the main goals of this study was to assess Amish patient satisfaction with genetic counseling services in one clinic. The GCSS was adapted for incorporation into the questionnaire for this study.

1.7 Rationale of the study

The Amish have historically been a useful population for the study of genetic disease due to genetic isolation, large family size, high rate of consanguinity, and excellent genealogical records (McKusick et al., 1964). The high rate of genetic disease in Amish communities creates the necessity for culturally appropriate genetic services for this population. Clinics such as the Clinic for Special Children and The Community Health Clinic have sought to meet this need. However, little research has been done on how the Amish perceive these genetic services and no research has been done specifically on how the Amish experience genetic counseling. This study aims to fill in this gap in our knowledge about how the Amish perceive genetic counseling. Understanding the Amish patient perspective will help to promote culturally sensitive genetic counseling service. The goal of this study was to assess Amish patient satisfaction with genetic counseling services and to gain insight into Amish perspectives of the genetic counseling process. The objectives of this study were as follows:

1) Assess patient satisfaction with their genetic counseling experience
2) Understand how the Amish perceived their genetic counseling session in their own words in relation to:
    a. The rapport-building aspects of the session

b. The cultural appropriateness of the session

c. The impact of the session on their medical decision-making.

Chapter 2: Amish Perspectives of the Genetic Counseling Process[1]

2.1 Abstract

The Amish are a population with a high concentration of genetic disorders who have informed our understanding of several genetic conditions. This culturally unique group has special need for genetic services. While clinics have been established to care for Amish individuals, such as The Community Health Clinic in Indiana, little research has been done on Amish perspectives of these services, specifically genetic counseling. Amish individuals who received genetic counseling from The Community Health Clinic were sent recruitment letters and a questionnaire via mail. The questionnaire consisted of demographic questions, a 7-item adapted Genetic Counseling Satisfaction Scale (GCSS), and open-response questions. Thirty-three individuals completed the questionnaire. The majority of participants were aware they were receiving genetic counseling (81.8%), and most participants received genetic counseling from a genetic counselor (54.5%) versus a geneticist (39.4%). The mean satisfaction scores for each 5-point Likert-scale question showed that overall, participants were satisfied with their experience with genetic counseling, with mean scores ranging from 4.58 to 4.77. Descriptive and univariate statistics showed some statistically significant differences in satisfaction when comparing males versus females, referral type, and whether the participant saw a genetic counselor or a geneticist. Open-ended responses also showed that participants were satisfied

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with their genetic counseling. These responses centered around three themes: rapport-building aspects of the session, the cultural appropriateness of the session, and the impact of the session on medical decision-making. In this first study to explore Amish perceptions of a genetics clinic tailored to their particular way of life, we found that the Amish served by The Community Health Clinic felt respected, that their care was culturally sensitive, and they were satisfied with the genetic services they received.

2.2 Introduction

Genetic counseling is a process inherently tied to the culture of both the counselor and the counselee. The term culture can be defined as, "shared values, goals, expectations, beliefs, perceptions, and behaviors" among a group of people who share similar origins and styles of living (Randall-David, 1989). Common topics addressed in a genetic counseling session include reproductive decision-making, health, and disability. Beliefs, practices and values surrounding these issues are strongly tied to a person's world view (Weil, 2001). Culture is an integral component of the genetic counseling process and a significant aspect in providing successful and effective care (Weil, 2000). The American Board of Genetic Counseling (ABGC) practice-based competencies include expectations about cultural competency and the ability to tailor sessions to meet the needs of culturally different patients ("ABGC - Genetic Counseling Standards & Best Practices | ABGC," 2015.). Cultural competency can be defined as "the ability to provide skilled treatment to members of diverging cultural backgrounds through the use and knowledge of differing cultures, and self-awareness of one's attitudes, beliefs, behaviors, and biases linked to culture and cultural differences" (Leroy, Veach & Bartels, 2010, p. 227).

The Genetic Counseling Competence Toolkit serves as a resource to improve cultural competency in the profession. The "culturally humble" genetic counselor is one who realizes that each cross-cultural interaction can be viewed as a learning opportunity and that lessons learned should be shared. Genetic counselors should be open and willing to try new approaches and ask their patients to share their stories and experiences with them (Warren, 2011).

Many studies exist in the literature that focus on specific cultural groups and how genetic services are perceived and delivered (Awwad, Veach, Bartels, & LeRoy, 2008; Baars, van Dulmen, Velthuizen, van Riel, & Ausems, 2017; Barlow-Stewart et al., 2006; Kinney, Gammon, Coxworth, Simonsen, & Arce-Laretta, 2010; Kowal, Gallacher, Macciocca, & Sahhar, 2015; Mittman, Bowie, & Maman, 2007; Thompson et al., 2015; Tsai et al., 2017). While it is impossible to summarize any particular culture, and people of a certain culture do not necessarily all share the same views and behaviors, it is still important to explore different cultural responses to genetic services. Several common themes emerge among the current literature on culture and genetic services.

A commonality among many non-Western cultures is community or family-based decision-making. Individualism is a major component of Western medicine, whereas many cultures have a strong sense of community that extends to health care (Weil, 2000). Southeast and East Asian women living in the U.S. that were interviewed with regard to prenatal genetic counseling said that pregnancy decisions would be made as a family rather than as an individual (Tsai et al., 2017). Similarly, Chinese-Australians interviewed regarding genetics services expressed that senior family members should be involved as they typically play a role in decision-making for the family (Barlow-Stewart

et al., 2006). In Orthodox Jewish tradition, the Rabbi provides mediation between modern health technology and ancient laws, and is therefore involved in health care utilization among this community (Mittman et al., 2007). Genetic counselors with experience working with Indigenous Australians said that engaging family members in decision-making dialogue can be important as many Indigenous people involve community elders in their decisions (Kowal et al., 2015). These studies emphasize the need for inclusion of family and/or community members in decision making among certain cultures.

In cross-cultural genetic counseling, establishing rapport plays an even more essential role than in a typical session (Weil, 2001). Cultural needs described by genetic counselors who have worked with Indigenous Australians include spending extra time on rapport. This can likely be attributed to the population's distrustful attitude towards health care services stemming from their previous negative experiences with government policies (Kowal et al., 2015). Turkish and Moroccan participation in genetic counseling in the Netherlands was also found to be affected by previous negative experience with health care services (Baars et al., 2017). It can prove helpful to ask about what the patient understands about the indication for their appointment as this allows them to tell their story. Additionally, if a patient voices any distrust, the counselor could respond in a non-defensive manner to help open conversation even further (Weil, 2001).

Communication issues can create barriers to genetic counseling. In a study focused on the Latino community and amniocentesis refusal, Mexican-origin women did not understand that the protein discussed in relation to prenatal screening is not affected by the protein in their diets. For example, one woman who screened positive for a chromosome abnormality said she would simply eat more protein. Additionally, the non-

directive nature of genetic counseling was a source of miscommunication for some women. One participant said that she did not want the amniocentesis because everyone was calm and reassuring (Browner, Mabel Preloran, Casado, Bass, & Walker, 2003). Similar communication issues were found in a study regarding Latinos' attitudes towards cancer genetic counseling. Participants suggested providing clear definitions of "gene" and "mutation," and to limit unnecessary information such as detailed information on genetics (Kinney et al., 2010). Participants of a another study on Latina perceptions of prenatal genetic counseling suggested providing information regarding what genetic counseling is and what to expect beforehand as well as providing written or print resources during the session as ways to improve communication issues (Thompson et al., 2015).

Certain beliefs among cultures can also affect genetic counseling. For example, Latinos' interviewed regarding cancer genetic counseling brought up cultural taboos about cancer. In their culture, cancer can be viewed as contagious and is often seen as a death sentence. There is also shame associated with cancer, thus it is often not discussed (Kinney et al., 2010). Chinese-Australians also mentioned that cancer is a taboo topic and that illness is often not discussed in their culture (Barlow-Stewart et al., 2006). In Turkey and Morocco, cancer is also not spoken of and is seen as a death sentence (Baars et al., 2017). These cultural beliefs can prevent patients from obtaining genetic counseling and are important to be aware of when taking a family history as individuals may not be aware of cancer in their families.

There are differing beliefs among different cultures regarding having a child with disabilities. Southeast and East Asian individuals interviewed about prenatal genetic

counseling described shame that would be associated with the family if they had a child with a disability and that community members might think that the child is punishment for a wrongdoing in a past life (Tsai et al., 2017). Native Palestinians responding to hypothetical prenatal situations expressed that they would not want to marry someone with a family history of intellectual disability (Awwad et al., 2008). Latinas interviewed about prenatal genetic counseling did express the desire for a healthy baby, however, they did not feel the need for prenatal diagnosis, citing the desire to leave it up to God (Thompson et al., 2015). Weil explains that the dominant U.S. culture perceives nature as something that can be understood and potentially changed by humans. Other cultures have a more accepting perspective of nature and thus are more accepting of those with disabilities and take issue with some of the testing options offered in genetic counseling sessions such as prenatal diagnosis (2000).

In many of the studies previously discussed, participants expressed that they saw benefits to genetic counseling. Latinos felt that cancer genetic counseling could open the doors to earlier treatment, provide information for family members, and encourage more frequent screening. Members of this community felt that cost would be a barrier to pursuing genetic counseling, however, as many Latinos are low-income and uninsured (Kinney et al., 2010). Turkish and Moroccan individuals in the Netherlands noted similar benefits to genetic counseling, such as knowing more about personal and family risks (Baars et al., 2017). In the prenatal setting, Latinas felt that learning the risk for a chromosome abnormality in their pregnancy was a helpful part of the session and made them feel reassured (Thompson et al., 2015).

Exploring and understanding these cultural beliefs is an important part of a genetic counselor's role. In order to best serve individuals of different cultures, genetic counselors need to know about the beliefs and values these cultures share as this promotes cultural competency and provides patients with the most appropriate care. One particular group of people with a unique culture, the Amish, have contributed a great deal to our understanding of genetics (Strauss & Puffenberger, 2009). The Amish are members of a Christian church formed out of the Anabaptist movement in South Germany and Switzerland in 1525 (Cates, 2014). The Anabaptists believed in adult baptism and that an individual should be able to make an informed choice about their faith as opposed to being born into one. As the Anabaptists were not Catholic or traditional Protestant, they faced persecution for their beliefs in the 16th-century. An early leader of the Anabaptist movement was Menno Simons and those who still follow his teachings are known as Mennonites (Francomano, 1996). Under the leadership of Jakob Ammann, the Amish separated from the Mennonites in 1693 because they felt they needed to be more isolated from those who did not follow their belief system (Kowal et al., 2015). Ammann advised a "sharper separation from the world and more severe shunning of unrepentant members." Because of the persecution they faced in Europe, the Amish accepted William Penn's offer of religious tolerance in Pennsylvania in the 1740's, and their last congregation in Europe died out in 1936 (Cates, 2014). Several other groups have roots in the Anabaptist movement. Today, Amish, Old Order and Conservative Mennonites, Old Order Brethren, and Hutterites are collectively referred to as "Plain" people, owing to their plain dress and simple way of life (Strauss & Puffenberger, 2009).

Today, there are 330,270 Amish individuals in North America, with 63% living in the states of Ohio (Holmes and Wayne Counties), Pennsylvania (Lancaster and Mifflin Counties), and Indiana (Elkhart and Lagrange Counties). ("Amish Population Profile, 2018," 2018; Francomano, 1996). Approximately 40 "orders" of Amish exist, each varying in terms of their degree of separation from the world, technology usage, and adherence to tradition, and over 475 settlements that span 30 states including Ontario (Cates, 2014). Among the 40 Amish "orders" there are Old Order Amish, Swartezentruber Amish, New Order Amish and Beachy Amish. The Old Older Amish is the largest group in the United States. The Swartzentruber Amish are more conservative than the Old Order Amish and are unlikely to utilize modern health care, while the New Order Amish are seen as less conservative. The Beachy Amish are the least conservative and typically are accepting of modern medicine (Francomano, 1996). Amish group range from "low" groups of Amish communities, seen as more traditional, to "higher" groups that have more interaction with the world. The openness a group of Amish might feel toward those of the outside world can depend greatly on whether they belong to a more traditional or more progressive group (Cates, 2014).

Amish culture is deeply intertwined with religion. Amish communities are divided into "church districts" that are typically based on the population of an area, and the congregation is known as the Gmay. A Gmay usually consists of 75 to 150 individuals. Four to five "ministers" lead each congregation, including a bishop, a deacon, and several preachers. The bishop is the leader of the congregation and is responsible for baptisms, weddings, and similar rituals. The deacon is responsible for financial aspects of the Amish community, including health care and fire or storm

damage (Cates, 2014). The clergy is elected by the community. In this patriarchal society, senior bishops hold authority among the clergy. The bishop's authority is granted by God and he is responsible for the physical and spiritual health of his congregation. If the bishop perceives an outside service to be in conflict with their religious views, he can prohibit use of this service (Miller-Fellows, Adams, Korbin, & Greksa, 2018). At home, the husbands and fathers hold authority, however mothers and fathers typically make decisions together (Francomano, 1996).

The core beliefs of the Amish center around living simply and separately from the rest of the world. The Amish typically avoid modernization and physical connections to the outside ("English") world. To accomplish this, they prefer no or limited use of electricity, wearing plain dress, and using horse and buggy for transportation purposes, and pacifism. The practices by which each congregation of Amish people live by is taught and upheld through *Ordnung*, which is an oral tradition that advises on how to live. This includes rules about clothes, technology, higher education and divorce, among other things. *Ordnung* details might be slightly different from community to community (Cates, 2014). Amish culture is rooted in ordinary, day-to-day life practices, and *Ordnung* provides instructions for these practices to help accomplish the goal of peaceful, supportive community (Francomano, 1996).

The pervasiveness of religion in Amish culture extends to matters of health. Any illness is seen as a reflection of God's will. Additionally, Amish do not see illness through the lens of symptoms, but rather as an inability to perform their typical duties such as farming or housekeeping. Therefore, Amish individuals may not present to medical care until an illness is severe. Modern medicine is not prohibited in Amish

communities, but typically home remedies and folk medicine are preferred. Health care professionals working with the Amish should therefore be accepting of their use of alternative medicine, as long as it does not cause harm. The Amish may choose not to use medical interventions, and typically weigh the degree to which an intervention would disrupt the family or community. An additional factor in Amish utilization of health care is their religious proscription against the use of insurance. The Amish believe that commercial insurance takes away from the concept of leaning on their own community for support (Miller-Fellows et al., 2018) This means that medical interventions can be costly. The Amish do, however, have a system in place in which the community pools finances to be used in case a member of the community needs medical care (Francomano, 1996).

Family is also an important aspect of Amish culture. Amish adults are rarely unmarried, and it is important to note that marriages are not arranged. The Amish do intermarry within their cultural group as they believe in staying separate from the outside world (Francomano, 1996). On average, Amish couples have five to eight children (Cates, 2014). The Amish keep meticulous genealogical records to honor their families and provide stability and foundation for where the current generation fits in. When entering church, the Amish walk in according to age, so an Amish individual will walk behind and in front of the same people their entire life (Francomano, 1996).

Amish education is guided by their culture as well. Typically, Amish schools consist of one room where children of all ages learn in English. Children are usually taught by an unmarried woman who has recently completed her education. Subjects such as reading, writing, arithmetic and world geography are taught until 8th grade. Once a

child has completed 8th grade, education moves into the home where girls learn about homemaking and boys study farming. In order to satisfy state requirements, children keep a journal of what they have learned until age 16 that a teacher will review. At home, a form of German called "Pennsylvania Dutch" is typically spoken and learned from parents, and at church a form of High German, distinct from Pennsylvania Dutch, is used. Therefore, many Amish individuals are trilingual (Francomano, 1996).

In a health care situation, there are several things to keep in mind to have a culturally competent interaction with an Amish individual. Amish individuals do not typically use formal titles and prefer the use of first names. Therefore, rapport can be built using a professional title and then first name, such as "Nurse Bill." Additionally, Amish children are often referred to as their father's name followed by their first name, for example "Eli's Adam." This can be helpful in distinguishing individuals who share the same name, especially since there are only a handful of last names in these communities. Health care professionals should also keep in mind that English is usually a second language for Amish people, so they may not communicate as well but this should not be conflated with a lack of intelligence or literacy. However, biological and medical principles are not an area of focus in Amish schooling, thus professionals should take the time to clearly define words and use visual aids when explaining concepts. Finally, photography is a taboo subject in Amish communities. Some Amish families will allow the use of photography if they feel it will only be used for medical purposes, but others refuse to allow it (Francomano, 1996).

With these aspects of Amish culture in mind, it is clear that health services in these communities must be provided by those who have a good understanding of the

Amish and their beliefs. When services are provided, the experience that Amish individuals have with this service spreads throughout the community quickly. Therefore, it is important to provide services carefully and thoughtfully, and potentially include family members and the bishop in these conversations (Miller-Fellows et al., 2018).

Cultural aspects of the Amish affect the uptake in medical services, including genetic services. One clear example is the use of newborn screening among the Amish. Newborn screening is difficult to accomplish in the Wisconsin Plain population, which includes both Old Order Amish and Mennonite communities, for a variety of reasons (Kuhl et al., 2017). The main barrier is the lack of medical follow-up due to lack of medical insurance, the difficulty of getting to a medical facility, and the idea that a doctor does not need to be seen unless a child is seriously ill (Kuhl et al., 2017). In order to gain a better understanding of newborn screening in the Wisconsin Plain population, Sieren et al. (2015) surveyed about their utilization of newborn screening and their attitudes towards it. In Wisconsin, around 2% of births occur outside of hospitals, and the majority of those occur among Plain communities. The study found that 40% of households screened all of their children, and of those who responded to the survey, most thought that the screening was important. Of note, one of the main reasons that individuals did not have their children screened was due to lack of awareness about newborn screening.

In an attempt to help alleviate the issue of the lack of newborn screening among the Amish, the Wisconsin Plain population was provided with carrier testing kits to be distributed via midwives and at community meetings (Kuhl et al., 2017). In order to build trust with the community, outreach activities with an educational component were performed that included community elders at community meetings. While the carrier

testing kits did not identify anyone that had not already been diagnosed, the study was successful at gaining trust and providing information about the importance of newborn screening to this community through their outreach portion of the methods. These findings speak to the idea that health care services must be provided in a culturally sensitive fashion.

Another study assessed cancer screening practices among the Amish in Ohio Appalachia (Katz et. al, 2011). Similar to the newborn screening survey, the study found that cancer screening among Amish was significantly lower compared to non-Amish individuals in the same area. It was found that Amish individuals did not think screening was necessary as they did not believe they were likely to get cancer. The authors believe this may be due to the hesitation the Amish have in utilizing preventative medicine.

The Amish are also known for having low immunization rates. A study done on the Amish population in Holmes County, Ohio aimed to discover the reason for these low rates (Wenger, McManus, Bower, & Langkamp, 2011). The immunization rate in this population at the time of the study was 45% as compared to national rate of 85%. The study found that the Amish weighed the possibility of adverse effects of the vaccines more heavily than other factors such as financial burden, accessibility or religious issues when deciding whether to vaccinate their children. Interestingly, many participants in this study allowed their children to have some vaccines but not all of them.

A special concern within Amish communities is health literacy. Formal education stops in 8th grade in Amish communities and continues at home with a focus on learning how to farm and run households (Francomono, 1996). Those who have a limited education usually have lower health literacy, and health literacy is likely a predictor of

how well individuals understand and utilize genetic information (Lea, Kaphingst, Bowen, Lipkus, & Hadley, 2011). Health literacy also has implications for how individuals utilize health services and how it impacts health behaviors. In order to assess health literacy in Amish communities in Ohio Appalachia, Amish and non-Amish individuals completed interviews and were given the rapid estimate of adult literacy in medicine (REALM) instrument to compare the two groups (Katz, Ferketich, Paskett, & Bloomfield, 2013). The REALM scores of the Amish individuals were significantly lower than non-Amish, indicating limited health literacy. The authors suggest that culture is a contributing factor to the limited health literacy, citing the unique educational practices among the Amish, and the limited exposure to media and technology.

In 1962, Victor McKusick learned of the Amish population's high incidence of achondroplasia. He recognized that two recessive conditions, Ellis-van Creveld syndrome and cartilage-hair hypoplasia, were actually present in the population and were incorrectly diagnosed as achondroplasia (Strauss & Puffenberger, 2009). This sparked the beginning of many genetic studies on the Amish population. McKusick (1964) realized that this population was an excellent source to study genetic disease due to their geographical isolation, high rate of consanguinity and recessive conditions, large family sizes, and excellent genealogy records. However, these initial years of studying the Amish did not have a focus on providing clinical services to the Amish, and many research subjects (affected with genetic conditions) passed away due to the lack of services (Strauss & Puffenberger, 2009).

In 1989, Dr. D. Holmes and Caroline Morton opened the Clinic for Special Children in Lancaster County, Pennsylvania. This clinic serves the Amish and Mennonite

populations of children affected by genetic conditions and operates on the idea that research and clinical care go hand in hand. The Clinic for Special Children has given the Amish a logistically feasible place to receive care. The Clinic has a history of incorporating advanced biochemical and genetic laboratory techniques into their care. Dr. Puffenburger, a molecular geneticist, was hired in 1998 to help advance the Clinic's molecular techniques. Dr. Strauss, the Medical Director, plays a large role in the research and clinical aspects at the Clinic for Special Children. The Clinic serves over 1,000 patients with over 264 known genetic conditions. Many specialty services are also made available to patients as part of the Clinic's vision to be a medical home for the individuals they treat ("History & Mission," 2016).

The Clinic for Special Children is funded through annual auctions, donations, collaborative relationships, and fees paid by patients ("Our Impact," 2016). Four annual quilt auctions are organized by members of the Plain community that contribute to one third of the budget. The Clinic has always worked to keep costs minimal for their patient population through efforts such as utilization of publicly available molecular information and reducing hospitalizations and laboratory costs. For example, in 2010, the $1.5 million dollar operating budget saved the community an estimated $20 million in medical costs. Keeping costs minimal is important for their patient population, as 95% are uninsured (Strauss, Puffenberger, & Morton, 2012).

In 2008, The Community Health Clinic (CHC) was established in Topeka, Indiana. Its inception was based on the need for a more logistically reasonable place for children affected by genetic disorders to receive care ("Our Story," n.d.). Since it opened, Amish individuals have been receiving genetic counseling from both physicians and

genetic counselors. The Community Health Clinic is similar to the Clinic for Special Children as many health care services are provided to Amish individuals in one place, including newborn screening services, speech therapy, audiology and nutrition services. They also maintain low costs for patients and rely on donations and fundraisers such as auctions to provide for their patients.

The CHC has also helped with other aspects of care. Within the community that CHC serves, a healthcare sharing ministry has been established called the Plain Church Group Ministry (PCGM). Amish deacons and members of this group negotiate with hospitals to obtain discounts for their community needs. For example, PCGM has discounted rates for formula that children with metabolic conditions need. They are able to get formula for wholesale price and then a 2% administrative fee is charged by the PCGM to cover their services. In order for PCGM to be formalized, Amish individuals must carry a card that shows they are members of PCGM. Many hospitals and medical groups, including CHC, send bills directly to PCGM. If an Amish family asks for help or the bill exceeds $3500, PCGM will contact the deacon to help with cost sharing. When the family receives the bill, they pay however much they can afford and then the rest is covered by a fund that PCGM has access to. If an Amish individual feels strongly that they do not want to bill through PCGM then they can pay a self-pay price (R. Evans, personal communication, April 8, 2019).

Another aspect that makes the CHC unique is the length of time of the appointments. A new patient appointment is scheduled to be 2-3 hours. This initial appointment may or may not include genetic counseling, and sometimes patients come

back for genetic counseling. Ultimately, the physician at the clinic spends a great deal of time with patients (R. Evans, personal communication, April 8, 2019).

As for testing, CHC does whole exome sequencing through the CSC on a research basis, so turnaround times can be lengthy. Other testing is done through standard labs such as Invitae that have reasonable turnaround times (R. Evans, personal communication, April 8, 2019).

While the established clinics provide much needed genetics care to Amish individuals, no studies to date detail patient perceptions of the genetic counseling process or their experiences with genetic counseling. One study, however, sought to examine the Amish population's general knowledge of genetic disorders and attitudes towards medical care, including genetic counseling by interviewing 17 Amish families were interviewed in Lancaster County, Pennsylvania (Brensinger & Laxova, 1995). Only three of 12 families with a child with a genetic disorder knew about genetic counseling services, and only two were able to provide an explanation of genetic counseling. The majority of those interviewed, however, would have considered genetic counseling and wanted to know more about their child's condition.

Many genetic conditions affect the Amish and Mennonite populations. Two conditions that were first treated in the Amish and Mennonite populations are maple syrup urine disease (MSUD) and glutaric acidemia Type 1 (GA1). MSUD is an autosomal recessive disorder in which the body cannot process leucine, isoleucine, and valine. The byproducts of these amino acids build up in the body causing a sweet odor in the urine. Poor feeding, abnormal movements, delayed development are hallmark symptoms, and if untreated, it can cause seizures and death. The general population

incidence of this condition is 1 in 185,000, while the Old Order Mennonite population has an estimated incidence of 1 in 380 (Strauss, Puffenberger, & Morton, 1993).

GA1 is also a recessive condition that affects the body's ability to process amino acids. Build-up of the amino acids lysine, hydroxylysine and tryptophan can cause damage to the basal ganglia and result in issues with movement as well as intellectual disability. The general population incidence of this condition is estimated to be 1 in 30,000 to 1 in 40,000, while the Amish population incidence is 1 in 300 (Strauss, Puffenberger, Robinson, & Morton, 2003, p. 1).

Exome sequencing has also recently been used in Plain populations to help identify pathogenic variants for conditions. The combination of SNP microarrays and gene sequencing have been used to map loci in individuals affected with genetic conditions. At the Clinic for Special Children, loci had been mapped for 28 genetic disorders found within the Amish and Mennonites since 2004, but for 11 of these conditions no causative genes were found. While genetic mapping is relatively easy and cost-efficient, the homozygous blocks typically contained hundreds of genes. A collaboration between the Clinic for Special Children and the Broad Institute using phenotype data, autozygosity mapping and exome sequencing resulted in the identification of pathogenic variants for seven disorders (Henderson & Anbar, 2009).

Through the study, a mutation in the *SLC6A3* gene was found to cause infantile Parkinsonism-dystonia syndrome, a progressive movement disorder characterized by difficulty walking, eating, and talking. Psychomotor retardation, epilepsy and craniofacial dysmorphism (PMRED) is a condition marked by psychomotor delay, dysmorphic features, focal or generalized seizures, and heart issues such as aortic stenosis. A

mutation in the *SNIP1* gene was found to be the cause of this condition. A mutation in the *CRADD* gene was found to cause nonsyndromic mental retardation. For many of the pathogenic variants, in vitro studies provided further evidence of pathogenicity. This study shows the clear potential for exome sequencing to elucidate pathogenic variants for conditions among the Amish in the future. (Puffenberger et al., 2012).

Given that new technology allows the study of genetic conditions in the Amish, it is important to consider the health literacy of this population and to deliver patient sensitive services. One way to ensure that the Amish population is receiving appropriate genetic services is to assess their satisfaction with genetic counseling they have received. Since only one study to date has focused on Amish perspectives, it seems pertinent to revisit this topic especially given the advances in technology.

Patient satisfaction is an important measure to consider for the continuous improvement of genetic counseling services. Previous studies have focused on counseling in the prenatal and cancer settings and have shown that satisfaction with genetic counseling is high (Bleiker et al., 1997; Bober, Hoke, Duda, & Tung, 2007; Kaduri, Zlotogora, & Peretz, 1998.; Nordin, Lidén, Hansson, Rosenquist, & Berglund, 2002; Tercyak, Johnson, Roberts, & Cruz, 2001). Several studies found that patients were satisfied with the counselor's ability to listen and thoroughly answer questions, and overall genetic counseling was found to be helpful (Bober et al., 2007; Tercyak et al., 2001; Veach, Truesdell, LeRoy, & Bartels, 1999.) Areas of dissatisfaction included patient perception that genetics professionals were not communicating information with other health care professionals and that the primary health care provider did not seem to be involved (Bleiker et al., 1997).

More recently, patient satisfaction has been assessed with regard to alternative delivery models for genetic counseling. Satisfaction was found to be high for group counseling in both the cardiology and prenatal settings (Otten, Birnie, Ranchor, van Tintelen, & van Langen, 2015; Cloutier, et al., 2017). Patient satisfaction with telegenetics (live videoconferencing) services has also been assessed (Buchanan et al., 2015). While patients were less likely to present for appointments, satisfaction was high among those who did attend. Additionally, no difference in satisfaction was noted between those who received in-person genetic counseling versus those who received telegenetics services.

A number of measures have been developed to measure patient satisfaction. These measures typically include qualitative and quantitative items. Many of these are long and take time to complete. The Genetic Counseling Satisfaction Scale (GCSS), a six-item Likert scale measure, was developed as a way to assess patient satisfaction in a simple, quick manner (DeMarco, Peshkin, Mars, & Tercyak, 2004). First used in the prenatal genetics setting (Tercyak et al., 2001), the measure was also found to be reliable in the cancer setting (DeMarco et al., 2004). The GCSS has also been used in the cardiology setting (Otten et al., 2015) and to assess satisfaction with telegenetics (Buchanan et al., 2015). The GCSS assesses patient perceptions regarding rapport, information provided, and counseling provided (DeMarco et al., 2004).

One of the main goals of this current study was to assess Amish patient satisfaction with genetic counseling services in one clinic. The GCSS was adapted for incorporation into the questionnaire for this study.

The Amish have historically been a useful population for the study of genetic disease due to genetic isolation, large family size, high rate of consanguinity, and excellent genealogical records (McKusick et al., 1964). The high rate of genetic disease in Amish communities creates the necessity for culturally appropriate genetic services for this population. Clinics such as the Clinic for Special Children and The Community Health Clinic have sought to meet this need. However, little research has been done on how the Amish perceive these genetic services and no research has been done specifically on how the Amish experience genetic counseling. This study aims to fill in this gap in our knowledge about how the Amish perceive genetic counseling. Understanding the Amish patient perspective will help to promote culturally sensitive genetic counseling service. The goal of this study was to assess Amish patient satisfaction with genetic counseling services and to gain insight into Amish perspectives of the genetic counseling process. The objectives of this study were as follows:

1) Assess patient satisfaction with their genetic counseling experience
2) Understand how the Amish perceived their genetic counseling session in their own words in relation to:
    a. The rapport-building aspects of the session
    b. The cultural appropriateness of the session
    c. The impact of the session on their medical decision-making.

2.3 Materials and Methods

This study used survey methodology. A paper questionnaire including demographic information, an adapted Genetic Counseling Satisfaction Scale (GCSS) and open-response questions were distributed to Amish individuals who received genetic

counseling through the CHC. The adapted GCSS consisted of 7 Likert-scale items (from 1 = "strongly disagree" to 5 = "strongly agree"), and there were 5 open-response questions. Mixed methodology was used to allow for data that could be analyzed statistically as well as to gain insight into Amish perspectives in their own words.

The original GCSS from DeMarco et al. (2004) was adapted in the following ways: the question, "My genetic counselor seemed to understand the stresses I was facing" was changed to, "My genetic counselor seemed to understand the stresses my family and I were facing," the question, "My genetic counselor helped me to identify what I needed to know to make decisions about what would happen to me," was changed to "My genetic counselor helped me to identify what I needed to know to make to decisions about what would happen to me/my family," questions 8 and 9 are original questions that were added, and the question "I felt better about my health after meeting with my genetic counselor," was omitted for this study.

To recruit participants, CHC staff accessed patient information, identified those who had received genetic counseling since 2013, and sent the recruitment letter and questionnaire via mail to 203 appropriate individuals including an additional envelope and stamp for return of the questionnaire. Upon completion, the participants mailed the questionnaires back to the CHC, and the CHC then forwarded completed questionnaires to the primary investigator for review. No patient identification was attached to the questionnaires received by the primary investigator. Questionnaires were stored in a locked filing cabinet and destroyed once analyzed.

The methodology for this study was both qualitative and quantitative. For questions 1-12, descriptive and univariate statistics was used to analyze the data. For

questions 13-17, grounded theory was used to analyze responses and identify themes. Data collection ended at the end of December and data analysis started at the beginning of 2019. The anticipated sample size was 20-36 individuals. The necessary sample was determined using G* Power, which gave the sample size needed to accomplish the goals of the study and establish significance.

2.4 Results

A summary of participant characteristics can be found in Table 1. A total of 33/203 individuals (16% response rate) completed the questionnaire and mailed it back to the CHC. All participants were considered in reporting these results, however the number of participants varied per question. Of the participants, 14 were male (42%) and 19 were female (58%). The majority of participants were Amish (N=32), and one participant was Mennonite. The minimum age among participants was 18 years old and the maximum was 75 years old, with a mean age of 38 years old. Six participants indicated an age less than 18, but the open-ended questions were clearly answered by a parent or adult; this presumably was the age for whom genetic services were sought. For these participants, the age was removed and the rest of the data was analyzed.

All participants responded to the question regarding their awareness that they received genetic counseling. The majority of participants were aware that they received genetic counseling (N=27, 81.8%). Four participants were unsure if they were aware they received genetic counseling (12.1%). Two participants indicated that they were not aware they received genetic counseling (6.1%).

Of the 33 participants, 13 responded that they received genetic counseling affecting themselves (39.4%), 11 responded that they received genetic counseling

affecting their child (33.3%), 8 responded that they received genetic counseling related to a genetic condition running in their family (24.2%), and one person did not respond to this question (3%).

All participants responded to the question regarding who provided the genetic counseling. Of the 33 participants, 18 indicated that they received genetic counseling from a genetic counselor (54.5%), 13 indicated that they received genetic counseling from a physician (39.4%), and 2 indicated that they were unsure about who they received genetic counseling from (6.1%). The physician at this clinic is a clinical geneticist.

Table 2.1 Participant characteristics (*N*=33)

Participant Characteristics	*n*	%
Sex		
Male	14	42
Female	19	58
Group		
Amish	32	97
Mennonite	1	3
Aware of receiving genetic counseling		
Yes	27	81.8
No	2	6.1
Unsure	4	12.1
Referral reason		
Genetic condition affecting participant	13	39.4
Genetic condition affecting participant's child	11	33.3
Genetic condition running in participant's family	8	24.2
No response	1	3%
Received genetic counseling from		
A genetic counselor	18	54.5
A physician	13	39.4
Unsure	2	6.1

2.4.1 Participant satisfaction with genetic counseling. Participants were asked to respond to a 7-item Likert scale Genetic Counseling Satisfaction Survey (GCSS) as part of the questionnaire. These questions are summarized in Table 2.2 along with the

response rates. For each item, the participants selected strongly disagree, disagree, uncertain, agree somewhat, or strongly agree. The mean scores were calculated for each item of the GCSS. In order to calculate the means, the responses were assigned numerical values where strongly disagree=1, disagree somewhat=2, uncertain=3, agree somewhat=4, and strongly agree=5. To see overall mean scores for each item, see Figure 2.1.

Table 2.2 Genetic counseling satisfaction scale questions and response rate

Genetic Counseling Satisfaction Scale Questions	Response Rate (*n*)
My genetic counselor seemed to understand the stresses my family and I were facing.	93.9% (31)
My genetic counselor helped me to identify what I needed to know to make decisions about what would happen to me and/or my family.	93.9% (31)
I understood the concepts the genetic counselor explained to me in the session.	93.9% (31)
The genetic counselor allowed time for me to talk with family and community members before making a decision.	84.8% (28)
The genetic counseling session was about the right length of time I needed.	93.9% (31)
My genetic counselor was truly concerned about my well-being.	93.9% (31)
The genetic counseling session was valuable to me.	93.9% (31)

Univariate statistical analyses were conducted to determine associations between different participant characteristics and each Likert-scale GCSS satisfaction question. Statistically significant and non-statistically significant results are summarized in Tables 2.3, 2.4 and 2.5

2.4.2 Gender and satisfaction. A one-way ANOVA was conducted to determine if gender was associated with how participants perceived the genetic counselor's concern for their well-being.

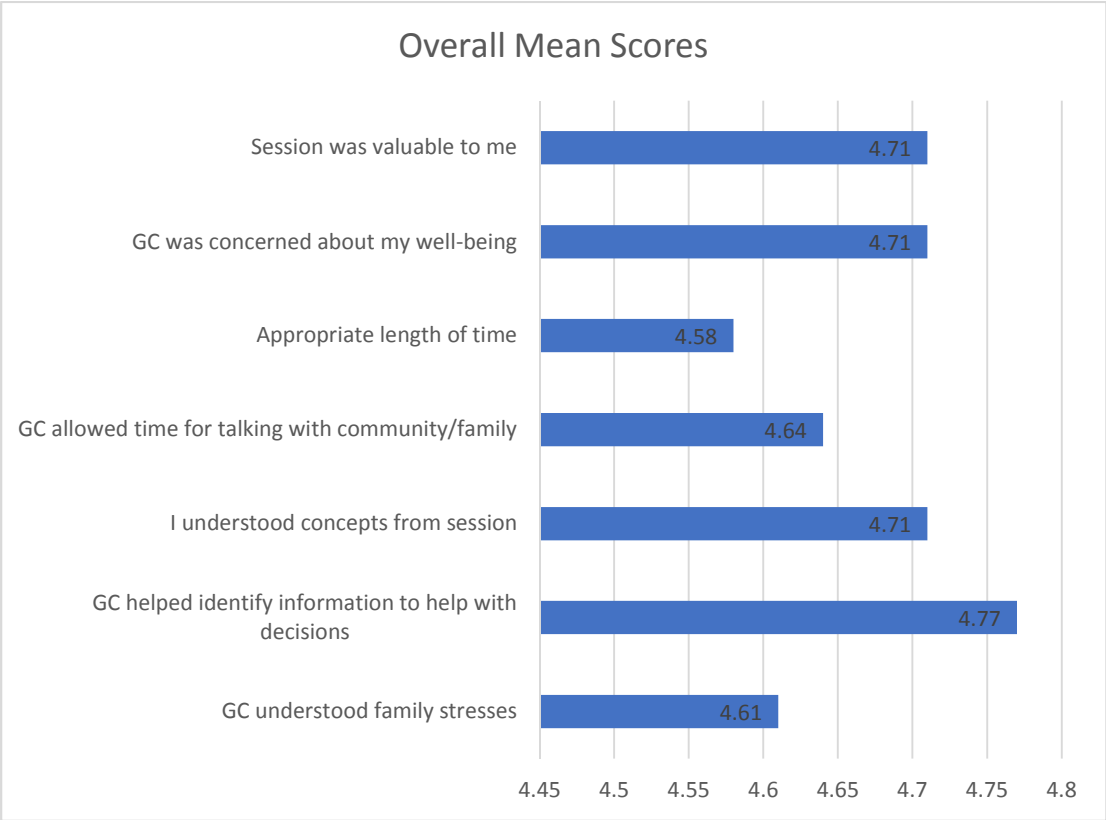

Figure 2.1 Overall mean scores for satisfaction

Participants were classified into two groups: male (n=13) and female (n=18). Data is presented as mean ± standard deviation. The mean satisfaction for the genetic counselor's concern for participant well-being increased from males (4.46 ± 0.78) to females (4.89 ± 0.32) and the differences between these groups was statistically significant, F (1,29) = 4.438, p = 0.044, η^2=0.133.

A one-way ANOVA was conducted to determine if gender was associated with participant perception of the value of the genetic counseling session. Participants were

classified into two groups: male (n=13) and female (n=18). Data is presented as mean ± standard deviation. The mean satisfaction for valuableness of the session increased from males (4.38 ± 0.96) to females (4.94 ± 0.24) and the differences between these groups was statistically significant, F (1,29) = 5.707, p = 0.024, η^2=0.164. Females also demonstrated higher total satisfaction and higher satisfaction on all other individual scale items but these were not statistically significant. Data for all items are summarized in Table 2.3.

Table 2.3 Gender and satisfaction

	Mean ± Standard deviation	***p*-value**
Total Satisfaction		
Males (n=13)	31.77 ± 4.36	0.464
Females (n=18)	32.67 ± 2.32	
GC understood family stressors		
Males (n=13)	4.46 ± 0.88	0.325
Females (n=18)	4.72 ± 0.58	
GC helped identify information to help with decisions		
Males (n=13)	4.69 ± 0.63	0.499
Females (n=18)	4.83 ± 0.51	
I understood concepts from the session		
Males (n=13)	4.69± 0.48	0.862
Females (n=18)	4.72 ± 0.46	
GC allowed time for talking with community/family		
Males (n=13)	4.54 ± 0.66	0.418
Females (n=15)	4.73 ± 0.59	
Session was appropriate length of time		
Males (n=13)	4.54 ± 0.66	0.754
Females (n=18)	4.61 ± 0.61	
GC was concerned about my well-being		
Males (n=13)	4.46 ± 0.78	0.044
Females (n=18)	4.89 ± 0.32	
Session was valuable to me		
Males (n=13)	4.38 ± 0.96	0.024
Females (n=18)	4.94 ± 0.24	

Table 2.4. Referral reason and satisfaction

	Mean ± Standard deviation	*p*-value
Total Satisfaction		
Condition affecting participant (*n*=12)	32.92 ± 2.27	0.595
Condition affecting participant's child (*n*=11)	32.09 ± 4.09	
Condition affecting patient's family (*n*=7)	31.14 ± 3.67	
GC understood family stressors		
Condition affecting participant (*n*=12)	4.92 ± 0.29	0.219
Condition affecting participant's child (*n*=11)	4.45 ± 0.93	
Condition affecting patient's family (*n*=7)	4.29 ± 0.76	
GC helped identify information to help with decisions		
Condition affecting participant (*n*=12)	5.00 ± 0.00	0.307
Condition affecting participant's child (*n*=11)	4.64 ± 0.67	
Condition affecting patient's family (*n*=7)	4.57 ± 0.79	
I understood concepts from the session		
Condition affecting participant (*n*=12)	4.92 ± 0.29	0.185
Condition affecting participant's child (*n*=11)	4.55 ± 0.52	
Condition affecting patient's family (*n*=7)	4.57 ± 0.54	
GC allowed time for talking with community/family		
Condition affecting participant (*n*=9)	4.78 ± 0.44	0.686
Condition affecting participant's child (*n*=11)	4.64 ± 0.67	
Condition affecting patient's family (*n*=7)	4.43 ± 0.79	
Session was appropriate length of time		
Condition affecting participant (*n*=12)	4.83± 0.40	0.025
Condition affecting participant's child (*n*=11)	4.64 ± 0.51	
Condition affecting patient's family (*n*=7)	4.00 ± 0.82	
GC was concerned about my well-being		
Condition affecting participant (*n*=12)	4.75 ± 0.45	0.886
Condition affecting participant's child (*n*=11)	4.73 ± 0.65	
Condition affecting patient's family (*n*=7)	4.57 ± 0.79	
Session was valuable to me		
Condition affecting participant (*n*=12)	4.92 ± 0.29	0.452
Condition affecting participant's child (*n*=11)	4.45 ± 0.93	
Condition affecting patient's family (*n*=7)	4.71 ± 0.76	

2.4.3 Referral reason and satisfaction. A one-way ANOVA was conducted to determine if referral reason for the participant was associated with satisfaction with the

length of the genetic counseling session. Participants were classified into three groups: genetic condition affecting self (n=12), child (n=11), or the family (n=7). Data is presented as mean ± standard deviation. The mean satisfaction for length of the genetic counseling session increased from the family (4.00 ± 0.82), to child (4.64 ± 0.51), to self (4.83± 0.40), and the differences between these groups was statistically significant, F (3,27) = 3.656, p = 0.025, η^2=0.289. Genetic counseling related to participant's own condition had the highest satisfaction rating on all items, but it was non-statistically significant. Data for all items are summarized in Table 2.4.

2.4.4 Provision of services and satisfaction. A one-way ANOVA was conducted to determine if the type of health professional participants received genetic counseling from was related to how participants perceived the genetic counselor's ability to understand family stressors. Participants were classified into three groups: genetic counseling from a genetic counselor (n=17), a physician (n=13), or unsure (n=1). Data is presented as mean ± standard deviation. The mean genetic counseling satisfaction for understanding family stressors increased from unsure (3.00), to geneticist (4.46 ± 0.88), to genetic counselor (4.82 ± 0.39), and the differences between these groups were statistically significant, F (2,28) = 4.371, p = 0.022, η^2=0.238. Other associations between healthcare professional type and total satisfaction and other individual scale items were not statistically significant but are summarized in Table 2.5.

2.4.5 Age and satisfaction. A linear regression did not find that age statistically significantly predicted genetic counseling session satisfaction, F (7,20) =1.04, p =.44 and age accounted for 27% of the explained variability in satisfaction.

2.4.6 How the Amish perceived their genetic counseling session in their own words. The questionnaire also contained five open-response questions, which are summarized in Table 2.6. For each open-response question, a different number of

Table 2.5 Provision of services and satisfaction

	Mean ± Standard deviation	*p*-value
Total Satisfaction		
Genetic counselor (*n*=17)	32.94 ± 3.07	
Physician (*n*=13)	31.54 ± 3.64	0.490
Unsure (*n*=1)	31.00	
GC understood family stressors		
Genetic counselor (*n*=17)	4.82 ± 0.39	
Physician (*n*=13)	4.46 ± 0.88	0.022
Unsure (*n*=1)	3.00	
GC helped identify information to help with decisions		
Genetic counselor (*n*=17)	4.76 ± 0.56	
Physician (*n*=13)	4.77 ± 0.60	0.924
Unsure (*n*=1)	5.00	
I understood concepts from the session		
Genetic counselor (*n*=17)	4.82 ± 0.39	
Physician (*n*=13)	4.62 ± 0.51	0.139
Unsure (*n*=1)	4.00	
GC allowed time for talking with community/family		
Genetic counselor (*n*=16)	4.63 ± 0.62	
Physician (*n*=11)	4.64 ± 0.67	0.852
Unsure (*n*=1)	5.00	
Session was appropriate length of time		
Genetic counselor (*n*=17)	4.59 ± 0.71	
Physician (*n*=13)	4.62 ± 0.51	0.647
Unsure (*n*=1)	4.00	
GC was concerned about my well-being		
Genetic counselor (*n*=17)	4.76 ± 0.56	
Physician (*n*=13)	4.62 ± 0.65	0.710
Unsure (*n*=1)	5.00	
Session was valuable to me		
Genetic counselor (*n*=17)	4.82 ± 0.53	
Physician (*n*=13)	4.54 ± 0.88	0.505
Unsure (*n*=1)	5.00	

participants responded. There were 35 total responses across the questions and the highest number of responses for a question was 13/33. Since the response rate was lower among the open-response questions and several common themes emerged across questions, the responses to the open-response items were analyzed as a whole as opposed to item-by-item. The responses were categorized into three mean themes: responses pertaining to the rapport-building aspects of the session, the cultural appropriateness of the session, and the impact the session had on medical decision making. There were also several responses that mainly provided feedback, such as suggestions for improvement or expressions of gratitude for the clinic, and these were categorized separately.

2.4.7 Rapport-building aspects of the session. There were eight responses that related to the rapport-building aspects of the session. Many of these responses indicated that the genetic counselor showed interest in them and handled their specific situation in a satisfactory manner. For example, with regard to the open-ended question, "How did your genetic counselor show you that they understood your specific needs and what mattered most to you?" one participant responded with, "*By showing compassion and caring in the time of distress for us*."

Another participant cited the lack of pressure to do testing as something they wanted to elaborate on in response to the GCSS. This participant said that, "*It was a relaxed, informative and interesting session*."

One participant described that genetic testing was scary to them, but, "*the whole staff did a good job of helping us understand what was going on and our options from here on. Thank you.*"

Table 2.6 Open-response questions from questionnaire

Open-response questions
Please feel free to elaborate on any of the answers provided above (Likert-scale questions)
Each individual or family who receives genetic counseling has different matters that are important to them, such as being able to share information with family and community before making a decision, religious considerations, or financial concerns to name a few. *How did your genetic counselor show you that they understood your specific needs and what mattered most to you?*
In many genetic counseling sessions, decisions about medical care are discussed. For example, someone might be trying to decide whether or not to have genetic testing, what medical treatment to pursue, whether or not to have more children based on the chance to have children with a genetic condition, or whether or not to be involved in research. *If any situation regarding decision making occurred in your session, how did your genetic counselor help you arrive at a decision?*
Sometimes health professionals don't realize the impact their service has had on families. *If there were any interactions you had that were not helpful or made you feel uncomfortable, please tell us about them here.*
The Community Health Clinic aims to provide genetic services that are tailored specifically to Plain populations. We want to make sure that we are providing the most appropriate and useful services possible. *Please describe what genetic counselors at the clinic can address or do differently to best meet the needs of your family and the Plain community.*

2.4.8 Cultural appropriateness of the session. Nine responses specifically related to the cultural appropriateness that participants felt in sessions.

A few responses specifically referenced God and the Amish church. For example, one participant stated that the genetic counselor, "*Respected our choice of leaving major decision making in the hands of our creator, 'God.*'" Another participant said, "*She helped explain our chances of having more children with PKU…but will let it be God to decide if we have more children*."

One participant mentioned they did not support the idea that the Amish church could help pay for formula, stating, "*we think there should be more options than just send*

bills to the church." Another response brought up this point, with the participant noting that they do not support sending bills to the Amish church.

Several responses referred to Amish values. For example, one participant stated that, "*She gently answered my questions and concerns. I was never pushed out of my comfort zone and she understood my values*." Another participant said that the genetic counselor listened to them and, "*considered our way of thinking*." One participant said, "*We felt comfortable with your helpful advice and we'd feel comfortable to send our friends and our family to you.*" One response was regarding the house visits that the clinic makes, and the participant expressed gratitude for this.

A few responses touched on more specific Amish values, such as photography and family planning. One participant suggested, "*to not focus too much on family planning, although necessary in some situations*." Another participant discussed pictures taken of their child's birthmark, and although the participant was uncomfortable with pictures, the genetic counselor was, "*respectful of taking it only of the birthmark*."

2.4.9 The impact of the session on their medical decision-making. A total of 13 responses were related to decision making, or the medical aspects of the session. Many participants commented on how the genetic counselor gave all the options, but ultimately left decisions up to the participant. One participant put it this way, "*She helped me by explaining everything first and then made suggestions of what I could do in advance. She narrowed everything down so I would know my options. The final decision was mine, if I wanted to act on it!*" There were five other similar responses.

A few responses related to how the counselor explained things. For example, one participant said, "*They done a good job in showing us how a genetic thing in the family*

runs and we now understand it better." Other participants noted the thoroughness of the healthcare provider's explanations.

Other responses related to how a condition would affect the participant. For example, one participant said, "*She helped me by trying everything to figure out what caused my condition and to maybe prevent it from happening to someone else in the family.*" Another participant said, "*I have been very grateful for CHC, it has gave me much peace to know what my condition is and to know what my future looks like. Thank you for all you have done*."

2.4.10 Feedback There were four responses that were suggestions or comments. There were two comments that expressed gratitude for the clinic and how helpful the services are. Other suggestions included, "*we would be very excited if the clinic would be able to get formula for a cheaper price. We get a better price if we order it by ourselves*." And, "*Shorter wait periods? For testing, etc*."

2.5 Discussion

The Amish are a culturally unique population that have a high incidence of genetic conditions. This population has been studied by medical geneticists since the 1960's, and clinics that provide services for this population have been established since the late 1980's. However, no studies to the principal investigator's knowledge have been conducted on how the Amish perceive the genetic counseling services available to them. Similar to previous studies on patient satisfaction with genetic counseling, responses supported that Amish individuals were satisfied with their genetic counseling experience (Bleiker et al., 1997; Bober, Hoke, Duda, & Tung, 2007; Kaduri, Zlotogora, & Peretz,

1998.; Nordin, Lidén, Hansson, Rosenquist, & Berglund, 2002; Tercyak, Johnson, Roberts, & Cruz, 2001).

This is the first study to address Amish perceptions of genetic counseling at a clinic that has been established specifically for this population. Although this was an exploratory study with a small sample size, it provides insight on Amish perceptions of aspects of genetic counseling sessions including rapport-building, cultural appropriateness, and impact on medical decision-making.

2.5.1 Practice implications. This study showed that, overall, satisfaction with genetic counseling was high among the Amish. There were some statistically significant differences when certain demographic variables were compared within the high satisfaction scores.

Firstly, while men and women were both satisfied with the genetic counselor's concern for their well-being and the valuableness of the session, women were more satisfied than men. Differences in roles between Amish men and women could help explain this difference. After 8th grade, girls are taught homemaking and boys learn farming (Francomano, 1996). Gendered difference in lifestyle could contribute to differences in how Amish men and women perceive the value of a service such as genetic counseling. Men and women in general tend to have different health practices, irrespective of religious practices. For example, men are less likely to participate in routine health care visits. One author proposes that this could be explained as men rejecting feminine ideals when it comes to healthcare. Women are more likely to be proactive with regard to healthcare, so to be more masculine, men forgo these health practices (Courtenay, 2000), which could also help explain the difference in satisfaction

between men and women with regard to the value of the genetic counseling session and the genetic counselor's concern for their well-being. Additionally, genetic counselors are trained on the importance of engaging each individual when counseling a couple. Doing so helps to establish rapport with both individuals and makes each person feel heard (Weil, 2000). Often, women are in charge of health information for the family, which could result in a counselor focusing on the woman in a session (Gaff & Bylund, 2010). It is also possible that this could help explain the gendered difference in satisfaction observed in this study. While there are gendered lifestyle differences among Amish men and women, there are also differences among men and women in general. Therefore, it is unclear exactly what contributed to the difference in satisfaction between men and women in the current study.

Satisfaction with the length of the session increased from a referral for a condition in the family, to a referral for a condition in the participant's child, with the most satisfied individuals being those referred for a condition affecting themselves. Due to the nature of the question, it cannot be surmised whether the differences in satisfaction were a result of a too lengthy session or one that was too short. One study that interviewed non-Amish individuals who had received genetic counseling found that all participants were pleased with the length of the session, citing that other health care professionals do not typically spend as much time with them. Genetic counselors interviewed in this same study, however, did note that the length of a session does not necessarily correlate with success (Bernhardt, Biesecker, & Mastromarino, 2000). Of note, the appointment times at CHC are longer than a typical appointment, with new patients being allotted 2-3 hours for an initial visit. Genetic counseling may or may not occur during this initial visit, however

the actual time for genetic counseling is likely not much different than at other clinics (R. Evans, personal communication, April 8, 2019).

While participants were satisfied with both the physician and the genetic counselor's ability to understand family stressors, satisfaction was higher for participants who received genetic counseling from a genetic counselor. Genetic counselors receive focused training on psychosocial techniques as part of their education. Additionally, the CHC intentionally hired a physician with excellent psychosocial skills, knowing that these skills would be particularly important for building relationships with the Amish community (R. Evans, personal communication, April 8, 2019). Thus, the difference in satisfaction is not likely due to a difference in psychosocial skills. When looking at the overall mean satisfaction score for the question regarding family stressors, it is the second lowest of all the questions. This implies that those delivering genetic counseling at the clinic could attempt to have a greater understanding of patient stressors and take time during sessions to explore these stressors.

The open-ended responses also indicated that the Amish were overall satisfied with their genetic counseling experience. Many responses indicated a general sense of satisfaction with the session regardless of culture. These responses reflect that those providing counseling in this clinic are skilled at genetic counseling. For example, many responses indicated that the genetic counselor successfully built rapport and helped with medical decision making, exemplified by participant statements that their counselor showed them compassion and laid out all options but ultimately left the decisions up to them. These responses align with the definition of genetic counseling developed by the National Society of Genetic Counselors (NSGC) Task Force:

Genetic counseling is the process of helping people understand and adapt to the medical, psychological and familial implications of genetic contributions to disease. This process integrates the following:

- Interpretation of family and medical histories to assess the chance of disease occurrence or recurrence.
- Education about inheritance, testing, management, prevention, resources, and research.
- Counseling to promote informed choices and adaptation to the risk or condition (Resta et al., 2006)

A similar definition of genetic counseling was provided to participants on the questionnaire.

Importantly, many responses also centered on specific cultural needs that were met by those providing genetic services at the CHC. Several participants expressed that the genetic counselor was accepting of their faith and the role it plays in medical decision-making. The importance of religion amongst the Amish population is well-described (Francomano, 1996; Cates, 2014). Responses from the current study support the idea that showing understanding and acceptance with regard to the religious aspect of Amish culture is appreciated when counseling these individuals. Other responses mentioned that the genetic counselor considered the Amish way of thinking, and one participant said they would feel comfortable sending friends and family to the clinic. These responses reflect the importance of understanding Amish values when counseling this population and also support that those performing counseling at this clinic were culturally competent.

One of the concerns that was brought up in the open-response portion of the questionnaire was regarding billing, which is also unique to Amish culture. The participant expressed that they do not support sending bills to the Amish church. Within the community that the CHC serves, a healthcare sharing ministry has been established called the Plain Church Group Ministry (PCGM). Most likely this participant is referring to the sending of bills to PCGM. It is unclear why the participant was upset with this process, since the establishment of PCGM was largely a community decision. Perhaps this individual is dissatisfied with how their community has chosen to handle billing. Given that there is a general proscription against insurance in the Amish community (Francomano, 1996), it is possible that this participant felt this is similar to insurance. Since this study used survey methodology, we could not follow up with this participant to clarify their statement.

Another participant expressed that they were able to get formula prices for cheaper on their own, and they would be excited if CHC could get the formula for a cheaper price. PCGM has negotiated for the cheapest possible price and are able to provide formula for wholesale price. There is a 2% administrative fee added on to this price that covers PCGM services, which could be what this participant is referring to. The participant is possibly getting formula at a cheap price directly through the formula company through a patient assistance program (R. Evans, personal communication, April 8, 2019).

Finally, a participant suggested that shorter wait periods for testing would be helpful. It is possible that this participant had experience with whole exome sequencing, which the CHC does through CSC on a research basis. Thus, the testing can take a long

time to come back. Otherwise, the CHC uses standard labs which do have good turnaround times (R. Evans, personal communication, April 8, 2019).

2.5.2 Study limitations. This study had several limitations. First, the study had a small sample size. Additionally, participants came from one clinic, the CHC. The CHC is one of several clinics that provides specialized services for Plain populations. While the CHC is similar to clinics such as the Clinic for Special Children in Pennsylvania, each respective clinic is unique with regard to the personnel, policies and services available to patients. For these reasons, this study is not generalizable to all clinics that provide genetic services for Amish populations.

In addition, the methodology of this study was a questionnaire, which did not allow the principal investigator to follow up on responses.

2.5.3. Future research. Future studies focused on Amish experiences with genetic services could aim for a larger sample size and include participants who have received genetic counseling from several clinics that serve the Amish. Additionally, future studies could focus on Amish who receive genetic counseling from clinics that do not primarily focus on the Amish. This could give more insight into this patient population as well as allow for more generalizability. Based on some of the responses from this study, it could be useful to conduct interviews as opposed to or in addition to questionnaires which could allow for more follow up questions. It may also be helpful to focus on specific conditions and Amish experiences with these conditions. Additionally, it could be helpful to interview leaders of the Amish community such as deacons who are often involved in decisions for individuals and families. Finally, it could be interesting to

survey genetic counselors who have had experience working with the Amish to gain insight on their perspectives.

2.6 Conclusion

Genetic counseling services provided at the CHC have been successful at meeting the specific needs and values of the Amish, given the overall satisfaction represented by this inquiry. Responses from this study support that genetic counselors have been able to adapt and meet the needs of this specific population. The Amish participants in this study were highly satisfied with their genetic counseling experience, and their responses highlighted several specific aspects of the session that they were satisfied with: rapport-building aspects of the session, medical decision-making portion of the session and the cultural appropriateness of the session. Importantly, the current study highlights the value of genetic counseling for the Amish community. While the Amish are often thought to not utilize modern medicine, the current study clearly demonstrates that this population can benefit from genetic counseling services and is accepting of these services when provided in a culturally competent manner. More broadly, the current study emphasizes cultural competence as an important factor in providing successful genetic counseling services to populations who may show reluctance or distrust towards the medical community.

Chapter 3: Conclusions

The Amish are a culturally unique group that have added significantly to our understanding of genetic conditions. Due to the high incidence of genetic conditions in this population, special genetics clinics have been established to meet the needs of the Amish. While many studies have been done on medical genetics in the Amish, only one study to date has assessed Amish perceptions of genetics. The current study was the first study to specifically address Amish perceptions of genetic counseling. The study assessed Amish satisfaction with genetic counseling services and gained insight into how Amish individuals felt about the rapport-building and medical-decision making aspects of a session, as well as the cultural appropriateness of the session.

While the sample size was small and the results are not generalizable, this exploratory study provides important insight into Amish reflections on genetic counseling. The Amish were highly satisfied with their genetic counseling experiences, adding to the body of literature showing high satisfaction rates with genetic counseling services. Additionally, Amish individuals expressed satisfaction in their own words though open-response questions. Importantly, Amish responses spoke to the cultural appropriateness of the genetic counseling they received.

This study demonstrated that genetic counseling services provided at the CHC met the needs of their Amish patients in a culturally competent manner and highlighted the value of genetic counseling for the Amish community. The current study also showed the significance of cultural competence in providing successful genetic counseling

services for minority populations who may show general reluctance or distrust of the medical community.

Similar studies could be conducted at similar clinics as well as at non-Amish focused clinics, and more in-depth studies could be conducted to gain further insight and to continue listening to Amish perspectives.

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Appendix A: Recruitment Letter

Hello,

We are writing to let you know that The Community Health Clinic (CHC) has been working with Brianna Teapole, a genetic counseling graduate student at the University of South Carolina, to help her complete her Master of Science thesis project. With our help, she is trying to learn more about Amish community members' experiences with genetic counseling, in order to assess and improve the care we provide to the Plain community.

Because you have received genetic counseling at some point in the past through the CHC, we invite you to complete the enclosed survey. The questionnaire should take approximately 5-10 minutes to complete, and there will be no compensation or payment for participation. However, we greatly appreciate your time and insight. We hope that the results of this study will benefit the genetic counseling experience of Amish individuals in the future.

Your participation in this study is completely voluntary; all responses are anonymous and kept confidential. Your decision to participate or not will in no way affect your medical care. Completion of this questionnaire implies you understand that it is being used in a study and that you give us permission to use your responses without your name attached to evaluate the genetic counseling process. You may stop completing this questionnaire at any time or choose not to answer specific questions. Once you complete the survey, please place it in the provided stamped and addressed envelope, and mail it back to the CHC.

If you have any questions, please do not hesitate to contact Brianna at 336-380-7219 or her faculty advisor, Janice Edwards at 803-545-5775. If you would like to learn the results of the study after its completion, please contact the CHC after May 2019.

Sincerely,
Jeremy Kauffman
Executive Director
The Community Health Clinic

Appendix B: Questionnaire

Thank you for your input on the best way to provide genetic counseling in The Community Health Clinic. Your ideas will help shape the service to best meet the needs of the Plain community.

Demographics

1) What is your gender? Male /female

2) What is your age?

3) With which group do you belong? Amish, Mennonite, or Other

Genetic counseling helps people understand how genetic conditions can impact an individual or family. Genetic counselors provide information such as how medical conditions are inherited (passed down through a family) and how likely it is for the condition to occur again in a family. They can discuss medical care and help patients understand genetic testing options that are available to them. The process of genetic counseling can be done by genetic counselors, who have a master's degree in genetic counseling, or physicians with special training in genetics. For example, you may have received genetic counseling from Rebecca Evans or Beth Woolley, who are both genetic counselors, or Dr. Zineb Ammous, who is a physician.

4) At one of your past visits at The Community Health Clinic, were you aware that you received genetic counseling? Circle one please:

   Yes/no/unsure

5) You received genetic counseling related to:

   a. A genetic condition affecting yourself
   b. A genetic condition affecting your child
   c. A genetic condition running in your family

6) You received genetic counseling from:
   a. A genetic counselor
   b. A physician
   c. Unsure

The following questions relate to your satisfaction with the genetic counseling services you received. Please circle the number that corresponds with how strongly you agree or disagree with each statement:

	Strongly Disagree	Disagree Somewhat	Uncertain	Agree Somewhat	Strongly Agree
7) My genetic counselor seemed to understand the stresses my family and I were facing	1	2	3	4	5
8) My genetic counselor helped me to identify what I needed to know to make decisions about what would happen to me and/or my family	1	2	3	4	5
9) I understood the concepts the genetic counselor explained to me in the session	1	2	3	4	5
10) The genetic counselor allowed time for me to talk with family and community members before making a decision	1	2	3	4	5
11) The genetic counseling session was about the right length of time I needed	1	2	3	4	5
12) My genetic counselor was truly concerned about my well-being	1	2	3	4	5
13) The genetic counseling session was valuable to me	1	2	3	4	5

14) Please feel free to elaborate on any of the answers you've provided above:

The following questions are free response so that we can understand more about your experience and ideas for improving genetic counseling at the CHC. Please provide as much detail as you are able.

15) Each individual or family who receives genetic counseling has different matters that are important to them, such as being able to share information with family and community before making a decision, religious considerations, or financial concerns to name a few. *How did your genetic counselor show you that they understood your specific needs and what mattered most to you?*

16) In many genetic counseling sessions, decisions about medical care are discussed. For example, someone might be trying to decide whether or not to have genetic testing, what medical treatment to pursue, whether or not to have more children based on the chance to have children with a genetic condition, or whether or not to be involved in research. *If any situation regarding decision making occurred in your session, how did your genetic counselor help you arrive at a decision?*

17) Sometimes health professionals do not realize the impact their service has had on families. *If there were any interactions you had that were not helpful or made you feel uncomfortable, please tell us about them here.*

18) The Community Health Clinic aims to provide genetic services that are specifically tailored to Plain populations. We want to make sure that we are providing the most appropriate and useful services possible. *Please describe what genetic counselors at the clinic can do differently in order to best meet the needs of your family and the Plain community.*

Appendix C: Responses to Open-response Questions

Please feel free to elaborate on any of the answers provided above (Likert-scale questions)

Dr. Z had explained about our genetic heart disease that is in our family. She did a good job in going through everything. Thank you for all that you do at the clinic to help us out!

I loved how my counselor thoroughly explained everything and took time to draw pictures and elaborate for better understanding. There wasn't any pressure to do further testing on my parents if they didn't want to. It was a very relaxed, informative and interesting session.

Genetic testing was new to us- kinda scary- the whole staff did a good job of helping us understand what was going on and our options from here on. Thank you

I have been very grateful for CHC, it has gave me much peace to be able to know what my condition is and to know what my future looks like. Thank you for all you have done.

We were wondering about our child. There was nothing serious going on. She needed other medical attention.

The answer to number 9- we had no immediate decision to make.

Each individual or family who receives genetic counseling has different matters that are important to them, such as being able to share information with family and community before making a decision, religious considerations, or financial concerns to name a few. How did your genetic counselor show you that they understood your specific needs and the what mattered most to you?

Counselor provided detailed information of why certain mutations are more prominent in Amish community (smaller gene pool) and further testing was optional and available but not demanded. I tested positive for the mutation but an echo showed no abnormalities currently and the clinic offered further testing and support but I declined because of distance (travel), time issues and everyone was understanding!

My counselor laid out the picture of the genetic disorder I was wondering about and sent in a blood sample to determine what's going on.

By showing compassion and caring in the time of distress for us.

She shared a few of her experience similar to mine so she could relate until what I was going through. She gently answered my questions and concerns. I was never pushed out of my comfort zone and she understood my values.

They done a good job in showing us how a genetic thing in the family runs and we now understand it better.

Respected our choice of leaving major decision making to the hands of our creator, "God."

Explained everything in detail.

The clinic is very supporters of sending bills to Amish church- we don't agree to this.

Showed interest and paid attention thanks!

Listened to what we have to say, considered our way of thinking.

She helped me by trying everything to figure out what caused by condition and to maybe help prevent it from happening to someone else in my family.

They took the time to listen and to take the cheapest route but still helps!!

Can't really remember, as I myself didn't have this genetic problem and all 3 of our children that have it were adults.

In many genetic counseling sessions, decisions about medical care are discussed. For example, someone might be trying to decide whether or not to have genetic testing, what medical treatment to pursue, whether or not to have more children based on the chance to have children with a genetic condition, or whether or not to be involved in research. If any situation regarding decision making occurred in your session, how did your genetic counselor help you arrive at a decision?

I was satisfied with what they found and that they did a good job of handling my situation. I also spoke to Dr. Z and she gave me some insight in my meds that I didn't need.

By talking about options we had and weighing the differences.

She helped me by explaining everything first and then made suggestions of what I could do to advance. She narrowed everything down so I would know my options. The final decision was mine, if I wanted to act on it!

She explained things and brought out points.

She helped explain our chances of having more children with PKU… but will let it be God to decide if we have more children.

She just explained everything thoroughly and the pros and cons of knowing or not.

She gives her advice but still leaves it up to us. She is not my way or know way!!

She gave her opinion and that helped make our decisions as she knows more about than us.

I was past child bearing age.

Sometimes health professionals don't realize the impact their service has had on families. If there were any interactions you had that were not helpful or made you feel uncomfortable, please tell us about them here.

We felt comfortable with your helpful advice and feel we'd feel comfortable to send our friends and family to you.

We are not supporters of the idea that the Amish church can help pay for formula…we think there should be more options than just send bills to the church.

Pictures taken of barbs birthmark made us uncomfortable but she was respectful of taking it only of the birthmark.

The Community Health Clinic aims to provide genetic services that are tailored specifically to Plain populations. We want to make sure that we are providing the most appropriate and useful services possible. Please describe what genetic counselors at the clinic can address or do differently to best meet the needs of your family and the Plain community.

Shorter wait periods? For testing etc.

To my way of thinking, everything was done satisfactorily and was satisfied how everything was handled.

To not focus to much on family planning, although necessary in some situations. Thank you.

We would be very excited if the clinic would be able to get formula for a cheaper price. We get a better price if we order by ourselves.

Your service was helpful to us and you know what you are doing. Keep it up.

They already come out for house visits and that sure means a lot with a family and the distance to travel! Thank you all a lot!!!

Thank you for what you all do to help us out!

At the time we were there the office gal was super nice but our financial payments were often mixed up or incorrect that we received via mail.