FACTORS AFFECTING MATERNAL COPING AND ADAPTATION FOLLOWING THE PRENATAL DIAGNOSIS OF A RARE BIRTH DEFECT, FIBULAR HEMIMELIA, FOR WOMEN WHO CHOOSE NOT TO TERMINATE

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Table of Contents

List of Tables

List of Figures

Chapter 1

Introduction

Birth defects affect 3% of all births around the world, and the rates appear to be rising, as stated by the World Health Organization and the International Clearinghouse of Birth Defects Surveillance and Research in their 2012 report. According to these statistics, in 2015, there will be approximately 120,000 American children born with a recognized birth defect. The burden of these conditions is high, affecting an individual's morbidity and mortality, impacting their daily functioning not to mention the overall cost of care. For the caregivers of these individuals, an enormous emotional, psychological and practical components are involved in understanding the nature of the birth defect and responding appropriately to the needs of their loved one while balancing the quality of their lives. Discovering that a child has a birth defect while a woman is pregnant can be a catastrophic and debilitating experience. How a woman responds, and what support she receives can impact her decisions about her future and the future of her child.

A prenatal diagnosis of a birth defect provides the mother with an element of forewarning and gives her an opportunity to either learn and understand the new reality she will be managing after the birth of her child, or the chance to decide to terminate the pregnancy on the basis of these findings. These women are subjected to numerous factors which may influence their decision to either sustain or terminate. Exploring these factors, and their impact on the mothers can help society to understand the psychological processes the mothers are undergoing and in turn provide support to those women who

choose to sustain their pregnancy, despite the enormous psychological and practical hurdles they may encounter raising their child with a birth defect.

Birth Defects

It is believed that most birth defects occur during the first trimester of a pregnancy, defined as the first 12 weeks post gestation. At this time, the anatomical development of all the major structures of the fetus takes place. An interruption in genetic coding or a teratogenic element can have catastrophic results on the development of a fetus at this point and result in a defect. Birth defects can vary in severity; defects can affect multiple organ structures and compromising the survival of an individual or go virtually undetected and insignificant. The etiology of some birth defects is well established and, through research and investigation, can be prevented as is the case with Fetal Alcohol Syndrome or Neural Tube Defects; however, many more defects have no identifiable cause and therefore no preventative protocols to protect against their incidence. Many birth defects are identified and possibly diagnosed in utero through the use of prenatal tests during a mother's obstetric care. A common example of this is Down's Syndrome or Neural Tube Defects and the use of maternal serum screening and ultrasounds. Nevertheless, many birth defects are not discovered despite prenatal testing until after birth (and sometimes even later in an individual's life when their effects upon an individual become apparent).

The Center for Disease Control (CDC) estimates that approximately 1 out of 33 births is subject to a birth defect. This is a rate which has been largely stable from 1978 and onwards, which is when the CDC began a novel program tracking the incidence of birth

defects in Metropolitan Atlanta. The Metropolitan Atlanta Congenital Defects Program (MACDP) is just one of thirty nine government funded programs throughout the world that contributes an annual report to the International Clearinghouse for Birth Defects Surveillance and Research (ICBDSR), a voluntary non profit organization affiliated with the World Health Organization established in 1974. The mission of this organization is to "bring together birth defect programmes from around the world with the aim of conducting worldwide surveillance and research to prevent birth defects and to ameliorate their consequences" (ICBDSR, 2012). Data collected by ICBDSR includes elective termination and stillbirth rates of pregnancies for 39 identified birth defects. ICBDSR members collect data only on isolated birth defects, with no other involvement. If a specific birth defect is a result of a syndrome or larger anatomical anomaly, it is not reported individually but rather as the larger causative condition.

Fibular Hemimelia (Postaxial Limb Reduction Defect)

Fibular Hemimelia (FH) is a rare birth defect, occurring in approximately 1/50,000 (Orphanet) births. It is categorized under a Post Axial Limb Reduction Defect in the ICBDSR. The name specifically refers to an underdevelopment of the fibula bone of the lower leg, usually only present in one limb. As with many birth defects, there is quite a wide spectrum whereby this defect expresses itself. Some children are born with a completely absent fibula, a reduced number of toes or rays on the affected leg, and an underdeveloped or mal-developed anatomy, both in soft tissue, vascular, associated neurology and orthopedic involvement of the entire affected limb. For some, Fibular

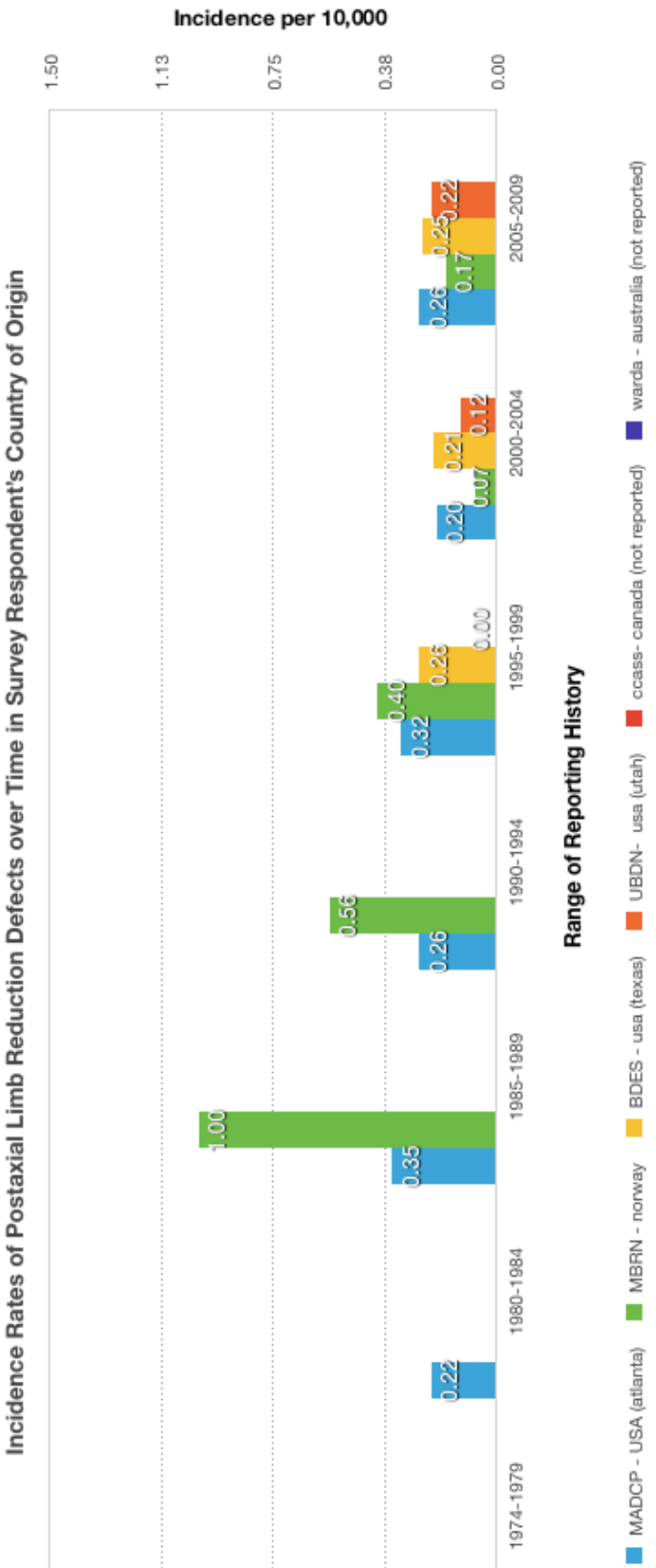
Hemimelia presents as a slightly shortened fibula in the affected leg with everything else somewhat intact.

The prognosis for FH also covers quite a wide spectrum for those affected. Surgical intervention is the norm for this condition with repetitive, intensive physiological limb reconstruction and lengthening procedures on one end, and various degrees of amputation and prosthetic usage on the other. Lifelong physiotherapy, impaired gait, arthritis, associated musculo-skeletal deficiencies and stressors, altered mechanics, orthotics and long term surgical and pain management can be expected for individuals with FH who undergo either the limb saving or amputation route as a treatment of choice.

The birth defect can be found in conjunction with other congenital abnormality diagnoses and syndromes, but most often it is isolated. Researchers, Lewin and Optiz, suggest that there may only be 0.8% of Fibular Hemimelia cases that are associated with other nonskeletal malformations. When it is isolated, it does not involve or affect any vital organ or neurological/cognitive function. The etiology of this defect is unknown, but is not believed to be a result of chromosomal or hereditary factors. There is no evidence of any connection to race, maternal age, environmental factors or influence in the prevalence of this defect. The incidence rate of the birth defect has been fairly steady since birth defect surveillance organizations started recording it in 1974 as seen in Table 1.

Fibular Hemimelia is very easy to identify and diagnose on a standard obstetric level one ultrasound; however, because it is a rare condition and therefore not often seen, radiologists and neonatologists use numerous differential diagnoses.(Radler et al.). These

Table 1


Incidence Rates of Postaxial Limb Reduction Defects over Time in Survey Respondent's Country of Origin
Incidence per 10,000
1.50
1.13
0.75
0.38
0.00
1974-1979
1980-1984
1985-1989
1990-1994
1995-1999
2000-2004
2005-2009
Range of Reporting History
0.22
0.22
1.00
0.35
0.56
0.26
0.40
0.32
0.25
0.00
0.21
0.12
0.20
0.26
0.17
0.25
0.22
MADCP - USA (atlanta)
MBRN - norway
BDES - usa (texas)
UBDN- usa (utah)
ccass- canada (not reported)
warda - australia (not reported)

differential diagnoses include Club Foot, Amniotic Band Syndrome, Proximal Femoral Focal Deficiency and Generalized Limb Dysplasia. Regardless of these initial differential diagnoses, the pregnancy and associated fetus has now been identified as having a Limb Reduction Defect of a Post-Axial Nature and it is now considered a potentially high risk pregnancy.

Termination of Pregnancy (ToP)

The International Clearinghouse for Birth Defects Surveillance and Research includes Limb Reduction Defects as a category in its standardized reports from global members. The prevalence rates remain relatively consistent among each of the 39 contributing programs around the world as reported in their latest compilation, 2012, and stable over the last 40 years. See Table 1. The data shows the average prevalence of Limb Reduction Defects at 5.3 per 10,000 births. This number is the total incidents of the birth defect and includes data from still births, terminations of pregnancies and live births. The reported rates of elective termination of pregnancies varies quite dramatically for this birth defect though, among the different countries and regions. The highest percentage of termination of pregnancy (ToP) rates for a Limb Reduction Defect is 46.5% reported in the REMERA program in Lyon, France. The lowest reported ToP is 3.2% by the Texas Birth Defects Epidemiology and Surveillance Branch. Of all the other contributing members of ICBDSR, the average ToP percentage for Limb Reduction Defects is 24.0%. This means that of all fetuses diagnosed with a limb reduction defect, including Fibular Hemimelia, worldwide, almost 1 in 4 of them will be terminated. See Table 2. The following table reports the prevalence and reported rates of termination of pregnancy for a specific birth

defect category, Limb Reduction Defects. It should be noted however, that this category is a broad one which includes five subcategories relating to the exact location of the anatomical birth defect. These subcategories are Transverse, Preaxial, Postaxial, Intercalary, Mixed and Unspecified. Fibular Hemimelia is only one condition that can be found under the subcategory of Postaxial Limb Reduction Defects. With this is mind, it further illustrates the rarity of this defect is, the difficulty in obtaining reliable data, and the limited medical resources and expertise on this condition. The table also highlights the potential for sense of isolation that patients and caregivers experience when diagnosed.What is it that is causing this dramatic difference in termination rates for this birth defect considering that the viability of the fetus is unaffected and the overall health and cognitive ability is not impaired of the affected individual? This question is a challenging one and potentially can only be theoretically postulated as women who have chosen to terminate would naturally be reticent of having their motives questioned about a decision they made to abort their fetus based on a defect. In light of that, it is worthwhile to consider the opposite. What are the circumstances women are subject to when the fetus they are carrying has been diagnosed with a Limb Reduction Defect, specifically Fibular Hemimelia? What factors influenced their decision to sustain the pregnancy? What challenges did they face during their pregnancy that they overcame and resolved them to have their children? What factors helped them cope with the diagnosis and allowed them to make a decision to not terminate? This research paper has identified a very specific population of mothers who received an in utero diagnosis of Fibular Hemimelia for their fetuses. These women chose to not terminate their

pregnancies based on the diagnosis. What follows is an examination of their experiences within the medical arena, their community and an introspective look at their individual,

Table 2

Total Limb Reduction Defects ICBDSR (2011 report, 2009 data)

Contributing Program	Country	Prevalence / 10,000	ToP%
WARDA	Australia	4.8	40.7
ACASS	Canada-Alberta	14.2	18.3
CCASS	Canada-National	3.9	nr
RECUMAC	Cuba	1.22	28.9
Czech	Czech	7.84	17.9
Finland	Finland	8.08	23.1
PARIS	France	5.89	43.1
REMERA	France	5.42	46.5
Strasbourg	France	9.14	31.0
Saxony Anhalt	Germany	5.81	36.4
Hungary	Hungary	3.65	18.3
BDRI	India	2.8	13.7
IBDSP	Israel	2.33	11.1
BDRCam	Italy	4.86	17.6
IMER	Italy	2.1	29.2
North East	Italy	1.54	23.4
Tuscany- RTDC	Italy	5.56	31.0
Northern Netherlands	Netherland	3.86	19.2
MBRN	Norway	4.58	37.3
MRRCM	Russia	2.03	15.4
Sweden	Sweden	5.53	18.7
CARIS	UK - Wales	9.68	35.4
MACDP	USA- Atlanta	6.69	10.8
BDES	USA_ Texas	5.45	3.2
UBDN	USA_ Utah	5.91	4.8
		5.3	24.0

ToP = Termination of Pregnancy
Prevalence = Live births + Still Births + ToP

personal characteristics in determining how they coped with the diagnosis and how they came to the decision they did: To not abort.

Research Methodology

This research seeks to examine the various factors in the maternal environment of a woman whose fetus is diagnosed prenatally with a congenital defect. A retrospective study was done on biological mothers of children who were diagnosed with the Limb Reduction Defect, Fibular Hemimelia, during their pre-natal period. These subjects made an informed decision to not terminate their pregnancies despite the option being available and offered to them.

The original research was conducted online through a secured, private, survey website, *Survey Monkey*, over a period of three months. Subjects were recruited from three social media support groups on Facebook: *Fibular Hemimelia Support*, *Fibular Hemimelia and Limb Lengthening Awareness* and *Fibular Hemimelia and Amputation Awareness*. These groups were a mixture of both member only and public access and their membership totaled over 2, 200 people both with FH and parents or loved ones of those with FH. Inclusion criteria for the survey were biological mothers that have a child with Fibular Hemimelia (FH) exclusively. The children had to have been diagnosed with a limb reduction deficit in utero and where pregnancy termination was a sanctioned option. Children who had significant co-morbidities or syndromes that included FH were disqualified as were children who had siblings with the same diagnosis as a genetic factor could therefore not be ruled out.

The survey consisted of 50 questions collecting various demographics on the participant mother, and exploring the topic utilizing multiple choice, rating and comment/ essay style questions. They were given the option of abstaining from certain questions that they were not comfortable answering. In total there were 16 respondents, only six of which met all of the above criteria and were included in final analysis. There was no geographical restriction on where the respondents reside. In light of this, we have data included from respondents in the United States, Canada, Australia, and Norway. There were significant limitations to the study, primarily due to the low incidence of Fibular Hemimelia in the general population. Internet and Facebook access, English proficiency and specifically requiring the subjects to have pre-natal knowledge of a limb reduction defect or FH diagnosis further restricted those able to participate and became a factor influencing the sample size.

Questions on the survey addressed three primary areas: Medical Arena, Community Resources, and Individual Characteristics of the mother. Respondents were asked detailed questions about their prenatal experience with their primary care provider and adjunct medical personnel, re-classification of the pregnancy post diagnosis and communication styles, referrals and resources provided to them by medical personnel. Within the subject's community, the research tried to identify the presence of an established support or information systems through organized religions or groups, employers and family/friend networks. The nature of the relationship between the mother and her partner was also examined. In addition, the concept of a virtual community was also addressed, to find if access to internet searches and online support groups proved

influential. Finally, the mother's individual personality and psychological make-up was probed to determine if there were any resonating characteristics that could be identified as influential on the respondent's experience, coping skills and ultimate decision to sustain the pregnancy.

Chapter 2

Medical Arena

Pregnancy and birth were the domains of women for centuries. Female attendants or midwives assisted at births and used wisdom acquired through the generations before them to deliver a mother of her child. Starting in the 17th century, men became interested and involved in obstetrics, introducing instrumental and surgical deliveries, and the concept of asepsis and anesthesia. However, it was not until the early 20th Century that antenatal or prenatal care, the oversight of the pregnancy itself, became an element in obstetrics.

The practice of obstetrics and prenatal care has made revolutionary advances in the last 40 years in Western Medicine. Prenatal testing is just one aspect of obstetrics that has seen dramatic developments. According to the American College of Obstetrics and Gynecology's (ACOG) guidelines, prenatal tests are primarily used to identify defects with the developing fetus and are divided into two categories: Screening and Diagnostic. Screening tests include complex maternal blood work analysis and ultrasound imaging and are used to identify possible defects. These tests pose no risk to the fetus and are largely non-invasive. Under certain conditions a mother, pregnancy or fetus may be considered to be at higher risk for birth defects or a screening prenatal test may come back flagged with an anomaly. It is at this point that diagnostic prenatal tests are utilized. Fetal cell analysis through placental, umbilical and amniotic tissue samples, targeted ultrasound exams, and genetic screening allow a clinician to have a much more thorough understanding of the fetus' health, development and genomic status. Thus, prenatal

testing has been an outstanding tool for early identification and diagnosis of possible complications in the gestation of the fetus and the presence of birth defects. The results can allow clinicians to tailor the care of their patient to optimize outcomes. It often also allows the mother with an early warning of a problem and the option to terminate her pregnancy based on the results or alternatively gives her time, should she chose to continue her pregnancy, to research and prepare for the birth of a child, potentially with special needs.

Perhaps one of the most influential and revolutionary tests used in prenatal care since the 20th century is that of the ultrasound scan. Obstetric ultrasounds became widely used beginning in the 1970s and represented the first time that a developing fetus could be imaged, physical development quantified and thus in turn diagnosed with certain anatomical anomalies, which would suggest specific birth defects. In the last 45 years, ultrasound scans have been part of standard prenatal care in the developed world and during that time extensive data has been collected on fetal physical development milestones and therefore observable/measurable anatomical defects or deficits. Ultrasounds have become such a common test performed during pregnancy that it has gone from a diagnostic and clinical tool to become an instrument for an emerging commercial entertainment enterprise. Private, non-clinical facilities now offer 3D and 4D ultrasound scan versions of a fetus to clients as mementos to take home to excited family and friends.

The caveat of all this prenatal testing is that once a mother undergoes these tests and is given the results, she is in a position to make choices. For women whose fetuses do

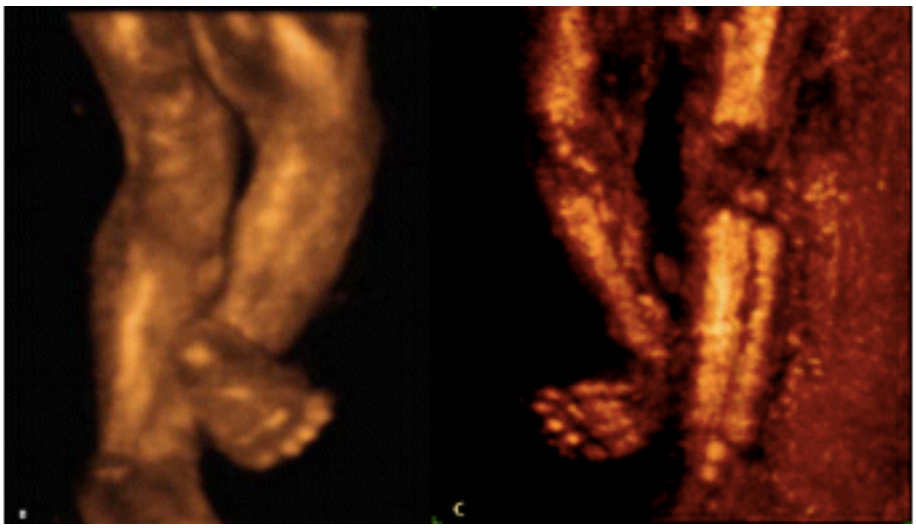
come back with anomalies or defects this process of understanding, acceptance, weighing options and making decisions can challenge the core of a woman's concept of motherhood and forces her to re-evaluate her role in relation to a child that is no longer perfect or necessarily healthy (Lalor). According to research done by McKechnie, "Following a prenatal diagnosis, expectant parents are at risk for clinically concerning levels of distress including episodes of major depression, severe anxiety, traumatic stress and grief. Both expectant mothers and fathers emotional responses to the diagnostic news are characterized as intense and consistent with a grief reaction" (McKechnie 37).

Ultrasounds and Fibular Hemimelia

At approximately 10 to 12 weeks of age, ossification of the long bones in the lower limbs begins and from previous membranes and cartilage, bones begin to appear in the fetus. Routine second trimester (14 -26 weeks gestation) ultrasounds look at long bone length in the limbs to accurately date and size a fetus (Bagnall et al.). The American Institute for Ultrasound in Medicine (AIUM) in conjunction with ACOG developed guidelines for routine obstetric ultrasounds after 18 weeks where fetal biometry measurements are taken, including the limbs and extremities of the fetus (AIUM, 2013). When there is a dramatic disparity between the limbs, a flag is immediately raised. Below is an example of an ultrasound that highlights a significant limb reduction defect in a 21 week old fetus (see Figure 1). It includes images of the lower limbs of a single female fetus, both with and without x-ray mode. The image clearly shows an absent fibula, bowed tibia, leg length discrepancy and outward deflecting clubfoot of the right leg only. The left leg is anatomical unremarkable. This clinical finding is indicative of

Fibular Hemimelia and is the beginning of the mother's experience of discovering her fetus has a birth defect.

Figure 1: Three Dimensional Ultrasound (with and without X ray mode) of Unilateral Fibular Hemimelia in a 21 Week old Fetus (Monteagudo)



When examining the influence of the medical arena on the experience of the pregnant woman and her ability to cope with the prenatal diagnosis, the survey examined two main areas: Prenatal Identification, Intervention and Medical Management of Prenatal Diagnosis of FH, and the Communication and Resources provided by Medical Professionals to Mothers.

These two factors combined to have an impact on the mother's ability to cope with the unexpected diagnosis of her fetus and in turn may have contributed to her decision to not terminate the pregnancy based on the diagnosis.

Prenatal Diagnosis, Intervention and Management

With the diagnosis of a birth defect during a routine second trimester ultrasound, the management of the pregnancy often takes a very different turn in the medical community. This new path often provides cues to the mother as to how significant the finding is and what level of concern should be attached to it. There were a variety of primary care providers for the mothers in this study. Two thirds of their pregnancies were managed by an obstetrician, while one third had either a midwife or family doctor. Upon the prenatal diagnosis of Fibular Hemimelia, half of the respondents stated that their pregnancies were considered high risk, while the other half did not. Of all the mothers though, only one had her pregnancy formally classified as high risk due to the prenatal diagnosis of her child and had her care transferred from an obstetrician to a specialized Fetal Medical Unit in a large teaching hospital. The other mothers had their pregnancies managed with their original care providers, obstetrician, midwife or family doctor, and the status of their pregnancies did not alter. It bears repeating that Fibular Hemimelia is not associated with any mortality of the fetus, nor does it confer any complications or risks on the pregnancy or delivery itself. The act of identifying the pregnancy as high risk, and a resultant transfer of care based on the diagnosis therefore could be interpreted as an excessive reaction and alarming for the mother.

All of the subjects were notified of a limb defect during a routine obstetric ultrasound. Technicians performing a standard second trimester obstetric ultrasound are trained to confirm and measure the size of the tibia and fibula bilaterally (Monteagudo 533); however, this job can be extremely challenging depending on the presentation of

the fetus at the time of the exam and the experience of the technician. There are detailed charts by which technicians are able to identify abnormal results and which aid them in flagging a potential problem. Ultrasound technicians are advised that "Long bone length may be used for monitoring fetal growth and for diagnosing bone dysplasia" (Exacoustos, 330). It can be theorized that due to the extremely low occurrence of Fibular Hemimelia within the population this birth defect has not been widely documented, and as a result, ultrasound technicians and radiologists are not able to identify it easily. To address this issue, Dr. John Herzenberg of The Rubin Institute for Advanced Orthopedics in Baltimore, Maryland, and a world renowned specialist in Fibular Hemimelia, believes that "they (ultrasound technicians) can easily diagnose it if they look for it. The reason why most (diagnoses of) clubfoot and FH and Congenital Femoral Deficiency and Proximal Femoral Focal Deficiency get overlooked is because, in my opinion, they don't make the time to look for it" (Herzenberg). Often the findings are labeled under a differential diagnosis related to a limb defect. Dr. Herzenberg goes on to explain the process by which an ultrasound technician is able to check for limb defects,

> Most cases of FH (Fibular Hemimelia) are unilateral, so the big thing is to look for LLD (limb length discrepancy), and to measure carefully the lengths of the tibias and femurs. FH is often associated with PFFD (Proximal femoral focal deficiency), so (they) need to look at both. FH often has less than five toes, so if it is possible to count toes that helps. Three dimensional ultrasounds are useful for foot issues, and FH often has

> an equinovalgus and occasionally equinovarus foot deformity. FH is rare, and is sometimes overlooked or misdiagnosed. (Herzenberg)

In the original research performed for this paper 67 percent of the subjects had an accurate primary prenatal diagnosis of Fibular Hemimelia, which was established through a routine ultrasound examination. The other differential diagnoses that were initially identified were club foot and a 3 ray foot (absence of 2 toes on affected foot). In light of the initial diagnosis, all of the mothers underwent additional high level ultrasounds while pregnant. These follow up studies of the anatomy of the fetus served to image the limbs more accurately and track the progress or any changes to the apparent defect over time. Christof Radler and his team at the Rubin Insititute for Advanced Orthopedics conducted a study on prenatal ultrasound diagnosis of congenital femoral deficiency and fibular hemimelia in an attempt to discover whether mothers preferred prenatal diagnosis of the defect or not. Surveys were sent to mothers who stated that they received a prenatal diagnosis of their child's limb defect. Results found that the mothers "appreciated the time to prepare, to research the condition and different treatment options and to become emotionally accustomed" (Radler 34). Herzenberg adds that in his opinion, "prenatal diagnosis allows parents to educate themselves," however, the caveat is that he is "fearful that prenatal diagnosis will also lead to elective termination of the pregnancy, which I personally find to be very sad" (Herzenberg).

In addition to high level ultrasounds to further elucidate the presenting malformation of the fetus' limb, one third of the mothers' pregnancies were further managed by undergoing high risk, invasive fetal tissue sampling as a result of the prenatal diagnosis.

Chorionic Villi Sampling and Amniocentesis were used to determine if there were any genetic abnormalities in the fetus that could account for the presence of the limb defect. Both of these procedures are associated with higher miscarriage rates, preterm birth and teratogenic trauma to the fetus (Golden; Tabor; Medda). There have been a number of studies done examining the self-reported levels of stress encountered by mothers undergoing invasive prenatal tests, specifically CVS and Amniocentesis. The findings suggest that the anticipation and performance of these tests on mothers increased their stress levels significantly. In addition, this increased level of subjective stress was not ameliorated by a supportive or involved spouse (Brajenovic-Milic et al.). For the women in this study, the management of a prenatal diagnosis of a rare birth defect during the pregnancy included the potential labelling of the pregnancy as high risk, possible transfer of care to a high risk obstetric management unit and in all cases additional high level prenatal testing, some of which included invasive Chorionic Villi Sampling and/or Amniocentesis. This creates in the woman an overwhelming sense of fear and anxiety surrounding her unborn child and her pregnancy. It is natural to assume her stress levels would increase under these circumstances and correlate with a possible deterioration in coping skills. As an aside, the results of these higher risk, invasive prenatal testing for each of the subjects in the study came back negative for any chromosomal or genetic abnormalities, giving greater validity to the evidence which suggests that unilateral Fibular Hemimelia, as found on a routine second trimester ultrasound is not associated with any genetic abnormalities.

Despite the increased stress levels induced by the prenatal practical management of FH, it is imperative that the research examines the role that communication between the medical care providers and the mother plays.

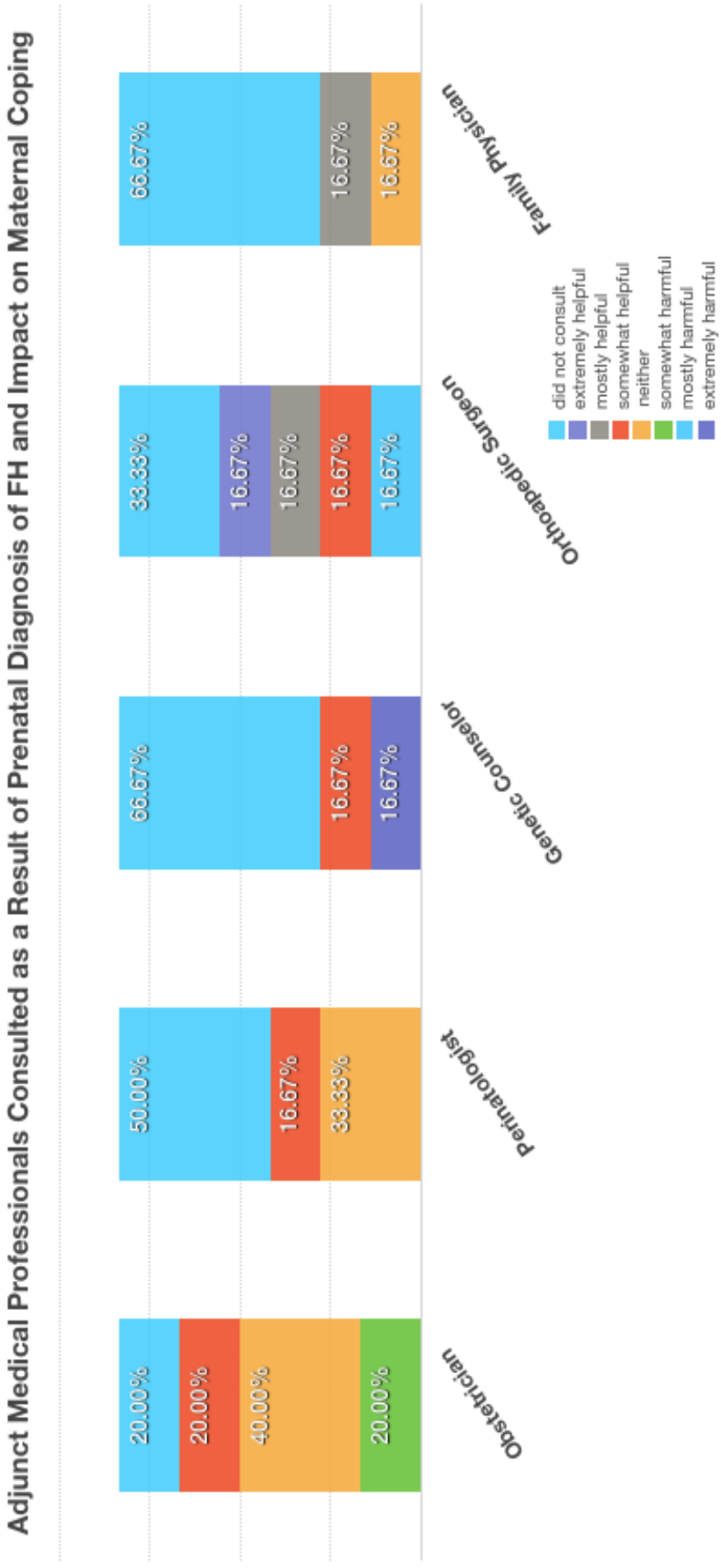
Communication and Resources

Communication between the mother and the various medical personnel she encountered during her prenatal period was a significant factor to explore. The medical personnel were her primary source of information and counseling and also served as an additional example of how the diagnosis should be received and reacted to. Often the reactions and attitudes of the “first responders" greatly impacted the mother’s stress levels and coping techniques. All of the subjects were first informed of the prenatal diagnosis of a birth defect by the attending radiologist and accompanying ultrasound technician who was involved in the testing and analysis. Generally, patients do not interact with radiologists at all during ultrasounds. Technicians perform the test and then the results are given to the attending radiologist who interprets the findings. Traditionally, radiologists then confer with the primary care physician who then relates the information or diagnosis and the implications to the patient. It is significant that for each of these women surveyed, a radiologist was the primary medical personnel who advised the patient. This would be an individual that the mothers would have had absolutely no prior relationship with, and a physician whose focus is not dealing one on one with patients. The occasion of a routine ultrasound during a pregnancy is often associated with excited, anticipatory feelings in a mother of seeing her fetus. Under these circumstances, the ultrasound would have taken extra time as the technician would be re-

checking measurements and attempting to get better visualization of the area in question and may involve calling in the radiologist to confer at the point of the exam. The subjects all reported increased levels of stress, anxiety and panic during this revelatory exam; however, it should be noted that all the women reported at their initial disclosure of the birth defect and their consult they felt that the radiologist was compassionate and patient answering their questions despite not being well informed about the condition. Respondents to the survey indicated that "*The radiologist was very compassionate, was very thorough.*" This experience of the initial findings of a birth defect, although difficult, was managed very well through the communication with the attending radiologist. It is worth noting that there appears to be a movement within the American College of Radiology (ACR) to increase direct consultations between radiologists and patients. The findings from the ACR's pilot projects have been very positive with patients reporting a 4.8 out of a possible 5 for usefulness of consult with a radiologist and 100% stating that would prefer to meet with a radiologist again in discussing their findings. The most significant and important benefit of the ACR pilot project clinic seems to be the impact it has on helping patients better understand their conditions. The ACR goes on to state that, "Many patients are visual learners, so if we show them their images … we can make these fairly abstract conditions more tangible. It empowers patients and improves patient engagement, which we anticipate will translate into improved patient outcomes" (ACR).

The FH survey respondents reactions to the initial diagnosing radiologists and the results of the ACR radiologist - patient consult pilot project raise a provocative point.

Table 3


Adjunct Medical Professionals Consulted as a Result of Prenatal Diagnosis of FH and Impact on Maternal Coping
Obstetrician
20.00%
20.00%
40.00%
20.00%
Perinatologist
50.00%
16.67%
33.33%
Genetic Counselor
66.67%
16.67%
16.67%
Orthopedic Surgeon
33.33%
16.67%
16.67%
16.67%
16.67%
Family Physician
66.67%
16.67%
16.67%
did not consult
extremely helpful
mostly helpful
somewhat helpful
neither
somewhat harmful
mostly harmful
extremely harmful

They suggest that the anecdotal response of the women in this study to the consulting radiologist may indicate a untapped medical resource which could prove invaluable to a mother's ability to cope with the initial diagnosis of a rare birth defect. It may suggest that if the initial diagnosis and communication was handled competently and thoroughly in direct consult with the radiologist performing the ultrasound, the first medical professional to identify it, that this may increase a mother's understanding and thus her trust and comfort levels in the management of the diagnosis, especially in the immediate period after diagnosis.

Along with their primary care provider, mothers were referred to multiple sources for clarification on the diagnosis, implications, outcomes, and management. The subjects consulted with supplemental Obstetricians, Perinatologists, Genetic Counselors, Orthopedic Surgeons and Family Physicians. There was general consensus among the subjects that the Orthopedic Surgeons provided the most valuable information about the significance and outcomes of the diagnosis. These medical professionals were instrumental in helping the mother relieve some of her stress and concerns about the quality of life for her child. The Orthopedic Surgeons were not able to give a comprehensive analysis of the severity of the defect or recommend a management technique (surgical reconstruction or amputation) would be ideal until after the baby was born when they could conduct a full radiological and physical exam. This however, did not hinder the positive impact that their consultation had on the mother's coping abilities. "*Meeting with the surgeon and understanding that the baby could and would still lead a normal life,*" was a comment left by a subject who listed this as the incident that was

most helpful in allowing her to cope with the diagnosis of the birth defect. In fact, the reassurance that "*this was something that could be taken care of managed after he was born*" seemed extremely helpful to another mother under these circumstances.

Other medical personnel who were consulted with by the mothers were evaluated as having none to somewhat helpful impact on the mother's coping abilities with the diagnosis. "*Obstetrician was sympathetic, however, he had never delivered a baby with FH.*" Of the mothers who did confer with a genetic counselor, their experiences did not fare so well. "*The genetic counselor could have a little more tact. She seemed excited that this could be a new syndrome."* This comment was left by a subject who listed it as the one thing she wished the medical care professional had done to help her cope. Another mother stated that she wished they (Genetic Counselor) had offered "*more emotional support vs. just talking to us in medical terms. At times they seemed frustrated with my emotion/reaction to seeing it for the first time.*" She went on to state that, "*There was also too much of '*I think*' or '*it could be*' which meant that I lost trust in them."* Although potentially well meaning, the other medical professionals consulted by the respondents seemed to lack both the knowledge and expertise to address the diagnosis as well as deficient in emotional consideration for the mothers.

In general, the response to additional resources and communication styles of the various specialists was not positive, except for the consult with the orthopedic surgeon who was able to more fully answer the mother's questions and hard pressed concerns: that of whether the quality of their child's life would be severely impacted (see Table 3).

Impact on Termination of Pregnancy (ToP) and Maternal Decision to Sustain

The subjects all chose to sustain their pregnancies despite the challenge of receiving a prenatal diagnosis of a rare birth defect and access to abortion. Half of all the mothers reported that the medical personnel they dealt with did discuss the option of terminating the pregnancy or adoption as a way to manage the diagnosis. One respondent expressed a very specific attitude toward abortion:

> *I wish they had not asked so many times about abortion. It came up over and over and I knew I wanted my baby and it almost seemed pushy. I wish they had asked once and then just let it go when I said I was definitely keeping the baby.*

The other half of mothers did not receive any counseling at all about these options. Despite these different experiences though all the women reported little to no influence from the medical community on their decision to not abort; instead, the subjects resoundingly state that they never considered terminating the pregnancy after they were given the diagnosis of a birth defect. No preference was noted between geographical regions, health insurance status, marital status or education and the discussed option of termination of the pregnancy. Further comment will be made in the following section looking at the community's influence on ToP rates.

Although we have subjects who participated worldwide, their interactions, experiences and opinions of the medical community in relation to the prenatal diagnosis of a birth defect were fairly consistent. All first received the diagnosis from the radiologist and felt that they were generally compassionate yet uninformed about the

condition. They did not find the additional consultations with adjunct health care professionals or testing particularly helpful, and under certain circumstances, found it to be harmful to their ability to cope. The only medical professional they found to be generally helpful was consulting with the Orthopedic Surgeon. In general, they felt the lack of knowledge from the other professionals was a hindrance to them, reducing their levels of trust and amplifying their sense of concern. As one mother stated, there was

> *Not much they could do, as we did not know what we would be dealing with. I feel they did all they could, though nothing really made me feel better about the situation.*

Chapter 3

Community Resources

The second area that was examined with mothers who received a prenatal diagnosis of a rare fetal birth defect was Community. The concept of community encompasses quite a wide range of sources; however, the research focused on four main areas: The environment of the actual Physical Community, Role of Friends and Family, Role of Partner/Spouse, and the Virtual Community hosted by the internet.

Country of Origin

The subjects who participated in the survey came from countries that contribute to the annual report in the International Clearinghouse for Birth Defects Surveillance and Research (ICBDSR), which was introduced earlier in this paper. Australia, Canada, Norway, and the United States of America all have well established monitoring systems in place for tracking the incidence of birth defects. These countries have advanced medical, specifically obstetric and orthopedic, services available to patients and have sanctioned medical abortions available to women whose fetuses test positive for birth defects. The data on this condition is shockingly limited though due to its low incidence rate, but when looking at each country's incidence trend and reported termination rate for this birth defect, a conclusion can be drawn that not many women are opting to terminate a fetus that is diagnosed prenatally with Fibular Hemimelia. It should be noted that the Canadian and the Australian National birth defect registry, did not report on the incidence of the specific subcategories for Limb Reduction Defects. Nor did they report the termination of pregnancy rates for each of these subcategories.

In the year 2009 (Norway data from 2008), there were a total of ten cases of live births of children diagnosed with Transaxial Limb Reduction Defects reported between the five surveillance organizations that represent the countries of Australia and Norway and the three that are in included from the United States, MADCP (Metropolitan Atlanta), BDES (Texas) and UBDN (Utah). Within these five organizations there was only one termination of pregnancy (ToP) reported in Norway over this time period, 2008-2009, for this subcategory of birth defect, representing a ten percent abortion rate. See Table 4. The low incidence of ToP for Transaxial Limb Reduction Defects may be indicative of a number of factors unique to these countries and in turn the women who live there.

Table 4

Postaxial Limb Reduction Defects Incidence Rates in Survey Respondent's Country of Origin (2009)

Birth Defect Program	Country of Origin	Live Births	Still Births	Termination of Pregnancy (ToP)	Incidence Rate (per 10,000)
CCASS	Canada	nr	nr	nr	nr
MBRN (2008) *	Norway	1	0	1	0.32
WARDA	Australia	0	0	0	0
MADCP	USA (Atlanta)	0	0	0	0
BDES	USA (Texas)	7	0	0	0.17
UBDN	USA (Utah)	2	0	0	0.37
Average for Reporting Countries		10	0	1	0.17
Rate of ToP in Respondent's Country of Origin for Postaxial Limb Reduction Defects				10%	

' no data was available for 2009
nr - not reported

(Courtesy of ICBDSR report 2011)

Access to advanced medical resources, a well-established social and legal system to protect the rights and provide opportunities for those with disabilities, cultural norms that embrace those with special needs, a higher than average Gross Domestic Product, which would suggest that these affected families may have financial resources that better allow

them to manage the condition are all factors that could potentially support a mother's choice to sustain the pregnancy and raise the child.

Immediate Community

A sense of connectedness to a person's community, neighbors, businesses, services, resources, religious organizations and schools often provide a feeling of membership and stability for an individual going through crisis. This stability could provide support for and be of benefit to an individual who is faced with a significant life stressors, such as receiving a diagnosis of a rare birth defect in their as yet unborn child (Ozbay). It was theorized early in this study that having a community where special needs individuals are included through independent residential homes, employment in local businesses and school integration policies would positively impact the coping abilities of the mothers. This exposure was thought to possibly provide, for the mothers, a higher comfort level with the presence of differently abled people and the community adaptations and de-mystify the stigma and discomfort which often accompanies interaction with a person with special needs. To that end the study sought to determine if a high self-reported sense of community connection and visibly integrated and non-discriminated residents within the community with special needs would foster a greater sense of comfort within the subjects surveyed, reduce their stress levels and aid in their coping of their fetal FH diagnosis.

Eighty three percent of the subjects in the study reported they had lived in their community a long time and had liked it. This implies a sense of connection to where they live and stability indicated by their residence there for "a long time." These respondents

however, did not report being highly involved or active within their community. Only one mother indicated that they were involved in a community aspect such as school parent teacher association, political action committees, recreational activities, or fundraising/volunteer efforts. The remaining respondents were either completely uninvolved by choice or had no particular investment in that aspect of their community living. These mothers did not know of any residents within their community that had special needs. If the community did have these individuals, they were not easily visible or the mothers were not aware or sensitive to their presence. When asked whether they knew of any resources within their school district that addressed and assisted children with special needs they were not able to identify any. This is an interesting finding because 67% of the mothers surveyed actually had older, school-aged children who would already be attending the local schools. It would seem to follow that the respondent would be exposed or have knowledge of a policy of integration of children with special needs or accommodations for differently abled students; however, they were unaware of them. It is possible that the mothers did not recognize or understand that their child may require special physical or curriculum accommodations at school, such as home-schooling and lesson flexibility during potentially extended periods of surgery or recovery, handicap accessibility, and physical education/sports participation accommodations. There is also the possibility that these mothers would not have qualified their child's physical birth defect as requiring special needs or place them in the same category as other individuals with associated mental or cognitive impairment. This finding will be explored further in the Maternal Characteristic section.

In general, the subjects exhibit a seeming paradox in the study, between liking their community, yet, not establishing a sense of community through interaction and knowledge of resources, residents or school district climate. The exception to this finding is for the two women who identified as having very strong connections to their churches. It is also possible that a lot of their free time and socialization would take place within their church to the extent where their wider community that they lived in was not a significant factor in their lives. With the exception of these two respondents who were actively involved in their church, the community, fellow residents and neighbors, schools, recreational or volunteer organizations within was never listed as a significant source of support or resource for helping the mother cope with the prenatal diagnosis of her fetus. The role of the church therefore was examined a little more closely to help identify its role in community resources for these women.

As stated, only two of the mothers surveyed reported a very high involvement with their church or religious affiliation. Often the sense of community within a church can confer an excellent source of support and be very empowering for an individual who is facing a life stressor such as the welfare of their unborn child. This phenomenon was examined by Donia Baldacchino in her research of an individual's spiritual coping strategies in relation to illness. Baldacchino found that "increased frequency of church attendance appears to be used as a means of socialization and support, which may decrease feelings of withdrawal and isolation" (836). The respondents in this survey identified their religious practices and church as one of the most helpful aspects in their community when coping with the prenatal diagnosis of FH. One mother responded that

one of the factors most important to her coping was: "*My church family as well as my family. Lots of prayer and support." "We found that our family and church community were very supportive and helpful."* Possibly the act of prayer and the sense of community experienced while engaging in religious practices at church allowed them to feel supported during this difficult time in their lives. Specifically, they were seemingly able to release their worries and lack of control or sense of helplessness to a higher power through prayer freed them from the inertia commonly created by fear of the diagnosis.

Family/Friends/Partner

As expected, there was a high correlation between time spent with positive, pleasant family and friends and on the subjects ability to cope with the prenatal diagnosis. One respondent indicated that she felt that the lack of a "*good local friend network*" was an element that she felt was missing from her community experience, and she believed that it could have helped her cope better with the diagnosis at that time in her life. Another mother felt that one of the most important things that helped her cope was her "*best friend who also has a child with special needs.*" Although friends were listed as very important, it was found that family was a more significant source of support for the mothers. Every single respondent in the survey reported that they mostly or completely agreed that they relied on their family for support and assistance coping during the prenatal period. "*Family was my main support."*

Eighty three percent of the respondents were married or in a domestic partnership at the time of the prenatal diagnosis. Of those, each one stated that their partner was instrumental in helping them cope with the prenatal diagnosis of a birth defect. They

reported that their partner actively participated in the decision making process with them regarding managing the diagnosis of their child. This strong partnership and sharing of responsibility between the mother and her partner can serve to lessen the emotional burden of a life stressor. Studies have proven that "marriage … can buffer against stress and result in lower physiological activation in response to challenges" (Maestripieri, 422). "*My partner has been my greatest strength,*" said one mother. Another stated that "**WE** *found that our family and church community were very supportive and helpful,*" suggesting that the journey and challenges she faced during this period in her life were shared by her partner.

Virtual Community

As these subjects were recruited from three online patient/parent run support groups for Fibular Hemimelia hosted by the social media site, Facebook, it was felt that the concept of a virtual community could be a significant factor in exploring how these mothers coped with the prenatal diagnosis of a rare birth defect. Fibular Hemimelia is as previously stated a rare birth defect, occurring in approximately 1 out of every 50,000 births. It is not associated with any particular geographical area, ethnic group or socioeconomic status, so it affects the entire world population equally. The relative low incidence rate makes it very challenging for people within this community to find others like themselves, let alone meet and share their stories. One way that this has been possible is through the internet where geography is not an issue.

It requires notice though that the internet is not an equal access resource. There has been (and continues to be) extensive debate concerning the digital divide. Open access to

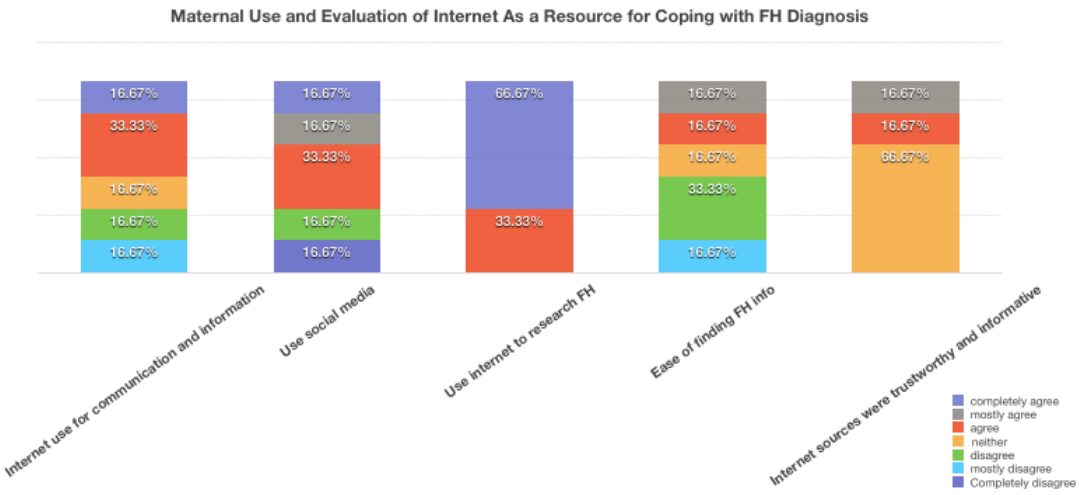
the world wide web and comfort with internet searches and social media is definitely limited by the economic, language, education and privilege status of a given user (Katz). This means that only those with the means, understanding, and time to use internet access devices would be the ones able to benefit from this technology. This is reflective of the survey respondents who all identified themselves ethnically as white. Of these respondents 100% of them indicated that they had at a minimum a post secondary undergraduate education, with at least half holding Master's, PhD or professional degrees. They all also all live in a politically and economically stable, democratically developed nation: United States, Canada, England, Australia, and Norway. All of these demographics indicate that the subjects were in a position to utilize the internet as a potential source of information and support for their FH diagnosis. With Fibular Hemimelia diagnoses throughout the entire world, this resource is truly for those most privileged. Nevertheless, the virtual community of the internet and social media bears exploration as a factor influencing the coping ability of these women whose fetuses have been identified prenatally with Fibular Hemimelia.

Three online support groups for Fibular Hemimelia, hosted by Facebook, served as the recruitment source for this study: *Fibular Hemimelia Support*, *Fibular Hemimelia and Limb Lengthening Awareness*, and *Fibular Hemimelia and Amputation Awareness*. In total, there are over 2, 200 members in these groups from around the world. These sites host an open forum for discussion between members and facilitates web based seminars with FH world wide experts. Groups also provide resources for members on surgeons, surgical facilities, practical coping measures, mentorship. Emotionally, these pages are

spaces for expressing sorrows and concerns along with the victories and milestones in the lives of an individual with FH.

According to the the study's findings, all of the respondents utilized the internet at the time of prenatal diagnosis in an attempt to find information about Fibular Hemimelia, to understand it more completely with the purpose of coping with the condition and improving their overall experience and outcome. Unfortunately for most of them, they were not able to find the kind of information they were looking for easily. Only 33% met with relative success in their initial internet search on FH. The majority expressed that they found it challenging to get to the type of medically credible sources they desired. When they did manage to successfully find sources on FH, they had mixed opinions on whether the sources were truly information and trustworthy. Not one of the respondents stated that they were completely confident in their internet sources for FH. Although women in the study were able to get some information and resources from the internet on this rare condition, they did not feel that it was an adequate source of support or that it contributed significantly to their ability to cope during their prenatal period. See Table 5.

Table 5



Physical FH Community

Despite the incredible resource and support that these virtual FH communities provide, there was a resounding desire for these mothers to physically meet other parents and children with FH. When the subjects were asked what they felt was missing during their diagnosis period and that would have helped them cope better, they predominantly wished for support groups.

> *Support groups and/or more visibility, the surgeon said there were a lot of kids with FH locally, but there doesn't appear to be any support networks for it for parents and families.*
>
> *Support group would have been helpful and probably informative.*
>
> *Would have loved to meet other people with the condition, who had not received surgery. (Would still love that).*
>
> *We couldn't find a community group for this type of condition. Also, we didn't know it was specifically FH until 2 weeks after birth, so we didn't know what exactly to search.*

For these mothers, being able to physically comprehend the nature of the diagnosis and finding someone who could understand their concerns and answer their practical questions was a crucial element that seemed to be elusive. Experiencing the prenatal diagnosis of Fibular Hemimelia places a mother in a position where she is dealing with the issue on a conceptual basis. Although ultrasound images and medical personnel can vouch for the birth defect, it is impossible to comprehend the full impact of the condition both from a physical stand point and an emotional one. Fibular Hemimelia orthopedic

surgeons are unable to make a full prognosis of the degree of defect from in utero exams and must wait until the birth of the child when they can do a hands on full exam (Herzenberg). This leaves the mother in a state of ignorance of what to expect after the birth of her child. By itself, this feeling, is a source of great stress for the mother. Amber Mathieson, at the University of Utah, found in a study evaluating parent to parent support networks that "Compassion, caring and connecting with others who have traveled a similar path may also help parents immensely during this time" (Mathieson, 3466). By physically connecting with a peer, an experienced caregiver of an FH child, or meeting children with FH, the mystery and fear of the unknown outcome of a prenatal diagnosis can be ameliorated. The social support that only a cohort can confer may have a positive impact on the coping skills of the subject to an acute stressor (Lepore). Having that sense of community is an invaluable tool for mothers; however, the nature of the condition makes it extremely difficult to connect these people who potentially live great distances from one another. As evidenced by the research and personal commentary, a virtual community is not able to fill the need for women. The act of physical connectedness was deeply desired and suggests that there is an essential emotional component that cannot be transferred or communicated through the internet or social media sites.

This research found that a lack of connectedness and support within the mother's community can often result in a sense of isolation, increasing her anxiety and stress levels and affecting her ability to cope. Organized religion, however, as an act of community, does seem to serve as an excellent source of support and strength for an individual going through this crisis. Engaging in ritualistic prayer and meditation common in religious

practices allowed these mothers a way to release their fears and deal with acceptance and moving past the shock of the diagnosis.

Interpersonal relationships with friends and family was extremely important for coping, but a strong relationship of the mother with her spouse or partner was found to be the most influential in helping her cope. It is perhaps because the partner is also heavily invested in the health and well-being of the developing fetus that the mother can feel a sense of camaraderie with her partner. The mother may feel that the father is perhaps the only one experiencing a similar sense of grief over the prenatal diagnosis of their child and this reduces her feelings of isolation, thus sharing the burden.

The internet proved a vital tool for seeking out information about FH as well as attempting to connect with others who have personal experience with the defect. Despite all of the respondents using this tool however, only a small percentage of them actually found it useful, trustworthy and reliable. Social media support groups for FH hosted by Facebook were instrumental in many of these women getting support through mentors and reducing feelings of stress and isolation. The gold standard for community support, for all of the respondents, was clearly identified as being able to physically meet other parents or individuals affected by Fibular Hemimelia. These findings suggest that the ability to physical connect and witness other FH individuals or families would prove to be the greatest sense of support and strength to these women struggling with the prenatal diagnosis. Fibular Hemimelia is a rare occurrence in the population and not easily visible necessarily, so for those women who are pregnant and struggling with the diagnosis of their fetus, the sense of isolation and the fear of the unknown is extremely poignant.

Having access to a FH "mentor" and being able to physically see the effects of FH on an individual can demystify the condition and in turn bring great relief to their highly personal struggle.

Chapter 4

Maternal Characteristics

A final factor that was investigated in the study was the mother's personal characteristics, beliefs, and personality type. In particular, the research sought to determine if specific elements of the mother's personality informed her ability to cope with the prenatal FH diagnosis and the ultimate decision to sustain the pregnancy and not abort. Subjects were asked to rank statements on a seven point scale in order of Completely Disagree to Completely Agree. The questions were formulated to assess four main components of their characteristics and personal belief system. The components were: Role/Concept of Motherhood, Concept of Their Child,Woman in Relation to Others, and the Woman as an Individual.

Concept of Motherhood

The concept of motherhood is a hugely political issue with many factors and philosophies that have contributed to the ideology. It is not the purpose of this research to investigate the origins and pitfalls of this ideology, but merely to acknowledge that it does exist. There are, of course, variations of this ideal that exist between cultures and communities, but by far the idea of a good mother is entrenched largely, if not wholly defined, by the act of dedication, self-sacrifice and focus on the welfare of their children, sometimes to the exclusion of others in their life and often to the detriment of the woman herself who seeks to achieve this ideal (Vigil, Shanahan). By examining the concept that these women had of motherhood, the survey hoped to provide some insight into how they coped and to see their rationale for the decisions they made in light of their prenatal

diagnosis. The first step that needed to be established was the importance of motherhood to these women. If the women felt very strongly in favor of being a mother and had a defined identity of themselves as such, then it would follow that the decision to terminate the pregnancy would not be an option highly regarded or easily made. If the mother was ambivalent about being a mother, or did not feel it was that important, then abortion may have been a more considered option. For the respondents, 83% completely agreed with the statement that it was important to them to be a mother and have children of their own. Seventeen percent were ambivalent about the importance of being a mother and neither agreed nor disagreed. This strong identity of being a mother can definitely be seen as a significant contributing factor to sustaining a pregnancy despite a known birth defect and not opting for a termination.

Widely held concepts or archetypes or ideologies of motherhood often include the belief in a "mother instinct." Mothering instinct can be defined as an innate knowledge or guidance within the woman to respond to and care for her child. It is often compared to a biological imperium where it resides naturally within the woman and is not the result of learning or training. Madoka Noriuchi, of the University of Metropolitan Tokyo, conducted some interesting research in 2008, utilizing functional magnetic resonance imaging on mothers who were presented with both their own infant and other infants who demonstrated two different attachment behaviors: smiling and crying. The purpose of this study was to test the validity of mother instinct and a woman's innate biological response to their own child's needs. Results were definitive and pointed to evidence that there is in fact a neural mechanism within a woman that "mediates maternal love and

diverse and complex maternal behaviors for vigilant protectiveness" (Noriuchi 415). Noriuchi comments on his finding and states that "maternal love is essential for maternal behavior in which the mother makes sacrifices that are necessary to care for her infant day and night. Loving the infant is the dynamo that empowers her to maintain never-ending vigilance and sustain exhausting toil for the protection and nurture of her infant " (Noriuchi 420). The women in this study seem to hold strong views on the role of the mother. When asked whether they relied on their mothering instinct to do the right thing by their child, 83% of them agreed with the statement. This suggests that their concept of motherhood and their ability to innately make the correct decisions for the child was believed to be a hard-wired natural response, similar to that found in Noriuchi's work. When the subjects were asked whether they believed that a mother should make "whatever sacrifices necessary for the wellbeing of her children," a full 100% of them agreed completely with that statement. This was the highest ranking they could possibly assign. These strongly held beliefs by the mothers would indicate that any sacrifice necessary should be made for their fetus. It follows then that the option of terminating the fetus on the basis of the prenatal diagnosis of Fibular Hemimelia would be anathema to these women, unless of course they believed that non-existence would be a better option for the child than a life with a FH diagnosis. These findings suggest though that perhaps abortion would never have been an option despite the diagnosis and that perhaps their journey through this prenatal diagnosis was more one of coping and deciding how to adjust to a new reality for their child and not about whether to sustain the pregnancy at

all. This is further confirmed in additional questions that were posed to the women in the survey.

When asked whether at the time of their pregnancy and the fetus' diagnosis they ever considered the option of termination, an emphatic 83% said no they did not consider it. Only one respondent revealed that she did consider it, but it was the lowest agree ranking that they could assign to a statement. The study went on to find the mother's response to the idea of giving their child up for adoption due to the prenatal diagnosis. Again, the response was clear. One hundred percent of the women completely disagreed that they considered the option of adopting out their child. It would only be reasonable to remind the reader that this was a retrospective study of mothers who already had given birth to their children with FH. Their relationship and bond with their children was already well established by the time the survey was taken and therefore this more than likely could have created a bias towards their reported responses to the idea of terminating their pregnancy or placing their baby up for adoption. It should also be noted that a decision to abort or adopt their baby could have also been influenced by their partner who would have an emotional and legal investment in the child. The majority of respondents reported a close relationship with their partner and listed them as integral source of support in the ability to cope with the situation, as well as their community that could have possibly exerted extreme influence on the acceptability of the options of abortion or adoption.

Concept Child

Following up from the concept of motherhood, maternal instinct and unlimited sacrifice for the good of the child, the study sought to examine a mother's projected

image of her fetus. It has been argued by scholars that for most women the concept of pregnancy, having the child and the construct of said child is well-established in many cases prior to becoming pregnant. Janet DiPietro of Johns Hopkins University, in her work on maternal-fetal relationships found that,"The earliest relationship does not begin with birth. Pregnant women construct mental representations of the fetus, and the feelings of affiliation or 'maternal-fetal attachment' generally increase over the course of gestation" (DiPietro 27). Polite society encourages mothers to not be concerned about details such as the physical attributes of eye and hair color, or who the baby looks like, but simply to be happy that your child has "ten fingers and toes" which is a reference to a healthy and perfectly formed baby. In addition to this, many women visualize and imagine what it will be like to be pregnant with their child, what the child will look like, who they will be as adults, and the goals and accomplishments they will achieve as they mature. This desire for a healthy, perfect child is ingrained in a mother's psyche. A women's emotional investment in her concept child is heightened by the principle of the Endowment Effect, primarily an economic term, introduced by Daniel Kahneman, Jack Knetsch and Richard Thaler. The mother has an extreme psychological attachment to her concept child and the endowment effect states that "people often demand much more to give up an object than they would be willing to pay to acquire it" (Thaler 52). For the mother, this means her concept child holds incredible value for her and letting go of this dream and facing a new reality will actually be is an incredibly daunting hurdle to overcome. The issue is, how does a mother psychological adjust to or accommodate a variation in her expected perfect baby. With a prenatal diagnosis of Fibular Hemimelia,

the mother's concept of her child has been violated and contravened. This single event, the unexpected diagnosis, can be interpreted as a catastrophic negative event affecting the mother on a very intimate and intense level. She is now forced to accept that the conceptual child that she has nurtured and invested emotionally, potentially over years, in fact, does not exist.

People by nature exhibit *Loss Aversion*, another concept and term coined by Kahneman and Tversky. Studies have suggested that losses experienced by people have twice the psychological impact as a gain; therefore, people will work significantly harder to avoid or avert these losses than to adopt a gain (Kahneman). This implicates again, a deep, emotional, and psychological struggle for the mother, on a theoretical level, to give up her ideal and accept the new child that has a birth defect. It requires much more effort for the woman to give up her concept child than it does for her to embrace her actual fetus/child. This is not a comment that these women are rejecting the fetus and their actual child, rather that the conflict is held on a more subconscious level of awareness. In fact, it is not unreasonable to argue that it is possible for the mother to embrace the new child on one level while still struggling with—and even keening—the loss of what the child will never be.

The research suggests that many of these women experience this loss as something akin to a death and exhibit a reaction similar to that postulated in the Kubler-Ross model of the stages of grieving. Denial, Anger, Bargaining, Depression and Acceptance are emotions that all of the mothers alluded to in their survey responses. They expressed surprise at the prenatal diagnosis of their fetus and experienced varying degrees of guilt,

shame and concern about the impact of the diagnosis on the quality of their lives and pre-existing relationships. None of the traditional sources of information or support in the form of medical consults, family, friends, spouse/partner relationships, internet research, community resources and social media support groups were able to fully reconcile their loss. One mother summed this up best by revealing that,

> *I don't think anything could have helped. The unknown is so scary and there's really no way to make that better. Once she was born, I felt better able to cope as I saw what her little food looked like and was able to start taking action to help her.*

This research participant expresses very clearly her struggle dealing with the diagnosis during her pregnancy. Her final act of acceptance came when her daughter was born, and she was able to have physical evidence of the defect, "*her little foot*," in front of her testifying to the new reality. Yet for many mothers the evidence and acceptance of a physical birth defect and the unconditional love and protectiveness that they have for their child does not necessarily translate into a full acceptance of their child as being disabled or having special needs like other disabled individuals in their community.

Special Needs/Disabled Value

Gail Landsman has conducted a number of expository studies on the concept of motherhood and the personal psychological challenges of mothers with a disabled child. For the purpose of this research and in response to a never ending social etiquette dialogue regarding the appropriate nomenclature used for individuals who have defects and require accommodations, "*Special Needs"* is defined by a person requiring corrective

measures or accommodations in response to a "*Disability"* where an individual is unable or disabled from participating in "normal" life activities. Often the two terms are used interchangeably and both are vulnerable to prejudice, bias and value judgement for the individual to which they are applied. Landsman discovered in her research that "there is often a distancing of the child from the label of disability by mothers whose infants have just been diagnosed as at risk for disability" (87).

In fact, she found that mothers were able to "simultaneously hold negative attitudes about people with disabilities and to attribute personhood passionately to their own children (who were classified as disabled)" (78). This can be interpreted as a manifestation of the Denial stage in the Kubler-Ross grieving process, but Landsman goes on to explain that "such women [are not] stuck in a stage of denial or rationalization in which they do not allow themselves to see the extent of their child's disabilities in order to cope better…rather, they have normalized both their children and their own experiences" (93) and do not consider the labels of special needs or disabled as appropriate to their case. The findings seems to suggest an interesting and complex psychological phenomenon at play here in the question of whether a mother identifies her child as having special needs. The survey probed the mother's reactions to the perceived impact that Fibular Hemimelia would have on their children's lives. They were asked to respond to the statement, "I thought that my child would have special needs." The data reveals absolutely no consensus amongst the survey participants, some agreeing as strongly as some disagreeing. When asked specifically about different challenges they thought their children might face with the diagnosis, they all responded rather uniformly

and attested to their concerns about their child experiencing a lot of pain and being unable to walk. These characteristics, chronic pain and inability to walk, would definitely qualify an individual as having a disability, requiring special needs accommodations. When asked again, further along in the survey whether they "thought that my child would be considered to have special needs," 83% of the mothers now strongly agreed. The latter question about special needs, asks the mother to evaluate her child's status in relation to how society would evaluate or categorize their children. The former question more directly addresses how the mother identifies her own child in relation to special needs. The mother does not necessarily believe that her child has special needs, but she does feel that others will think that her child has special needs. There is an apparent dissonance between the mother's concept of her child with FH and how she feels her child will be perceived or labeled by others in society. This finding, although striking, is not really that unusual.

Landsman found that in developed nations, mothers of infants with birth defects "construct narratives of hope, attributing to their infants culturally valued qualities… [and have] faith that their children will become the children for whom they had hoped and prayer for" (Landsman 78). Although, their children are not physically "perfect" their attributable characteristics of bravery, toughness, perseverance and specialness help the parent recreate their infant in an image that asserts their personhood and value and overlooks the labeling of their child as disabled or special needs. This finding is supported by statements made by the mothers in the survey in response to their reaction to parenting their child with FH.

I never expected my daughter to be as amazing as she is. I have been terrified for her in so many cases only to find that she shines every setting. She never gives up and is an inspiration.

We are very grateful to have our son in our life. he is amazing, strong and helped us become a closer family. We wouldn't change a thing.

Woman In Relation to Others

These women all expressed very close and supportive relationships with family, friends and partners. They all believed that this community of people was instrumental in helping them cope with the prenatal diagnosis of their child. The survey attempted to examine in depth the interpersonal factors in their relationships that may have impacted her management and decisions. All of the mothers responded strongly that they did not feel that the prenatal diagnosis of their fetus would negatively impact their relationships with their friends, family or partner. In a somewhat paradoxical finding, the self-reported level of confidence in knowing where to turn for emotional support was mixed with only 50% agreeing that they did know. That is only 50% of the subjects felt confident in where to turn for emotional support. The other 50% were unsure or were patently not confident in who to turn to. Eighty three percent of the mothers also reported that they did not rely on other people to help them make decisions regarding the welfare of her fetus with FH, despite having reported great confidence in others.

These findings may suggest that despite the fact that these women had very strong interpersonal connections in their lives, they did not feel a sense of confidence in who to turn to for support and therefore did not rely on anyone else. Their experience of the

prenatal diagnosis of a birth defect in their fetus was an intensely personal challenge. A challenge that they subconsciously felt they must manage alone and as was described earlier in this paper, is a response to the loss of their concept child. A woman has an intimate and personal relationship with her fetus that is not at all possible with anyone else. Friends, family even a partner or father cannot experience the connection or oneness that a pregnant mother does. The creation of life and nurturing it within a woman's body is an immense responsibility and an overwhelming concept for many women. An unplanned disruption in this process is nothing short of an assault on the relationship between a mother and child. For all the other people in the women's life this disruption can only be experienced second-hand through the mother. Although these mothers reported their relationships with others as being of great support to them during the prenatal period, it inevitably falls to the mother alone to experience most acutely the pain of a prenatal diagnosis of a birth defect. It is perhaps for this reason that she feels she was not sure where she could find the emotional support she needed and that she did not rely on others to help her make decisions regarding the welfare of her child and the management of her pregnancy.

Woman as an Individual

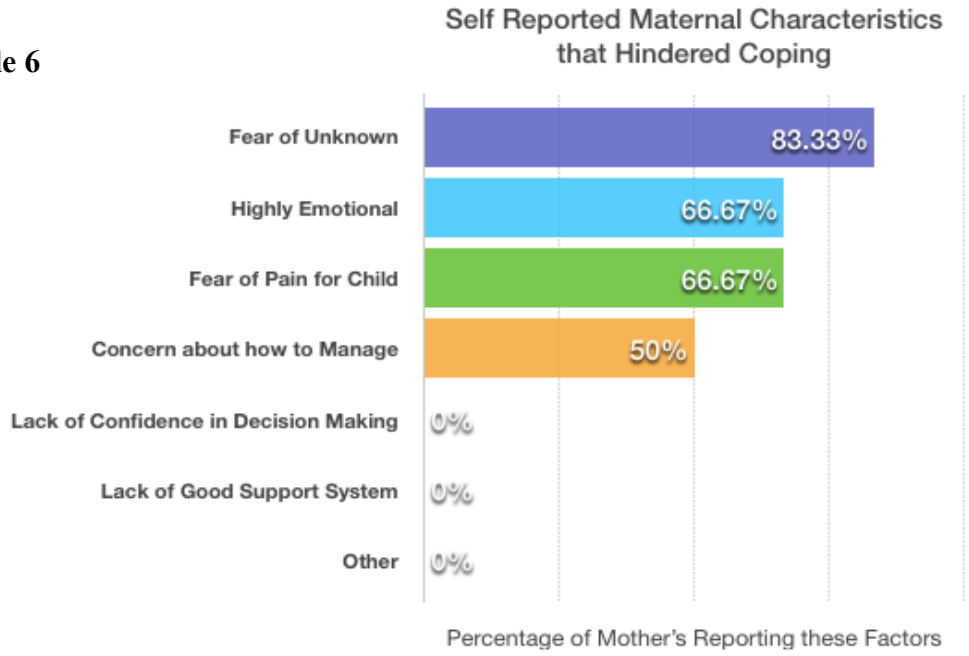
The subjects engaged in this retrospective study attempted to recall their coping experience while they were pregnant; however, their individual personal belief systems and characteristics were already developed, ingrained and evident independent of their relationships with others, with their pregnancy or with their role in society well before

this episode in their lives. The study sought to examine if there were any reported characteristics of these women that influence how they coped with the prenatal diagnosis.

When asked to evaluate how important it was to the respondents to maintain their independence and pursue their own goals after becoming a mother, 33% agreed that is was important. This answer is in direct conflict with a previous query where 100% of the women completely agreed that a mother should make whatever sacrifices necessary for the welfare of her children. Presumably "whatever sacrifices are necessary" would include their independence and own goals. Eighty three percent of the respondents felt overwhelmed by the unexpected diagnosis, with 33% and 67% stating that they felt, at the time, unable to handle respectively the emotional and financial burden of a child with special needs. Only half of the women felt confident in where to go for emotional support and information. At least half of them felt personal guilt over the diagnosis of their fetus and at the time blamed themselves for it. These facts perhaps contributed to 33% of them feeling that they may be judged negatively by others. When subjects were asked to identify their personality factors that they felt negatively affected their ability to cope with the diagnosis while pregnant, the element of lack of control seemed to be a significant theme. Fear of the unknown, highly emotional personal state, fear of pain their child would experience and the concern about how they were going to manage everything all reflects back to control issues. Parents are expected to protect and guide their children. They should be wise and act with deliberation and purpose to achieve a good outcome. These formerly listed factors expressed by the subjects indicates that they believed this parental role was not able to be fulfilled under the circumstances. It is for

this reason, this lack of control or sense of not being able to deliver in their role as parents that these mothers struggled most. See Table 6.

Table 6

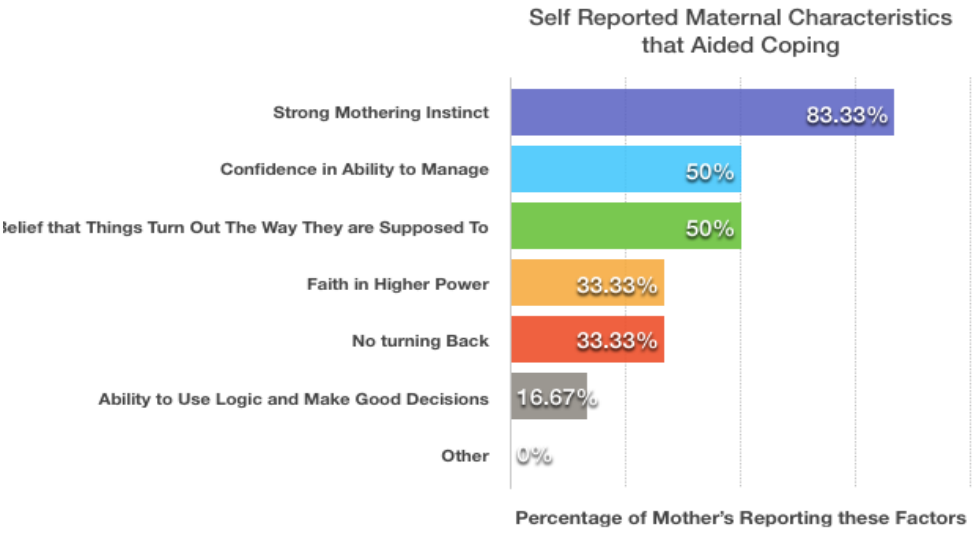


Despite these grave statistics though, 67% of them felt confident with their ability to make the correct decisions about the treatment of their child after birth and not one of the respondents expressed concern that the circumstances would negatively impact the quality of their lives. How did they manage this optimism in the face of these significant personal challenges?

The answer may lie in one resounding finding that was repeated numerous times throughout the study. These mothers had confidence in their ability to manage because of their belief in factors outside of their control. They did not feel that conducting good research and using logic to make high quality practical decisions was the most important

factor that helped them cope. Instead it was the combination of the nebulous and unquantifiable concept of mothering instincts, belief in fate, karma, faith in a higher power and the permanence of the status quo that helped them cope. See Table 7.

Table 7



The challenge comes in reconciling this seeming paradox between similar quality factors hindering as well as helping the mother to cope with the situation. In dealing with an unexpected, stressful situation such as the prenatal diagnosis of FH, a mother is unable to find reason or fault. The uncertainty of the situation is a direct result of the limited medical resources and lack of good information on the condition and the fact that FH is a random birth defect that is not linked to any teratogenic or heritable factor so there is no definable reason for the occurrence other than the fetus is victim to a random chaotic event. Chaos has intruded on their conceptually blissful experience of bringing a child into the world. *Why has this bad thing happened to me and my child?* Instead of struggling to find answers, make sense of the situation, or achieve control, these mothers

have ultimately given up on that quest and used an adulterated concept of the attribution theory proposed by Fritz Heider: to foster acceptance and release their need to control. By relinquishing control over their circumstances, the individual achieves a sense of psychic and personal relief in relation to the situation and thus are able to move forward. Attribution theory suggests that there is a cognitive bias among people who try to make sense of events that impact their lives. Positive events that happen to people are often attributed to something that the individual has done and therefore earned. Negative events, such as the prenatal diagnosis of FH in a fetus, is usually interpreted by an individual as being caused by something outside of their locus of control (Heider). It follows then, that not only is this event (FH diagnosis) perceived as something outside of the control of the mother, but that it in fact has absolutely no basis in reason or logic. The mother cannot control the event, the event is unexplainable and random; therefore, the only option for coping with it would be to embrace the unexplainable as the solution to the problem. They have given up their need for control to a higher power and released their personal responsibility. By allowing a greater force to be present in the situation, one that they have no control over, they have enabled themselves to move forward in the process of acclimating to the situation, not be stuck in a continued search for meaning.

Similar findings are reported by R. Otto in his book, *Idea of the Holy* and Baldacchino's *Spiritual Coping Strategies: A Review of the Nursing Research Literature,* in reference to the utilization of spiritual coping strategies during illness. Donia Baldaccino of the University of Malta, researched this phenomenon as a result of her nursing experiences. She witnessed the impact of spiritual assistance on her critically ill

patients. “Illness may render the individual, being a believer or non-believer, to realize the personal nothingness and lack of control over his/her life…The use of spiritual coping strategies may help the individual in self-empowerment leading to finding meaning and purpose in illness, achieving a sense of personal wholeness by unifying the bio-psycho-social perspectives” (Baldacchino, 838). This concept is mirrored in the findings highlighted earlier in this research paper regarding strong religious affiliation and spiritual practices aiding women in coping with the prenatal diagnosis of a birth defect. These women were able to “give up control” to a higher power, or supreme being, which brought them comfort and allowed them to proceed with managing their circumstances. In light of this, perhaps religious and spiritual practice can be seen as a culturally acceptable form of releasing control of a situation, attribution theory in the face of catastrophic, negative circumstances.

A strong identity of motherhood, including natural instinct, proprietary relationship with their fetus and protective self-sacrifice for their child worked in conjunction with a psychological release from the fear and need to control the unexplained and allowed these women to transcend their experience of a prenatal diagnosis of their fetus and manage their circumstances. These women were not inevitably victims of inertia, stagnated to suffer under the fear of their prenatal experience and the projection of possible outcomes for their child. They accepted and then released their emotions in an act of true self-sacrifice and proceeded to work towards the goal of what they felt would be best for their child, credited to an intensely intimate relationship that was initiated at gestation and continues through their lifespan. In light of this commitment, the option of

terminating the pregnancy would never have been considered despite the external influencers and challenges that they faced with the medical arena, community and relationships with others.

Chapter 5

Conclusion

This original research sought to elucidate the experience of a mother who discovers her fetus has been diagnosed with a rare birth defect, Fibular Hemimelia, and the various factors which may have influenced her ability to cope. The study recruited subjects from three online social media support groups, hosted by Facebook, which focused specifically on those families or individuals affected by FH and specifically looked at women who had chosen to sustain their pregnancy and have their children despite access and opportunity for an abortion. Three environments were examined in relation to this period of time in their lives: the Medical Arena, Community, and the Characteristics of the mother. This retrospective look by the subjects was reported through a 50 question survey that used multiple choice, ranking and comment/essay-type questions.

Within the medical arena, the subjects found the lack of information and access to resources about Fibular Hemimelia very discouraging. Fibular Hemimelia, although rare, is relatively easy to identify during a standard obstetric second trimester ultrasound. The diagnosis was often managed through additional prenatal testing, some of which was highly invasive and carried significant risk to the fetus. Primary obstetric caregivers, radiologists and genetic counselors were not able to provide their patient with much useful information about the diagnosis. They were also not helpful in supplying resources for these women. The genetic counselors who were enlisted in the management of the diagnosis were found to negatively impact the mother's coping under the circumstances. Their lack of perceived empathy and focus on statistical

analysis, medical terminology, and abortion as an option made the situation significantly more challenging for the mothers. By far, the greatest positive impact within the medical community, on these mothers after the initial diagnosis was meeting with an experienced orthopedic surgeon. The surgeon was able to provide concrete answers to what the diagnosis was and its prognosis for the child. This access to an expert allowed the stress associated with the rare nature of the diagnosis and the unknown outcome to be somewhat alleviated. It provided the mother with an opportunity to have her questions answered and be somewhat prepared for the management of Fibular Hemimelia after their child was born.

The interaction between the mother and her community was extremely significant in how she managed the prenatal diagnosis. Strong community links and relationships with others has been proven to provide support for better stress management (Ozbay). These findings were supported by this original research. Active involvement with a church community was a highly influential factor in some subjects coping abilities. Church involvement allowed these participants to feel supported and comforted during this stressful period of their lives. This supported provided them with the confidence to make the decisions required and manage the outcome of the diagnosis. Prayer was listed as a particularly effective management strategy, where they were able to be meditative and release their concerns and worries to a higher power. Strong, positive, relationships with family, friends and partners also played a huge role in ameliorating the stress of the prenatal diagnosis for the mothers. Having a network of individuals invested in the well-

being of both the mother and fetus provided support and reassurance that help was available to manage the circumstances.

The mother as an individual was a final factor that was investigated in an attempt to see if certain characteristics contributed to her overall coping ability. The study examined four aspects of the subjects. The subjects self reported identity as a mother, the concept of her unborn child, the mother in relationship to others in her life and her as an independent individual. All of the subjects reported a very well-established identity as a mother. They adhered to the concept of maternal instinct as a driving force in how they made their decision to manage the prenatal diagnosis of their fetus. The option of termination of the pregnancy was adamantly rejected fairly early in the coping process. They reported a strong sense of protectiveness towards the fetus and had fears that their child would experience significant pain and physical disability throughout their life. They did not, however, categorize their children as having special needs related to Fibular Hemimelia. This finding may be related to a mother's psychological adjustment to having a child with FH and their inability or unwillingness to categorize them as others who have been labelled as requiring "special needs".

All of the women in the study reported very high rates of positive interpersonal relationships with family, friends and partners. Yet despite this, findings stated that 50% of the women were not confident in who to turn to for support. It was unclear who they felt would provide this support, but throughout the study, it is suggestive that the subjects having access to other mothers or individuals affected by Fibular Hemimelia would provide them a cohort by which they could feel a kinship and confidence in their support.

On a personal level it was reported by all of the mothers that their fear of the unknown of Fibular Hemimelia and their emotional state was a huge detriment on their ability to cope with the diagnosis. The mothers did not know what the diagnosis meant, what to expect or where to get their questions answered. They were concerned about how they would manage the condition and the impact it would have on their children and families. These fears were debilitating to most of the respondents. When asked though what was the single largest personal factor that helped them cope with the diagnosis, they all responded that it was not their confidence in the ability to get answers and make good decisions which helped them the most, it was a uncompromising belief in their instincts as a mother and that things would turn out the way they should that comforted them the most. This belief allowed them to transcend the fears that crippled them initially, let go of factors outside of their control and focus on moving forward to care for their child.

Limitations of Research

There are a number of limitations to the study and research that was conducted, and thus the conclusions that were drawn.

- Firstly, the study was retrospective and so naturally there is an element of reporter bias that is unavoidable. Although this bias may be unconscious. it is important to be aware that it exists.
- Due to the rare nature of the birth defect and the difficulty of discovering suitable candidates with which to conduct the research, the sample was extremely small and not necessarily reflective of a wide range of experience.

- The survey was conducted online and in English, further restricting those who do not have access and opportunity to using social media and proficiency in the language. The subjects were relatively homogenous in terms of ethnicity, education, wealth, marital status and community life. This again would limit the amount of women able to participate and prevent the research from getting a fuller picture of the elements at play.

Recommendations for Future Research

This original research represents the first time an examination of the factors that influence the coping strategies of women whose fetuses have been diagnosed prenatally with Fibular Hemimelia, a rare birth defect has been conducted. Although this study only focused on one very specific birth defect, it is possible to imagine that women who receive an unexpected diagnosis of any birth defect in their fetus would experience similar challenges in coping with the news. Shock, despair, fear and desperation are emotions that seemed universal to each of these mother's experiences. How the medical community and society manages these events will become more significant as prenatal testing advances and early diagnoses of birth defects become more prevalent in developed countries. It is possible to follow trends around the world correlating prenatal diagnosis of birth defects and termination of pregnancy rates. However, for those women for whom abortion is not an option, society must be able to respond to their needs and provide a safe, supportive environment in which to deliver their children and to raise them with an exemplary quality of life within their community. To achieve these goals, further research is required.

Follow-up focus groups or one-on-one interviews would enable a researcher to more fully explore some of the concepts suggested at and unearthed in this paper. Areas of interest to focus on would be:

a. Role of communication with medical care providers.
b. Role of the radiologist in an initial consult with mother's who's fetuses have been diagnosed with an anatomical birth defect.
c. The role of the Orthopedic Surgeon in prenatal diagnosis and a communication strategy for the mother.
d. Analysis of access to trustworthy, high value resources for mother.
e. Exploration of the role of faith in helping mothers cope with an unfavorable prenatal diagnosis.
f. Role of ultrasounds in establishing a closer mother-child bond and outcomes on termination rates.
g. Role of attribution theory in response to catastrophic, negative circumstances in a pregnant woman whose fetus has been diagnosed with a birth defect.
h. Stigmatization of those identified with so-called "special needs" and its impact on maternal-child bonding.

Recommendations for Improving Maternal Experience

Two primary recommendations may prove useful in aiding a mother who has recently received a diagnosed of a birth defect of their fetus.

1. An independent, impartial patient advocate to bridge the gap between the medical arena and community resources for mothers and also serve as a source of

nonjudgmental, unbiased strategies to help a mother deal with the challenges of the circumstances.

2. A not-for-profit, online resource center to provide all women regardless of diagnosis, race, socioeconomic status, geography with high quality access and funding to resources and specialists able to provide reliable, valuable information regarding the specific birth defect affecting the fetus and nonjudgemental options for managing the diagnosis.

Chapter 6

Post Birth Commentary

The women who participated in this study bravely and generously examined a very challenging moment in their lives. They were confronted with a diagnosis of a rare birth defect in their fetus and chose to sustain their pregnancies and not opt for a termination. Medical Arena, Community Influence and Independent Characteristics of the mother were examined to determine which factors negatively affected the woman's experience and which factors were helpful for her. It is of interest then to move to the current situation with these women and examine how their management of the diagnosis and perception of the outcome did in fact turn out. The research asked the mothers to evaluate their current management of Fibular Hemimelia, how it has impacted their family and affected their children's lives, and their treatment option and rationale. It is worth noting that 80% of the survey respondents believed that the reality of dealing with their child's Fibular Hemimelia condition was less difficult than they expected while pregnant. They feel that their child had not suffered emotionally or socially because of the defect, although they do recognize the impact of the condition on their physical wellness. None of them would have changed their mind and considered a termination of the pregnancy in light of their current knowledge of the defect. Sixty percent of the women reported that the diagnosis had not placed a financial burden on the family, but there is no consensus among the respondents when asked whether it had been an emotional burden on the family. Family continued to be extremely important as a source of support for them, and all reported positively in regard to the medical team that treats their children. The greatest

source of information about Fibular Hemimelia was reported by 67% of the respondents as the internet, while the remaining stated that their hospital or surgeon provided them with the best information. Despite these positive outcomes for the mother and child, the mother still felt that she was inadequately prepared for the condition prior to the birth of her child and still continues to want more information about the defect. When projecting themselves into the future and considering the long term prognosis, 60% believed that their child will have more physical complications related to Fibular Hemimelia.

There are three options for the treatment of Fibular Hemimelia and rely primarily on the severity of the presentation and the treating physician. A mild case of FH could be left alone, with no intervention necessary. Through observation of the child's growth and their adaptation to the condition it, would suggest a surgical or mechanical intervention if necessary. There are two very distinct and dramatically different surgical options: Limb Amputation and Limb lengthening and reconstructive surgery. These options tend to be heavily debated and tread on potentially political and emotionally treacherous ground. Limb amputation is suggested in many cases and is usually considered dependent on the severity of the presentation, but other factors, such as geography and access to advance FH surgical procedures and surgeons, can play a role in the decision. The Fibular Hemimelia amputation patient traditionally undergoes a Syme or Boyd amputation, which involves the disarticulation of the ankle and removal of the forefoot. This is generally done earlier in a patient's life, prior to the age of two. A below knee prosthetic is then fitted and worn. The limb-saving option is usually quite complex involving

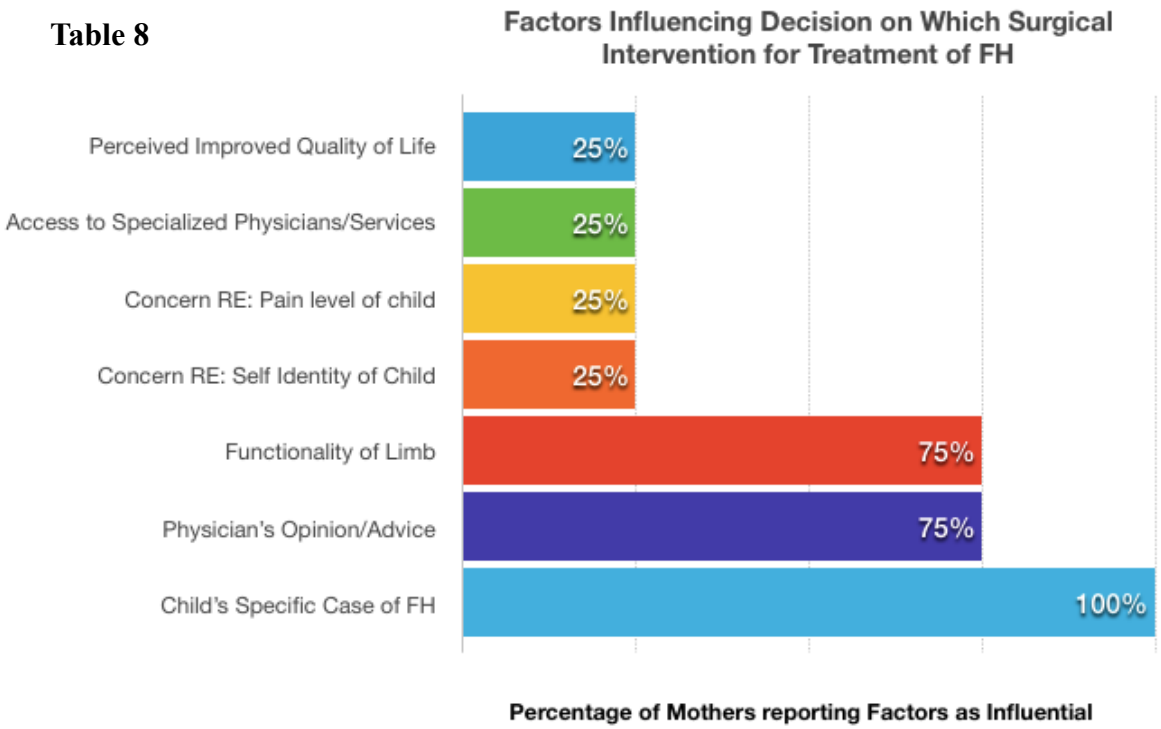
multiple, staged surgeries from year one of a child's life through puberty. In may involve ankle reconstruction, multiple limb lengthenings, knee stabilization and reconstructive surgery. Dr. Minoo Patel, along with a panel of additional world renowned orthopedic surgeons and FH specialists conducted an examination of the surgical options for the treatment of FH. "The prognosis for both options is quite good and there is usually fairly good motility and positive outcome reported by patients and their caregivers" (Patel). As can be imagined though, making the choice between the two options can be very difficult for a parent. There exists the conflict between saving the limb and allowing the child to make the decision to amputate when she or he is old enough to understand the consequences or subjecting their child to possibly numerous intensive surgical procedures with the possibility of chronic pain and limited functionality of the limb.

For the subjects in this study, 60% chose limb amputation and prostheses for their children, 20% chose limb saving and reconstructive surgery and the last 20% had decided not to have any medical intervention to date. Regardless of the treatment for their child's Fibular Hemimelia, all respondents reported the same factors as influential to their decision. One hundred percent listed their child's specific presentation of Fibular Hemimelia as the reason that they chose their surgical option. Seventy five percent of the mothers listed the physician's opinion and concern about the functionality of the limb. Other factors that were influential were pain level of their child, concerns about how the child would feel about themselves and access to specialized physicians and services. See Table 8.

Herzenberg, a renowned expert in limb saving reconstruction for FH patients, makes a case by stating that "in cases of very high grade deformity in which I feel that there is little hope of a successful reconstruction, and for families in which the social situation precludes the time/effort/expense of reconstruction" that amputation may be a reasonable option (Herzenberg). The mothers of children who underwent amputation believe completely that they made the right management decision for their children. The mother who chose the limb-saving option was unsure of her decision at times.

> *I believe we made the right decision for our son, however, there are days that I question if it was the right thing to do. It doesn't last long though, after decision is made you need to just move ahead and not second guess yourself.*

Table 8



As a final note, the women were asked to share any words of advice that they would offer someone who was in the same situation as they found themselves. Their responses were overwhelmingly positive and encouraging:

> *That children are resilient and that it is amazing to see what a child can do within a supportive environment.*
>
> *The diagnosis will affect you more than it will affect your child :)*
>
> *Take a deep breath and breathe. Although it seems like devastating news,there are so many options out there to help your child. Would suggest doctors and pray with them. FH children are resilient, determined and inspiring. Once you meet that little person, they will melt your heart.*
>
> *The child will be a blessing to their family. Whatever medical course they take to help the child's quality of life (amputation, lengthening etc.) is unique to them. It is a decision that needs to be made on your knees, in prayer. You will know what is best for your child. Don't let others tell you what to do. Take their advice and make your OWN decision.*

Epilogue

My daughter was diagnosed with a left club foot during a routine second trimester ultrasound. I was referred to genetic counselors, and high risk obstetricians for consultation. My experiences with these professionals was not particularly informative or reassuring. I was pressured to consider additional invasive prenatal and genetic testing of my husband and I to screen for rare syndromes to explain the finding. This entire process was extremely stressful and changed what should have been a normal, exciting pregnancy into one of fear and concern for the well-being of my child.

The greatest piece of advice that I received at this time was from my family physician who had known me for years. She said: "If all the tests results in the world do not change the fact that you want and love this child, then why would you do them? You will deal with it after she is born." This brought me immeasurable comfort and resolve for the remainder of my pregnancy. I was given permission to let go of control. Needless to say, I did not undergo any additional tests.

My daughter's club foot was always complicated, with numerous other unexplained features and was very reluctant to respond to any surgery. When she was seven years old, after undergoing four previous surgeries starting at the age of six months, we were finally given a diagnosis of Fibular Hemimelia. She underwent an extensive limb lengthening and de-rotational foot procedure at that time. She has worn ankle foot orthotics her entire life and has always been under the care of a physiotherapist to reduce pain and improve her gait. She is now sixteen years old and completed an additional reconstructive surgery on her knee and foot a year ago, related to her birth defect. She is an extremely active

and well-adjusted young adult, playing lacrosse and field hockey for her school and horse riding; however, she has chronic pain in her foot and knee, an aberrant gait and experiences fatigue very easily with her leg. She often expresses frustration and anger that her leg and foot do not work properly, that she has to wake up every day with pain and occasionally feels self conscious about the scars covering her limb. She is now considering amputation of her foot to improve the quality of her life. Her greatest desire is to one day be able to wear high heels.

APPENDIX A: Drew University IRB Approval

DREW

Institutional Review Board
Drew University
36 Madison Avenue
Madison, New Jersey 07940

Kate Ott, PhD
Chair, IRB
Asst. Professor Christian Social Ethics
e. kott@drew.edu
p. 973.408.3298
drew.edu

October 23, 2014

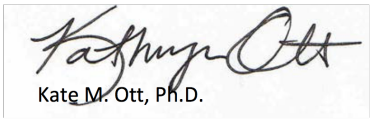
Dear Yvette Vieira,

The Institutional Review Board has conducted an expedited review and approved your research entitled "Maternal Rationale and Coping Strategies Following Prenatal Diagnosis of a Rare Birth De-fect, Fibular Hemimelia, for Women who Choose to Sustain the Pregnancy and Raise the Child" with the following conditions:

- Include in your proposal and in the consent forms how long the data will be kept. In other words, you must tell the committee and participants when the data will be discarded.
- Also, please change throughout the document that Dr. Kate Ott is the chair of the IRB, not Dr. Bill Rogers.
- For clarity of reading, please proofread one more time before finalizing and adding to your research portfolio. There were a number of typos throughout.

The approval for your research protocol and the human participants' component of your work is valid through 23-Oct-15. If you plan to continue this research past this date, you will need to reapply for IRB approval. If you make any modifications to your research, you will need to obtain IRB approval for those changes as well.

Sincerely,



Kate M. Ott, Ph.D.

APPENDIX B: Letter of Permission to Solicit Subjects

Permission for Solicitation of Subjects from Facebook Group

- Fibular Hemimelia and Amputation Awareness (open group)
- Fibular Hemimelia and Limb Lengthening Awareness (closed group)
- Fibular Hemimelia Support Group (open group)

My name is Yvette Vieira. My fifteen year old daughter has type 4 Fibular Hemimelia and I have been an active member of your Facebook group for the past few years. In addition, I am also a researcher / graduate student at Drew University, Madison, NJ seeking my Master's in Medical Humanities.

I am interested in conducting research into the experiences of mothers who have had their fetus' diagnosed while pregnant with Fibular Hemimelia, or a differential diagnosis of a limb reduction defect which ultimately was confirmed as FH. These mothers must have sustained their pregnancies and chose to raise their child.

As a researcher, I seek your permission, as the facilitators of the group, to solicit from your membership potential subjects that meet the requirements as noted above. Solicitation would involve me posting a formal invite to your membership at large. Interested parties would be contacting me via private email to confirm interest. All communications, consent, surveys, interviews, debriefing would take place in a confidential, closed, private environment not associated at all with Facebook, or your support groups at all.

The potential benefits from this research include,

1. Better understanding of the cognitive processes of women who are faced with a prenatal diagnosis of birth defect in their fetus
2. Identifying various factors which contribute to better management of information and resources for mother
3. Identifying various factors which negatively influence the coping strategies of the mother
4. Recommendations for protocol surrounding management of a prenatal diagnosis of birth defects within the medical community
5. Recommendation for resources within the community to provide support to mothers of fetus' diagnosed with a birth defect
6. Bring more awareness to the birth defect, Fibular Hemimelia, in order to share outcomes, information, improve resources and strengthen the affected community.
7. Provide a narrative for women under these circumstances to feel a greater strength of support and understanding in order for them to cope better with the circumstances.

APPENDIX C: Letter for Solicitation of Subjects

Solicitation Letter for Subjects
Posted in Facebook Social Media Groups

- Fibular Hemimelia and Amputation Awareness (open group)
- Fibular Hemimelia and Limb Lengthening Awareness (closed group)
- Fibular Hemimelia Support Group (open group)

My name is Yvette Vieira. My fifteen year old daughter has type 4 Fibular Hemimelia and I have been an active member of your Facebook group for the past few years. In addition, I am also a researcher / graduate student at Drew University, Madison, NJ seeking my Master's in Medical Humanities.

I am conducting research into the experiences of mothers who have had their fetus' diagnosed while pregnant with Fibular Hemimelia, or a differential diagnosis of a limb reduction defect which ultimately was confirmed as FH. These mothers must have sustained their pregnancies and have chosen to raise their child.

As a researcher, I am looking for subjects that meet the requirements as noted above.

The potential benefits from this research include,

1. Better understanding of the cognitive processes of women who are faced with a prenatal diagnosis of birth defect in their fetus
2. Identifying various factors which contribute to better management of information and resources for mother
3. Identifying various factors which negatively influence the coping strategies of the mother
4. Recommendations for protocol surrounding management of a prenatal diagnosis of birth defects within the medical community
5. Recommendation for resources within the community to provide support to mothers of fetus' diagnosed with a birth defect
6. Bring more awareness to the birth defect, Fibular Hemimelia, in order to share outcomes, information, improve resources and strengthen the affected community.
7. Provide a narrative for women under these circumstances to feel a greater strength of support and understanding in order for them to cope better with the circumstances.

APPENDIX D: Informed Consent

Imbedded in the online survey

CASPERSEN SCHOOL OF GRADUATE STUDIES
DREW UNIVERSITY, Madison NJ
Masters of Medical Humanities, Thesis

INFORMED CONSENT FOR ONLINE SURVEY FOR MOTHERS OF CHILDREN

WHO WERE IDENTIFIED PRENATALLY WITH A LIMB REDUCTION DEFECT,

RESULTING IN A DIAGNOSIS OF FIBULAR HEMIMELIA

Principal Investigator : Yvette Vieira
Name of Organization: Caspersen School of Graduate Studies, Drew University, Madison NJ
Academic Advisor: Dr. Philip Scibilia, Director of Medical Humanities Program, Drew University

You will be provided a copy of the full Informed Consent Form via email upon request.

Information Sheet

Purpose of the Study. This research study examines how mothers whose fetus' are identified prenatally with a limb reduction defect, resulting in a diagnosis of Fibular Hemimelia cope with the news. The study will explore how the medical arena, community resources and an individual's personal characteristics influence their coping skills and impact their rationale for sustaining the pregnancy and raising the child.

What will the study involve? The study will involve an anonymous survey/questionnaire which will be accessed online. The survey will take approximately 15 - 25 minutes of your time to complete. There will be approximately 20 subjects involved in this research

Why have you been asked to take part? You have been asked because you have identified yourselves as mothers of children who have Fibular Hemimelia. In addition, you have identified that while you were pregnant, pre natal testing diagnosed your fetus as having a significant limb reduction defect.

Do you have to take part? You do not have to take part in this study. Participation is entirely voluntary. You will be asked to read through this consent form and sign it before participating. You have the option of withdrawing before the study, even if you have previously agreed to participate. You can also opt out of the study or discontinue at any time after you have started the survey.

APPENDIX E: Debriefing Document for Subjects

Maternal Rationale and Coping Strategies Following Prenatal Diagnosis of a Rare Birth Defect, Fibular Hemimelia, for Women who Choose to Sustain the Pregnancy and Raise the Child

Respondent Mothers,

I would like to take this opportunity to thank you for contributing your time and efforts to this study. The study was designed in an attempt to capture the complex and emotionally challenging experience of discovering while you were pregnant that your child was diagnosed with a birth defect. For some of you that original diagnosis may not have been Fibular Hemimelia, but you were probably informed of a significant limb defect. For some of you the diagnosis may have been given to you early in your second trimester while others received the news further along into your pregnancy. Regardless of these factors though, you did, as mothers commit to seeking information and resources for your child. You did have your child and commit to raising them. This process of figuring out a way to cope and how you personally adjusted and accommodated to this new reality for your child is what interests me.

As a mother of a child with Fibular Hemimelia who was also diagnosed prenatally I recall how difficult it was dealing with a diagnosis, trying to find good information and understanding what this would mean for the future.

I attempted to examine three different areas that I felt could contribute to how you managed the diagnosis: the Medical Arena, Community Resources, and you as an individual. I would like to understand in greater depth what factors helped you in your journey, what factors negatively affected your experience. After collecting this information, I am hoping that this will allow me to formulate some suggestions on policy changes, resource referrals and greater community and interpersonal support systems for future women faced with this situation.

I do recognize that some of the questions in the survey may have brought back uncomfortable or distressing emotions or memories. If this is the case, for this I am very sorry, however, I felt I needed to fully understand the impact these circumstances had on your life so that I may be able to help future generations of women. I would suggest, that if you are having difficulty reconciling these emotions or feel in need of discussing them further that you follow up with some of the support resources listed on the attached page.

I would like to remind you that your responses to the survey are fully confidential. The responses in general will be published as group data. I will also only include excerpts from your answers in my publication if you have given permission for me to do so in the informed consent you signed. These excerpts again will remain anonymous and protect your identity.

APPENDIX F: Subject Survey

1. Description of Study

CASPERSEN SCHOOL OF GRADUATE STUDIES
DREW UNIVERSITY, Madison NJ
Masters of Medical Humanities, Thesis

INFORMED CONSENT FOR ONLINE SURVEY FOR MOTHERS OF CHILDREN WHO WERE IDENTIFIED PRENATALLY WITH A LIMB REDUCTION DEFECT, RESULTING IN A DIAGNOSIS OF FIBULAR HEMIMELIA

Principal Investigator : Yvette Vieira
Name of Organization: Caspersen School of Graduate Studies, Drew University, Madison NJ
Academic Advisor: Dr. Philip Scibilia, Director of Medical Humanities Program,
Drew University

Information Sheet

Purpose of the Study. This research study examines how mothers whose fetus' are identified prenatally with a limb reduction defect, resulting in a diagnosis of Fibular Hemimelia cope with the news. The study will explore how the medical arena, community resources and an individual's personal characteristics influence their coping skills and impact their rationale for sustaining the pregnancy and raising the child.

What will the study involve? The study will involve an anonymous survey/questionnaire which will be accessed online. The survey will take approximately 15 - 25 minutes of your time to complete. There will be approximately 20 subjects involved in this research

Why have you been asked to take part? You have been asked because you have identified yourselves as mothers of children who have Fibular Hemimelia. In addition, you have identified that while you were pregnant, pre natal testing diagnosed your fetus as having a significant limb reduction defect.

Do you have to take part? You do not have to take part in this study. Participation is entirely voluntary. You will be asked to read through this consent form and sign it before participating. You have the option of withdrawing before the study, even if you have previously agreed to participate. You can also opt out of the study or discontinue at any time after you have started the survey.

Will your participation in the study be kept confidential? Your participation in the study will be held in confidentiality as will your responses to the survey. No clues to your identity will appear in the research publication and no one will be able to infer your identity from any of your responses that may be quoted. The survey responses will be collected online through the site, survey monkey and is password protected. The head researcher, Yvette Vieira, is the only individual who will have this password to access the data.

What will happen to the information which you give? The information you provide will be kept confidential. It will be shared with an academic advisor and the Institutional Review Board, which is the human ethics committee at Drew University.

What will happen to the results? The results from the survey will be included in the research publication and will be available in the Drew University Library.

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December 2015
VITA

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Place and date of birth: Umtali, Zimbabwe October 15, 1971

Parents Name: Stella Krause and Raymond Kennedy

Educational Institutions:

School	Place	Degree	Date
Secondary: Loyola Catholic SS	Mississauga, Ontario Canada	HS Diploma	1989
Collegiate: McMaster University,	Hamilton, Ontario, Canada	BA	1994
Graduate: Drew University,	Madison, NJ	MMH	2015