PATIENT PREFERENCES MATTER: A QUALITATIVE INQUIRY WITH PATIENTS ABOUT THE SURGICAL DECISION FOR OSTEOSARCOMA IN THE LOWER EXTREMITY

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Purpose

The experience of surgical patient decision-making for amputation, rotationplasty, or limb salvage surgery for the lower extremity was explored with osteosarcoma survivors and parents. Issues and patient concerns were examined prospectively in Facebook posts and retrospectively in personal interviews. Recommendations were sought for the development of a decision aid.

Methods

A qualitative inquiry was conducted in two studies: 1) a content analysis of 15 Facebook posts on the Osteosarcoma and Ewing's Sarcoma Support Group about the surgical decision, and 2) a narrative analysis of 20 interviews with survivors and parents about their lived experience. The interview guide was informed by a knowledge translation theoretical model. In-person or video interviews included 29 parents or adolescent-young adults between the ages of 14-71 across 15 states.

Results

Participants were concerned about making the best decision for themselves or their child. Having little time to make the decision contributes to feeling overwhelmed and uninformed. The lack of information about options, potential limitations, prosthetic needs, and long-term functional outcomes impacts the decision-making process. Resources for finding information are limited. Shared decision-making approaches were preferred though many decisions were made by the surgeon. Patients felt unprepared for complications encountered after limb salvage.

Conclusions

Limb salvage surgery remains the gold standard for most providers; participants felt there was a bias on the surgeon's part for salvage. Potential complications and uncertainties of each option were largely omitted in consultations; information about living with a prosthesis is an unmet need for decision-making. The decision affected their quality of life in ways they did not anticipate, resulting in additional decisions.

Implications

The findings of this study can be used in medical education for orthopedic surgeons to understand the long-term outcomes of limb salvage surgery. Patient preferences matter in the decision and patients who are involved in the decision are more satisfied if they are prepared for the unknown and potential complications. Recommendations for a decision aid include an online format, content that includes a discussion of uncertainties as well as complications of each option, patient stories for long-term outcomes, and resources for psychosocial support.

Elizabeth Goering, PhD, Chair

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LIST OF ABBREVIATIONS AND TERMS

AMP	Amputation
AYA	Adolescent-young adults
Differential	Leg length difference
Distal	Further from the torso or center of the body; distal femur is closer to the knee.
Endoprosthesis	Metal implant for the limb salvage surgery
Femur	The upper part of the leg above the knee
LSS	Limb Salvage Surgery
MAP	High-dose Methotrexate, Adriamycin, Cisplatin are the standard chemotherapy agents for the treatment of osteosarcoma.
MIB FACTOR	Annual osteosarcoma conference, Make It Better, Funding, Awareness, Collaboration, Trials, and Osteosarcoma Research
NED	No Evidence of Disease
Necrosis	Dead cancer cells from a tissue sample
OS	Osteosarcoma
PROs	Patient-Reported Outcomes
Prosthesis	Artificial leg or “prosthetic device”
Proximal	Closer to the torso or center of the body, proximal femur is closer to the hip.
Recurrence	The return of cancer (also referred to as relapse)
SDM	Shared Decision-making
Tibia	The lower part of the leg below the knee.
LSS	Limb salvage surgery
RP	Rotationplasty

CHAPTER ONE
INTRODUCTION

Osteosarcoma

Osteosarcoma is a rare cancer, but it is the most common type of bone cancer in children and young adults and the third most common of all pediatric cancers (Durfee et al., 2016). It usually occurs around the knee during periods of rapid bone growth in adolescence with a peak between ages 10-14 (Misaghi et al., 2018; Robert et al., 2010). Osteosarcoma is a very aggressive type of cancer; one third of all patients with successful chemotherapy who have not metastasized are expected to have a recurrence (Crompton et al., 2006). That said, it is still considered curable for most patients with a combination of chemotherapy and surgery; the 5-year survival rates are around 65% (Durfee et al, 2016; Zhang et al, 2018).

Surgical options for osteosarcoma in the lower extremity. Depending on the location of the tumor in the lower extremity, there are several options for surgery that include amputation, rotationplasty, and limb salvage surgery, each of which has risks and benefits and very different appearances (Zhiqing et al., 2019). The surgical decision is considered preference sensitive - one in which clinical evidence does not indicate a clear right or wrong option - making it optimal for shared decision-making (Charles et al., 1997). The shared decision-making approach brings the doctor’s expertise in treatment options and evidence-based outcomes together with what the patient knows best – their preferences, values and beliefs (Frosch & Kaplan, 1999). True shared decision-making occurs when individuals decide which treatment option best supports their physical and psychological well-being and they feel supported to make decisions that are right for them (Elwyn, 2006; Pierce, 1993).

A pediatric cancer diagnosis causes feelings of fear and uncertainty for the patient and parents alike and threatens the family’s sense of safety. Choosing one of the options is an extremely difficult decision, especially for surrogate decision makers like parents of young children. This mother posted her struggle with the long-term outcomes of the surgical decision for her young son on the public Osteosarcoma and Ewing's Sarcoma Support Group Facebook page:

> How do you make this life changing choice for a 5-year old? What if you make the wrong choice? What if there’s a way to save the leg and you don’t take that chance? What if you decide to try, and it only causes pain and a life of additional surgeries?

This dissertation is the formative research needed to develop the Amputation Patient Decision Aid for Osteosarcoma in the Lower Extremity, part of a five-year cooperative agreement between the Administration for Community Living and the Amputee Coalition. Chapter One includes the rationale for the research study, an introduction to the theoretical framework, and a note on my reflexive role in the community.

Rationale

The development of resources to aid families when the decision for surgical intervention is being made for osteosarcoma in the lower extremity is important for several reasons, which are outlined in this section.

Patients are unprepared to make surgical decisions. Research indicates that patients may feel intimidated by the complexity of medical decision-making (Frosch & Kaplan, 1999). Many families report feeling uninformed and confused about their options, such as this parent on the public Osteosarcoma and Ewing's Sarcoma Support Group:

> i honestly don't know. we were a little blind sided when his surgeon spoke to us a couple weeks after our initial meeting. his first reaction was the internal then came back and told us he thought rotation was the way to go. been so caught up on trying to get other opinions with this we didn't know where to look or what to ask. doctor offered no information and told us to do the research.

The manner in which patients or parents have communicated with doctors prior to the diagnosis can also influence their expectations for decision-making responsibility for osteosarcoma. Traditional passive patient roles and deference to the physician's expertise can prevent patients from participating fully with providers in decision-making (Frosch et al. 2012). From the patient's perspective, engaging in a medical decision, much less a surgical decision of this magnitude, can be uncharted territory.

Patients may have different preferences for whether they want to take an active or passive role in making the decision though most patients want to be involved in treatment decision-making (Chewning et al., 2012). The role of surrogate decision-making is particularly problematic for parents who make the decision for their children. Decisional burden can occur when parents are expected to be in a decision-making role that they don't want, particularly for children with cancer (Sisk et al., 2019). This type of decisional burden has long lasting psychological effects on surrogate decision-makers and can lead to role stress and depression (Hickman & Pinto, 2014).

Expectations for decision-making are unclear. The surgeon's role in making the decision can vary widely between providers. One surgeon might authoritatively decide which procedure is best for the patient in a patriarchal model of decision-making. For surgeons who follow an informed decision-making model, the decision is entirely the patient's preference if the surgeon views their role as strictly to provide information and necessary scientific knowledge (Eddy, 1990; Mooney & Ryan, 1993).

Surgeons sometimes engage collaboratively with patients and families about their preference for a treatment option in a shared decision-making model. Carol Morris, MD, Johns Hopkins, Division Chief of Orthopedic Oncology, shares her perspective about the importance of engaging with patients for decision-making in osteosarcoma surgery:

> The most nuanced part of what we do is matching the right operation with the right patient. There are lots of options out there and all of those options probably will work fairly well. But if the patient doesn't want the option, then it doesn't work at all (Johns Hopkins Medicine, n.d.).

Most people do not have a history of contributing to surgical decisions, much less one of this magnitude, for themselves or for a child. The uncertain roles of both patients and providers make this traumatic diagnosis even more overwhelming, as described by this father on Facebook, “We are trying to make an educated decision while moving a million miles an hour.”

No decision aids exist for osteosarcoma surgery. In the United States, there are no patient decision aids available to families when making a surgical decision for osteosarcoma in the lower extremity. This lack of comprehensive information about risks and benefits of each option represents a missed opportunity in shared medical decision-making that has lifelong ramifications for function and quality of life in osteosarcoma survivors.

Outside of the U.S., a decision aid for osteosarcoma decision-making was developed in the Netherlands at the Amsterdam Academic Medical Center. One of the developers, Dirk Ubbink, MD, PhD, is a surgeon and professor in Evidence-Based Medicine and Shared Decision-Making whom I met at the 2019 International Shared Decision-Making conference. He shared the decision aid with me and although it is in Dutch, I could translate the content to English with Google Translate. It is an excellent example of a decision aid and includes photos, animations, videos, and knowledge/preferences questions; it has been used with a small group of surgeons at his institution but data had not been collected prior to the completion of this dissertation.

Knowledge Translation

Understandably, families are not equipped to make surgical decisions and are unsure of their decision-making role for osteosarcoma surgery. Understanding their needs and concerns at this pivotal time is paramount to informing the development of a decision aid. Knowledge translation is a theoretical framework that grounds applied research in the need for knowledge by users (patients) from producers (surgeons). Knowledge translation (KT) arose out of translation theory from the field of applied linguistics and communication to describe an iterative loop between knowledge producers and knowledge users in various stages of KT (Davison, 2006). It is defined by the American Institutes for Research as “strategies that move research into practice by improving the relevance, reporting, accessibility, interpretation, and application of research results."

There are many models of KT but the one that best drives the qualitative inquiry of lived experience for surgical decision-making is the interactive framework of knowledge translation (Jacobson et al., 2003). The interactive framework has five domains: user domain, research domain, issue domain, researcher-user domain, and dissemination domain. As modified and applied to this study, the user is the osteosarcoma patient/family, research is the knowledge/information about surgical options, issues are concerns that impact the decision, researcher-user relationship includes interactions between the patient/family and any person who provides information, and dissemination is the recommendations on the decision aid for future families.

Combined with the high-level objectives of knowledge translation as defined by AIR, the interactive model is the best framework for the personal interviews as formative research leading to implementation with a patient decision aid. A more complete overview of this model will be included in the methods section of Chapter Four: Study 2.

Reflexivity

For full transparency, I am the parent of a child who was diagnosed with osteosarcoma at the age of 12. Zoe had a limb salvage surgery and 11 years later, she had an above knee amputation at the age of 23. I have been active in the osteosarcoma support community for all since her diagnosis in 2007 and I am trained as an Ambassador Agent, a certified peer visitor with the MIB Agents, a leading pediatric osteosarcoma nonprofit dedicated to Making It Better for other families. As a member of the community, I am aware of the reflexive role I play as both a parent of a child who survived osteosarcoma and as a researcher gathering data on the decision-making experience. While one of the benefits is that I share a similar lived experience with the participants, I am also mindful of subjective conclusions drawn from the results. By using narrative methodology to report the results in their own words, the participant's voice is uninterrupted, and conclusions are based on their concerns as expressed verbatim.

CHAPTER TWO

LITERATURE REVIEW

This section looks at several overlapping areas of decision-making from patient involvement in surgical decision-making and more specifically in osteosarcoma surgery decision-making to the role of parents and children/adolescents as decision-makers. An overview of decision-making models for making decisions (doctor vs. patient vs. doctor/patient) is included along with a review of decision aids and the use of qualitative research for patient-reported outcomes.

Surgical Decision-Making

Decision making with patients is an inherent part of orthopedic surgery, and researchers have explored shared decision making and the use of decision aids in areas of orthopedics such as spine surgery (Nwousu et al., 2017), knee replacement (de Achaval et al., 2012), back surgery (Deyo et al., 2000), and hip surgeries (Arterburn et al., 2012). However, research shows that surgeons and patients approach surgical decision-making differently. Surgeons rely on several factors when determining which type of surgical procedure to recommend. These can range from patient factors such as age, gender, and patient preferences to how comfortable the surgeon feels about recommending a newer technique as opposed to a “tried and true” method (Gunaratnam & Bernstein, 2018). Patients, on the other hand, consider daily quality of life considerations and how much they trust their physician as well as the desire to be informed and involved though they might prefer a more passive role when making a surgical decision (Cranley et al., 2017). For orthopedic decision-making, “decisions often involve possible elective quality of life surgery rather than treatment or screening for immediately life-threatening conditions” (Slover et al., 2012, p. 1049), making the need for most orthopedic surgeries very different than the need for osteosarcoma surgery. Therefore, the next section focuses specifically on scholarship related to osteosarcoma surgical decision-making.

Osteosarcoma Surgical Decision-Making

Patients and orthopedic surgeons alike often refer to the surgical options as these three: amputation, rotationplasty, and limb salvage surgery (see Figure 1). However, rotationplasty is a type of amputation and within each option is a great many variances. Rotationplasty is an amputation that involves the removal of the knee where most tumors occur, then rotation the lower leg below the knee 180 degrees, and reattaching it to the upper leg with the attached foot pointing in the opposite direction (Betsy et al., 2005). The ankle becomes the knee and the foot supports a prosthetic device.

Figure 1

Surgical Options

Rotationplasty Amputation Limb Salvage Surgery

Amputations can vary from below the knee to above the knee depending on the location of the tumor. These will vary in appearance, depending on the amount of bone left, affecting the length of the residual limb. Another type of amputation used with osteosarcoma is the tibia turn up plasty. In a tibia turn up plasty, the tibia or lower leg is removed, turned upside down and attached to a high amputation of the femur or thigh; the knee, foot, and ankle are discarded (Tronzo & Janek, 1995). For patients who have limb salvage failures that would leave them with a short stump with amputation, a tibia turn up plasty gives them a longer stump that allows for better weight-bearing when wearing a prosthetic device (Lim et al., 2019). This procedure is being seen more often in the osteosarcoma community when survivors choose amputation for a better function years after a limb salvage surgery that limits their reconstructive options.

Limb salvage procedures can include many types of biological reconstruction using a donor bone from a cadaver or the patient’s own transplanted bone. These are biologic replacement and are called allografts and autografts (Misaghi et al., 2018). Expandable implants are also an option and there are many different systems to choose from; however, some are associated with high rates of failure. The expandable device known as the Repiphysis Limb Salvage System allows for noninvasive lengthening but is associated with a high rate of failure. After 6 years, 10 pediatric patients with a Repiphysis had 13 implant failures and 37 complications related to the surgery (Cipriano et al., 2014). On the basis of their findings, these researchers advised surgeons to decide on an individual basis if the device should be used, concluding that “surgeons should recognize the potential for significant bone compromise limiting revision options and consider other options” (p. 831).

Patient-provider communication is paramount to this decision-making process, yet little is known about the factors considered by patients and families when making treatment decisions for osteosarcoma in the lower limb. Patients with osteosarcoma in the upper limb preferred limb salvage but the evidence for patient preference in the lower limb was unclear (Barr & Wunder, 2009). Four issues for surgeons to consider when making medical decisions were identified by orthopedic surgeon Michael Simon (1991) as:

1. Is there any decrease in survival rate after limb salvage compared to after amputation?
2. How do the immediate and late complications compare?
3. How does function compare?
4. Are there any psychosocial benefits for one over the other?

Factors such as the location of the tumor, how much tissue needs to be removed to get clear margins, infections, hardware issues, etc. all impact the mobility and limitations of the patient. For example, the risk for recurrence is slightly higher in limb salvage compared to amputation (Anderson, 2016). The need for understanding the complexity of this decision is best described by Polonet et al. (2017):

> Limb-threatening conditions and actual loss of limb are complex situations that require an appreciation of the treatment options as well as potential outcomes and treatment courses associated with therapeutic interventions. Decisions about salvage and amputation have lasting effects on the patient and, in various settings, can take a patient down a pathway that can have excellent or catastrophic results (p. 217).

Research indicates that information needs for cancer patients are highest near diagnosis; as patients gain a greater understanding of treatment information, they continue to need additional information (Matsuyama et al., 2013). The challenges of osteosarcoma specifically may lead to additional informational needs and psychosocial support online (Lam et al., 2013). According to the standard protocol for osteosarcoma, surgery is scheduled ten weeks after clinical diagnosis. This leaves little time in which to make a major life-changing decision. However, some adolescents and parents indicate that at the beginning of diagnosis, they find it difficult to participate in decision-making due to a perceived lack of options when confronted with established protocols for cancer treatment (Day et al., 2016). It has been suggested that offering patients additional consultation session with time in between to think about their options may help achieve patient preferences (Herrmann et al., 2019).

Skilled surgeons in osteosarcoma recommend lifestyle considerations when counseling with families about surgical choices: “Reconstruction should be individualized to the needs of the patient keeping in mind the oncological, functional and social requirements” (Tiwari, 2011, p. 8). Lifestyle is also a predictor of success for limb salvage surgery. Misaghi et al. (2018) posit that limb salvage has been demonstrated to lead to an improved quality of life. A study in which 27 osteosarcoma patients with LSS who played sports before surgery continued to play sports five years later (Lang et al., 2015). The conclusion was that if patients played sports before LSS, that they could continue to do so if they were active in sports before surgery; the potential to be active rested more on the individual's activity before surgery than with any complications with the LSS. Limb salvage is indeed considered the preferred treatment, "the gold standard" of care by surgeons (Misaghi et al., 2018, Stevenson et al., 2016). It is considered common and is performed for up to 85% of children with osteosarcoma (Grimer, 2005). Despite the significant percentage for children who receive limb salvage surgery, the decision often defaults to the parent to make the decision for which option is best for their child. Parental participation in decision making is often expected when decisions are being made for children.

Parental participation in Decision-Making

Promoting parental engagement in decision-making for hospitalized children has been increasingly recognized as a key component of family-centered care and is endorsed by the American Academy of Pediatrics' Committee on Hospital Care and the Institute for Patient-and Family-Centered Care (2012). They issued a policy statement recognizing that "the perspectives and information provided by families, children, and young adults are essential components of high-quality clinical decision-making, and that patients and family are integral partners with the health care team" (p. 394).

Research indicates that parents want to participate in medical decisions for their children (Dadlez et al., 2018) but when parents are in the decision-making role, they may or may not be in the role they prefer (Sisk et al., 2019). Robertson and colleagues (2019) conducted a qualitative study with pediatric oncology patients in which 25 parents and 5 adolescents that information alone is not enough to facilitate parental participation in decision-making. While parents wanted to be involved in their child's treatment decision, they trusted the oncologist with decisional priority to do what they thought was best. Furthermore, parents were satisfied with the decisions made if there was a positive outcome, as opposed to feeling like it was a well-informed or "right" decision.

The American Academy of Pediatrics (Katz & Webb, 2016) makes these recommendations for surrogate decision-making for parents who make decisions for their children:

> Parents should generally be recognized as the appropriate ethical and legal surrogate medical decision-makers for their children and adolescents. This recognition affirms parents' intimate understanding of their children's interests and respects the importance of family autonomy.

> Surrogate decision-making by parents or guardians for pediatric patients should seek to maximize benefits for the child by balancing health care needs with social and emotional needs within the context of overall family goals, religious and cultural beliefs, and values. (p. 6)

Training health care providers to discuss values and quality of life goals with parents when making decisions for treatment is recommended by Swota and Bradford (2015). They suggest using influential factors found with other parents as a foundation to foster the skills needed to provide support, "A valuable part of training health care providers could start with providing data from the literature on the factors identified by parents as central to their decision-making process" (p. 34). Concerns for parents making decisions for their children often include when and how much to share with their child to involve them in the process.

Child/Adolescent Participation in Decision-Making

The participation of children, adolescents, and young adults in health care decisions is often discussed, especially in oncology. The National Cancer Institute defines the adolescent-young adult (AYA) age group as between 15-39 years (National Cancer Institute, 2018) while other organizations have the start of adolescence earlier as the age range between 12-21 years (Hardin & Hackell, 2017). The AYA group is encouraged to participate in decision-making according to a clinical report from the American Academy of Pediatrics, "Children, when cognitively competent, should be involved in decisions about their care…. Providers should partner with adolescents and parents to solicit preferences about capacities and preferences for involvement…" (p. 5-6). A decision-making tool for AYA patients, their parents, and their doctors has been created by Snaman et al. (2019) called the *MyPref* ACA survey. This tool is designed to help AYA oncology patients identify and communicate treatment preferences and is one way in which researchers are addressing the unmet needs for this population. Family decision-making patterns, factors that influence involvement in decisions, and attitudes were found to be key components of clinical trial enrollment with AYAs and their parents (Barakat et al., 2014) but less is known about how adolescents feel about cancer treatment decisions retrospectively.

Both parents and youth prefer the involvement of their doctors when making treatment decisions and "feelings of distress, inadequacy, and lack of choice impacted decision-making participation" (Robertson et al., 2019, p. 4331) and it has been noted that youth with cancer are making decisions at a time when they may also be experiencing pain or discomfort from treatment as well as a disruption to their normal lives (Boles & Daniels, 2019). However, AYAs do want to be involved in decision-making, and the importance of the patient perspective for them is vital (Mobley et al., 2018). A recent study with 203 AYA cancer survivors found that the majority wanted to share decision-making with the doctors while half wanted limited involvement from parents, and nearly one-fourth expressed regret about the initial treatment decisions (Mack et al., 2019). While shared decision-making is a preferred model of communication for many doctors and patients, there are other types of decision-making taking place including one that excludes the recommendation of the doctor and one that excludes the preference of the patient. To better understand how and when these are used, the next section includes a review of three models of decision-making.

Models of Decision-Making for Medical Treatment

Several models of decision-making for medical treatment have been developed over the last three decades. The three models most relevant to this research study are those in which the physician makes the decision exclusively, the physician provides information for the patient to make the decision exclusively, or the physician and the patient make the decision together: these are the patriarchal model, the informed model, and the shared decision-making model (Charles et al., 1997; Charles, et al., 1999; Emanuel & Emanuel, 1992; Klifto et al., 2017).

The patriarchal decision-making model. In the patriarchal model, the physician is seen as “dominating the medical encounter and using his skills to diagnose and recommend tests and treatments for the patient” (Charles et al., 1997, p. 682). In the patriarchal model, the physician either authoritatively decides which option is best for the patient or chooses selected pieces of information to share and encourages the patient to do what the physician thinks is best (Emanuel & Emanuel, 1992). The patient’s involvement in the decision is thus limited to consenting to the treatment being chosen or advocated by the physician (Emanuel & Emanuel, 1992). In the patriarchal model, “the physician does what he thinks is best for the patient, without eliciting the latter’s preferences” (Charles et al., 1997, p. 682).

The informed decision-making model. In the informed model, decision-making as a process is independent of the physician; a decision is made by the patient with no input from the physician beyond medical information provided by them (Klifto et al., 2017). The physician’s role in this model is strictly for knowledge translation; they only provide technical or scientific knowledge but do not engage in sharing their opinions (Mooney & Ryan, 1993). As such, “the physician's treatment preferences for the patient do not enter into the decision-making process” (Charles et al., 1997, p. 683). The physician does not make a treatment recommendation “for fear of imposing his or her will on the patient and thereby competing for the decision-making control that has been given to the patient" (Emanuel & Emanuel, 1992, p. 2225).

The shared decision-making model. In the shared model, a balance is sought between the one-way exchanges of the paternalistic and informed models, to a two-way exchange involving both the patient and the physician who share the decision-making together (Klifto et al., 2017). The physician educates the patient on treatment options and the patient shares their preferences and values as they relate to the options. Patient outcomes for satisfaction, understanding, and trust are greater when they feel like they are engaged in making medical decisions (Shay & Lafata, 2014) and evidence of poor shared decision-making is associated with worse patient-reported outcomes (Hughes et al., 2018). For example, breast cancer patients who believed they had some input into the treatment decision reported a higher quality of life than patients who believed they had less control over the choice (Street & Voigt, 1997).

In a study on how shared decision-making impacts patients in orthopedic surgery, the recommendation for SDM and decision aids will improve patient satisfaction and engagement in decision-making though further research is needed in this area (Slover et al., 2011). One of the challenges of the shared decision-making model is providing sufficient quality information so that patients understand the risks, benefits, and outcomes between each of the options (Charles et al., 1999). The use of decision aids is one way to fill the need for comprehensive information while improving satisfaction and engagement when different treatment options are available.

Decision Aids

The International Patient Decision Aid Standards, commonly referred to as IPDAS, identify six elements of shared decision-making as situation diagnosis, choice awareness, option clarification, discussion of harms and benefits, patient preferences deliberation, and making the decision. Decision aids include this information as well as patient testimonials and exercises that help patients clarify their personal preferences (Barry, 2002). In a pediatric cancer study, parents indicated that decision-making should focus more on involving the families early in the deliberation phase and not just when making a treatment decision (Roberston et al., 2019). Decision aids can address this need for more information about treatment decisions, particularly during early diagnosis when families are in shock over the diagnosis and sometimes have little time to decide between options. The International Patient Decision Aid Standards (2017) define decision aids as the following:

> Patient decision aids are tools designed to help people participate in decision-making about health care options. They provide information on the options and help patients clarify and communicate the personal value they associate with different features of the options.

Some researchers have stated that it is unclear whether decision aids should support all of the IPDAS criteria and choose to define a decision aid as “any tool designed to support SDM” (Joseph-Williams et al., 2014; Wieringa et al., 2019). Regardless, decision aids are not intended to persuade users to choose one option over another or replace talking about options with their doctor; their intended use is to help patients be better prepared to make informed decisions based on their values with their doctor (Wieringa et al., 2019).

Decision aids have been developed for a myriad of medical conditions. One study found 107 decision aids for prostate conditions, cardiovascular disease, colorectal cancer, breast cancer, endocrine disorders, congenital disorders, infectious diseases, diabetes, psychiatric conditions, neurologic conditions, dental conditions, health behaviors, vasectomies, acute appendicitis, obesity, pulmonary disorders, end of life care, and orthopedic conditions (Leinweber et al., 2019). Many of the decision aids identified were focused on oncology such as cancer screening, cancer care, genetic testing, radiation, and deciding between two surgical options. The researchers also found that "decision aids in cancer care help patients make difficult decisions where a "right" answer often does not exist, and therefore choices must be governed by individualized patient values" (p. 360). In a similar review for decision aids that used the six key elements of shared decision-making, researchers found that these were mostly designed for transferring knowledge about options and their risks and benefits (Wieringa et al., 2019).

Decision aids can be in many formats from paper brochures and booklets to apps on a smartphone and interactive websites with videos. The use of the internet for health information in general has become widely acceptable with both patients and providers. Despite the perceived risks associated with seeking online health information (Benigeri, 2003) and with social media in particular (George et al., 2013), research indicates that patients who seek online health information feel empowered and expect to be more engaged in medical decision-making (Tan & Goonawardene, 2017). As a form of knowledge translation, online information is not seen as a threat by general practitioners and leads to a better understanding of the symptoms and illness (Van Riel et al., 2017) while women with breast cancer value, use, and trust "Dr. Google" for information (Marcu et al., 2018).

The dissemination of online decision aids is now supported by hospital systems such as the Mayo Clinic's Shared Decision Making National Resource Center (Mayo Clinic, n.d.) that offers many decision aids and companies like Healthwise (n.d.) that help health care systems design decision aids using shared decision-making methods. The Veteran's Administration has one for PTSD that explains different types of psychotherapy like cognitive processing therapy, eye movement desensitization and reprocessing, and prolonged exposure to trauma as well as antidepressant medications (Veteran's Administration, n.d.). Online decision aids have also been used in prostate cancer screening decisions with providers and led to increased knowledge, reduced decisional conflict, and reduced the providers role in the decision-making process (Baptista et al., 2018). They were found to be low cost, anonymous, and always available for patients with no differences between printed decision aids and slightly lower decreases compared to video decision aids (Baptista et al., 2018).

Decision aids make it easier for doctors and patients to talk about options and help patients make decisions that include their personal values and preferences, but they do not replace the decision-making collaboration between doctors; rather, they supplement it. Research on surgical decisions found fewer decision aids than for other treatment decisions, “Decision aids designed to assist patients in choosing between competing surgical interventions remain less common and represents an important pathway forward in helping patients make the best quality decisions related to their surgical care” (Leinweber et al., 2019, p. 361).

Despite the connection between SDM and positive outcomes for patients, the evidence for better patient outcomes when SDM and decision aids are used together is weak (Shay & Lafata, 2014). This may be due in part to their lack of use in clinical practice. Researchers in oncology and health behavior have suggested a next steps design to help facilitate the implementation of decision aids that includes outcomes measures, control group testing, and effectiveness trials (Herrmann et al., 2019). While the correlation between SDM and decision aids may be limited, there is evidence that decision aids inform patients about their options better than what generally occurs during consultations with physicians (Barry, 2002). The benefits of using decision aids when making decisions for orthopedic surgery such as intervertebral disc herniation, spinal stenosis, and knee replacement have been established (Klifto et al., 2017). A study that compared the use of a decision aid to the usual education for patients who were considering surgery for a total knee replacement showed that patients who used the decision aid had higher decision quality based on knowledge and the decision was more consistent with their values for option outcomes (Stacey et al., 2014).

Decision aids have demonstrated benefits for patient satisfaction, engagement, and information provision. However, it is important to understand what patients themselves are saying about outcomes in their own words in addition to quantitative measurements and assessments.

Qualitative Research for Patient-Reported Outcomes

Patient-reported outcomes (PROs) are widely understood to come directly from the patient about their health condition without an interpretation of their response by a doctor or anyone else. The exposure of patient reports in social media have opened many windows for researchers to peek into the world of PROs. Facebook specifically has been used to explore how and what people communicate about when they or a family member are diagnosed with a chronic health conditions such as mental health disorders, cancer, and diabetes. For example, a content analysis of user-generated posts and the resulting comments between parents and/or caregivers of children with autism spectrum disorders was conducted to better understand social support messages (Roffeei et al., 2015). An analysis of posts during breast cancer month was analyzed to identify functions and uses of the Facebook site for health promotion (Abramson et al., 2014) while another used Facebook and other social media platforms to examine prostate cancer resources in the community (Struck et al., 2018). Content analysis of existing communication between patients has been explored by using Facebook posts to analyze user-driven conversations about dialysis (Ahmed et al., 2017), support and education about epilepsy (Meng et al., 2017), supportive communication for multiple chronic diseases (Torre-Díez et al., 2012), online support between parents of premature babies (Thoren et al., 2013), and to examine social networking between patients with diabetes (Greene et al., 2010).

Qualitative research with the AYA population impacted by pediatric cancer have been conducted to look at how the cancer experience affects the post-treatment lives of AYA survivors (Belpame et al., 2019), boys who survived testicular cancer (Carpentier et al., 2010), and in explorations with adolescents about their sense of self and quality of life as a cancer survivor (Woodgate, 2005). Qualitative inquires have been used to examine how AYAs experienced the health care system (May et al., 2018) and how parents and children with cancer make treatment decisions (Robertson et al., 2019). The use of “flexible and enjoyable methodology” such as those employed in personal interviews is one way to connect with this unique population to ensure that their voices are heard (Boles & Daniels, 2019). An engaging interview approach is sometimes used in studies by starting with an open narrative section asking the participant to share their story about how the condition started before asking more structured questions (Angel et al., 2017).

Qualitative osteosarcoma research. Qualitative inquires with osteosarcoma patients is limited in the literature to date; only one qualitative study was found specifically with osteosarcoma patients, survivors, or families. This population is usually included with other types of bone cancer with the primary site in the upper limb or hip, both of which have a different set of challenges due to location. A Canadian study utilized in-depth interviews to explore redefining and returning to work with 14 participants identified only as ‘bone tumour patients’ (ages18-38) including upper limb (Parsons et al., 2008). A UK study with 12 mothers of youth (ages 6-22) diagnosed with osteosarcoma and Ewing’s sarcoma used interviews at 6 months post diagnosis and 12-18 months later to assess their children’s participation and coping strategies in sports, school, social activities, and mobility (Earle et al., 2005).

Norwegian researcher, Lena Fauske, PhD, Oslo University Hospital, Department of Oncology, has co-authored several qualitative research studies with bone cancer survivors, including the only study found that is specific to osteosarcoma. The osteo-specific study used in-depth interviews to look at negative and positive outcomes of lower extremity treatment decisions for long-term osteosarcoma survivors (Fauske, Bondevik et al., 2015). Fauske also used interviews with survivors of primary bone cancer to explore changes in body image (Fauske et al., 2016), functional impairment and missed opportunities (Fauske, Bruland et al., 2015), and psychosocial challenges (Fauske et al., 2019). The inclusion of osteosarcoma participants in these studies was limited. Like this dissertation study, Fauske's dissertation was a qualitative study with 18 bone cancer survivors (ages 18-60), 10 of whom had osteosarocoma, to examine the changes and losses for primary bone cancer survivors (Fauske, 2016).

No qualitative studies were found with osteosarcoma patients, survivors, or family members about the experience of decision-making for surgery in the lower extremity in the US or elsewhere.

Overview and Research Questions

A decision aid is an imperative resource for surgical options for patients and families to participate in shared decision-making. Knowledge translation indicates that the key to developing a decision aid is understanding the needs of patients. To do that, we need to understand how decisions are made specific to osteosarcoma surgeries in the lower limb. A gap in the literature exists for qualitative research with osteosarcoma patients for surgical decision making. Therefore, I designed two studies to understand these needs both prospectively before surgery and retrospectively after surgery.

Chapter Three: Study 1 is a content review of posts on the public Osteosarcoma and Ewing's Sarcoma Support Group on Facebook. Study 1 reviews posts that indicate a current need for information from the members about a decision or options for surgery of the lower extremity. A total of 15 posts were analyzed for common themes and knowledge gaps to answer this research question:

RQ1: What are patients asking other patients on Facebook when making the surgical decision for osteosarcoma?

Chapter Four: Study 2 builds on the results of study 1 with an in-depth look at the lived experience of surgical decision-making in personal interviews with survivors and families. The interview guide was informed by themes found in Study 1 and by the interactive model of knowledge translation. Twenty interviews with 29 adult survivors, youth, and parents are presented in short stories or "vignettes" highlighting their needs and decision aid recommendations for the decision aid to answer these research questions:

RQ2: What is the lived experience of surgical decision-making for patients and families?

RQ3: What dissemination recommendations would survivors and families make for the development of a decision aid?

Finally, in Chapter Five: Discussion and Conclusion, the two studies will be discussed together and conclusions will be drawn for the design of the decision aid. Theoretical implications and medical education implications are presented as well as strengths, limitations, and future directions.

CHAPTER THREE

STUDY 1

Study Design

We know that a key to shared decision-making is translating knowledge from providers to patients; however, it is largely unknown what newly patients and families need to participate in this particular decision. To better understand the unmet needs of this unique population, a qualitative research design was chosen to examine Facebook posts on the Osteosarcoma and Ewing's Sarcoma Support Group page. This design utilizes recommendations for qualitative research with Facebook by taking a “passive analysis” approach, using pre-existing content with no researcher interaction in status updates, a common type of post that is disseminated to all members of the group (Franz et al., 2019). There are three Facebook groups exclusive to osteosarcoma and Ewing’s sarcoma; this study was conducted using the only public osteosarcoma Facebook group to answer this research question:

RQ1: What are patients asking other patients on Facebook when making the surgical decision for osteosarcoma?

Data Collection

To answer the research question, the search bar on the group page was used with the search words *decision*, *choice*, and *option* to filter for posts related to surgical decision-making. Those posts were then filtered again for lower extremity surgical decisions in the tibia or femur. Other types of decisions such as those for chemotherapy options, second opinions, or upper limb surgery were excluded. Posts were limited to the last two years to reflect the most current options and concerns related to this type of surgical decision. This study reviewed only the initial post from the individual or family member to the group to answer the research question.

Demographics

Parents posted the most in this data set (11/15) followed by individuals with osteosarcoma (3/15), and one partner of an adult with osteosarcoma (see Figure 2). There were five fathers and five mothers and one father/mother combined post. All of the adults who posted for themselves or for their partner were female. While the majority (9/15) were female, the father/mother postings were equally split with 5 posts from each plus one from both. For those who included an age for the patient (13/15), the range varied from 5-33. The oldest patient is a self-post; two adults did not state their ages. Data collection for this study took place from June-September 2019 and was IRB approved as exempt by Indiana University.

Figure 2

Study 1 Participants

ID# #	*Role*	*Gender*	*Age of Pt*	*Year*	*Responses*
1	Self	M	28	2019	55
2	Mother, son	F	5	2017	74
3	Father, son	M	5	2017	84
4	Mother, son	F	10	2018	42
5	Father, daughter	M	7	2017	28
6	Mother, son	F	9	AUG 2017*	48
7	Partner/spouse (f)	F	Adult	2019	47
8	Mother, daughter	F	14	2018	76
9	Mother, father, son	F, M, M	5	SEP 2019*	20
10	Father, daughter	M	13	2018	38
11	Father, son	M	14	2017	107
12	Father, son	M	8	2019	91
13	Self	F	33	2017	51
14	Mother, daughter	F	20	2017	107
15	Self	F	Adult	2018	56
				*earliest-latest	

Data Analysis

Fifteen unique posts (not duplicated by the same member) were eligible for content analysis according to the criteria from August 2017-September 2019 when data collection ended. The posts were transferred verbatim to individual Word documents, deidentified, and imported into Nvivo 12 qualitative software to facilitate organization. Content analysis was used using the defined characteristics of content analysis (Vaismoradi, 2013). This method of analysis is often used for exploratory research where little is known for simple reporting of common issues mentioned in the data (Green & Thorogood, 2004). Starting with the first five posts, overlapping themes were coded. Once those were identified, the remaining posts were coded, adding new themes as they emerged.

Results

Four main themes emerged in the fifteen posts: 1) knowledge of the condition, 2) feelings/emotions, 3) concerns, and 4) experience/expertise from the FB group. All the posts stressed some knowledge of the condition. Most posts have strong emotions associated with the surgical decision. All the posts have concerns about long-term outcomes such as risk of recurrence. Most questions have a direct ask for similar experiences or expertise from members of the group. A more detailed description precedes the results of each theme along with verbatim examples (including typos and grammatical errors) from the posts. Some posts are duplicated with there are overlapping themes.

Knowledge of the condition. All the posts indicated some knowledge of the osteosarcoma diagnosis and treatment plan. Three subthemes were identified in the posts as part of their introduction: tumor size/location, surgical options, and the decision. All of the participants had some understanding of where the tumor was located and, in some cases, the size. This information can impact the options and decision. They also understood that options were available – this is primarily the information that brings them to the group. They also understand that a decision must be made, whether that is by the surgeon, the patient, the parent, or some combination of the three.

Tumor size/location. A basic knowledge of the tumor emerged when individuals describe the reason for their post; it often sets the background for the question. Tumor size or location is mentioned in nearly all of the posts (12/15). The posts might be succinct, such as "a tumor in his knee" (4) or "my tumor is very close to my ankle joint" (13) or they might be more detailed, "localized OS in her proximal Tibia, she does have tumor that protruded through her bone and into the muscle" (8). Some posts give more detailed information, "It's located in his right distal femur and goes into his growth plate" (12) or indicate the staging of the tumor, "osteosarcoma in the left distal femur stage 2 b" (6). Other posts directly connect the location of the tumor to the options, "We already knew the options and the one that would likely be recommended for our son, based on his young age and the size of his tumor" (2).

Surgical options. In addition to information about the tumor, all fifteen posts clearly state that the person posting understands there are surgical options. Some posts about the surgical decision are very broad, "Do we do limb salvage, rotationplasty or just amputate?" (11) while others narrow down their options, "Our two options are LSS or amputation" (5).

Some parents know there are options but aren't quite sure what they are, "So many whys I don't want the surgery done until I feel that every option has been looked at" (6). Similarly, this adult wants to know all her options even though her medical team has made a recommendation, "My oncologist and surgeon… both feel that amputation above the knee will be my best option. We are currently seeking another consultation to survey all options" (15).

The role of the individual varies as well. Adults might be looking at options for themselves, "(I'm) faced with making surgery decisions. I'm considering rotationplasy" (9). Most of the questions come from parents. Some describe letting their child make the decision, "our 13 years old hero is probably going for LSS and hopefully not amputation" (10) while others are making the decision with their child, “The first option we have decided not to go with and (son) agrees…That option is limb Salvage surgery….The second option is amputation…The last option is rotationplasty” (8). Some parents question the strong opinions of their medical team, “The Drs have been pretty forcefully promoting limb-sparing surgery. But we have been reading that's not always the best option” (4).

The decision. All of the individuals who posted understood that a decision could be made and/or mentioned who that decision maker would be. The adults who posted stated that they or their partner were in the role of the decision-making according to the surgeon. This 28-year old man states that he is making the decision for himself, “The surgeon gave me 2 options” (1). This post states that her partner was given options and making choices, “He’s been given two options he can choose for his surgery” (7).

Parents often state that they are the surrogate decision makers for the child and struggle with the difficulties of making a surgical decision for someone else. Here a mother/father post talks about making that choice for their 5-year-old son, “I don't want to make the wrong choice for him” (9). Another mother wrestles with the long-term ramifications of being the decision maker for her son: “How do you make this life changing choice for a 5-year-old? What if you make the wrong choice?” (2). Similarly a father describes how he and his wife feel about being decision makers, “I know that this is the right decision for him, but just the fact that this is a decision we have to make at all is messed up” (3).

Some parents feel they are making the decision with their child. This father describes the decision as being made by the parents with their 8-year-old, “(Son) and us still have a week to make the final decision” (12). Another father makes the decision with his 13 year old son:

> I understand of course that if amputation is the only life saver then we will go with it but I need to understand if we have the opportunity to choose is it the norm that LSS comes with much trouble later? (10)

This post states that while the surgeon is adamant about LSS, the parents and their 14 year old daughter still feel they have choices and are making the decision:

> He is very very adamant that he does not want to amputate, however, my daughter is more concerned with reoccurrence and Mets with the LSS compared to amputation. I get this feeling that if We choose to keep the leg I am choosing quality of life over her actual life she feels the same way. The surgeon feels that saving the limb is what’s best for her and has no problem telling me that I am the one that has to make this choice as my daughter is “only 14” and doesn’t have the ability to fully make this choice. So I’m asking what helped you guys make the choice? (8)

Contrary to the position of that surgeon, this father feels that the decision ultimately belongs to his 14-year old:

> When I try to talk to my son about this he shuts me down and doesn't what to talk about it. This is very frustrating because this decision needs made soon. I go back and forth between limb salvage and rotationplasty, but ultimately this decision belongs to (him). (11)

This mother of a 20 year old daughter appears to be making the decision for her as a surrogate decision maker although her daughter is an adult:

> I don't know if I want my daughter to get LSS surgery or amputation… I wish I was 100% sure that after my daughter's LSS surgery, OS will not come back in the same area. Putting my daughter through another surgery to amputate after LSS is heartbreaking if I had the choice of amputation initially. (14)

Some posts do not clearly state who the decision maker is but describe options or decisions to be made. Other posts imply the decision could be made by them despite strong medical opinion for one surgical procedure over the others. Here the parents are reading that the option preferred by their doctors isn't always the best and they want to better informed about options to ask their surgeon:

> Second, we are talking with his surgeon tomorrow. The Drs have been pretty forcefully promoting limb-sparing surgery. But we have been reading that's not always the best option. What would be some good questions to ask our surgeon about lss vs. amputation? (4)

One post indicates that the surgeon was the decision maker. Here the mother talks about wanting a bigger role in making the decision even though the decision appears to have already been made by the surgeon:

> Everything has just happened so quick he has an amputation surgery on the 12 of September. The orthopedic surgeon hasn't saw him since she did the biopsy I have questions I need answers to the hem/onc doctors can't give me the answers I want. The questions I have is why can't he get LSS? Why is his only option amputation? Why can't a cadaver bone be used? So many whys I don't want the surgery done until I feel that every option has been looked at. Am I being rediculos over this? (6)

Feelings/Emotions. Not surprisingly, feelings and emotions emerged as a frequent theme in the posts. Feelings are the mental reaction to an emotion, such as confusion; emotions are physical and instinctive response to a feeling such as scared. Both can be prefaced by "I feel…" and therefore are include together as people post how the decision-making process affects them. While some are specific to the surgery, others are overall descriptors of the heightened emotions during this early period of diagnosis prior to surgery. Emotions range from fear and worry to confusion and feeling overwhelmed.

Three of the 15 posts do not include emotional content; they are framed primarily as information seeking queries. This post is asking for input, "Any and all input would be greatly appreciated!" (1). Another asks readers to private message them with information, "Looking for info if anyone wants to PM me please I don't want to make the wrong choice for him" (9). And the third one is asking for more general information: "If anyone has helpful resources or can share any similar experiences, your input would be greatly appreciated" (15).

Twelve posts in sum have either parental or self-reports of emotions or relate the emotional state of the child or partner. Parents frequently describe their emotions as being afraid or fearful. This father describes how he feels in spite of doing everything he can to make the right surgical decision for his son, "I'm struggling today. I'm sitting in my office off and on crying. Pretty much every time I think about my son, what he's been through already, and what's coming" (3).

A mother describes feeling both worried and scared about the long-term outcomes of the surgical options:

> She is done with only three treatments. Her overall health condition is excellent but I still worry. I don't know if I want my daughter to get LSS surgery or amputation. I know LSS will give her better quality of life but having the possibility of it coming back in the same area with a risk of it spreading to the lungs is scary. (14)

Some parents report feeling confused in their posts. This parent describes her emotional state as being lost, "This post is going to be long, but I'm so lost at the moment" (8).

Here a father feels both worried and confused about long-term outcomes relating to surgery:

> I have been seeing a lot of LSS related issues in the group which makes me worried since our 13 years old hero is probably going for LSS and hopefully not amputation …I was under the impression that LSS is obviously a less painful solution on the long term .. now I am confused. (10)

Some parents describe emotional states around the surgery itself. This parent is very specific about her concerns as they relate over time to recurrence, "I'm very nervous about cutting into tumor" (5). Another mother describes her emotions when thinking that a decision to save the leg she makes might still result in amputation later as heartbreaking, "Putting my daughter through another surgery to amputate after LSS is heartbreaking if I had the choice of amputation initially" (14).

This adult with osteosarcoma feels overwhelmed with making the decision:

> I had another consultation with my surgeon today and am facing an impossible decision… If anyone had to make this decision how did you do it? I feel so overwhelmed, I almost wish I didn't have options. I hate making big decisions and this is HUGE! (13)

A father sums up the daily emotions around early diagnosis and decision-making, "This sucks. I hate it. Every single day" (3).

Parents also describe their child’s emotions during this decision-making period. One mother describes her autistic son’s emotional state as ranging from cheerful to irate and finds it difficult to communicate the complexities of treatment with him. Here she describes his emotional state and asks the group for their personal experiences and expertise in handling it:

> First, my son is on the autism spectrum (high functioning), which adds a whole new layer of difficult to everything. He was really cheerful going into the hospital this time, but today (after receiving methotrexate last night), he was irate he has to stay in the hospital, screaming and crying. He knows he's usually in for five days with methotrexate, but today it was like he forgot. He's insisting he should go home tomorrow, and gets stuck on a loop asking when he can go home, over and over. I feel like I'm not talking to the real him when he's upset like that. Do you think any of his being upset or stuck could have to do with methotrexate side effects? Have any of you experienced mood/behavioral changes to in your children? If so, what helps? (4)

Another father describes how his son felt on the day he and his wife told him why they felt amputation would be best for him: “Well yesterday was a pretty hard day for all of us… He was very upset but took it well” (12).

Concerns. Concerns are the pulse of the posts and primarily center around quality of life long-term outcomes. All the posts analyzed are concerned with getting the information they need to feel educated; their specific concerns include reoccurrence, chronic pain, additional surgeries, and mobility issues that would impact their daily living. The presence of uncertain outcomes and the relative absence of certain outcomes make this decision multi-layered and complex; therefore it was not possible to separate these concerns out as subthemes.

Concerns for making a life-changing decision weeks after diagnosis is complicated by complications of early treatment. For example this father is concerned about making an informed decision while navigating unexpected side effects of chemotherapy:

> We are trying to make an educated decision while moving a million miles an hour. He has missed much of the first three weeks of school. His chemo started Sept 6 and he is having a very rough time with nausea and vomiting. He's having trouble going number 2 and also seems to be suffering from migraines, which was not a side effect we were aware of. He has been taking all the prescribed drugs for nausea and for going number 2 but these don't seem to be helping. And if all that wasn't enough we are trying to make the best decision regarding what to do with his knee/leg. (11)

Sometimes people weigh form over function concerns with quality of life activities such as running and jumping. An adult who is making the decision for herself is concerned with balancing the pros and cons of LSS and amptutation– one in which she saves the leg but loses function or loses the leg but gains function:

> I had another consultation with my surgeon today and am facing an impossible decision. My tumor is very close to my ankle joint and my surgeon told me that honestly in my case with LSS I will not be able to regain full functionality and

> will not be able to run or jump again. My other option is a below the knee amputation that would be much more functional, but obviously involves losing your leg. If anyone had to make this decision how did you do it? (12)

Some posts list specific activities as being quality of life factors to their decision-making process but not having enough information to guide them in the decision. This man is concerned about his long-term outcomes for LSS and more high level activity: “I'm a very active person (run, hunt, gym, kayak, backpack, stand 12 hours a day for work, etc.) and I've heard LSS does not hold up very well to certain and/or vigorous exercise” (1).

This post relays concerns from a partner about the differences between LSS and rotationplasty – one in which his mobility might be compromised while the other will afford more functionality, “He’s looking for the pros and cons of each procedure as he's in two minds now of which one to have done. He'll apparently be a lot more mobile with the rotationplasty but it wouldn't have been his first choice” (7).

Parents have quality of life concerns about the likelihood of uncertain outcomes. The mother of a 5 year old son wonders aloud how much of a chance she is willing to take to save her son’s leg against not knowing the risks of pain and more surgeries:

> What if there’s a way to save the leg and you don’t take that chance? What if you decide to try, and it only causes pain and a life of additional surgeries? My husband and will do whatever gives him the best quality of life, but just need to hear from everyone. (2)

One family is concerned about the uncertain LSS long term outcomes of limitations and additional surgeries in addition to reoccurrence, “Surgeon is looking so far for an internal prosthetic hip replacement and partial femur but will have multiple future surgeries and limited to activities and no guarantee the cancer won't come back” (9).

This father reviews all of the options relative to the uncertain risks. His concerns are his son’s long-term quality of life, mobility for an active lifestyle, need for additional surgeries, appearance, and energy exertion required for everyday living:

> [LSS]: Too many risks involved and he wouldn't be able to have an active lifestyle, that currently at 8 years old he loves doing. Running, jumping, riding atvs, just being a boy.
>
> [Amputation]: He is young he will adapt and only need one surgery.
>
> [Rotationplasty]: The only thing is his foot would face backwards... The bonus to this is if you get this done, it takes alot less energy in a person to walk and they get tired less. (12)

The risk of recurrence as an uncertain outcome in a concern for parents. This mother wishes there was a statistical certainty of knowing which option had the least risk for recurrence, “I wish I was 100% sure that after my daughter's LSS surgery, OS will not come back in the same area" (14). Other parents also list the risks of LSS: "[He] will have multiple future surgeries and limited to activities and no guarantee the cancer won't come back" (9).

The father of a 13-year-old daughter has concerns about recurrence, wondering if amputation is the only way to save his daughter's life. He compares the benefit of LSS (less pain) to the potential long-term issues later:

> I have been seeing a lot of LSS related issues in the group which makes me worried since our 13 years old hero is probably going for LSS and hopefully not amputation. I was under the impression that LSS is obviously a less painful solution on the long term .. now I am confused .. I understand of course that if amputation is the only life saver then we will go with it but I need to understand if we have the opportunity to choose is it the norm that LSS comes with much trouble later? (10)

Another father lists very specific LSS concerns (long rehabilitation, growth plate removal with significant leg length differences) against amputation concerns (psychological challenges) under the overriding concern of recorrence (getting clear margins and risks of cutting into tumor):

> Our two options are LSS or amputation. Her age and tumor location on tibia makes LSS challenging. We are talking about millimeters of clear margins, a year of rehabilitation, removal of growth plate, lose of 4 inches possibly, growth plate in other leg removed after chemo to make legs even. Amputation although difficult would be less challenging however psychologically more challenging. Little of leg bone would be left for LSS/allograft. It's still possible to have two good limbs but shorter in overall height. I'm very nervous about cutting into tumor. We've gone down pros and cons. (5)

The father of a 5 year old son expresses his concerns about the decision for his son's upcoming rotationplasty and rehabilitation as well as the success of the overall treatment, "Then there's the constant fear that none of this treatment is going to work, and he will die. Even after everything we are putting his little body through" (3).

Concerns arise when the surgeon promotes one surgery strongly over another, prompting people to wonder about other options. Some individuals are concerned that the doctor's strong position of one surgery might not be the best option. This mother expresses these concerns after reading about other options, "The Drs have been pretty forcefully promoting limb-sparing surgery. But we have been reading that's not always the best option" (4).

One adult patient is also concerned that the recommendations of both her oncologist and surgeon might not be the best or only option for her, "After speaking with my oncologist and surgeon, they both feel that amputation above the knee will be my best option. We are currently seeking other consultation to survey all options" (15).

This mother reports that her 14 year old daughter is most concerned with reoccurrence, "He is very very adamant that he does not want to amputate, however, my daughter is more concerned with reoccurrence and Mets with the LSS compared to amputation" (8).

The same mother shares her daughter's concerns but is also concerned she herself is over reacting:

> I get this feeling that if We choose to keep the leg I am choosing quality of life over her actual life she feels the same way… Am I just over reacting with the LSS because I want the cancer to be gone? (8)

This mother is also concerned that she is over-reacting and wonders if she's being ridiculous, "So many whys I don't want the surgery done until I feel that every option has been looked at. Am I being rediculos over this?" (6). Another mother struggles with concerns over the risk of reoccurrence concerns as well as potential decisional regret if she chooses LSS and later needs an amputation:

> I realized the LSS would give her better quality of life but amputation will eliminate any chance of reoccurrence in the same area. As a parent I don't know what is the better option…Taking a chance with LSS and hoping it won't come back in the same area; which can spread to the lungs… Putting my daughter through another surgery to amputate after LSS is heartbreaking if I had the choice of amputation initially. (14)

Experience/Expertise. Parents and individuals alike post to the Facebook group because they are looking for someone with a similar experience, such as the location of a tumor on a very young child. They also more broadly seek the group's expertise when making the decision such as what questions they should be asking the surgeon or potential complications associated with the options.

Thirteen posts are direct asks for information, framing their posts around seeking personal experiences and/or expert advice. The other two queries indirectly value the group as a passive expert audience for their post. The theme of experience/expertise further branched into four subthemes for information seeking: long-term outcomes, location impacts decision, managing provider expertise, and right or wrong decisions.

Long-term outcomes. Long-term outcomes include personal experiences or expertise from the group. This man is making the decision for himself and focuses on specific long-term outcomes. He seeks personal experiences to help him make the surgical decision when weighing the benefits of LSS against potential complications:

> Hi all! Im 28 y/o and was diagnosed December 2018 with osteosarcoma in my proximal tibia including knee joint. The surgeon gave me 2 options: limb salvage or above knee amputation...I'm a very active person (run, hunt, gym, kayak, backpack, stand 12 hours a day for work, etc.) and I've heard LSS does not hold up very well to certain and/or vigorous exercise. What experience has everyone

> had with LSS and regular to vigorous exercise? Has anyone had any long or short-term complications? Has anyone opted for amputation right off the bat to avoid further surgeries? (1)

Other questions more broadly request expertise in long-term outcomes such as this father who requests information on long-term issues associated with LSS:

> I have been seeing a lot of LSS related issues in the group which makes me worried since our 13 years old hero is probably going for LSS…I understand of course that if amputation is the only life saver then we will go with it but I need to understand if we have the opportunity to choose is it the norm that LSS comes with much trouble later? (10)

Posts reflect feeling conflicted about choosing one surgical procedure over another and not understanding the pros and cons of each option. This father asks for specific expertise from the group on the risks and beneifts of LSS and rotationplasty, “Do we do limb salvage, rotationplasty or just amputate? I was hoping that this group could provide some pros and cons between limb salvage and rotationplasty?” The partner of an individual making the decision also seeks expertise as well as support (help) in learning the pros and cons of LSS and rotationplasty:

> This is my first post on this group and I'm looking for some advice and information if any of you lovely people can help. I'm posting to see if anyone has had either option done and looking for the pros and cons of each procedure as he's in two minds now of which one to have done. He'll apparently be a lot more mobile with the rotationplasty but it wouldn't have been his first choice. Any help or information would be greatly appreciated. (7)

Other queries address the direct value of the experience/expertise of the group such as this post from a mother who also seeks both information and support:

> What are the struggles you have gone through whether it’s been with amputation or LSS? Thanks so much for the understanding and support. It’s hard to talk to other parents/families that haven’t faced this choice and haven’t dealt with OS. (8)

Some questions are more existential in nature, probing deeper with more theoretical questions to the group that can’t really be answered. This mother poses these questions to the group:

> How do you make this life changing choice for a 5 year old? What if you make the wrong choice? What if there’s a way to save the leg and you don’t take that chance? What if you decide to try, and it only causes pain and a life of additional surgeries? (2)

Location impacts decision. While the location of the tumor and staging dictate which surgical options are clinically possible, some queries name the location of the tumor as impacting their role in making the decision. Posts that pinpoint the osteosarcoma location as a problematic factor sought expertise from families who those who had similar experiences, such as these parents for their son, “I'm considering rotationplasty but has anyone else had it where the tumor is up at the top?” (9).

A father posts that the location of his daughter’s tumor impacts the decision and weighs the psychological adjustment of amputation against limb differences associated with LSS as he seeks someone who has made a similar decision:

> Amputation although difficult would be less challenging however psychologically more challenging. Little of leg bone would be left for LSS/allograft. It’s still possible to have two good limbs but shorter in overall height. I’m very nervous about cutting into tumor. We’ve gone down pros and cons. Looking for some feedback on any similar situation. (5)

Managing provider expertise. While most posts come to the group to seek their expertise, some name provider communication as a failure to provide information needed to make an informed decision. Parents in these posts state explicitly that they are seeking expertise online because they aren’t getting what they need from their providers and express frustration, such as this mother who can’t get answers to her questions:

> Idk why but tonight my mind is running a mile a minute and I guess I need reassurance I figured I might be able to get that here…The orthopedic surgeon hasn't saw him since she did the biopsy I have questions I need answers to the hem/onc doctors can't give me the answers I want. The questions I have is why can't he get LSS? Why is his only option amputation? Why can't a cadaver bone be used? So many whys I don't want the surgery done until I feel that every option has been looked at. Am I being rediculos over this? (6)

Sometimes parents struggle with surgeons insisting on only one type of surgery when they know other options are available. This mother wrestles with the surgeon’s strong position that he does not want to amputate in spite of his own failed LSS and subsequent amputation after OS diagnosis. She also expresses recurrence concerns for her daughter if they decide to save the limb. Here she seeks experience in clarifying values when making the choice when framed against the surgeon’s surgical preference:

> We were given the options of amputation and LSS. Her surgeon is an OS survivor, he had LSS then later after many surgeries ended with amputation. He is very very adamant that he does not want to amputate, however, my daughter is more concerned with reoccurrence and Mets with the LSS compared to amputation. I get this feeling that if We choose to keep the leg I am choosing quality of life over her actual life she feels the same way. The surgeon feels that saving the limb is what’s best for her and has no problem telling me that I am the one that has to make this choice as my daughter is “only 14” and doesn’t have the ability to fully make this choice. So I’m asking what helped you guys make the choice? Am I just over-reacting with the LSS because I want the cancer to be gone? (8)

This mother also struggles with the surgeon’s strong position toward limb salvage surgery and also asks directly for questions to ask the surgeon about LSS and amputation:

> We are talking with his surgeon tomorrow. The Drs have been pretty forcefully promoting limb-sparing surgery. But we have been reading that's not always the best option. What would be some good questions to ask our surgeon about lss vs. amputation? (4)

Right or wrong decisions. Posts for expertise raise concerns about making the right or wrong decision for the surgical option. Some families frame the conflict as wanting to make the best decision for their child such as this dad who posts, “I know that this is the right decision for him, but just the fact that this is a decision we have to make at all is messed up” (3).

Others struggle with making the right or wrong decision such as this father who prospectively doesn’t want to pick the wrong option, “Looking for info if anyone wants to PM me please I don't want to make the wrong choice for him” (9).

Parents also think about the dichotomy of the decision retrospectively, as this mother does since she can’t know the choice was wrong until after it’s been made, “What if you make the wrong choice?” (2).

Parents also confront the diametrically opposed good/bad labels associated with decision-making such as this mother who struggles with knowing there is no right or wrong choice for her daughter, “There is really no right or wrong. And I get that” (8).

The two posts that indirectly value the expertise of the group are two fathers who discuss making the surgical decision for their sons. This father’s post shares a personal experience with the group and draws on a common metaphor of illness as a battle, with the implication that the group would understand, “I told him he has to think of this as he is now a "Warrior" and when you go to battle as our Military does, most times you come back with some battle scars from battles you fight!” (12). Another father also indirectly values the group’s experience and expertise more as understanding as he vents his frustration and heightened emotional state:

> I don't really know which cancer group I should post this in, but I'll just drop it here. I'm struggling today. I'm sitting in my office off and on crying. Pretty much every time I think about my son, what he's been through already, and what's coming... I don't really know which cancer group I should post this in, but I'll just drop it here. (3)

Summary

The results of Study 1 will be discussed together with Study 2 in Chapter Five: Discussion and Conclusions; this summary provides a brief overview of the findings.

The answer to RQ1: What are patients asking other patients on Facebook when making the surgical decision for osteosarcoma? is complex. Armed with a plethora of knowledge from the tumor size and location, families understand that options are available, whether they’ve been offered them or not. They also realize that someone must make the surgical decision, whether that’s the surgeon, the parent, the patient or some combination of those.

Patients and families are under an incredible amount of stress and express emotions that reflect their shock and confusion. Their concerns around making the decision are also multi-faceted and include risks of recurrence as well as the potential for chronic pain, additional surgeries, and mobility issues.

Patients and families describe knowledge gaps and unmet needs; they seek the expertise and experience from the Facebook members to answer questions about what to expect with each option. They seek other people for whom the location of the tumor impacted the decision. They seek advice on how to manage the recommendations of providers and how to choose the right option for themselves or their child.

The existing literature and knowledge translation indicate that what we need to develop useful decision aids is an understanding of what people need when they are making this decision. Study 1 answered the research question, but additional research is needed to identify knowledge gaps and unmet needs so future patients will have access to that information. In-depth interviews with survivors and families after the surgical procedure has been made will provide insight into the lived experience as they look back on the period of decision-making as well as unforeseen knowledge they did not know they needed at the time. Their experiences will drive the content of the decision aid.

Strengths

A strength of this study is the first-hand analysis of the unmet needs for decision-making from a newly diagnosed population of a rare cancer. As a retrospective study of existing posts, the users are largely unfiltered and lead to an uncensored opportunity to understand their concerns around making the decision for surgery.

Limitations

This study is limited to individuals who are internet savvy with the navigation skills to find support groups online, use Facebook, and who are comfortable posting personal health information on a public page. This content analysis only reviewed the initial user-generated post; a limitation of this study is only capturing one side of the interaction between the user and the other members of the group. A further limitation is the use of only one coder, eliminating the opportunity for deliberation and consensus.

While only the initial post was reviewed in the Facebook study, there were significant responses to each post. Of the 15 posts reviewed, there were 924 responses – two questions had 107 responses each. There is potentially rich data in these responses, including additional questions that arise either from the original poster or between people who are responding. Future research is recommended using the interactive model of knowledge translation for thematic analysis in the responses to the posts. A recommended research question for this secondary study is - What are patients telling other patients on Facebook when making the surgical decision for osteosarcoma?

CHAPTER FOUR

STUDY 2

Study 1 gives essential information about the kinds of information patients and families are seeking when making the decision. However, to build a useful decision aid, more specific information to provide the content needed for future families. More research is needed to expand on the broader knowledge gaps identified in Study 1 as well as recommendations survivors would include for the decision aid itself. The lived experience of surgical decision-making for osteosarcoma patients and families is largely unknown; no patient-reported outcomes have been published to help guide the development of this tool.

Study Design

A qualitative research design was chosen with in-depth semi-structured interviews to explore the unmet needs of osteosarcoma survivors and parents at the time of the surgery and upon reflection, after surgery. Interviews also allow the opportunity to ask what would have been helpful to them when the options were either being discussed or when they became aware of other options for surgery. Study 2 was conducted to answer these research questions:

RQ2: What is the lived experience of surgical decision-making for patients and families?

RQ3: What dissemination recommendations would patients and families make for the development of a decision aid to help future families?

Knowledge translation. Interviews have been noted to be useful for research in knowledge translation to increase familiarity with user groups. Knowledge translation (KT) was chosen as the theoretical framework for this study. The interactive model of knowledge translation played a key role in the design of Study 2.

The needs for KT – the timely translation of research into clinical practice has been recognized for over a century (Grimshaw et al., 2012). During that time, more than100 terms have been used to describe the construct, including “knowledge implementation,” “knowledge transfer,” “quality improvement,” and “knowledge dissemination” (Mckibbon et al., 2010). KT evolved over time and was formally established by the Canadian Institutes of Health Research (CIHR) in 2000 with this definition (Government of Canada, 2020):

> Knowledge translation is defined as a dynamic and iterative process that includes synthesis, dissemination, exchange and ethically-sound application of knowledge to improve the health of Canadians, provide more effective health services and products and strengthen the health care system.

Additionally, there are two types of KT recognized by the CIHR: (1) the “end of grant KT” in which study results are tailored for dissemination for stakeholders and (2) “integrated KT” in which stakeholders engage from the onset of a study in collaborative research with providers, other patients, and even policy makers (Azimi et al., 2015). Integrated KT (IKT) is sometimes referred to as participatory research and promotes the movement of patients as passive recipients to active partners in the conceptualization of problems to implementation (Gagliardi et al., 2017).

KT is now embraced widely internationally and within the United States. Within the last 20 years, KT has been adopted by organizations such as the National Institute on Disability, Independent Living, and Rehabilitation Research (NIDILRR) and the World Health Organization (WHO). The definition that is most applicable to this research study is from the American Institutes for Research (AIR) which also includes their applied research goals (American Institutes of Research, n.d.):

> Effective knowledge translation requires expertise that begins with refining the questions that specific audiences need answered and finding relevant research studies that answer those questions. The process then moves to translating evidence into user-friendly products customized for specific audiences and providing technical support to apply the information to make individual, practitioner, and systems change. (para 1)

Furthermore, AIR's objectives begin with formative research such as the two studies with osteosarcoma community in this dissertation:

> AIR begins this process with stakeholder engagement (providers and consumers), using formative research to learn about the questions they want answered and the best way to engage, inform, and reach users. AIR then conducts systematic literature reviews and meta-analyses to find relevant research studies and other literature that answer their questions. (American Institutes of Research, n.d., para 2)

The development plan for the osteosarcoma decision aid project mirrors AIR's implementation strategies, "From a synthesis of the findings, our knowledge transfer and communication experts create online and print products including social media that communicate the results and disseminate them through traditional, web, and social media channels to specific audiences" (American Institutes of Research, n.d., para 3).

There are many different models of KT to choose from, each with different approaches: the classic, knowledge-driven model, the problem-solving, policy-driven model, the interactive model, the enlightenment model, the political model, the tactical model, the dialogical model, the coordinated implementation model, the knowledge-to-action model and the Ottawa Model of Research Use (Armstrong et al., 2006; Graham et al., 2006; Sudsawad, 2007). Of these two are good options for this project. The knowledge-driven model for research dissemination follows a pattern of basic research > applied research > development > application and is based on the premise that the very existence of knowledge moves it forward to development and use (Weiss, 1979). However, the knowledge-driven model is linear and assumes a steady progression from formative research with stakeholders to dissemination. The results of the study #1 on Facebook demonstrate non-linear knowledge-seeking as expert patients use a variety of resources to get the information they need to feel informed. Therefore, the interactive model of knowledge translation is the best framework for Study 2.

Interactive model of knowledge translation and the interview guide. The interactive model of KT uses an approach that moves from general information about the decision-making experience to the specific information needed for dissemination. It posits that knowledge translation does not follow a straight path and is part of broader and more complex journey to knowledge that involves multiple inputs (Estabrooks et al., 2006).

The interactive model is designed around five domains: user domain, issue domain, research domain, researcher-user relationship domain, and dissemination domain. The theorists have suggested questions to go with each domain to discover information about the intended users of evidence, in this case patients and families, and to identify missing information (Jacobson et al., 2003).

Each domain is described below according to the framework along with its application in designing this study as a model for the osteosarcoma lived experience. Included are sample questions suggested in the model and an adaptation of the questions for the interview guide (Jacobson et al., 2003).

User domain. The user group domain focuses on understanding several aspects of the user group by asking questions about access to information sources and expectations towards research and researchers. For the purposes of this research study, the survivors and family members are the user group and anyone who has information that the user group does not have is in the role of the researcher. This might include surgeons and oncologists but also can include expert patients or friends who have osteosarcoma expertise.

Questions. Samples of questions for this domain include:

What kinds of decisions does the user group make?
What is the user group’s criteria to make decisions?
What actions are available to the user group?
What is the user’s expectations of the researcher?
What is the user’s expectations of the knowledge translation process?

Adaptations of the sample questions above for osteosarcoma survivors and families are:

What surgical decisions were you making?
What was most important/least important to you when making this decision?
What role do you expect to play regarding the decision?
What are your expectations of the surgeon?
What did you expect to learn from your surgeon or other resources?

Issue domain. The issue domain is what is unknown and necessary for the user and is about the context in which they become part of a user group domain; the issue can be new or recurrent, certain or uncertain (Jacobson et al., 2007). Another way to think about the issue domain is that it "focuses on the characteristics and context of the issue intended to be resolved through the knowledge translation effort" (Sudsawad, 2007, p. 5). For the personal interviews, the issue domain includes concerns expressed by the participants when making the decision, such as understanding risks, benefits, and uncertainties for long-term outcomes. These concerns may be retrospective or current, occurring at the time of the interview or many years after the decision was made.

***Questions*.** Samples of questions suggested for this domain include:

Are things changing with the issue?
How much uncertainty surrounds the issue?
How much conflict surrounds the issue?
What risks are associated with the issue?
Is it necessary to have particular expertise in order to understand this issue?

Adaptations of the sample questions above for osteosarcoma survivors and families are:

Did things change with the decision-making?
How much uncertainty surrounds the decision?
What conflict did you experience when making the decision?
What risks are associated with the decision?
Do you feel like you had the expertise necessary order to understand the decision?

Research domain. The research domain centers on the research characteristics such as how relevant it is, if it is consistent with other research, and if it applies to the user's situation (Sudsawad, 2007). The developers (Jacobson et al, 2003) describe the role of the research domain for users as one that is "broad, provides answers, and is action-oriented," understanding that users will evaluate and use it in different ways. For the purposes of this study, research is defined as both evidence-based research studies as well as patient-reported outcomes, such as what patients and families share in the Facebook support group. Study 1 indicated that newly diagnosed patients and families may find that sorting information from online research and social media can be overwhelming; therefore, the interactive model of KT posits that they will focus on what is the most credible and how it relates to their unique situation.

Questions. Samples of questions suggested for this domain include:

What research is available?
What is the quality of the research?
Is the research unambiguous?
Is the research consistent?
What is the source of the research?

Adaptations of the sample questions above for osteosarcoma survivors and families are:

What research was available to you when you were making the decision?
How did you feel about the quality of the information you received?
Were the resources you used clear and easy to understand?
Was the information for options consistent from different sources?
What is the source of the (amputation/limb salvage/rotationplasty) research?

The elicitation of patient values is a pivotal component of shared decision-making and is included in the interactive model. These sample questions from the interactive model suggest ways to elicit patient values related to quality of life:

How relevant is the research to the user group?
Does the research have implications that are incompatible with existing user group expectations or priorities?
Would these implications disrupt the user group's status quo?
Do these implications suggest changes that the user group believes are politically feasible?

Adaptations of the sample questions above for osteosarcoma survivors and families are:

How relevant did the research feel to you or your child?
In what ways were the options in your research incompatible with your expectations or priorities?
Would those incompatibilities have disrupted your (your child’s) quality of life?
Are the implications for quality of life aligned with what you believe the surgeon can do? (Not all surgeons are trained in rotationplasty, for example).

Researcher-user relationship domain. The relational language around the patient-provider relationship itself reflects the researcher-user relationship. In this study, the researcher is defined as anyone who has knowledge of osteosarcoma surgical decisions – that might be the orthopedic surgeon or the oncologist but can also include friends, colleagues, or family members or expert patients found on social media. According to the theory developers, relationship is a key concept in the interactive model of knowledge translation; the relationship can enhance or inhibit the impact of information sharing. The timing of the relationship is also important as encounters that take place early and often can positively impact knowledge translation. For a new diagnosis, frequent face to face appointments that take place regularly can build a trusting relationship.

The interactive model embraces the reflexive nature of the researcher as part of the research; therefore users (research participants) and researchers (interviewer) have a relationship because they understand each other’s needs and circumstances. This is particularly relevant in this study because of my reflexive role, as described in Chapter 1.

Questions. This set of sample questions considers the relationship between informants and the user:

How much trust and rapport exist between the researcher and the user?
Do the researcher and the user have a history of working together?

How frequently will the researcher have contact with the user?
Have the researcher and the user agreed about the desired outcomes of knowledge translation?
Have the researcher and the user agreed about the responsibilities each will have during knowledge translation?

Adaptations of the sample questions above for osteosarcoma survivors and families are:

How would you describe the trust and/or communication between you and the medical team?
Did you know any of your medical team prior to diagnosis?
How often did you meet with that person? Did you meet in person or talk by email or phone?
Did you and the surgeon agree on what the goals were when making the decision?
Did you and the surgeon agree about who would be making the final decision?

Dissemination domain. This domain focuses on practical suggestions and strategies for disseminating knowledge.

Questions. Sample questions for the dissemination strategies domain are:

What is the most appropriate mode of interaction: written or oral, formal or informal?
What level of detail will the user group want to see?
How much information can the user group assimilate per interaction?
Should the researcher pretest or invite feedback on the selected format from representatives of the user group before finalizing presentation plans?

Adaptations of the sample questions above for osteosarcoma survivors and families are:

What would be the most appropriate way to receive the information, written or oral, formal or informal? (follow up with video, audio, interactive module, etc)
How much detail would you have wanted to see? (surgical details, outcomes, etc)
How much information do you think you could absorb at the time each time you used it?
Should we test a prototype with the osteosarcoma community before implementing it in practice?

All questions were formatted in a sequence of domains for the interview guide (see Appendix A).

Recruitment

After receiving expedited approval from the IRB, Study 2 recruited participants from the same Osteosarcoma and Ewing's Sarcoma Support Facebook group as Study 1. A “research self-identification” approach was used to identify myself both as a researcher and as a member of the community as a reflexive disclosure recommendation for qualitative research (Franz et al., 2019). After the study received expedited approval from the IRB, participants were recruited from the Osteosarcoma and Ewing's Sarcoma Support group for osteosarcoma as Study 1.

The recruitment notice (see Appendix A for recruitment notice) was posted five times between June-September 2019. Recruitment included snowball sampling, allowing group members to share the recruitment notice elsewhere. Interested participants emailed or texted to set a time for a phone screening to confirm eligibility, explain the goals of the study, and answer any questions (see Appendix B for screening interview guide). Selection was purposive, with participants selected to represent a broad range of geographical areas, genders, ages, levels of satisfaction, and surgical decisions. If selected, participants received an invitation to participate and schedule a time at which the in-person or video interview would take place.

Data Collection

Data collection took place at a location agreed upon by the interviewer and participant. Criteria for inclusion in the study included the ability and willingness to either meet in person or via Zoom for a video interview. Face-to-face interaction for relationship-building is part of the interactive model of knowledge translation (Jacobson et al., 2003); it is a logical extension of the framework to extend this requirement to the interviews. Eight interviews took place in hospitals (3) and participants' homes (5); 12 interviews used Zoom video. Participants were emailed a $25 Amazon gift card at the conclusion of their interview as a thank-you for their time.

Interview procedures. The semi-structured interview guide (see Appendix C) was used as intended for the first three interviews. The first question is open and broad, asking participants to share their story. Participants were aware that the purpose of the story was their experience at the time of surgical decision-making; they were prepared to talk about that at length, often starting with their lives just prior to the diagnosis. They shared concerns about the decision, how they felt about their doctor, and researching for more information or talking to providers about options. Therefore, most if not all the questions were addressed in the telling of their story during this decision-making period, making the questions repetitive and thus affecting the tone of the interview. After the third interview, it made sense to move to a narrative interview approach, often used to better understand illness experiences. Narrative interviews represent people's lives and let the participant control the direction, content, and pace of the interview (Anderson & Kirkpatrick, 2015). Participants were informed that an interview guide might be used but most questions were being answered organically as they told their story. This shift also allowed the participants to share the experience in their own words, thus sharing power in the interview to create a space that fosters reciprocal interaction (Cohn & Lyons, 2003). The natural back and forth process of asking questions as they emerged was a better fit for this rare cancer population, anxious to share their experiences to benefit future families. The dissemination domain questions remained at the end of the interview as planned; those questions were not part of the lived experience though participants often returned to their lived experience to illustrate the need for their recommendations.

A 5-item decision regret scale was included in the interview guide to gauge regret for the first decision made for or by survivors (Brehaut et al., 2003; O'Connor, 2003). Participants often continued to reflect on the decision when asked how likely they were to agree or disagree with statements like *It was the right decision* or *I would go for the same choice if I had to do it over again*. After the dissertation domain questions were answered, demographic data was collected for age, race, gender, marital status, education, and household income. Participants were also asked if they would be willing to participate in additional research for the decision aid and if they would like to be notified about the results of this study.

Demographics. In qualitative research, data saturation is the point at which continued interviews will no longer lead to new themes beyond those already identified. While some qualitative researchers suggest data saturation can be reached in as little as six interviews (Guest, 2006), others claim there is no one-size-fits-all method and that finding a balance between the quality (richness) and the quantity (thickness) is more important that having a predetermined number (Fusch & Ness, 2015). With over sixty inquiries to participate, twenty were selected for each of the three options with differing levels of satisfaction and a broad range of youth and adult participants. The participants were chosen from 15 states that were representative of the affected population with parents and youth/adult survivors. The interviews took place from August-November 2019.

Twenty interviews with 29 participants (see Figure 3) were conducted and included one father/son dyad, two mother/daughter dyads, two mother/son dyads, and two mother/father/son triads. One father and one mother took part in the interviews without their children. Ages ranged from 14-41 for osteosarcoma survivors (11 females, 9 males) who were between 10-33 at the time of diagnosis, all in the age range for the adolescent and young adult patient population as defined by the National Cancer institute (2018). Parent participants ranged in age from 36-71 and all identified as white. Interviews ranged from 51 minutes to 1 hour, 40 minutes with an average time of 1 hour, 33 minutes. Eight interviews were conducted in person, twelve were conducted via Zoom as video interviews. Participants were from fifteen states: Arizona, California, Delaware, Florida, Georgia, Illinois, Maryland, Michigan, North Carolina, Ohio, Pennsylvania, Texas, Utah, Virginia, and Washington.

Figure 3

Study 2 Participants

Participants (n=29)* *Vignette #/Pseudonym		***Role***	***Age at Diagnosis***	***Age Currently***	***Surgical Decision***	***Years Post-op***
1	Aretha	Self	15	41	LSS	26
2	Bert	Father				
	Benjy (nonparticipant)	Son	10	14	LSS	4
3	Cleo	Self	12	31	LSS	19
4	Diana	Self	18	20	LSS (AMP date TBD)	2
5	Ernie	Father				
	Frank	Son	14	15	RP	1
6	Gloria	Mother				
	Holly	Daughter	13	14	LSS, then AMP	1
7	Ingrid	Self	21	23	LSS (RP date TBD)	2
8	Joy	Mother				
	Kent	Father				
	Levi	Son	10	25	LSS	15
9	Mona	Mother				
	Mandy (nonparticipant)	Daughter	14	18	AMP	4
10	Nerisa	Mother				
	Nate (nonparticipant)	Son	13	17	LSS	4
11	Opal	Mother				
	Olivia (nonparticipant)	Daughter	10	13	LSS, then RP	3
12	Phoebe	Self	15	27	LSS, then AMP	12
13	Quentin	Self	15	32	LSS	17
14	Ramona	Mother				
	Ray	Son	10	16	LSS, then RP	6
15	Shelley	Mother				
	Ted	Son	18	33	LSS	15
16	Uma	Mother				
	Vera	Daughter	12	14	LSS	2
17	Wilma	Self	30	36	RP	6
18	Xena	Self	33	35	AMP	2
19	Yates	Father				
	Zora	Mother				
	Arthur	Son	11	17	RP	6
20	Brody	Self	28	30	LSS, then AMP	2

The aim of purposive sampling was to provide a variety of decisions for different surgeries. However, it was not revealed until the interview that some participants had a surgery prior to the one they stated they had in the screening. There were 8 participants who were living with LSS as their first surgery but of those, one was planning a rotationplasty early in 2020 and the other an amputation for the summer of 2020. There were three other participants who had LSS then later had an amputation and two who had LSS and later had a RP.

Data Analysis

The interviews were transcribed and deidentified in individual Word documents by a professional service, then reviewed for accuracy, and imported into NVivo qualitative data analysis software, Version 12 to facilitate organization. All participants were given a pseudonym and names of other people and locations have been changed to protect their privacy. All doctors are Dr. X or Dr. X and Dr. Y if two are being compared.

The data from 20 interviews was substantial and spanned 382 pages, therefore steps were taken to condense the stories while maintaining the participant's narrative in their own words. All five domains from the interactive framework of KT were embedded in the interview guide but the issue domain is the primary focus of the interview. The resulting knowledge of these lived experiences are presented as "vignettes", defined by Merriam-Webster (n.d.) as "a short descriptive literary sketch." Each vignette is a decision-making window into one patient, parent, or family's unique experience, either retrospectively, or in current time if they are making a second decision, or prospectively as they anticipate making another decision in the future. The user domain is summarized at the beginning of each vignette; both the research domain and the relationship domain emerge organically as people talk about where they sought information and how they felt about their surgeon. Each issue domain is highlighted in bold font prior to its discussion to draw the reader's eye to an upcoming issue or concern; there are several issue domains for each participant. The vignettes are each briefly summarized. A further thematic analysis synthesizing the findings for discussion in both Study 1 and Study 2 will be in Chapter Five: Discussion and Conclusion.

As a qualitative inquiry, the best approach to the data was a modified thematic analysis of narrative analysis. This differs from the traditional thematic analysis of grounded theory in that narratives do not "fracture" the data by including the voice of the researcher during analysis (Riessman, 2008). The importance of understanding the user context and issues in the interactive model of knowledge translation (McMaster University, National Collaborating Centre for Methods and Tools, 2017) is the best method in which to become familiar with the patients and parents in this user group. The modified thematic analysis requires some editing and set-up to tell a cohesive story in the participant's own words and therefore are not strictly narrative. The themes here are person-centered rather than thematically organized across the stories. The dissemination domain is a more traditional thematic analysis and follows the vignettes with its own section within this chapter.

Results

Vignette 1: Aretha

Aretha was 15 when she was diagnosed with osteosarcoma of the distal femur and had a limb salvage surgery. She was a dancer and played softball and basketball. Since the internet wasn't readily available 26 years ago, Aretha did not have access to online information like she does now. At 41, Aretha is currently struggling with some of the long-term complications of LSS and is researching her options for a different surgery. She lives in Arizona and we arranged for a video interview.

Issue domain: No options. The original surgical decision for LSS was not an option when she was diagnosed 26 years ago; it was presented as, "This is what you do. We're going to do the limb salvage." She felt that amputation was brought up as a worst-case scenario instead of a viable choice:

> …this is what we were told to do. You do the limb salvage and if it doesn't work, then they have to amputate, but that's what you do is the limb salvage, so. Amputation was kind of, like, ooh, that's if it's really bad and we can't fix it type thing, not oh, that could be something maybe better quality of life.

Issue domain: Passive patient as child. Aretha describes herself as "clueless" about interacting with doctors at 15, "My dad was a chiropractor and, honestly, we never went to doctors." She later regretted not being more involved at the time:

> I think the way I coped was just kind of ignored everything and just went along. So, I really didn't ask a lot of questions. I didn't know a lot of details. I kind of just ignored everything and just waited for it to happen to me. I didn't know a lot about what was happening. I remember years later being embarrassed that I didn't even know details about what they did or what was in there or anything. I think it was willful ignorance and my parents went along with it.

However, she felt that the expectations at the time were to accept the expert decision and not question it, especially at her age:

> …at 15, it really felt like it was kind of not up to me. It kind of just felt like I did what I was told. I don't feel like it was like, "Here, what do you want do?" It was like, "Here, this is what you do." I didn't know there was a choice.

Issue domain: Amputation was terrifying. The surgeon did ask her to sign off on an amputation if there were complications in the operating room, "I had to sign something that if something went wrong in the surgery that it was okay to amputate it. I remember having to sign that. That was just like a bad, you know, worst case scenario thing." The possibility that they might have to amputate was frightening to her at the time, "I remember talking and, you know, and being worried about what if they have to amputate and that kind of terrified me at 15. But there wasn't any decision going on. It was just preparing for the surgery." Aretha did ask her parents if they asked about other options, but her mom told her, “We didn’t even think, why would we want to choose something else?”

Issue domain: Unexpected complications. Aretha had just had her tenth surgery and was on her fifth surgeon when we talked:

> I’ve just had a lot of problems with it… My first limb salvage lasted four years. Then, I had to have the whole thing redone. Then, I just kept having things go wrong with it. Like, hinges cracking… So many different things. They had to realign the bottom part and then it caused my ankle to be out the way they realigned it differently. Then, I ended up having ankle surgery. So, it’s just a lot of problems with it.

Aretha had some good years with her limb salvage:

> I had four years of [being] incredibly strong, it was the best four years I had. I was able to hike. Then, I was doing Zumba. I was teaching Zumba. It was wonderful. Then, the hinge broke and then—so I was probably maybe overusing it, I don’t know. But anyway, besides those four years, I mean, I had no concept that I was going to be so limited.

Some of the problem stemmed from unexpected growth, “They like said I wasn’t going to grow anymore and then I did. So, it was off but when they had to redo it four years later, they kind of adjusted.”

Issue domain: Bias to save the leg. Not having options available to her was a missed opportunity to discuss the pros and cons of other surgeries and too much focus is on saving the leg at all costs:

> I feel like surgeons, they don’t think past the surgery. They think my success is getting this leg replaced and haha, it’s a success. They don’t think about how it’s going to actually function for your life. So, they don’t even—I don’t think they think of other options. They think, well, an amputation or a rotationplasty would be like a failure. No, I’m going to fix the leg…

Issue domain: Knowledge gap for functional limitations. Aretha felt that she was missing information about what to expect after her limb salvage and that more focus in general is on appearance rather than function:

> I just don’t even feel like they know or are aware that they should be thinking about quality of life, not just what your leg’s going to look like. I wish I would have of known, because I had no concept that I was signing myself up for a life of surgeries. If I had, I don’t know.

Admittedly though, prosthetic technology as we know it wasn’t as available 26 years ago, “It was hard back then too, though, because amputation—I don’t know if they had the great prosthetics that they do now.”

Issue domain: Making a second decision. Now Aretha is thinking more about what type of surgery would best for her next:

> I think in this day and age, it’s so important to discuss quality of life past the surgery, not just the leg looking good and fixed, but what about in the future. What about how long is it going to last? What are you going to be able to do? Now, researching more and more, I see more and more people able to do way more with their amputation or their rotationplasty.

She wanted to be part of this research study so her experience after 26 years of living with the limitations of LSS would benefit others:

> So, I just I wanted to participate in this to emphasize how important it is to let these patients know, think about beyond this year, next year. Think about 10 years from now, you know. What are you going to want to be doing? I’ve seen a lot of young kids now that are like smart and think, well, I want to do sports. So, maybe I should do the amputation because I can do more with a prosthetic. Because the limb salvage is usually so limiting. I was told to do nothing or you’re going to break it.

Issue domain: Seeking information online. Most of Aretha’s information came directly from her providers in the early years of her limb salvage. She only recently started researching online:

> Not even that long. I think four years ago. I started kind of researching and wondering if there is some kind of osteogenic group out there or something. I started researching and I stumbled upon, or someone sent me a video of someone with a rotationplasty. I was like, whoa, what is that, you know? So, I slowly started delving into research and seeing what else was out there. I joined an amputation group and learned a lot…it took me forever just to slowly start researching that, hey, there is other options out there. That’s when I was getting to the end of my rope and thinking about it for myself, but yeah. I might still end up with that, we’ll see. The jury’s out on this surgery.

Issue domain: Lack of long-term outcomes research. Now that she’s online more, she is using the internet to seek out research studies. Aretha says research at this level with longitudinal, patient-reported outcomes are imperative:

> I know they are all a grain of salt, but with my current surgery, it was kind of discouraging with this Compress unit, there really wasn’t a lot. The most there’s been a 10-year study out. So, yeah, I think it is valuable to see, Okay, well, 50 percent of these people had this happen. I think that’s just something to kind of base on, help make your decision. So, I do think it’s valuable. The more studies they have showing, following people. That’s another thing my hospital or surgeon didn’t have that I see some people do. They do long-term studies and follow the people. That is what we need.

Issue domain: Research not understandable to patients. Aretha had trouble finding research and when her doctor helped her find it, it was indecipherable:

> I tried to find it, and then I asked him because I wasn’t really finding anything on that besides from the manufacturer. He gave me a couple of very hard to understand medical jargon articles. I could kind of see the numbers on the 10-year study, but it was really impossible to read. So, yeah, he gave me a couple

> studies but there just wasn't much out there. I gave it to my physical therapist and he kind of interpreted it for me.

Translating research to something patients can understand and use to inform decisions is another priority; the one she tried to read was intended for the clinical audience: "…straight from a medical journal...meant for doctors. So, it was really hard for me to understand, but that's all he had to show who had been followed so far and the percentages of failure and everything."

Issue domain: More surgeries. Aretha had just had another surgery to fix her limb salvage leg – this time with a new type of implant:

> …this last time I spent two years on crutches and they couldn't figure out what was going on. I just couldn't put weight on it. Two years of awful, trying to just decide if I could live like that because I couldn't figure it out…my doctor here just pretty much gave up on me. So, I went (to another doctor). He proposed that the only thing to do really, is just replace the whole thing with a whole new type unit. So, I have like a Compress now. It's somewhat experimental. They weren't sure if I would even have enough bone to use. So, the second option was going to be a rotationplasty if I didn't have enough bone to use for the Compress. Because it's really high up, like right by my hip… Still not sure, you know, we'll see. I had to do three months zero weight. That's really affected now. I'm still can't get off the crutch because it's my muscles are just atrophied. The bone, I need my bone to grow around it for it to be very successful and it hasn't yet. He thinks, maybe, because it's so damaged from the other. So, we'll see.

If this doesn't work, she's ready to consider rotationplasty:

> …that could be coming, if this is unsuccessful. I mean, I'm going to let it go for a while. I'm not in a hurry to do another surgery any time soon. Yeah, if this doesn't work out, I think that would be what I would want to do next for sure. Because I wouldn't—just straight amputation, I wouldn't really have much of a limb to work with for the prosthetic. So, he would need to do and he was willing to do it, but I think he's done a few. I think I'd want to find a more expert in rotationplasty. So, but he was willing.

Issue domain: Rural area impacts decision. As she thinks ahead long-term to rotationplasty as an option, she realizes that since she's in a rural area, she'll need to travel more for rehabilitation – no easy feat with kids in tow. She says this was one of the factors she considered when deciding to have this last surgery to save her leg:

> I had to stay there for two weeks, you know, after the surgery and just travel. That was really hard with the kids …that's one of the things that worries me about the rotationplasty. If I do that, it's going to be hard because there's no one here for the follow-up. I don't even know for physical therapy. So, yeah. That was one of the reasons, I was swayed to just trying one more time to keep the leg.

Aretha did reach out to a surgeon to discuss rotationplasty but was told it's not for adults:

> When I asked her about it a few years ago, she was like no, I don't think adults can do rotationplasty. I think that's just for kids. That's blatantly wrong. She didn't even, you know. I didn't know at the time. I was like, oh. Now, I've

> researched more myself and that was totally wrong. She just didn't know. Again, I'm going to go back to the ego thing. Can you tell I'm biased against surgeons?

Issue domain: Options impacted by surgical training. She felt strongly that knowing all the options is paramount for families when making decisions for kids, not just the options that the surgeon wants or can do:

> I think if they're also not maybe wanting to give the other options to kids, maybe that is not something they do. If they do rotationplasty and they don't do amputation, why are they going to give that option? Which is not right, patients should have the options even if it means they don't do it. I know that happens.

Aretha felt that if she had been offered options and felt informed, she might be in a different place now, "Just kind of being aware of that, because I really wasn't aware. Looking back, if I had been, I really feel like maybe I would have made a different decision."

For Aretha, the most significant issue at the time of her first surgery was her age and passive acceptance for the recommended surgery. While options were not offered 26 years ago to a 15-year old, she felt she should have been more aware of what was happening for her future self. She did not know about the risks for limb salvage complications (besides limiting activity) and felt unprepared when she had surgery after surgery to repair broken parts. Aretha felt that the absence of information on long-term outcomes makes it challenging to feel informed for future decisions, including the surgery she had earlier in the year. Now, as she considers the needs associated with rotationplasty, factors such as limited prosthetists in her rural area impact her decision. Aretha scored high on the decision regret scale. For the statements, *I regret the choice that was made* and *the choice did me a lot of harm,* she agreed. For the statements, *I would go for the same choice if I had to do it over again* and *the decision was a wise one,* she disagreed.

Vignette 2: Bert (Benjy)

Bert's son Benjy was diagnosed with osteosarcoma of the right distal femur and knee at the age of 10. Benjy lived with his parents in New York and was not athletic at the time though he does swim now at 14; he did not wish to participate in the video interview. Benjy was offered all three options and had limb salvage surgery which was accompanied by complications immediately afterwards. At the time of the interview, more decisions were being considered for Benjy, although he was not interested in being part of in the decision-making process.

Issue domain: Complications from limb salvage surgery. Bert explained that Benjy "suffered substantial damage in the surgery" and has drop foot, an inability to life the front part of the foot, with limited feeling due to nerve damage. The family is at the point where a decision may have to be made again:

> We're now told that Benjy is going to be very tall and that the prosthesis will probably not expand enough. So, we have two issues. First, whether we want to

> replace the rod and his prosthesis to make it even bigger. Second, it's now clear that his tibia is not growing. So, if we continue to expand this prosthesis, his knees are already not lining up but the differential will become more pronounced…There are options—because he's so tall, the doctor thinks a good option would be to stop the growth in his other leg, which Benjy is understandably very opposed to. He doesn't want anybody claim to his good leg.

Issue domain: Child does not want amputation. This family faces several decisions in current time, including the option for amputation, an outcome the parents knew might be a possibility if the limb salvage failed:

> Quite frankly, a lot of the medical staff that we met with, they postured amputation as a second alternative if the limb salvage surgery failed. When we met with the doctor at [hospital], he encouraged us to go forward and do the limb salvage. He said, if that fails, you can always do the amputation later on. That's not something that we've ever raised with Benjy. I think that him having another surgery, at this point, is not something he really would want to consider. That's not really in the cards right now.

Issue domain: Common complications not discussed. Bert felt that while some general uncertainty about LSS was discussed, much was left out, "I was somewhat shocked afterwards when Benjy had no feeling in his foot and his leg. At that time, again, we were very hopeful that it would come back, but now I understand it's not an uncommon thing." This left him feeling uninformed later about potential complications when he had worked diligently to be informed:

> …the sense that I got from the surgeons is that they were presenting limb salvage surgery as the gold standard and they told us that it might fail, generally, but we never knew about the more common problems and challenges of limb salvage surgery survivors experience. I understand that drop foot and nerve damage is not uncommon. I also understand that it's the standard that there are going to be multiple revisions. We never really had those conversations. I don't think that it would have changed our decision in any way, but it's certainly something that I would have like to have known.

Issue domain: Lack of information affects trust. While their relationship with the surgeon has been positive, "He always makes time for us," Bert worries that information on all available options at this point are not forthcoming:

> Are there other options out there that we have not explored in terms of trying to regenerate his nerves? That's always something that bothers me. I don't know. I think that when we force that subject with him, he's not been a fan of that information.

So the issue for the family now is deciding which option is the best for Benjy, in the absence of his willing participation and based on information they suspect it is not unilaterally exhaustive, "It's a decision that's going to have to be made soon. We've only dusted the surface of that with Benjy. It's not very good when we talk about it."

Issue domain: Passive patient. The diagnosis of cancer at any age is traumatic and frightening; patients of all ages have different preferences for how much they want to be involved in medical decisions. At the age of 10, Benjy did not want to participate in making the initial decision for surgery. Bert felt that it was too much for him to deal with at the time, which he understood, though he tried to include him:

> We told him what was going on. He really deferred and relied on us. He didn't want to be involved in the decision. So, he had very limited participation. We invited him and certainly wanted to hear what he was thinking. He just wanted us to make decisions and be told what he had to do.

Bert's empathy for his young son was clear as he imagined how his son must feel:

> They're scared of adults. Even though these doctors did their best to speak directly to him. You know, they're way older people that walk in with white lab coats. You know, he's already going through the scariest time in his life. So, I would be quiet unless it was something that he was emotionally prepared to deal with.

Issue domain: Surrogate decision-making. When making the decision for Benjy, Bert and his wife thought about values and what Benjy would consider as a good life for himself:

> I was, from my perspective, trying to think what Benjy would want if he were ten years older and looking back on it. You know? Benjy was not an athletic child. He didn't play sports. That limitation made the decision easier. Because the doctors that we met with said, both of them said, Benjy is never going to play soccer with the limb salvage surgery. He's probably not going to be playing baseball and other things...That wasn't an issue for Benjy, because he didn't play those sports anyway.

Issue domain: Self-esteem. While these values were understandably important to Bert and Emma, they also worried about his adjustment to limb loss: "We were very concerned with his self-esteem and his self-view about losing the leg to amputation. Both my wife and I felt that if we could save his leg, that was something he would want."

Bert and Emma felt overwhelmed but leaned on the expertise of the surgeons they met with to discuss options when making the decision:

> I don't think we're qualified necessarily to make the decision but somebody's got to make the decision in this. From what I can tell, there are pros and cons of the various options. I wouldn't want any surgeon to make the decision for us. Again, if Benjy was in the position where he could vocalize what he wanted, that to me would have been the end of the story, but he wasn't. It was overwhelming for us to have to make this decision but giving that with the two pretty well-respected surgeons that we met with were both telling us the same thing, that's helpful.

Retrospectively one of the most challenging aspects for this family was making the surgical decision for a child who could not state any preferences, but they felt like they had enough information at the time to make the decision:

> I feel like we were as well informed as most parents can be. I don't, again, I don't know that there's a whole lot that speaking with more physicians would have helped. If my son was a little bit more mature and was able to voice his needs a little better, that would have certainly been helpful. I'm not sure that there's a whole lot of data that would have changed our decision or influenced it.

Issue domain: Recurrence risks. When Benjy was first diagnosed, the primary concern was to be cancer free. As he weighed the different options, Bert was concerned about the validity of online information and relied on the surgeons to give him reliable data:

> The most important thing to me was me to ask repeatedly is there any – and I asked several people with this, is there any differential in terms of the survivability in beating the cancer, whether he has limb salvage surgery or an amputation? Because, you could find information online that says everything. The oncologist, both the surgeons we met with said that the statistics don't show any difference in terms of recurrence of the cancer. So, that was by far the most important thing.

Even though Bert and his wife were concerned about the nerve damage from the beginning, their "biggest concern was that clear margins were achieved in the surgery, that the tumor was out."

The best chances for success in chemotherapy is following an approved protocol: evidence-based research was pivotal to Bert in understanding that process and making decisions for Benjy. When Benjy's endoprosthesis wasn't ready in time following the chemo schedule, he researched to see if that could impact the success of the chemo if they went slightly off protocol, "There's no statistics you know that would tell us that this is a problem. That's what we did." He also looked to the Osteosarcoma Facebook group for information on recurrence risks with the different surgical options though he placed far more credibility on this issue in the hands of the physicians:

> You know what, when we looked at the Facebook page, there was some people there who were pretty insistent that if you do a limb salvage surgery, you were increasing the likelihood that the cancer will return. That was always our first question to every physician we met with. They were all three insisting that was not the case. Whether we do a limb salvage surgery or rotationplasty or amputation was an orthopedic question. It would not impact the likelihood that this cancer would return. That was our primary concern at the time.

Issue domain: Unmet psychosocial needs. While he had met an osteosarcoma survivor with LSS, he felt it was a missed opportunity not to meet with people who chose other options:

> …we did not meet with any amputees or people who have had rotationplasty. If that were available and I was able to meet with either the patients or the parents or family and talk to them, that would have been very helpful to me. I find the personal interactions very helpful… Face-to-face is always better, but cyberspace is second. I mean, just reading you can gather a lot of statistics, but you can tell a lot about how somebody feels by actually speaking with them and seeing them.

Bert sometimes referred my experiences with my daughter. This connection is part of the researcher-user relationship with a shared understanding of the osteosarcoma experience. He compared being marginally involved in a medical decision for his mother and how that differs for your child, “So, I had some experience in dealing with this but quite frankly and I don’t have to tell you this but when it’s your child, everything’s different.” Bert also reflected on a talk I gave about how survival concerns for parents often trump long-term late effects: “As you said, there are long-term other things on the priority, but no. At that time, our worlds revolved around that.”

The connection and rapport between us as parents were particularly moving in this interview. Bert talked about how meaningful it was to meet other families like my daughter and me at the osteosarcoma conference years ago:

> I don’t know if this is helpful in your study but as a personal matter, when it came to the MIB conference and I met you and I met Zoe and I met others like you, that was the most help for me. I mean, meeting people who have walked through the fire and been through exactly what we have—as far as I know, there was nothing like the MIB conference before. Whereas I go and I sit through all the lectures and I take away some of it, to me the best part of that is being with others in the community that have been through something similar.

The decision for Benjy’s surgery is both retrospective and prospective. The current issue of what to do next to manage the limb salvage complications is the next decision. The issue that impacts this decision the most is Benjy’s preference to have someone else make medical decisions. The SDM communication style between the surgeon and family places Bert and his wife in the position of the surrogate decision-maker. The primary consideration for the first decision was to make sure that any orthopedic decision they made did not impact Benjy’s risk for recurrence. However, they were unprepared for common complications that continues to affect Benjy’s function. The overarching issue for these parents in both decisions is choosing a life-altering surgery for their son who is prefers not to engage in the decision-making.

Vignette 3: Cleo

Cleo was 12 and living with her family in a very small town in the Great Lakes region when she was diagnosed with osteosarcoma in her left distal femur. Even though Cleo had a limb salvage surgery, she continued living an active life and played volleyball in high school, though admittedly she was told not to. Now 31 and married with two children, Cleo joined me for a video interview from her home in Michigan.

Issue domain: Decision-making at 12. Cleo’s options for surgery were amputation or limb salvage surgery; rotationplasty was not mentioned. Her parents asked her what she wanted to do. Cleo talked with survivors as she was considering her options:

> When I was making those decisions, they had a survivor who had limb salvage come speak to me about the pros and cons, too….that was really beneficial to me to hear someone from the other side talk about what life was like. She seemed to be active, she was pretty positive that that was the right decision for her…There were a few other kids going through the same thing in the children's wing. I think it was about 50/50, half of us had limb salvage, half of us had amputations. So, it was—we all got to see at different times people going through different things.

Seeing peers make surgical decisions helped Cleo feel more comfortable as a decision-maker, "It was, I think, easier to make a decision when there was other people going through it as well."

Issue domain: Weighing risks and benefits. At the time of diagnosis, Cleo played sports recreationally but not competitively. She remembers talking with her surgeon about her options, "It was really just like we believe we can give you the best odds at survival either way… so, I wanted to try to keep my leg." Cleo based her decision, in part, on appearance, "I didn't want to not look like my friends. So obviously that played a big role. I don't think I ever wanted to amputate." Cleo said her parents were on the same page but felt that if she had chosen differently, they would have supported her decision.

Issue domain: Amputation risk during surgery. Few uncertainties were discussed at the time, but one was the possibility that the surgeon might have to amputate during the limb salvage surgery, so she really had to consider both options seriously since she had no control over the outcome:

> I was told that they couldn't tell exactly how high up the cancer was. So, if they got in there and it was too high to tourniquet that they would just have to amputate. So, I kind of just had to make peace with both options before I went in… It wasn't what I wanted but I also wanted to live, so you know, was out of my hands… I remember when I woke up from surgery, the first thing I did was feel to see if my leg was there or not.

Issue domain: Fertility risks. The uncertainty around fertility was implied by Cleo's doctors but she didn't feel she was ready to hear that at the time:

> …they did, but it was very lightly. Like, "Because you've already started your period, it may not come back." You know, but I feel like they did the right thing by not making a big deal out of it because I would not have had the mental capacity to even think about that with everything else going on…. Thankfully, it didn't for me but I know that there are others that it does. I think it was just a crapshoot, basically.

However she also says that if her fertility were affected, she might feel differently, "Maybe I would have wished that they would have made a bigger deal out of it, but at that time—when you're 12, you're not thinking about kids at all." Understanding the difference of potential side effects and expected side effects was outside her skillset at 12, "I didn't have the perspective on life to be able to differentiate between potential side effects and this is definitely going to happen to me. I'm glad…that it was mentioned, but not focused on." Cleo added that as an adult, she would want to know potential side effects.

Issue domain: Uncertain risks associated with necrosis. Necrosis refers to the percentage of cells that die as a result of chemotherapy; a good response is over 90% while a poor response is below 90%. Cleo had the same roommate throughout her treatment who had a LSS, then had a recurrence and an amputation, and later passed away. She also became friends with other osteosarcoma kids, and it was terrifying for her when they later had a recurrence and died:

> …one girl that I was friends with, she had amazing percentages when they did her original operation. She was 99% necrosis. I think I was only 80%. So, that was like terrifying that if it came back so aggressively with her, I was petrified to go to follow-up appointments because I was just sure that it wasn't going to be good.

She tried to understand what the doctors were saying about necrosis rates:

> They're saying it doesn't impact survival but they're also saying that if your numbers are higher that means you don't need as much chemo. So, they can say that it doesn't, but to you going through it, it feels like obviously it is a factor. So, it kind of, it's the opposite.

Issue domain: Life expectancy of limb salvage implant. Another uncertainty was how long the endoprosthesis would last; this information left her fearful, with alarming answers to her questions:

> I actually remember being petrified, like, when we were talking about limb salvage. I forgot about this. Because they did tell me, the knee lifetime is only 20 years. That was best case scenario. I remember being petrified. Like, how do I find out if it's broken? Does metal just come through my leg, like a traumatic experience? They're like, well, "Your bone could snap but generally we'll know it's wearing out and we'll be able to fix it." I don't know. I feel like when you're so young, it is scary to think about all the things that could go wrong but I also feel like—I don't know. Normally, as an adult, I'm like yeah, I would want to know. As a 12-year-old, I had to do what had to be done. That was my only, like my whole focus. I have to do what has to be done.

Cleo has had the same surgeon since her LSS; she felt that her relationship with her surgeon positively impacted her outcome:

> I had my orthopedic oncologist who I still would trust with my life. I really always completely was comfortable and trusted her and her decision of what she thought was best. So, I feel like that is a key. Also, if you don't trust your doctor or if you don't feel comfortable with them, it's hard to take what they're saying.

Her surgeon's communication style felt tailored and genuine to Cleo:

> Honestly, I feel like she was just super honest. Even though I was young, she never sugar-coated anything but she also wasn't scary about it. She was very matter-of-fact, which works for me. I'm very logic-based. So, when she laid out the facts for me, she did it in a way of I knew she cared but she was going to be as honest as she could be….I just trusted her because she didn't try to push an

> agenda….She told me what she would do if I was her child and what she thought the best outcome would be for me… I love her.

Cleo's surgeon built a relationship with her by sharing not only personal information like playing hockey but also involving 12-year-old Cleo in the surgical process. She "brought all of this stuff in for me before, like the rod, the knee…let me play with it, let me feel it, explained in-depth pretty much what she was going to do. I always appreciated that. I really did."

Issue domain: Poor provider experience prior to diagnosis. While Cleo's relationship with her surgeon was positive, it was an early experience with a physician who was dismissive and threatening that remained part of her story:

> He told me that it was just I needed to wear a knee brace, that it was a cartilage issue. If I kept coming back, he was going to send me to PT which stood for pain and torture. I didn't want to come back. So I kind of just sucked it up until I couldn't sleep, I couldn't eat, I was in so much pain.

Nearly 20 years later, Cleo finds herself delaying her scans, referring back to that earlier experience as the trigger for her apprehension:

> Just have anxiety about doctors. I always feel like it's going to be the worst…the one doctor that put me in a brace and told me that I was going to be tortured at physical therapy if I didn't suck it up, I'll always remember his face. I'll remember his voice, because he was—like, I just feel like that wasn't the right approach. I'm sure he sees thousands of people that are just fine, you know.

Cleo said she wished her parents had taken her back to see that doctor so he'd know she had cancer all along, but they never go around to it.

After Cleo's treatment ended, her parents encouraged her to resume her activities, "Do everything you would do before you got sick…don't let this limit you." So all through high school, Cleo continued to play sports. Her surgeon had mixed responses, "I wasn't really supposed to, but—I played volleyball…my orthopedic surgeon wasn't the biggest fan. She plays hockey, so she understood and gave me ways to try to be safe."

She had no issues with her LSS until 12 years later when she had pain and subsequent surgery to replace some screws that had worn out in the knee. Five years after that, she started having more pain and needed a revision.

Issue domain: Limb salvage complications – pain. Cleo's pain was so bad at that time that she considered amputation:

> Before my last revision, I told my husband if I had to deal—I was in so much pain, like constantly. I couldn't take my kids to walk. I couldn't stand up and walk around the park. I would hurt so bad after. I was like, if this is how it's going to be for the rest of my life, I just want to amputate it. I just want it gone. Then, since I had the revision, though I've been like, I honestly don't have—my other leg hurts more than this, than the left one. Like, when I do a lot of walking

> or whatever, I'm just—it's been night and day. So, now, I would not change a thing. A few years ago, my answer would have been different.

Issue domain: Replacement parts unavailable. What she could not have anticipated 17 years earlier was that the manufacturer of her endoprosthesis was no longer in business, forcing her to undergo a much more extensive surgery than needed:

> Unfortunately because I am so old, the person who manufactured my original part, is no longer in business. So, we had to have it custom made. Next time, she said, they'll have to replace the rod too because there won't be anyone to make the pieces that fit. Well, that kind of is frustrating to go through this a few more times in my life. Hopefully, the next time they'll be able to keep the manufacturing available.

Prior to her revision, she discussed amputation with her husband due to the pain, "He was like, if that's what's going to help then you got to do it. He has a trach tube, so he's very, you know, whatever's going to make us happiest and healthiest, we just got to do it." However, her pain was resolved when her leg was lengthened during the revision. Looking back, Cleo thinks this might have been the cause of pain for years before: "I think a lot of my issues with pain before were from me being so off kilter because my back and my hips were always hurting. Since she lengthened it, it's been great."

Issue domain: Weight gain, pregnancy and obstetrical care. The pain increased around the same time as her last pregnancy; Cleo thought that her pregnancies compromised the integrity of the endoprosthesis, contributing to the pain:

> I also gained a lot of weight. I feel like that impacted it. I feel like my leg being off an inch and my hips. Then, your hips and back hurt anyways when your pregnant. I feel like that just added more pressure. I really feel like that was a big deal.

She felt that her obstetricians were unprepared to understand a patient with LSS: "They honestly weren't much help. It's hard. I don't think many OBs are used to dealing with people of child-bearing age having artificial joints or limbs."

Cleo's issues at the time of decision-making were primarily around appearance and uncertainties such as if an amputation would be necessary during the initial surgery, how the recurrence risks related to necrosis rate, and the life expectancy of the implant. Later complications developed when she had pain so bad that she considered amputation. She also struggled with pregnancy care and obstetricians who she felt were unprepared to deal with a cancer survivor who had an endoprosthesis. Most of Cleo's information came directly from her surgeon whom she trusted but also from friends she made in the hospital who also were making surgical decisions. The internet as not readily available in 2000. Looking back, she is glad because she thought researching on her own now would be scary for a 12-year old.

Vignette 4: Diana

Diana was 18 and away at college when she first noticed a bump on her knee. She played varsity volleyball and her athletic trainer sent her to an orthopedic clinic. Within a month she was diagnosed with osteosarcoma. Diana moved back home with her parents in Washington state to an area she described as very rural and had limb salvage surgery. Now 20, she is back at college but due to pain and functional limitations, she is considering amputation next. After juggling schedules with different time zones, we met for a video interview while she was between classes.

Issue domain: No options. Diana said she did not have any options at the time she was diagnosed, "We asked about amputation but my surgeon was concerned that I wouldn't have enough femur, and less stuff left to really work with a prosthetic….He doesn't do rotationplasty…" Diana asked her surgeon if she could play volleyball again, "He kind of just shrugged and said, 'Maybe.' I think that's just because of the nature of limb salvage, it can either really work for some people or it can really not."

Issue domain: LSS complications. Diana had a LSS two years ago but her rehabilitation did not go as hoped. Diana had issues with the build-up of scar tissue that affected her mobility and caused pain. Her surgeon was as frustrated as she was about the scar tissue but additional surgeries seemed to be her only option, "He was talking like we could just keep doing scar tissue resections. Ten or 15 later, you still don't know if it's even going to be any better. It's kind of a shot in the dark, really."

Issue domain: Honesty about uncertainties. Diana attributes her acceptance of the complications to her surgeon's honesty about the unknown and potential risks:

> I think it's just really a matter of being straight up and being honest about things. A mentality in the medical field that's starting to become a little bit better but having doctors who are able to say, "I don't know," I think is really going to be important. You know, patient trust and things moving forward with confidence in our medical professional. Just because sometimes, I mean, we're human, you know. Sometimes people just don't know things. I think the important part to put in there would be, "I don't know but let me figure it out for you. In this case, I don't know but I still think you have a good chance of having success." I think it's really about being really up front and honest about things.

Diana thought honesty also helps to build a relationship of trust between doctors and patients:

> I think in some ways, you know, it's kind of like a reverse psychology where it builds more of a trust because that physician will come across as more human, you know. In that way, people will be more comfortable with them instead of coming across as this magical doctor robot who knows everything and operates like a machine. I think it's important to still have that human aspect these days in the medical field. Just because, I do think that trust comes out of showing human attributes…

She speculated that it could be hard for some – and maybe even most - people to hear about uncertainty:

> I think it could be tough for some people who aren't very good at taking honesty and hard truths. That could really drag some people down and make their recovery process a lot tougher. For somebody who can't, hearing the fact that this could not work out for you, could really send them into a crazy panic and they won't want their surgery anymore. They are going to be terrified, lose hope, or stuff like this. So, it's hard because I think you have to tread lightly with the majority of the population for people who can't really handle tough situations.

But in her case, it was better, "For me, I am a very level-headed person. I can see both sides of things."

Issue domain: Complications lead to second decision. At the time of Diana's interview, she was turning 20, back at college, and actively making the decision for another surgery since she was so unhappy with her LSS, "…it's holding me back in a lot of ways. Pain wise, movement wise, happiness." This dramatically affects her quality of life, "I used to be super outdoorsy, like mountain biking. You know, I did hiking, everything I could get my hands onto outside, I would do it. I just can't do those things anymore. So, it's pretty tough." There were two meaningful points that led her to seek other options. One was the summer she spent working at a national park lodge and she was unable to participate in the things her friend were doing; if she tried, she was in terrible pain afterwards. The second was being a part of the sitting volleyball team and getting to meet amputees, "see their prosthetics, see how they move, and how happy they are and how much they can do really. I think those two things together helped me realize maybe amputation is the best option at this time for me." Although rotationplasty is a viable option, it is not a good option for her, "Part of the reason I don't really necessarily want rotationplasty is because the aesthetics of it. I'm not really comfortable with that."

Issue domain: Planning for amputation. Moving from frustration with her LSS to thinking more about amputation has been a process for Diana:

> I did the research and then I brought it up to my surgeon when I had my most recent scans which were in July. He was able to tell me a little bit more and tell me how much stump I would have left and all that. Then, he scheduled a amputee clinic appointment for me. That was a couple weeks ago. That was when I think I became more comfortable with the idea. That's when I was just like, okay. I think this is going to be good for me. The amputee clinic appointment was with a prosthetician and an orthopedic surgeon who wasn't my original surgeon.

Diana is anxious to be active and pain free again; she looks forward to scheduling her amputation and talked about knee shopping for prosthetics. She met several peers on the U.S. Paralympic Volleyball Team who had limb salvage surgeries and later opted for another surgery but she was the only one who still had the endoprosthesis, "Everybody else who has had limb salvage in any way, shape, or form, has had amputations." Meeting people who had been in a similar situation and had the same quality of life goals helped make Diana feel more comfortable with her decision. One person has the same high level above knee amputation she anticipates she'll get:

> …she's a softball coach and she works for a prosthetics and orthotics company…. She has a really cool prosthetic. We were talking and she probably has about the same amount of stump as I would have. So, she was a really good model for me to see what things could be looking like.

Issue domain: Preparing for pain. Diana has done some research on tactics to manage phantom pain such mirror therapy and is preparing by asking questions:

> Then, also, I guess it helps if you desensitize the nerves at the end, if you do a lot of massage, and you really use your prosthetic. The doctor who I talked to at the amputee clinic, told me the people who struggle with phantom pain the most are those who don't use their prosthetic enough.

She is resolved to accept that phantom pain may be a part of the amputation process, "I'm not really concerned about it necessarily because it's just going to be a part of what I'm going through."

Diana's issues were mostly around making the decision for a second surgery – amputation after limb salvage. The impetus for amputation is the uncertainty about the success of continued scar tissue surgery. If she chose to continue, it could take 10-15 more procedures and it would still be a "shot in the dark" whether it would help or not. While she was not offered other options for her first surgery, she attributes her lack of regret to her surgeon's honesty about the unknown; namely, that the LSS might not work. Diana trusted and liked her surgeon who shared in her frustration about the scar tissue. He continued to give her medical information and a referral to meet other specialists, including a prosthetist. Diana feels she was prepared for a possible poor outcome and is now being proactive to attain better function and quality of life with an above knee amputation. After her LSS, she joined the U.S. Paraolympic Volleyball Team and now has significant peer support for amputation, some of whom also had failed LSS. She feels she is preparing herself for managing phantom pain and getting fitted for a prosthesis. Diana was looking forward to her new life as an amputee, scheduled for nine months after her interview. Although Diana was planning an amputation after LSS, she scored low on the decision regret scale stating that she highly agreed with these statements, *It was the right decision* and that *The decision was a wise one*, and highly disagreed with the statement, *I regret the choice that was made.*

Vignette 5: Ernie & Frank

Ernie's son Frank was 14 was he was diagnosed with osteosarcoma in the proximal tibia. They live in near the mountains in Utah where Frank and Ernie enjoy hiking and backpacking. Frank was now 15 and had a rotationplasty less than a year ago. They both joined me for a video interview.

Issue domain: Two options. According to Frank, "the surgeon said we have three options. One, we could do amputation and then he said, "We're not going to do that." Then, he gave us the option for limb salvage or rotationplasty." Ernie's first reaction when the surgeon described rotationplasty was, "In my head I thought no self-respecting parent would ever let him do that. That's just too off the wall." But Frank's parents encouraged him to make his own decision:, "They had their opinions but they let me make the decision on my own."

Issue domain: Missing information. The biggest issue when making the decision was finding the information needed to make it. Initially Ernie expected to get more answers from Frank's doctors than he did:

> I would talk to the doctors about everything. You know, it would start out as, "Oh yeah, that's a great question." Then, it would kind of just they'd direct the conversation to where it would go vague. They'd kind of ramble for a while then. You could tell they didn't really want to say anything.

Ernie asked for research advice, "'Well, where do we go to research things?'" The only thing the surgeon told me is he's like, 'There's videos on YouTube.'"

Issue domain: No psychosocial support. Ernie also asked about peer support from other parents:

> …this was the part that in my mind, it's still really weird. I don't like that this is how it happened. I'm not sure what the actual doctor, either the oncologist or the pediatric surgeon could do about this. We're like, "Can we talk to people?" They're like, "Oh, yeah. The social worker will get you in touch with people."

But those connection to other parents never happened. Ernie and his wife hoped to meet other parents organically at the hospital and asked the social worker about a specific family:

> My wife and I would go in there to eat lunch and there's nobody in there. We're like, we know there are other people who are being treated for Ewing's sarcoma or osteosarcoma, and we can't find them. We did hear about one young girl, actually a few years younger than Frank. She had done the surgery a month before Frank. We tried to get in contact with her and her parents. The social worker was just like, "They just, they don't want to talk." Just trying to get in contact with parents and have conversation…. I'm like, this is crazy. I'm like parents need to be able to talk to other parents, understand what's going to happen.

Frank liked the surgeon and felt like he was included in discussions: "My parents were in the room, but he spoke directly to me and listened to me about everything. It seemed like he was more on me than my parents, which was nice." Ernie liked both Frank's surgeon and oncologist but struggled with how little guidance they were given to make such a big decision; he thought working with a therapist would have helped too:

> I loved the surgeon. I think he did an outstanding job. I think he's a fantastic doctor, same with the oncologist. Looking back on it, I'm like, why do they not tell us more about the options and go through it with therapy. I don't know. I have no idea as to why they did it or not.

Issue domain: Gathering information. Since Ernie could not get information from the surgeon, oncologist, or other parents, he began researching the options online, forwarding videos and articles to Frank. As an IT professional, Ernie was comfortable navigating with technology. He even set up Skype meetings and in person meetings with other patients and families so Frank could ask questions in person. Frank talked about meeting a boy with limb salvage:

> Frank and I love to hike and backpack. He said, "I know I'm never going to hike (mountain range) again. I'm totally okay with that." He is, like, he's the kind of boy who that isn't a big deal for him. When he said that, I kind of like, was like, oh no. What else goes on—what are some of the other cons from a limb salvage? At first, I was thinking, wow, this is great. The only thing he's got is this scar that runs the length of his—if that's the only con then maybe this is the right thing. When he mentioned that, that's when I started to really gravitate towards what are the limitations physically of either option.

Ernie joined Facebook support groups, read through blogs, and research studies in journal articles; he talked about the challenges of searching online for information from ground zero:

> …the very first thing I came across was there is a rotationplasty.org super old website that has not very good information. I started doing research, I started finding there were - 10 years ago, 15 years ago, there were some online websites that allowed people to make personal blogs about their journey through cancer. I started to find stories about people who had sarcoma specifically. I started to learn, I found the right terms to search for. Just by accident, I would find medical journals. I found several journals that had done studies on osteosarcoma and the outcomes of each of the different subtypes, what was it that they knew and they had bibliographies. So, I started to find key words to search for. As far as the procedure, I started to find those as well. So, it started to amass the right—I learned the right phrases to search for, the right things to search for. Being a computer scientist, I knew how the clarity engines worked, so I exclude certain terms and include other terms and try to narrow things down. The more I found, the more I could start to branch out and find more stuff. It was really just a process. It was extremely slow and painful at the beginning.

Issue domain: Patient-reported outcomes and functional risks. Ernie recognized that not all outcomes would be out there but began to see a pattern of risk associated with LSS that wasn't present with rotationplasty:

> For rotationplasty, if you had a great experience, you blogged about it or you posted YouTube videos about it. If you didn't have a great experience, there was nothing out there. I don't know that people that had a negative experience with

> rotationplasty, are as vocal online in any form. So, I definitely could tell I was biased by what I was finding. I'm like, okay. One group is generally positive and one group is split fifty-fifty. So, there's a risk on the limb salvage side.

Ernie struggled with feeling like information was being withheld when risks were not timely shared:

> I knew from my research that one of the biggest issues with rotationplasty is, for twenty-four to thirty-six hours after surgery, they have to make sure that blood is going into the foot. The surgeon, the first time he talked to us about that, was like the day before. Like, why are you waiting? Like, we couldn't handle that up front or—I don't know. I don't know why they wouldn't have explained some of this in more detail. We didn't get that detail. I got that detail from talking to other people online, from research.

Issue domain: Lack of clinical support. He knew that collectively the surgical team had over 35 years of experience; he wished they could have shared in a general way what the surgical outcomes were like:

> I would have loved to have had a sit-down conversation with the surgeon. Just, "Hey, talk to me. Don't identify anybody but let's talk the last two to three years surgeries have you done of this Van Nes procedure. How are the kids doing today? How many really didn't work out as well as you thought they would? How many did?" Just see where that conversation goes, just have this nice frank conversation…. There's a lot of experience here doing this procedure. I would have loved to have known how things worked out.

Ernie felt that the overall lack of outcomes research was a significant knowledge gap for patients and parents when making the surgical decision:

> He's like, "We don't know what's going on in Phoenix. We don't know what's going on in California. It's very hard to share that information in the medical community. I'd love to say, here's, in general, here's what's going on. I don't have access to that information." That was really eye-opening to me. I was like, man, we've got to fix that. You've got to be able to not identify individuals but identify groups and outcomes and give that to parents.

Issue domain: No surgical recommendations. Retrospectively, meeting with the surgeon before the decision was made would have been welcomed, "Yeah, totally would have been useful, yeah. Yeah, just to even confirm some of the research that I had done, right?" Also looking back Ernie described both the oncologist and the surgeon as guarded but that changed after a decision was made:

> Once Frank had made his decision, then the oncologist was like, "Hey, Frank, that's what I would have chosen." All the sudden, he would let us know, "I'd seen patients in here for years. They do a lot better. If I had to make that decision, that's what I would have made." ….I can only attribute that to they don't want to be seen as the influencer or let us do whatever the choice. Then, once we've made the choice, then they're more open to talking.

Ernie felt that by not giving patients information, they were avoiding influence and blame if something went wrong:

> So, the doctors themselves, I really feel like they were trying to stay away from convincing us or influencing us too much. They were just saying, hey, if something were to go wrong here. We want it to be your choice, not our choice. That was hard, because I was like, wait. We're in a new world here. We need you guys to talk to us about the pros and cons. I would say, you know, there's a limited chance when we do rotationplasty that a nerve get cut. They'd say, we're not going to cut the nerves or the blood vessels, but there's always a chance that this might happen or that might happen.

Ernie felt that sharing the information wouldn't have changed their minds about choosing rotationplasty: "…love to have known more information about how things could potentially turn out, what their experiences are. I'd probably still—it wouldn't have changed my mind I don't think, but would have felt more comfortable with the openness."

Issue domain: Conveying support to child. Another concern Ernie had was making sure that Frank understood he would fully support him for life regardless of his decision:

> …the biggest thing I wanted to share with Frank was, "Hey, we're not dropping you off at the pool, and go have fun." I wanted Frank to know, whatever choice you choose, I want you to feel confident that your dad has researched both options. Dad knows what to do to try and help. The kids that we did meet with that had done limb salvage, I felt like I knew what they were trying to accomplish from a physical therapy, from a stronger standpoint. I wanted him to know, if that's what you choose, I have this by knowledge now, but I'll go become an expert. I will figure it out and I will work with you, same thing on the rotationplasty side of things. That's what we conveyed to him is, I'm trying to educate myself. I'm trying to not be ignorant of what's going to happen here. I know I don't know everything. I know we're going to learn a lot just because there's no way to learn without doing. That's what I wanted to convey to him was, you're not alone. I'm going to be here. We're going to work through this. We'll get wherever you want to get to. So, I'll help you get there.

Frank said that one of things that made him feel more comfortable with his rotationplasty decision was the support he received from his friends and family:

> It was hard, but I'm just glad I was able to choose the decision. It looks weird. I mean, it doesn't really bother me that much. I'm kind of in a good area where I've got people who support me and everything, so. That kind of helps, I guess….I think it was the right decision.

For Frank and Ernie, the issues were mostly around not having enough information to make the decision and not getting any recommendations or support from the surgeon beyond looking at YouTube videos. Ernie was able to research options and outcomes and was comfortable doing that with his professional training but never understood why the surgeon didn't get them more guidance. At 14, Frank was involved in the decision from the beginning and felt supported throughout the process by his parents. Of the 20 interviews, this family was the newest from their first surgery; they scored low on the decision regret scale and both were pleased with the surgery and recovery at the time of the interview.

Vignette 6: Gloria & Holly

Holly was 13 when she was diagnosed with osteosarcoma of the proximal tibia and had limb salvage surgery. She was a busy teenager, riding her bike, and jumping on the trampoline but didn't see herself as active or into sports at the time. Holly's surgeon made the decision for LSS when she was 13 though Holly wanted a rotationplasty or amputation. Holly later made the second decision for amputation when she was 14. Holly and her mother joined me for a video interview from their home in Pennsylvania.

Issue domain: No options. Holly said her first problematic issue was not having any options presented by the surgeon, "He never really talked about anything. It was very much like, "Okay. This is what we're doing." There was no options presented."

Issue domain: Options are discovered. Holly and Gloria researched and found that there were options besides limb salvage surgery:

> The options that we knew about which was the rotationplasty, which she actually would have loved to have done. We did our own research at home, through Facebook groups and through—I mean, there's so many people who put their story out there. At this point, that's really good information. So, we just did our own research. We knew it was going to be between amputation and rotationplasty for her. That's what her two options were, that's what she wanted. He refused. Every time we'd bring it up, he wouldn't acknowledge it… if he did, he would just say, "Oh, but I think we're going to do this." Then, he'd just leave or start talking about something else. There wasn't even any discussions about the limb salvage procedure itself.

Issue domain: Long-term outcomes. Gloria and Holly read more about LSS and were finding mixed reviews on long-term outcomes:

> I really think that the lack of communication with the mobility aspect of it, is a huge problem. There is no conversation about what—even when you ask, it's, "Oh, the sky's the limit. The more you work, the more you do this, the more…" It doesn't work that way. There are kids that put in and put in and put in and don't get past 90 or don't get past 35 for the bending of the knee. You got to 10, 15 tops, sweetheart… I want to see the percentages on that….still, still, there is some people that have really great results.

Issue domain: Recurrence risks. The risk of recurrence was also a factor in their decision-making as they found conflicting information between what the surgeons said and what they read on the rates of recurrence for each:

> There are surgeons that seem to be giving the information out that there's no difference between a rate of reoccurrence with limb salvage comparatively to amputation. I have yet to see a single shred of anything to back that up. Everything that we were given, was the opposite. It was very much that the

> amputation was further away from the site. That, in relationship to the rate of necrosis, really played a huge difference to the rate of reoccurrence. She was like, you know, I want a really good chance of not having a reoccurrence. I want to make sure we've got it all. That's a really good way of doing that.

Holly had concerns early on about limb salvage after researching with her mom. According to Gloria, Holly said, "I do not want to have to have surgery after surgery after surgery. I don't want to run the risk of having all of these infections. I want the highest possible margins."

Issue domain: Gender bias. When they brought up rotationplasty, the surgeon touted it as not for girls:

> Rotationplasty, for us, was like a dirty word… He had said to her at one point, not liking to do rotationplasty especially with a girl because of body image issues. Then I was like, you've got to be kidding me? He just said that.

Issue domain: False hope for function. I asked Holly about her goals for activity when she was diagnosed, "I wasn't overly active. It was mostly just the basic things and also having the option to do something if I ever decided that I wanted to." Gloria added, "Yeah, she liked to take walks, ride her bike, jump on the trampoline with friends. She was not athletic by any means. She's a book worm." They asked about jumping on the trampoline, something Holly wanted to be able do:

> We asked about it and he's like, "Well, I mean technically you're not supposed to but I don't want to hear about it." That's so backwards. It's like it gives you that little bit of hope that, okay, well maybe. That's not the case."

Issue domain: Poor communication and no values elicitation. Holly described her surgeon's communication style, "It kind of was like he was talking to a brick wall. Like, he wouldn't face anybody. He would just start talking and then, he would leave." There was no values elicitation about what Holly like to do or wanted to do; in fact, according to Gloria, "I don't think he ever asked a single personal question, definitely not because that would have been an interesting conversation."

They tried to get a second opinion, someone suggested by their first surgeon, "Come to find out, her surgeon called the second opinion doctor before we met with him. So, he would not discuss the amputation with us either." Ironically, the first surgeon was recommended because he also had osteosarcoma:

> That was why our oncologist referred us to him to begin with. He was in the same area, he was well-known in the community because of everything. So, he was like, "This will be a great fit because he's been through what you're getting ready to go through."

Interestingly, the surgeon had limb salvage complications, "He never ended up healing. He ended up with infection after infection, revision after revision after revision, to the point where they told him he was done, he had no other choice other than amputate."

Issue domain: Limited time for decision-making. Holly wanted an amputation but it was too late to get the second opinion. Ultimately, the first surgeon made the decision to get the LSS due to the timing of surgery:

> I got a phone call back from them, probably about 30 minutes after she was taken back to the OR. It just didn't come in time. Like, at that point, we had to go. You have to make a decision. You only have this tiny little time frame filled with chemo craziness. It was never, "Make your decision." It was, we were told, this will happen."

Issue domain: Incorrect implant. Holly had complications immediately following limb salvage surgery. Gloria said this was predicated in part on an incorrectly sized implant:

> He's technically not a pediatric orthopedic. Like, he operated on everybody… so her implant wasn't a pediatric implant. So, it was too big around for her tibia. That was the smallest one he could find. When he implanted it, it cracked the tibia down. He told us, he was like, "Well we cracked the tibia." I was like, "Okay." He was like, "It won't be a big deal. It'll heal." Nothing heals on chemo, buddy. Nothing.

After Holly developed more complications, Gloria knew they had to make a change, "I couldn't fire him myself. We literally were like, 'No, we're done'…our oncologist emailed him and was like, no more. He was like, 'Well, she's never going to find a surgeon in the world to amputate that leg.'" With the endoprosthesis visible under the skin, the leg turned orange and a raging infection that alerted the CDC; they were told Holly was toxic. Holly's oncologist supported her desire to amputate and set the family up with another surgeon that both Holly and Gloria described as amazing.

Issue domain: Surgical skill impacts options. Retrospectively, Gloria felt that the surgeon's skills should not affect the patient's options:

> This sounds probably really rude on some aspects but it needs to not be about the surgeon. If you can't do something, send these kids to somebody who can. Don't let them settle just because you're not to that level, yet. You know, like with Zoe, she had the tibia turn back. I said that right, right? So, she had that but if she would have had a surgeon that wouldn't do that, she would have ended up with a shorter limb, the whole nine yards. Don't do that. There's a lot of times where I feel like with the rotationplasty, it wasn't really given even a thought because he was like, "Oh, I can't do that." You know what I mean? So, it's like, okay. I don't know if somebody could. You don't have the time. I know a lot of the oncologists do Tumor Board where they go in and they discuss the patients in that pass. I know sometimes it's the surgeons that are involved in that, also. I don't think on that same level where it's treating them as a community and not just, "Oh, well. I'm the surgeon."

Issue domain: Prosthetic needs. As Holly moved forward with plans for amputation, more decisions had to be made. At the time of their interview, it had been months since her amputation and they just met with their prosthetist:

> That's the other thing. I had no idea walking into the amputation. We were set on the amputation, we knew there were things she was going to be able to do. We just met with the prosthetist two weeks ago. That aspect of it is still very, very new. We have no idea. How long can you wear it? You know what I mean? We have no idea, we'll get there, but it's a learning curve. No one just tells you…

Gloria and Holly experienced poor communication issues with the surgeon, describing him as both non-communicative and authoritative. An incorrectly sized implant further complicated issues post-operatively. Adopting an informed decision-making style, they were proactive in seeking a surgeon who would amputate. Even though the amputation was very much desired, they felt unprepared for the next steps – meeting with a prosthetist. Not surprisingly, Holly scored high on the decision regret scale.

Vignette 7: Ingrid

Ingrid had been diagnosed with a benign fibroma at age 18 which was removed surgically and not monitored afterwards. Three years later at age 21, she was diagnosed with low grade osteosarcoma and had a limb salvage surgery a week after her diagnosis; she did not receive chemotherapy. Ingrid has extensive experience in health and medicine; in addition to a degree in public health, she juggles two part time jobs in medical arenas while going to nursing school. She has a first-degree black belt in taekwondo and "loved being active, going on walks, hiking, riding bikes, being out in nature, and just living life to the fullest." But she had chronic pain and mobility issues and was exploring other options for surgical intervention. Ingrid was 23 and living at home with her parents in Florida when we met for her video interview.

Issue domain: Complications from LSS. Ingrid's surgeon offered all three options at her consultation but she and her parents waffled at the unusual appearance of rotationplasty and the invasiveness of amputation:

> So, of course, as my parents and I are thinking about it, obviously save the leg is going to be the first line of defense. My doctor said that's the one that he believed that was going to work best. He knows how to do those, you know, that's what he does. So, that was how we made that very quick decision was because it was just brushed over, the rotationplasty and the amputation. The rotationplasty was a creepy looking surgery at first, you know what I mean? When you first look it up, you're like, "Whoa. What is that?", you know. Even though now knowing better, knowing that maybe I should have chosen something like that and I'd be able to lead a more active life and live the way I'd like to versus with being stuck with my leg. I'm in so much pain, I can't ever run or jump again. I can't go to a trampoline park. I can't climb. I can't do anything. So, that's kind of hard.

Issue domain: Limitations not explained. Ingrid's surgeon did caution her against some activities though she was unaware that limb salvage would affect her life as dramatically as it did:

> I was very transparent about everything that I did. He told me I probably wouldn't be able to go skiing. I probably wouldn't be able to do hard activities on my leg. That's all it was explained as was hard activities. It wasn't really explained that, "Hey, you really won't be able to run" or "Hey, you're going to be two years out of this and you still have to go up and down stairs one leg at a time because you can't bend that knee to put weight on it or else it will collapse." So, it was very much a "I don't know if you'll be able to do taekwondo again. Probably not, just the twisting". It was just, like kind of wishy-washy, not really clear like, "No, you will never be able to jump on it. You will never be able to do this."

Issue domain: Incorrect implant. While many people have to wait for a custom sized implant, Ingrid's was ready at her hospital. As an adult, she didn't question the readiness of the endoprosthesis and was anxious to have the tumor removed. In the absence of the need for chemo to shrink the tumor, it made sense to move forward quickly with the LSS. Soon afterwards she began experience pain in her knee, "Doctor didn't believe me when I said my kneecap is killing me…. So, we found that my prosthetic had ground off all of the cartilage on the back of my kneecap. So, it was just metal on bone." She had that cleaned out, replaced plastic parts, and added a button to the back of her patella. Ingrid felt that the implant was larger than the bone it replaced, causing early wear on her cartilage and lengthening that leg by one inch.

Issue domain: Chronic pain. That was almost a year ago; she was still having issues and sought additional pain management, "So, I have been rehabbing since then. My pain hasn't gotten better…. I ended up having to get a spinal cord stimulator put in to try and see if it would help manage the pain for my leg." But the stimulator was unsuccessful, and Ingrid was feeling trapped with no good options for keeping her leg:

> Didn't do anything. So, here I am with half a metal leg, and a battery pack on my side with electrode wires going into my spinal cord that I can't take off. I just feel like I'm robotic now, completely messed up, nothing's working. That's kind of my story. That's where I am right now. I just had the spinal cord stimulator put in about two to three months ago.

A surgeon who was further away said she was eligible for a rotationplasty if she could get there. But he also wanted to try some other pain relief options first; Ingrid felt like her options were dwindling:

> He does want to try some genicular nerve blocks first to see if those would take care of the problem of the pain in my knee. They believe that I developed complex regional pain syndrome. So, since they believed that I developed that, they're like "Well, we don't really know what to do with that now." So, either I go for a more radical approach and go for an amputation or go for a rotationplasty, or I just try and live with this leg that is killing me 24/7.

Issue domain: Impact on daily living. The impact of these complications was dramatically affection Ingrid's quality of life:

> Now, it's exhausting for me to walk from my car to inside a store, never mind try to get around a store to do to my shopping, to do my grocery shopping. I'm just too exhausted because I'm in so much pain. Now my limitations with my life are, you know, I can't be as active as I want to. I'm in a lot of pain all the time, so it's really hard for me to go out and enjoy things. Even just sitting at the movie theater, I can't have my leg bent for that long. I can't sit still for that long. The weather is bothering me today, so trying to walk around. I try to sit as much as possible.

She's tried using arm crutches to increase her mobility but didn't like how people responded to her: "as much as it helped, I didn't like the way people treated me at work."

Issue domain: Unprepared for limitations. Ingrid never knew about the uncertain outcomes of LSS until she went to the osteosarcoma conference and heard speakers talking about it:

> I went to the conference and basically all these doctors were saying how awful limb salvage was and how rotationplasty was the way to go. I'm sitting there like, "I wasn't told I wouldn't be able to do this." They said one story about this guy who had a limb salvage, everything was fine, he walked into the store and it fractured on him. He had to cut his leg off. That was it. That obviously terrified me, being two weeks out of surgery. So, yeah, no I felt like I wasn't really given the, "Hey, you're really not going to be able to do any of this ever again." It was never really conveyed to me how walking on a limb salvage leg is like walking on a loose door hinge. Like, it collapses all the time. If it's not locked up completely straight, it will collapse on you. Then, you're at risk of falling. If you fall, that's definitely bad.

Issue domain: Genetic risks for offspring. One of the reasons Ingrid attended the conference was to learn about genetic risks for osteosarcoma: "…at that point, basically there's cancer genetics that I was interested in. I didn't know if I would be able to transfer this to my children or something like that. That was something I was very concerned about."

Ingrid was also interested in anything she could learn about limb salvage surgery:

> I was like, "I want to know more about it. I want to know more people who have this and stuff like that." I went to anything that said limb salvage, or even just interesting ones to me. They did a reproductive health after chemo with these kids. That's something a lot of people don't think about. So, for me, that was something I was just interested in general.

Issue domain: Patient reported outcomes and peer support. Ingrid's search for long-term outcomes and connection with peers was also a knowledge gap that she hoped to fill at the conference:

> So, I was looking long-term and I was also looking to connect with more people who had limb salvage surgeries. A lot of the kids that I saw at the conference had rotationplasties or amputations. I didn't really get to meet another person that had

> a limb salvage. The only other person was this really, really little girl. She was not interested in talking to me in a wheelchair. So, that was a little hard. You know, it's just that kind of thing. I just wanted more information to see if there was new technology coming out, if there was anything to make it better or what kind of direction limb salvage surgery was going.

Issue domain: No pain resolution. Another issue for Ingrid was the lack of pain resolution for her chronic and debilitating condition. Her pain was so bad that she attempted suicide earlier in the year. Unfortunately this was the outcome that garnished the most attention from her surgeon:

> So, that's why my thought was, "Okay. This limb salvage, they say nothing's wrong with it. They say everything's perfect." It's clicking, it's popping, it's grinding, it hurts. I'm miserable every single day and I don't want to live my life like this. I can't walk on the sand in the beach. I can barely walk in the grass because it's uneven terrain. Plus, having a leg that's a little bit longer is really difficult. So, for me it was more of a, "I'm fed up and I'm so depressed from having to live my life this way" that I can't do it anymore. I straight up told my doctor, "Look, I attempted suicide in February." That's when he really understood. That's when he really understood that, "Hey, okay. This is really mentally affecting her."

Ingrid's doctors did not seem to take her pain complaints seriously and that was a regretful for this vibrant young woman:

> I'm frustrated that my doctor blew off my symptoms for so long. Oh, it might be a fracture. It might be this. Let's do all these other things to try and treat it. You know, I was in a full hip to ankle locked out brace for three months. Three months, living like that with my leg completely straight because he thought it could be fractured or something we couldn't see. Instead of just doing the arthroscopy, going in, taking a look, you know. I feel like doctors, in some cases, are too careful. I think for my situation, he was definitely way too careful. I had suffered for way too long. Even the limb salvage surgery, with him knowing my history with how active I was and everything, just not really giving me that reassurance or that guidance there. I really think being too conservative especially when it comes to a cancer like this, can be a detriment at times. Especially with the limb salvage surgery, if something's going wrong, if you're saying something hurts, don't blow me off and say, "No, it's looks fine. It looks perfect, prosthetic's perfect." It's not perfect if it's causing pain.

Ingrid wishes there had been more transparency about the complications that can come with LSS:

> Probably just all of the limitations, because I don't feel like those were adequately conveyed to me about all the limitations that I would have and how much it would affect my life. So, if there was a little bit more honesty surrounding that.

Issue domain: Decision regret. Looking back and with the knowledge she has now, Ingrid said she would have chosen rotationplasty from the beginning:

> Limb salvage…if everything looks fine and technically works fine, they're not going to declare it failed. So, then insurance companies are not going to cover

> rotationplasty or anything. So, I definitely would have liked to consider the option before I got my limb salvage. I would have rather gotten a rotationplasty as a result of the cancer to get rid of the cancer….it would have given me a natural joint that already knows how to move…The artificial hinged knee is not working. I'd rather have something, even though it looks bizarre, even though people would give me weird looks and all that kind of thing, I would rather have a natural joint to be able to move on with my life….So, yeah, that definitely would have been my number one choice, knowing what I know now.

Issue domain: Second decision. Ingrid's relationships with both surgeons was good. Even though her limb salvage surgeon did not forewarn her of limitations and potential complications, he recommended the rotationplasty surgeon and said he'd attend the procedure. He supported her values in making the best decision for her:

> My doctor said, "Look, if this is what you need to do, if this is what's going to help you, I'm going to come watch the surgery because I've never seen one before. You need to do what's going to be best for your life and your lifestyle."

But Ingrid felt like her second surgeon really understood her and believed her pain experience:

> I got a very good vibe from him. I got from him that he was telling me what he would do for his daughter. You know what I mean? I felt that kind of personal connection where he really saw me, really saw what I was going through.

She believed he was trustworthy and was empathetic to her plight: I really trusted that other doctor because he looked me in the eyes and said, "I'm not going to let you suffer. I'm not going to continue to let you suffer. I will do it."

Retrospectively, Ingrid felt that all information, including the unknowns and risks, should be discussed due to the invasiveness of the disease:

> I think the more information you can give a patient, the better, especially when you are dealing with something as lifechanging as losing a limb or dealing with bone cancer. Bone cancer is absolutely one of the worst there is just because of its effect on your system. It's your structure, your core structure are your bones. When that's affected, it can definitely limit your mobility, your arms, anything. I mean, poor people with it in their jaw. Its lifechanging, so I think as much information as we can give patients to understand what we don't know and what we do know. All of that for a better, I don't think that can be a bad thing.

Ingrid was unaware of the potential complications associated with LSS when she chose that option. She was unprepared for the limitations, frustrated with her mobility issues, and so debilitated from chronic pain that she tried to end her life. Her medical team was not responsive to her deteriorating condition, leaving Ingrid feeling unheard because the leg "looked" good. She trusted her second surgeon when he said he would not let her suffer anymore. Finding someone who would believe her desperation was problematic. She tried multiple interventions to manage the pain and improve mobility but ultimately chose rotationplasty as the best second decision for her. In time, she felt supported by her first surgeon who would attend the rotationplasty surgery to learn more about the procedure.

Vignette 8: Joy, Kent, & Levi

Levi was an active 10-year-old when he was diagnosed with osteosarcoma in the proximal femur. He came from a close family of six, played football on his school team, and had dreams of being a fireman. Levi's options were amputation, rotationplasty, and limb salvage surgery; he and his parents decided that limb salvage surgery with an allograft was the best choice for him. Now 24, Levi and his parents, Joy and Kent, invited me to their home in New York for an in-person interview. I was greeted by the family dog and fresh coffee as we sat at their dinner table.

Issue domain: Insurance coverage. Once a diagnosis of osteosarcoma was confirmed, Levi's oncologist recommended a surgeon at a highly regarded hospital system, Levi remembered his words, "If this was my child, this is the only doctor I would go to, which is Dr. X for the situation." But Joy had concerns about going out of network:

> Dr. X doesn't take our insurance. I'm like—now, of course, you can't help but think, "I got to stop working now." You can't help but think, "I've got four kids. I've got bills to pay. He doesn't take our insurance. How we going to pay for this?"

The interpersonal connection between Dr. X and the family was warm from the beginning; despite the financial concern, Joy felt good about the surgeon, "We just felt really, really comfortable with him from the moment we met him. I can read people really, really well." She was even more heartened when he said his team would take care of the finances, "He's like, 'Don't even worry about the money.' He's like, 'We'll deal with it. We'll take care of it.'"

The early building of this relationship also helped to establish trust in the surgeon to whom Joy would rely on for research, "We also had three months. It's not like, 'All right. You're diagnosed. You get your leg tore up tomorrow.' So, I looked into it. I did my research talking to Dr. X."

Issue domain: The options. The family was given four options: rotationplasty, amputation, limb salvage with a metal endoprosthesis or limb salvage with allograft or donor bone. Joy described her reaction when the surgeon explained what rotationplasty was like:

> I'm like, "Hell, no. I'm not doing that to my son. No, absolutely not…We did meet a girl at Camp Sunshine that had it done and this poor girl looked absolutely miserable. I mean, everybody was looking at her. You know, here she is a young girl with her foot on backwards. It's just—it's weird.

Joy was concerned about her son's self-esteem and self-image with respect to the unusual appearance of rotationplasty, "For kids, it's hard, you know? How do you meet somebody when you get older, you know, get married? I'm sorry, people are cruel, they just are…we weren't going for that." Levi remembered hearing his mother's response to amputation as he waited inside the exam room, "I overheard them outside talking about amputation and her. My mother speaks freely and loudly, okay. Through a closed door, I heard, 'Absolutely, not. We will not be opting for the amputation.'

Issue domain: Weighing risks and benefits of two options. Fifteen years ago, leg lengthening surgeries were performed to accommodate the growing limb of a child until the child stopped growing. The intervention was more invasive then than it is now, Joy said, "They would have to remove it and put in another rod that was," Levi finished, "…at the pace of my growth plate." While some children this age are tall, Levi was not, "I was short and stubby." This was a concern for Joy who didn't want her 10 year old son's life interrupted regularly for surgical intervention – every six months with a 1-2 month healing period, "For one, the metal rod, twice a year, he would have to go for surgery. That's ridiculous…I wanted him to go through the least amount of surgeries." In addition to adding to the surgical load, she worried about the stress of repeated interventions, "we just felt that that was another nightmare for him to put him through."

Issue domain: Values and patient preferences. Another factor in making the surgical decision was Levi's long-term quality of life. Dr. X shared stories of other patients who were active with the vascular fibular allograft which alleviated Joy's concerns:

> He had a lot of previous patients before you that played all different types of sports. He would show us. Remember every time we saw him, he would show us his other patients, where they were at, at what stage they were back to playing sports…

Issue domain: LSS as two-step procedure. Levi remembered seeing pictures of kids playing lacrosse and another kid playing soccer. Joy said this was the same two-step type of allograft LSS that was on the table for Levi; she felt these success stories spoke to the skill of the surgeon, "Same type of surgery, yeah… the man knows his stuff. He definitely knows his stuff. I know people come to see him from all over the country. I know that for a fact." Levi was involved in the decision-making process. Kent felt it was important that he be included in the discussion, "You have to say, they have to have some say. It's their body."

So Levi talked to Dr. X about his concerns and while high impact sports was not an option with the first step initial allograft, with the later vascular fibular allograft, he would have a stronger leg:

> I had fresh thoughts of wanting to be a fireman. I'm never going to be able to do that. Football, I'm never going to be able to do that again. So, as I started to speak to him and kind of ask him about those things, he would explain to me the—what's the best word for it? The delicacy of the femur that they had placed

> into my leg, which again I referenced what he referenced basically to a sheet of glass with a metal support. So, it could—a wrong twist, it could break. Wrong amount of pressure, it could break.

Although the donor bone allograft was “delicate” and Joy said, “We were told it’s possible it wouldn’t work or it would break,” the family felt this was the best decision. Due to chemo, Joy said they knew they would have to delay on the more extensive vascular fibular allograft:

> If he wasn’t so weak and all that because of the chemo, the vascular allograft would have been done right away, from the get-go. Because it was such a long, extensive surgery, we did the allograft first. We knew it was going to come. So, we were told everything from the get-go.

Levi explained that the allograft donor bone is considered a dead bone, “So, there’s no calcium, there’s no blood flow, there’s no bone marrow. It doesn’t build itself back up. It’s basically just for structural support with the metal plate that they installed in my leg as well.” He was in a wheelchair for six months after the surgery and then on crutches for awhile but his parents felt it was “the best option that you could have for you to have your leg and be a normal kid again once everything is healed up.”

Issue domain: How long would the allograft last? Levi continued to live his life for the next 8 years knowing that one day the surgery involving both legs in the vascular fibular surgery using his own bone would be necessary:

> In the back of my head, because I felt normal. Because of the original limb-sparing surgery I had, everything healed. I could run and jump for the most part. A little loss of mobility in the amount I can bend my knee or certain ways I could jump. Other than that, I could be kind of like was making up for my childhood…

Levi played handball and worked three jobs during high school. His passion for the Fire Department was realized when he joined the junior company at 14 and participated twice weekly. He knew the allograft wouldn’t last forever but didn’t know how long it would last.

Joy said it was uncertain how long the donor bone would last but Levi felt prepared for the inevitable, “With the allograft, he wasn’t able to do everything he wanted to do because it was such a fragile bone. We knew eventually it was going to break, it was just a matter of time.” Levi added,

> It was one of the inevitable things. I remember him saying, ‘Some people’s cadavers last a year, last six years. It could last 15 years. It could last six months.’ I knew eight years later. Eight years later, I still knew. Okay, when I was 10 and we did this surgery, it was explained to my mother and my father and me.”

Framing the break as a complication that was expected helped the family feel more prepared. Joy said that knowing the allograft would eventually break, lessened the blow when it did, “We knew. We already knew what was going to happen which we were informed, so it wasn’t a shock for us.”

Issue ddomain: Broken implant. When Levi turned 18, he was in the Fire Department on probationary status, a particularly grueling time for new members when they had to grunt work and push themselves above and beyond. It was then that Levi realized his leg had broken. Joy rcalled his reluctance to go in, "He finally came to me and you know he put it off for weeks to even say anything to me." Levi owned up:

> The reason that I personally pushed that off for so long was because in the back of my mind, I knew what the next surgery was, was the 14-hour to remove the bone. I really don't know all the specifics at that point. But I knew it was big and I was going to be put off my feet for a while and it was going to suck.

This surgery would be Levi's eighth surgery; it was extensive and long, involving both legs, but it was expected. The procedure is a vascular fibular allograft where the surgeon transferred the fibula of Levi's good leg over to his salvaged leg and wired it together. They shared a picture of the wire wrapped bone with me. This was a very special moment for both Dr. X and for the family according to Levi, "One of the reasons he calls me his miracle patient is because he did that surgery."

Issue domain: Drop foot. Immediately after his vascular fibular allograft, Levi had knew he had complications:

> Moving the lower right fibula to the left leg and fusing everything, putting the veins, the arteries, the muscles, everything together right. Everything just went absolutely miraculous. Woke up, couldn't feel my leg, basically from my knee down. Couldn't move my foot. So, I ended up suffering from what was called a drop foot… absolutely unexplainable reason why it was happening.

Levi and his family searched for answers: "I went to doctor to doctor to doctor…I went to a neurologist. Yeah, they ended up putting me in a brace to hold my foot up so the tendons didn't get stuck down. They couldn't figure out, couldn't figure out." Time is of the essence in for nerve repair:

> The scary part about that was, I went nine and half months with a drop foot. They say, typically, when you have zero reaction in a nerve—so, it was a perineal nerve of my left femur spreading down where it branches down through your lower leg that was cut off. It was literally a tiny, tiny pinch that was cutting it off…Twelve months after no activity through a major nerve for 12 months. It basically will never come alive again.

The surgeon decided to explore the nerves and in preparation for that surgery, he moved the wire out of the way that was close to the nerve. Suddenly Levi could move his foot, "My foot started move like a quarter of an inch. I remember him like a kid in a candy store, 'Nurse! He moved his foot.' It was such an awesome moment." Dr. X later told Levi, "I had a feeling before we went into this aggressive intrusion of a surgery looking at every nerve and you know, we'll try the wire."

Joy felt that the problem-solving was due to Dr. X's expertise: "That's just the insight in how smart this man is, like, 'Maybe it's the nerve pinching the wire.'" But Levi disagreed:

> Hold on; pause. I love Dr. X. I won't give him that credit because he had no idea. He'll say it today. He'll own up to it. He will. He will. He had no idea that that had anything to do with the reason that my nerve was pinched off. That was in preparation so it wasn't in the way for that exploratory surgery. The fact is, like, he's like, "I've never had that in any surgery that I've ever done before."

One of the building blocks to trust in this patient-provider relationship was honesty, even in times of not knowing the cause (dropfoot) or outcome (lifetime of the first allograft).

Issue domain: Late-term side effects and long-term outcomes. While Levi had a successful experience with limb salvage surgery, he was well aware that this is not often the case:

> Very, very few cases have I heard of but there definitely has been a few cases that I've spoken with people in regards to their limb salvage surgery that they were just super unpleased with….Splints in the bone, fractures in the bone, a year later, six months later.

As a firefighter, Levi was in a unique position as both a service provider and a pediatric cancer survivor. He felt called to offer support to other kids in similar situations when asked:

> Few friends from the Fire Department or somewhere along the line, somebody know my story and was like, "Hey, I know you're always about helping kids that are battling cancer, that having a struggle spiritually and mentally and with attitude. Can you talk to them?" You know, sure.

He has an understanding for how these experiences affect him and is careful to protect himself when helping others:

> I try not to overload myself because I started to realize working with too—I don't know if it sounds bad, but working with too many kids at once and then, constantly seeing these kids lose their lives or just not the outcome that I want to see was mentally impacting me and completely throwing me off course in my life. So, I was like, as much as I want to do that, I got to kind of veer away from it a little bit so I could get things right with me first. If I can't help me, I can't help anybody else.

Some long-term side effects of chemo remain unknown and are still monitored 15 years later: "That's why he's got every year he goes back, he could have issues with his heart. He's got slight upper tone hearing loss because of chemo. You know, like really high-pitched stuff." Levi also has issues with his hip and back due to leg length discrepancy, "The only permanent deficits, I guess you could say, is the shortness of my leg and a little bit of loss of mobility in the bend of my knee." He attributes some of this to rigorous training as a member of the Fire Department and jokingly says it's their fault:

> Well, I mean, I train a lot. So, not a lot—enough to where I'm having hip and back problems. The next conversation is, hey, if I end up needing a hip replacement by 26 years old, it's going through the Fire Department. If they

> don't pay for it, you know like, I'm just going to sue the Fire Department because of that.

Looking back, Levi is glad he was involved in making the surgical decision at the age of 10:

> I think the most beneficial factor out of that, like, for parents is making sure their kids have a choice. If they're old—you know, obviously if their kid is three years old, their kid's not going to have any idea of, you know—if your kid's three years old, they're not going to have any idea of what to say and how to react. If your kid is 10 or 12 or 14 years old, I think it's very imperative that they allow them to have some type of say and make sure everything explained in detail what's going on.

His father, Kent, summed up the family decision-making experience:

> I'll tell you what, the biggest thing—the biggest thing about it is, any decision was all made by the three of us, not one, all of us. We knew exactly what we were going to do, what's best for him…We want him to have every chance that he could to be himself.

Levi and his parents made a decision for surgery that they knew would fail at some point before they could move to a planned second surgery. While they were reconciled to living well with this uncertainty, they knew that the second step would be more intense and involve both legs. Their surgeon communicated clearly and effectively to them and expressed appropriate uncertainty, leading the family to trust him more for his honesty. They felt prepared throughout the process by a surgeon who kept them informed about what to expect. When the break did come, they were ready for another surgery. Although Levi had had many surgeries to repair minor breaks and some alarming complications like drop foot, he scored low on the decision regret scale.

Vignette 9: Mona (Mandy)

Mandy was 13 when she was diagnosed with osteosarcoma in the distal tibia near her ankle and had an amputation. Mona and her husband had busy lives with three teenage daughters (13, 16, 17); they were all athletic and active in sports. Mandy was a competitive swimmer when her tumor was discovered. Mona and I agreed to meet at the hospital in Delaware where Mandy had an appointment; she arranged for us to have a private room.

Issue domain: Two options. Given the location of Mandy's tumor, rotationplasty was not an option. Rotatationplasty would be an option only when the knee joint needed to be replaced by an internal or external prosthetic joint. Mandy's options were below the knee amputation or a cadaver bone allograft limb salvage.

Due to timing, Mona and her husband ended up meeting with the orthopedic surgeon before they knew the confirmed diagnosis:

> The orthopedic surgeon all of a sudden had availability and could see us. So, we didn't find out anything from oncology but when we met with the orthopedic surgeon that was the beginning of the relationship with him.

Issue domain: Down syndrome limitations. Mona described his unique patient-centered approach, "He was really good even then about keeping us focused… when we were with him, we always felt like we were the only people he was working on, you know what I mean, treating." He even drew a picture to help Mandy understand her diagnosis – Mona pulled it up on her phone and shared it:

> I have it on my Facebook page, this picture that he drew. I can show it to you, but I think the visual really helped us understand how big the tumor was and what it meant and where it was on the leg. He just sat there, freehand drew it. That really helped us, to make him do that visual. It definitely helped Mandy because with her having Down Syndrome that visual always helps.

There were ways that the surgeon demonstrated a desire to build a trusting relationship with the family, such as empathy and active listening, "Even when he first came in and had this, you could just tell he was in pain having to tell us. He answered all of my husband's questions. He was responding. He was respectful." The surgeon continued to build a relationship of trust with Mandy and gave her control over unwrapping her bandages, even if it took hours:

> She's limited, not limited vocabulary, but a little spotty when you meet her with her vocabulary because she has Down Syndrome, in thinking about what she wants to say. She was just like, "My rule is one person touches my leg and that's me." I mean, that was it. Like, nobody was touching her. He would come in and he would be like, "Okay, Mandy, we need to look. We need to change the bandage." He's like, "You unwrap it and I'll come back." He's like, "I'm going to go make a couple phone calls, see some patients." He would come back forty-five minutes later, come back and she's still doing it. He's like, "Okay, keep going." He was very patient with her. He would come back and it would take him two and a half hours to change her dressing from start to finish. He would go do work in between, and then come back and check on her. It was just his personality.

Issue domain: Elevated infection risks and recurrence risks. Mandy's diagnosis of Down Syndrome included physical limitations too; there were complications to consider when weighing the risks and benefits of her options. She has a compromised immune system in addition to having cancer, so the cadaver bone was an elevated risk. The surgeon presented the research to Mona, her husband, and all three girls:

> He rattled off some statistics on what the infection rate is. To throw that general infection rate in, then add having the cancer treatments and then having her have her compromised immune system, you're taking an already risky situation where there would be a risk of infection and throw in the cancer and Down Syndrome on top of it. It just felt like there was going to be a really tough battle for her if there was an infection. That could really impact how successful it would be if we did limb salvage.

Mona’s husband saved his notes:

> There are essentially two options for addressing the tumor. Remove the tumor and all affected bone, including clear margins with the bone and soft tissue. This option would involve inserting a metal rod from the bottom of the foot through her tibia then fusing the foot to the leg bone, thus immobilizing the ankle and the foot. In place of the bone that is removed, a cadaver bone would be surgically implanted. There is a 25 to 35 percent risk of infection, along with the potential of the rejection of the implant. Additionally, there is a risk of not removing all the cancer.

He compared this to the risk of infection with an amputation – which also impacted the risk for recurrence:

> He marked that the risk of infection is in the range of 1-2 percent when you do the amputation, because you’re getting rid of all the cancer. You just know you got rid of it. There’s not this hoping you get the clear margins.

Issue domain: Short term vs long term quality of life concerns. Mandy also had metastases to the lungs at diagnosis which further impacted the surgical decision. If the family chose limb salvage, Mandy’s ankle would be fixed, “That would impact her ability to move around and do things. Then, honestly what did it for us—what the kicker for us was that she was not going to be able to bear weight on her foot and do a whole lot for a whole year.” Mona and her husband were concerned about Mandy’s quality of life given her advanced stage of disease:

> At that point, it’s February and we don’t know how much time we have left…you’re in survival mode. If you don’t know what you’re looking at, why do you want to not be able to move around and bear weight and do things? She was a swimmer. She was athletic. We liked to do things. You know, we did a lot of activity. We just couldn’t imagine that she would just have a fixed ankle and not be able to put weight down on it, and what kind of life would that be.

Mona also worried about what it would be like long-term for Mandy if they chose limb salvage, “If her ankle was fixed, she would be dragging the leg around. I see it now with people. People who end up being uncomfortable not being able to do what they could do, not being able to run.” While the evidence was starting to mount for one option over the other, Mona wanted to be clear that they were given choices, “We were presented with options. I feel like he didn’t come and say, ‘This is what we’re going to do.’” Mona and her husband decided to bring Mandy and her sisters together with the surgeon so he could explain it to them, knowing there was no good choice, “We knew what we wanted to do, but then we brought the girls in and he explained it all. Mandy just was like, ‘Let’s do the amputation.’ It was that easy for her to make that decision.”

The bulk of the research, from statistics on the risks of infection to the risks of recurrence, came directly from the surgeon whom the family trusted. Mona said, “I feel like he did present it and I really believe that he told us the facts. I think that he was very honest” but she also said, “He may have wanted to do the amputation and thought that it was the best thing” though he never said so.

Their surgeon also advised them about changes in the field of prosthetic devices. He said that Mandy’s options for being mobile have never been better, “He mentioned, given the Gulf War and what type of technology changes there have been, the world of prosthetics has definitely changed from the old days…you can run and do a lot of different things with that prosthetic leg.” He also reminded them that that the goal of treatment was to get rid of the cancer and Mandy still had metastases in her lungs:

> …now that I’m thinking about it, that was probably another thing that he mentioned, that we need to kill the cancer. That’s the first thing that we need to do. We need to get rid of it. We need to kill it because we still had stuff in her lungs that we were trying to get rid of.

Mona reviewed all of her concerns about choosing limb salvage over amputation while thinking about what the surgeon told her:

> So, I think it was more we just didn’t want her walking around with a fixed leg. One, not being able to bear weight for a long time. Whether that was an exaggeration, I don’t really know. He was thinking she wasn’t going to be able to bear weight for a year. So, for her to—you know, maybe that was a lot, maybe not. I don’t know how long it was going to take her to heal. Any type of a little infection, would then impact her ability—that was the other thing. Any time there would be an infection that’s going to impact her ability to get her chemo. If your body’s busy fighting that, you can’t douse it with chemo or it’s not going to fight the infection. Any infection that she would get, would have to get healed before the ability to do chemo.

Issue domain: No physical therapy. Unmet needs for physical therapy was problematic after surgery, including the basics of moving her around after her amputation:

> Right when she got her amputation, we’re back in the room and I have an aide and a nurse. I have no idea what to do with my kid who now doesn’t have a leg. How do I get her to the bathroom? She doesn’t know what to do. How do we move her from the bed to the bedside commode? I had aides and nurses who didn’t really know what to do. We’re on the oncology floor. We’re not on the rehab floor. I don’t know what other hospitals do…they’re like, “Well, how do you want to do this?” I’m like, “You all should know how to do this.” Nobody knew. So, then I’m like, All right. Then, get away from my kid. Then, I don’t want you near my kid. If you really don’t know what to do, I can’t afford to have you drop her leg on the floor. I can’t afford to have you mess up.

Mona was shocked and disappointed that Mandy didn’t get regular physical therapy, “I had to beg for therapy. I had to beg for it… Not getting the physical therapy was huge.”

Issue domain: No prosthetic guidance. Mona was equally frustrated by the lack of prosthetic guidance she got, saying she didn’t know what she didn’t know getting a prosthesis: Mona thought about her goals for Mandy and resuming her quality of life:

> I said, “Okay. Well, I want her to be able to swim with her foot on.” They looked at me, and they said, “Well, that can’t happen.” I’m like, “I just looked it up the past couple of days on the internet.” Then, they take me and they’re like, “Oh, well let’s go see.” They start looking it up on the internet. I’m like, and they’re like, “Oh, well I guess you could.” I’m thinking, “Yeah, I don’t think—if you don’t know what your competition is doing, even if you don’t have it, how do—” I’m like, “Yeah, I don’t think so.” Then, I come back up here. I said something to the social worker. I said, “I think I need to find another prosthetist.” She said, “Let me look into it for you.”

For Mona, involving Mandy and the rest of the family in the decision helped pave the way for the amputation decision. They had a trusting relationship with their surgeon who provided the bulk of the information but did not make a recommendation. Thinking through their values and what made a good life for Mandy helped the family feel more comfortable with what ultimately was Mandy’s decision. Retrospectively, Mona talked about the challenges of thinking long-term with our kids when we’re in survival mode and just trying to get cancer-free. Unlike my daughter, Mandy had metastasized to the lungs at diagnosis, making her prognosis less hopeful. Mona said, “I wanted what time left we had to be good.” At five years post diagnosis, Mandy was still doing quite well, and Mona was thankful for their new moniker, “The fact that we’re here five years later is amazing...we’re an outlier…we’re a huge outlier.”

Vignette 10: Nerisa (Nate)

Nate was 13 and living in Texas with his parents and 10-year-old brother when he was diagnosed with osteosarcoma in the distal femur. Nate was an athletic boy and had been playing basketball at the time of his diagnosis. Nate’s mother, Nerisa, joined me for a video interview though Nate, now 17, declined to participate.

Issue domain: Information overload and the options. Neither Nerisa nor her husband or parents (who often accompanied her to appointments) had a medical background. Nerisa said she had zero background and it was all new to her, “I hate doctors and hospitals. I just learned as much as I could.” When she and Nate when to his first orthopedic appointment not knowing what to expect, they were told he had osteosarcoma and a hospital room was already prepared for him. She said that she actually passed out when she was told her son had cancer, “I’m not proud of that. My body, I just started sweating and shaking and I passed right out. I felt bad. Nate was sitting there, you know. Here I am, on the floor.”

One of the first issues she had in the decision-making process was feeling like information was being thrown at her:

> So, the surgeon, when he sent us over to the hospital and once we got settled in the room, he showed up that afternoon again to talk to us. He threw a whole bunch of information at us. One of the biggest pieces of information he said that day was, “It’s going to be best to amputate his leg.” He said, “I’ve been to

> multiple conferences across the world. There's just a better success rate of it not coming back, if we just amputate the leg." All in one day, he told me all this.

Nate's surgeon also mentioned limb salvage surgery:

> …he brought it up. Initially—it's a little bit foggy. You know, so traumatic. I just remember specifically that very first day him going through all the information very quickly. I think he did say limb salvage or amputation, and I recommend amputation. His reasoning was because there was more success in the osteosarcoma not coming back if they amputated the leg.

There is some question as to whether or not rotationplasty was an option or not after they said they were concerned about amputation:

> Once we expressed concern about doing that and what are all our options, he explained only the two. I think I mentioned that to you before. I had never even heard of rotationplasty. My husband says, he mentioned it but he doesn't remember it being an option perhaps, I don't know. I don't remember him talking about it at all.

Issue domain: Appearance concerns. Retrospectively, Nerisa thought it wouldn't have been a good option for Nate due to thee appearance, knowing him and what his quality of life priorities were:

> Well, I don't even know much about it. I've just seen a lot of things on Facebook about children who have had that option. I didn't look much into it because maybe I didn't want to go back and see what could have been. Although, I see the way that it looks and I could confidently say that Nate would have said, no…He would have said no and I wouldn't have blamed him one bit…I could have known that it was the greatest and best decision I could have made but he would not have wanted it and I would have honored what he wanted.

Issue domain: Lack of empathy from provider. Nerisa described the surgeon's bedside manner as "scary" and lacking in empathy:

> Like I said, when he told us what was going on, it was just within five minutes of sitting down and pulling up the MRI. It was very straight forward. There was no 'I'm sorry to break this to you' or not even a tone of that. It was, "This is what it is. It's bone cancer." Very straight-forward, there was a—I feel like he's that typical idea of a doctor that lacks a human component, not very compassionate or I don't know. There were times I saw that with him, as we got to know him more, but he wouldn't hesitate to tell you how it was and how it was going to be.

The surgeon had a physician's assistant (PA) who softened the blow of his delivery:

> …the surgeon had this PA who was a completely different person than him. She was very kind and connected with us more than he did, I feel like. She was more understanding and could see our reactions, whereas he was not seeing anything… it was nice to have her there. I feel like that was beneficial. Having someone else, not just him, to give feedback about what was going.

The manner in which the information was presented was problematic but once the surgeon knew the family was leaning toward limb salvage surgery, he was less aggressive about amputation, "I think his bedside manner, initially, I think he was hot off a conference. He just said, immediately, 'We need to amputate the leg.' Once he could tell we were leaning in that direction, he was onboard and supported us."

Issue domain: Gathering information. The family relied on the surgeon almost exclusively to provide medical information; neither Nerisa nor her husband did research online. The questions they had for the surgeon arose partly from Nerisa's parent's questions: Nerisa knew her mother seeking information online. Nate was present for these decision-making meetings with the surgeon but did not participate much:

> I think we had a lot of questions, so he talked to us directly a lot of the times. When he wanted to say something important about the surgery and the options, he would look at Nate too. I think he was welcome or maybe offered an opening for Nate to participate with any questions or the conversation. Nate didn't say much to him or ask many questions.

Nate did however do some online research of his own, "I just know he was immediately on his phone looking up to see what was going to happen to him. He wasn't trusting the faces in front of him for some reason. It was better online."

While the surgeon's bedside manner was alarming at first, in time they felt they trusted him to do what they wanted him to do:

> ...anybody probably thinks to himself, we should get a second opinion. I did think about that. So, I looked up his credentials and what people said about him online. He had received a lot of rewards nationally and internationally and had been doing it a long time. I did feel confident in his ability to do what we decided to do… his matter-of-fact approach, direct approach, did give us confidence.

Once they decided they wanted to move forward with limb salvage surgery, the surgeon became less aggressive about amputation, "So, the more information we got once we had all the scans and everything, more information at the hospital that first day, he was more open to what we were looking for." He reassured them that he would only do it if he felt it were the right choice, "He said, 'I won't do a limb salvage if I don't think there's a possibility that it's going to be the right choice.'"

Issue domain: Surrogate decision-making. While Nate was involved in the decision-making, he was still only 13 and his mother was mindful of her responsibility to think about his goals and what he would want, "I was trying to think about how Nate would feel. I never thought about anything else but that really, how he would feel about how he looked or what he could do." Her sense of uncertainty around the decision continued to the day of surgery, "Really, up until the day before, I was really conflicted. We all were." That sense of information overload and indecision led her to reach out once more to her son on the night before his surgery:

> It's just too overwhelming. I mean, we did get to the point—even the night before, I said I remember talking with Nate and I said, "We do not have to do that surgery tomorrow, if you don't want to. You're not locked in. I will not bring you there in the morning if you don't want to do it. If you want to amputate, I want you to feel really good about what we're going to do because it's not going to be easy either way." So we had some definite heart-to-hearts like, "You really know, like this is what's going on." So, yeah, we were comfortable going in with that decision and Nate was too.

I asked if they ever considered that they could do limb salvage first and amputation later, "I'm surprised I didn't, but I never thought that. It was just all too much, I think. I was just kind of like, 'We're going to do this. This is going to work. This is what's going to be best.'"

The idea of doing what's best or what's right came up a few other times. Once when they were making the decision but did not have the pathology results back yet, "we didn't know if we were doing the right thing, didn't know, didn't know up until the night before." Once when the surgeon said it, "When we got the pathology results of the tumor, he said we made a right decision." He also said, "I won't do a limb salvage if I don't think there's a possibility that it's going to be the right choice."

Issue domain: Recurrence risks. With the research cited by the surgeon as coming from conferences, Nerisa and her family struggled to make sense of what the recurrence risks were for each of the two options:

> So, I guess you don't really have an idea. You have these benchmarks. You don't really know if it's going to—you don't know if it's going to come back. One of the determining factors, from my understanding, is once they do the surgery and take the tumor for pathology, after treatment has started, at that point they can determine how much of the cancer has been killed from the treatment. So, that determines where you're at in your treatment, which way they're going to go with the drugs, and your prognosis, what percentages they're giving you.

Nerisa wishes the risks had been communicated in a visual way to help them understand the differences between the options:

> I, personally, would have like to have seen written statistics that I could visually see. Something that gave me information about children who have limb salvage versus amputation. Something like that rather than just him saying those things. If he had given us something to look at visually on a piece of paper, that would have been helpful.

Nate participated in the discussion about options but was torn between his desire to keep his leg and his desire to reduce risk for reoccurrence:

> We were very up front with him. He was there for anytime we talked to the surgeon. We told him exactly what the options were and what did he think and what did he feel. He had some input. You know, he would go back and forth just like we did. Mainly, because amputation gave him a better chance of it not coming back according to Dr. X. So, that was our, "Okay, well then we want to

> amputate." I don't think he ever really gave us any solid numbers about it. So, there wasn't a measurement in our minds about that or how many people have a reoccurrence. It was our understanding that those things were—the reoccurrence wasn't solely about whether we amputated the leg or not. It was about how the drugs worked. So, that weighed a little bit in on the decision. He, like I said, went back and forth, Nate did. Really, he really wanted to keep his leg and not deal with an amputation and all the things that went along with it. I mean, that was his inclination, "I want to keep my leg." That's how we felt too.

Issue domain: Unknown necrosis. With the understanding that a low necrosis rate might make amputation feel like a better decision, she felt much better having made that decision and then finding out after the surgery that his necrosis rate was high:

> So, I felt at that point, didn't know if we were doing the right thing, didn't know, didn't know up until the night before. When we got that news, a couple weeks after the surgery or whenever it was that they brought us those results, that the ninety-nine percent necrosis rate made me feel better in the decision that we made.

Feeling uncertain about the decision hinged on knowing the necrosis rate:

> I think we were unsure, but felt it weighed more on feeling that we were going to move forward with the limb salvage. We were unsure but my only uncertainty was I thought that the reoccurrence was mainly based on the necrosis of the tumor. I figured when we found that out, I would feel better or worse about our decision. It wasn't until we received those results that I felt confident.

The surgeon agreed, reinforcing her understanding of necrosis and how it informs recurrence risks, "When we got the pathology results of the tumor, he said we made a right decision. I felt better, more confident at that point." She didn't know what would have happened in terms of surgery if the necrosis was low and the decision was for LSS:

> I guess my understanding from the surgeon and the oncologist was, if the necrosis rate came back low, what the decisions were as far as the MAP at that point. I didn't think about what they would do surgically. I never thought of that.

Nate was active in sports and had been playing basketball when he was diagnosed. Nerisa said that the doctor had informed them of the limitations of limb salvage surgery and his advice post operatively remained the same:

> …his surgeon doesn't recommend that he does any sort of sports like that. He says he can ride a bike. Nate does do some of that. The information he has given us is that it's not going to be good to risk injuring himself in a contact kind of sport. The prosthesis will hold up as long as you are careful about—I don't know careful, but you wouldn't want to do those things because it would reduce the lifetime of the prosthesis.

Issue domain: Chronic pain. When they were making the decision, the surgeon warned of the limitations knowing that Nate was athletic and played basketball, "He did say, 'You're not going to be doing those things.' We knew. Either way, I mean, even if they had done an amputation, there would have been challenges with that." What they didn't know was potential complication of daily pain:

> Had I really had an understanding of the pain that Nate would have to deal with on a daily basis, I may have made a different decision…. I'm not sure that the surgeon and the oncologist didn't do their jobs, exactly. I don't know that I could say that. I really don't remember them really explaining that part of it very well. I mean, I knew that they might have some discomfort or might get achy. That's how he said, you know, it might be but there are days that is hard for him, really hard. I think it really compromises the quality of his every day. Sometimes, he might have a few bad days in a row. If I had understood that, I don't know if I could have wrapped my head around that even if they said it. I'm not sure that we would have made the same decision. I don't know.

While Nerisa was not active online in support groups during the decision-making process, she is now as she seeks non-invasive, non-pharmacological interventions for pain management:

> I went on actually to find out what people were saying about how they treated the pain. I'm always looking for new homeopathic, healthy ways to deal with pain, or alternative, supplements, things of that nature. That was why I went on… It was mainly I went on to find out about how people dealt with pain. I just want to stay current on anything that comes of research or treatment and long-term things. Just so I know what to keep an eye out for or to think of or to share with Nate if I think that it's necessary.

It is in these groups where she also hears of pain free outcomes with LSS and wonders about the long-term effects:

> Then again, I see people online who say they have no pain at all. Then, some people say, "No. I have pain every day." So, I think it's really split. So, I don't know if the surgeon really knows. I think they're really focused on what they can do with the affected bone and leg, and that's what they're going to do. I don't know if they had enough information for us on Yeah, I guess I think the more information you have the better equipped you are to make a decision. So, I think I would have liked him to say, to have told us more about that possibility.

Making the decision was problematic for Nate and his family due to an overwhelming amount of information framed with a surgical recommendation for amputation, an unclear link between the options and the risks for recurrence, and missing information about chronic pain as a long-term complication with limb salvage. Nerisa said a general lack of information or processing of the information shared led to feeling surprised an unprepared for the complications that came later:

> Yeah, I really felt like we were just kind of, the whole things was like, "Cross your fingers. Okay, this is what we're going to do, all right." So, with something so drastic, I think the more information you have the better. Maybe, then again, you don't hear everything. So, I don't know. I don't have recordings of those meetings. Perhaps he said that, at some point, I never got that feeling. I think it would have caught on. I would have remembered it because I am really surprised at what we deal with now.

Ultimately and except for the chronic pain, the family is satisfied with their decision and the information they received to make it:

> Other than what I've mentioned about knowing more about what the pain was going to be like, the potential of it being a daily thing. That's the only thing I can think of. Everything else, there's nothing that I've ever said, I wish I knew then. I feel like everything just worked out how it was going to work out. We made the decision how it was supposed to be made. I feel, don't regret anything.

Vignette 11: Opal (Olivia)

Olivia lived on a farm with her parents and eight siblings in Ohio. She was an active child, playing in the woods, and helping to weed the fields when she was diagnosed with osteosarcoma in the distal femur. Olivia had a limb salvage surgery at the age of 10. Olivia's mom, Opal, and I arranged for a video call – our interview was peppered with little voices in the background and a new puppy who sometimes found his way into the camera field. Olivia was almost 14 at the time of the interview and recovering from rotationplasty. She was on the sofa nearby and sometimes made comments directed to her mom but otherwise did not participate in the interview.

Issue domain: No options. Olivia did not have any surgical options presented to her for her first surgery although her parents brought it up, "We had asked about amputation and the doctors were all like, 'No, no, no. You don't want to do that.' So, yeah, very overwhelmed and just kind of, 'Okay. Well, I guess you probably know what's best.'" Apparently asking about amputation was an anomaly:

> One of the doctors was shocked we even asked about amputation. Told the nurse, I've never had a parent ask me that the first time I've talked to them. Most the time, they're like anything, "You do whatever you have to do, to not amputate."

Opal was aware of rotationplasty – she was a pediatric nurse and a friend's son had it - but didn't think to ask about it as an option for Olivia:

> I don't know that it crossed my mind for her at the time. I think just because I was thinking there was so much cancer in her knee and in her entire joint. So, I wasn't thinking of that as an option even.

At the time of diagnosis, Olivia was aware that a surgical decision was being made but she didn't state any preferences:

> She did know. I think, just probably all of us were really overwhelmed. You know, you kind of wonder, "Am I thinking straight? Am I not?" So, she didn't really say either way, like what she wanted. She knew we had asked about amputation. It wasn't until after the surgery and we came home and she was really upset and wished that we had just done an amputation.

Issue domain: Complications from limb salvage surgery. It was over 8 months after her limb salvage surgery before Olivia was given clearance to attempt walking again. By then, it became clear that most of Olivia's quad muscle had been removed during surgery and she did not have the structure necessary to bear her weight or walk. Olivia had gone from a busy little girl, playing with her siblings to not mobile at all:

> She couldn't really do anything. Yeah, I think we got her back on a horse with help, which she wasn't even supposed to be. With all that stuff and hardly any bone, she was a super-high risk of fracturing it. It just got to the point where it was more important for her to do something she liked, than worry about saving a leg that didn't work anyways.

Opal described what it was like to find out later that Olivia's quad muscles were no longer there:

> We were not prepared. I think they told us maybe six months, until she would be able to walk on them again. Nothing was ever mentioned about losing all of her muscle and the possibility like when you lose your muscle. I know this stuff but when your child's diagnosed, you don't think right. So, it was months later when her physical therapist said, "You know, she doesn't even have her quad left to build muscle up." So, she's like, "No matter what we do. It's never going to get stronger. She doesn't have the muscle to build up." We felt very frustrated. I mean, I think they even gave us a six-month handicap placard. I went back after six months, I was like, "I need a five-year permanent one." So, yeah, it was just very vague and "Oh, yeah, this is the best thing." Not really any mention about—they said, "Oh, it's probably in the soft tissue. We won't know how much we have to take out until we get in there." Not like, "If we have to take all the muscle out, she really isn't going to be able run or anything."

Opal speculated on why no other options were offered the first time. She said even when the surgeon was preparing for her recent rotationplasty, he was still questioning the family's decision:

> I really felt like all the doctors just thought that it was so important that her leg looked normal, I guess, if you will. I don't know that any of them could get past that. The surgeon that did the rotationplasty was the one that did the limb salvage. So, he does these surgeries. Was still like, "Oh, I don't know if you really want to do this." What if you can't do this or do that? I'm like, "She can't do any of that stuff, now." She's in pain all the time. So, I guess what do we have to lose, you know?

Issue domain: Gender bias. She also felt that Olivia's gender and appearance were contributing factors to the surgeon's reluctance, "I feel like everybody we asked about it was concerned about the appearance being really harmful to her. It was always, 'Well, she's a young girl.' Yeah, it did seem to be really important to everybody." Olivia's biggest concern was function.

Issue domain: Lack of empathy from provider. Opal didn't feel like Olivia's surgeon had a good understanding of her complications or limitations due to the limb salvage surgery:

> When we went and talked to him and told him we wanted the rotationplasty, he was like, "Well, you're going to go from having a big thigh muscle to using your calf muscle." I said, "Well, she doesn't have a muscle there. Like, she doesn't have a muscle in her thigh." He's like, "Oh, yeah. I guess you're right."

Opal felt that the lack of empathy came from not having a full picture of how chronic pain impacts everyday living for these kids and their families:

> I think at the end of the day, no doctors, no matter how much they care, they don't live with the child. They're not the one that is day-in, day-out dealing with the pain and having their kid not being able to sleep because they're in pain, and not being able to do really basic things because of pain. You know, just not thinking through everything. I'm thinking a calf muscle is an improvement over what we had. It is, when it came down to it. They kind of tried to really discourage her from it.

Issue domain: Searching for options. Olivia was declared incurable 15 months prior to our interview and had never been without tumors since her diagnosis. With a short life expectancy, Opal helped Olivia do the research to find better options for her remaining time. They considered both amputation and rotationplasty. Opal said she had seen my daughter's video on the Facebook page:

> I saw Zoe and I'm like, "Hey, I wonder if my daughter could do that?" Just because in my mind I was thinking, if we amputated, it would be she would lose her knee, her ankle, and her hip. I thought if we could save her hip and at least give her a stump to work with, that would be better than losing her hip too. From there, I know my friend—she has two kids that are amputees and they travel all over the country and do events and stuff. She had mentioned several times, "There's all these osteosarcoma patients there that have amputations." So, anyways, when we started looking at the amputation, I think once I realized rotationplasty might be an option.

Opal researched rotationplasty by joining a Rotationplasty Facebook group:

> I just read in the thread and watched videos. Everything I watched and read, I'm like, "You know what? These people are doing so much more than she can do." Especially like, all the people who had had the limb salvage, then done the rotationplasty. I think only one person had a regret and it was that they didn't do it sooner. That was the only negative thing anybody could tell us.

Opal felt like the outcomes were genuine and did not feel like the posters had an agenda, "I'm like, These people, they're not having anything invested in it. There's no reason to not be honest about it. So, I don't know, we just knew."

Issue domain: Pain management. Olivia's main goal was to get rid of the pain:

> The debilitating pain that she was in all of the time was awful. She's not a complainer at all, so it wasn't that but it was, you know, just—I don't know. When your kid can't do normal things because they're exhausted from the amount of pain that they're in day in and day out, it's awful.

Issue domain: Incurable. Opal and her husband wanted Olivia to make the decision on her own and in her own time:

> Last year they told us there was really nothing else they could do, nothing curative they could do for her. We really just said about every step of the way, "Here's your options. It's totally up to you." You know, "What do you want?

> Whatever you decide, we're behind you one-hundred percent." It needed to be up to her. She would ask us, "Well, this is what I want to do, do you think that's a good decision?" We would just say, "We feel like it's your decision. Any decision you make is good."

Olivia chose rotationplasty. Her parents did what they could to prepare her:

> We did meet with another family locally here who had rotationplasty. We had gone to Hanger, to the clinic, talked to a couple prosthetists. They sat down with her and just explained the whole process and what to expect. Then, probably the biggest, the neurovascular doctor from the plastic surgeon that actually did her surgery, was the most positive about it. Just really took a lot of time with her and was encouraging, you know. She really felt like it was going to improve her quality of life….I just kept asking her, "When do you want me to talk to the doctors?" It took several months, then she was like, "Okay, I'm ready now."

Opal felt fully prepared to advocate for a rotationplasty on her daughter's behalf:

> We were very confident in our decision. I had done my research and I knew what to ask him, stuff like, "Well, we're going to go from a big thigh muscle to a calf muscle." We were far enough out from treatment, I was able to think it through better, and be able to stand my ground and know this is what we need to do. This is what I have to do for my child to have a better quality of life.

At six weeks post op, Olivia's pain is gone, "Completely gone, no more pain at all." The impact of pain on Olivia's daily life was profound – even more so as she looks back to six weeks ago:

> I would say just even with the rotationplasty and not having a prosthetic, that she's able to do more. She has more energy and she says, "I feel like I'm getting stronger than before." Just from not being in pain, so.

Her next goal is to run.

Olivia's complications were not limited to post-operative; Opal had a bizarre story to share and wanted me to see Olivia's implant– "We have her implant. I'll show it to you." The implant was huge and looked really heavy. Opal said then that it was her first implant – she'd had two:

> She had her limb salvage surgery and they went to put the implant all together, and it didn't fit together. She was in surgery, opened up, all done, and the implant would not fit together. Somehow, all the pieces didn't get sent. Every implant is really personalized. So, they all have different pieces. It's all sterile so there's no way to open it up and look at it, you know, there in the OR. This is the actual one. It was an ordeal. We ended up the implant company has been involved with her whole journey since then. Anyways, but yeah, it was her knee and the top part she didn't have a screw.

The implant she was showing me was actually the first one – she had to have a second one with the right pieces implanted later, "…so she had a limb salvage revision five days after the original one. They put the missing piece in and then, yeah, so. This implant is like eighty thousand dollars or something."

Olivia had a preference for surgery from the beginning, but the surgeon was authoritative in his position that he would save the leg. He made the patriarchal decision to do limb salvage surgery regardless of what the family wanted; eventually they gave up and figured he knew what was best. The three main issues around the making the surgical decision were not being offered options even though they knew they were available, complications from limb salvage (loss of muscle tissue and debilitating pain) and ultimately an incurable status that led them to seek a surgical intervention for a better quality of life. Opal had mixed feelings looking back on their decision-making journey with Olivia but seemed resolved that the process was something they had to go through to get where they are now:

> I think it's okay—I know it's okay, because I don't know that we would have done rotationplasty or known to push for that in the beginning. And also, it showed me just talking to other families…You know, they were recommending amputation to them like, "Look. Your kid will be walking way before they would with the limb salvage." They're like four months and they're in a prosthetic and walking. So, everything happened how it was supposed to.

Vignette 12: Phoebe

Phoebe was 15 when she was diagnosed with osteosarcoma in the distal femur. She was very active in sports and loved to play baseball. She had three limb salvage surgeries before choosing to amputate at age 18. Phoebe is now 27 and lives in California. She wants to be a pediatric oncologist and was attending medical school in Arizona when we met for her video interview.

Issue domain: No options offered. When Phoebe was first diagnosed, she was not offered any options for surgery, "I know it's so fast in treatment. You're so sick, but if you would just take 10 minutes to just tell me, 'Here's your three options.'" She looks back on that time and wishes she knew how to advocate for herself then, "I felt like sometimes when you're so sick and you just don't know, you just don't fight. You don't know what to do. So, you just let it happen." But no options were mentioned at all: "Rotationplasty wasn't even offered. It was only limb salvage and that was all I was going to get. So, I'm angry at him for that. I really am."

Phoebe wanted to know the potential complications as well, "I would want to know everything. I would want to know the unknown, too. 'We don't know that this would happen but this could be a possibility.'" She was completely unprepared for additional limb salvage surgeries, "I didn't know that there would even be another limb salvage. I wish I would have known that this could not work. That was never expressed to me."

Although she's not sure what she would have chosen at age 15, discovering that she had options later her left her feeling uniformed for making future decisions:

> Amputation was not even discussed. I don't know in that moment if I would have been ready for amputation at 15. At least if I would have been educated on my options, I felt like I could have made a more sound decision. Now, I wish I would

> have amputated sooner because I would have had a bigger bone. So, now I have a very short bone.

Issue domain: Excluding teen patient from discussion. Phoebe understood age might affect information shared, “I think it just depends on the age too of the child. I think, I mean, everyone usually likes to be informed…they like to know what’s going on.” She discovered discussions about treatment took place without her and felt she was not privy to that knowledge due to her age and gender:

> I think it’s different for each person. I’m someone that likes to know what’s going on, shoot it to me straight. I don’t like to not know. So, I think that’s what happened a little bit. They thought, “Oh, she’s a 15-year-old girl. She can’t handle the truth.”

She understood being protected but resented being left out of conversations that affected her for the rest of her life:

> In treatment and through everything, they communicated more with my parents too, which was another issue with me…My mom asked about it but he just said this would be my best option. My mom, I asked her, she didn’t want to broach the subject of amputation at that time. Looking back, I wish she would have because—I think everyone, they’re just trying to protect you. I think if I knew more about amputation and all the surgeries that could happen with limb salvage, I probably would have chosen amputation.

Issue domain: Bias to save the leg. Retrospectively and through the lens of a medical student, Phoebe both emphasized and criticized her surgeon’s position:

> I think it’s a cultural thing…being in medical school now, it’s really hard not to let your own beliefs dictate your patient’s care. Him too, it’s like “save the leg, save the leg, save the leg.” Well, I think each person has their own case and their own what they want out of life. I think he just generalized me as saving leg without even saying, “Well, maybe amputation, she’d be okay with that.” I feel he was using his own beliefs of saving the leg instead of just discussing with me my own beliefs.

Issue domain: Multiple surgeries– infection/pain/broken implant/shorter femur. Phoebe’s first limb salvage surgery was fraught with complications almost immediately. After just a few months, she had an infection and the implant broke. For the second limb salvage, more of her femur was removed and a new endoprosthesis was implanted. A few months later, it broke again. Her surgical team did a third limb salvage – this time her nerves were accidentally cut and she had no feeling from the calf down. Phoebe decided to try to save her leg and spent a year and a half on antibiotics and doing hyperbaric work while using opioids to manage the pain; she developed another infection, “I had gotten to a point where either the next step to remove the infection would be another limb salvage or more bone taken. Didn’t really want to take that gamble... they wanted to make my limb longer." Phoebe had been struggling to save her leg for three years; she was ready to be done with the surgeries, infections, and pain:

> At that point, the only thing they had was extending the bone for a year. I didn't want to do that. I wanted to move on and go to college to become a doctor. I wanted to be more self-sufficient and able to live on my own and do my own thing.

She made the decision to amputate but the surgeons at her home hospital were not supportive, "I kind of felt like they were projecting their own opinion on to me. You know, I mean, no one wants amputation but that was my best option." She felt that some medical errors made there impacted her later treatment options, "The only surgeon there was the only one that did it and he missed the infection big time. It was a big whoops… I feel like what I was getting from them was, they just wanted me to go away." He refused to do an amputation.
So Phoebe "doctor shopped" to find someone who would.

Issue domain: Finding a surgeon who would do elective amputation. Phoebe and her mother researched online to find a doctor who would listen to her. She says one was dismissive of her complaints, "He came in and said, 'Just be happy you're alive' - kind of like deal with it. He did not want to do the amputation." Her search proved more challenging that she thought it would be – "some thought I was crazy" - as doctors refused her appeals again and again. Finally Phoebe met a doctor who got it, "Dr. X was the first doctor that actually had a conversation with me about what I wanted out of life and what I was going through."
Phoebe told me with great emotion what Dr. X said to her:

> This doctor, X, I voiced my feelings and what I wanted. He told me it was like dragging an anchor. He said, "Are you ready to get rid of that anchor?" It was the first doctor that really understood that I was in so much pain.

Issue domain: Patient-provider communication. Looking back on the communication episodes with her surgeon, she felt there was a missed opportunity to engage with the patient about their values, "I'm a huge athlete. I played softball and I was a runner. I was never once asked any of that. That's one of my angers, too. I was never asked what I wanted out of life." Phoebe's office visits with her surgeon were very short, maybe 10 minutes long; she was not told until after her surgery that she would not play softball again:

> I asked if I could play softball on my limb salvage and he said, "No. You have to be careful." So, I was really pissed about that because I didn't know that before. I really wanted to get back into running and stuff and I just couldn't with my limb salvage leg.

Phoebe felt that in the absence of eliciting a patient's values and preferences for a good life, personal identities are impacted by their inability to resume their lives:

> I think what’s so important is a conversation and saying kind of like what the person believes in and what are their interests. I think so many times decisions were made without looking at me as a person, like who I was. For me, for instance, with the limb salvage and stuff, I was such an athlete and such an avid person that loved to work out. Having limb salvage took that identity away from me, and kind of took who I was. I was so in a fog and stuff. I think it’s just important to look at each person as their own person and you can’t lump people into being the same.

After Phoebe had her amputation, she resumed some of the activities that made her who she was, “I did a swim triathlon. I played sitting volleyball. I work out every day. Sometimes, my socket annoys me but that’s just part of it. I feel much happier. I really thrived in life after I had the amputation.”

Phoebe pointed out the importance of listening with patients, “I think listening is huge as a healthcare professional… Just listening to their story, you can learn a lot about their problem and who they are.” Phoebe thought that communication skills were especially lacking with surgeons:

> I feel like all orthopedics need it. I’ve had a hard time with orthopedics. I feel like they lack the empathy. I don’t know what happens in their training or what happens with that. I think they need to remember that it is a person. I think that we need to, in any level as a doctor, need to ask people what they want out of their life with the disease.”

She wishes she knew how to advocate for herself more back then:

> I will say throughout my treatment with the orthopedic that treated me, it was never a discussion. It was always, you’re going to have this and this is what we’re going to do. I wish, at that time, I knew to voice my opinion more. As you know, as you go through it more you learn you have to be your own advocate.

Issue domain: Finding the right prosthetist. Phoebe has been an amputee now for nine years but felt it was a real learning curve to understand how the prosthesis was made and how to communicate effectively with a prosthetist:

> What I wish I had done is go to a prosthetic company and learned about the prosthetics before I had amputation, and the process and stuff. I didn’t know that you could have different prosthetic prosthetists. I wish I would have known and met different prosthetic companies.

As a new amputee, Phoebe learned ways to increase her mobility with a prosthetic, “I had struggled a long time because my bone is so short. Then I got into working out and stuff, so I’ve gotten more physically fit to walk.” The challenges with prosthetists included issues with insurance providers:

> I’ve learned to become a huge advocate for my socket. Insurance and knowing what I want and what I like, but it took a while. It just takes time. It’d be nice to have somewhere you can go and be educated on it.

Even her prosthetists seemed confused about the insurance process, “The guy had no idea different insurances say yes to different stuff… and you need an in-network letter of agreement, which I never knew.”

Not getting a good prosthetic fit and having limited mobility sometimes led her to question her decision, “When I got my sockets and they were terrible, I was like what did I do? Like, why did I choose amputation? I just kept falling and it just didn’t fit right, you know, we need the right fit.” In the beginning, her sockets were never long enough for her needs with the short femur:

> The problem is because I don’t fit (the prosthetist’s) molds. They’re nice people. It’s just they can’t think outside the box which I think is a hard thing with medicine, too. It’s like you learn one way and then, someone that’s very complex like me—I have a very short femur, a lot of soft tissue. So, they were making my sockets too short.

Once she found someone who would work with her special needs, the prosthesis experience became much better:

> I finally found someone that we worked together. I like do the elliptical. I can do a ton of stuff and walk really well. I wish I would have been more educated on after amputation of different prosthetics. I didn’t even know there was different knees or a micro-processor knee or anything like that…. I wish I would have met different prosthetic companies before I had the amputation.

Getting a good socket fit was problematic because her residual limb was so unique; Phoebe compared this to her overall experience with limb salvage surgery – it’s not a one size fits all for either the surgery or the socket:

> I think with limb salvage and even like sockets and how my prosthetic was made, I was always generalized with the public of this works but maybe this doesn’t work for everyone. I think it’s like a case-by-case thing. That’s hard for medicine. We are really into generalizing, where each person has genetics. Everything’s different in each person and you have to take that into account.

Looking back, Phoebe wishes she had done more research on prosthetics, “My biggest thing is I wish I would have gone and saw how a socket’s made, how they make it, kind of like prosthetist shopped like you doctor shop.”

Issue domain: Missing information for post-amputation. The role of weight fluctuation was also a learning experience for Phoebe, “I had no idea about having to maintain my weight the best of your ability, because then your socket doesn’t fit.” Phoebe needed more information about what to expect after amputation. While it wouldn’t have affected her decision to amputate, it would have provided the knowledge she needed to plan and manage her new life as an amputee:

> I wish I would have known. I still would have made my decision because it was the pain. Just to know less uncertainty about the future, would have been nice. I made that decision kind of on a whim. It was a gamble. It could have worked, it could have not. So, that's why I think this is so important to have more facts.

It took some time and going through nine prosthetists but as with finding the right surgeon, Phoebe finally found the right prosthetist for her, "You've got to meet someone that you gel with and you just know..."

Phoebe looked back over the telling of her story and all the challenges she had – inadequate prosthetic information, ineffective provider communication, broken implants, infections, and pain and finding a surgeon who would amputate but overall her biggest issue was not having options at the beginning:

> I don't know why they're so limb salvage heavy. I really don't… I know a lot of doctors don't do amputation unless they have to… Yeah, when I say elective amputation, people are like, "What?" …no one wants to be missing a limb. Sometimes, you have to.

Vignette 13: Quentin

Quentin was 15 when he was diagnosed with osteosarcoma in the distal femur and had limb salvage surgery. Now an athletic trainer, he was 32 when he joined me for a video interview from his home in Michigan. He said he was looking forward to talking with me, "It's kind of neat that I get to tell my story to someone other than students."

Quentin started his story by saying he understood the seriousness of his condition when he first met his surgeon:

> She said, "People die from this." I'm not going to lie, we're a Catholic family. We had our priest come over before I went down. We said prayers. I gave my confession, because I didn't know if I was going to make it or not.

Issue domain: Amputation vs. limb salvage. Quentin's surgeon explained his options and he weighed his risks against his life goals:

> Dr. X said, "We can give you a total knee replacement or if you don't want to deal with the multiple surgeries that may be ahead," she said, "we can also amputate." So, of course, I look at my mom and my dad. My biggest fear was, I was like, well—being 15, you think, "Man. If I amputate, then I can still do a lot of things that I wanted to do." My doc told me, she's like, "If you play sports, if you want to continue playing football, if you want to continue playing basketball, if you want to be a cop," she said, "choose that, I can amputate. Because then if you break that prosthetic, you can fix that."

Issue domain: Uncertainties. He said Dr. X did warn him about possible complications, "I think the most she said was you know, there could be possible other surgeries down the road. Didn't really know how many, didn't know what possible surgeries." Quentin understood that amputation was the option that would give him the most function and the least uncertainty for more surgeries. But he had other concerns too.

Issue domain: Phantom pain. Quentin was worried about the risk for phantom pain; his mother's background in nursing and his surgeon's reassurance moved him more toward LSS:

> My mom said, she goes—being a nurse, she says, "There's issues with phantom pain. There's issues with, you know, maybe you have infection and stuff like that." Dr. X was a big part of it, too. She said, "You know, you came to the right hospital to salvage a joint. You still have our knee, still have your bone, and still have your muscle and all that stuff, other than the one section that will be gone.

Issue domain: Chemo or no chemo. At first Dr. X thought they would just do the surgery and not chemo since he had low grade osteosarcoma. She talked to an oncologist at a nearby city and they said chemo would give him "a 95% chance of never having the cancer come back again." Quentin said his risk for recurrence was more like 55-65% if he didn't do the chemo. Dr. X asked the oncologist about recommendations, "Doc said—she said, "What would you do if that was your child?" She said, "I would treat, just because of the side of caution."

Ultimately it was Quentin who decided to do the grueling treatment. He was worried that one of the cancer cells got released into the blood during the biopsy and "who knows what it could do. It could travel and metastasize somewhere else."

Issue domain: Growth. As Quentin was reviewing his options, he described his biggest concern and how Dr. X reassured him:

> So, my biggest concern was, what about growth plate and stuff, too. She said, "No, no." She said, "You still have your growth plate in your hip." So, I was like, "Okay. That's not too bad, then." So, she actually got it pretty even when she did my knee replacement. She did tell me, she said, "There could be complications along the way with growing, and having a retread job, like take the plastic pieces out and redo the surgery." She did an awesome job in telling me everything that could happen.

Issue domain: Identity. Quentin recalled how he felt when he understood he had a life-threatening condition:

> The things that flash through my memory of "Man, I'm never going to be married. I'm never going to graduate high school. I'm never going to have kids. I'm never going to be able to do this. I'm never going to be able to do that." Those are like, being 15, you're thinking about that stuff, too.

Quentin struggled with his long-term goals and not really knowing what his future held, "I think the biggest decision was obviously giving up on my dreams of things I wanted to do versus still keeping my leg and all this stuff." He had to shift gears and rethink about what he wanted to do and how that would align with what he could do with a limb salvaged leg:

> So, because I couldn't play sports no more, I had to pick a career that I would maybe enjoy. When you have your heart set on something, it's hard. All of a sudden, now I have to figure out what I'm going to do.

At 15, Quentin was the one making the final decision, though he felt supported by his family:

> …then I talked to my mom and my dad, and I had to kind of make that decision myself. My doc said, "We're here. This hospital is known for limb salvage." She said, "What do you want to do?" I said, "I want to keep my leg." I had to make a tough decision at 15, either keep my leg or give up on a lot of dreams. So, I had to make that decision at a very young age.

He came a conclusion that led to his decision, "I think I kind of wrapped my mind around it a little bit by that time. I think it was where I say, you know, having my knee is more important than losing it…"

Quentin was excited to show me his original implant and held it to the camera of his laptop, it was very shiny, "I wanted to show you…That's the rod that went into the femur and stuff….she gave it to me. She cleaned it up. They sterilized it before they gave it to me." He brings the implant out when he teaches sometimes, "I use it a lot for a lot of visual purposes when I do my talks at the college classes with the athletic training students." Quentin has a great sense of humor, "I joke around. I say, 'Oh, I can make it a gear shift for my truck or whatever.'"

Issue domain: Muscle weakness. Quentin had some issues with his quad muscle after surgery. Dr. X told him she had to remove more muscle than she expected:

> So, that was one of the big things that I had an issue with, is still trying to redevelop that muscle and stuff again. I didn't realize that much of it. She said, because when she actually—she didn't know how much she was going to have to remove.

He turned the camera around and showed me the atrophied area on his leg, "You can see how much muscle that was taken out right there."

Issue domain: Complications and surgery…four times. Quentin's first implant broke soon after his surgery. He thought it was due to a combination of bone weakness from the chemo and weight he gained while in chemo. Dr. X did surgery and Quentin went off to college. He experienced his second limb salvage break while in college; he attributed the cause this time to overuse. Then a few years after that, he started to have pain and needed a full revision. She told him his femur bone was wearing out quite a bit at this point. The fourth break happened a few years ago. This time his femur cracked while he was hunting, "Then, I shot an eight point in October. We were dragging it out of the woods, and I think I got my foot stuck in some mud. I felt something pull. I was like, 'Ooh, that didn't feel good.'"

At this point Quentin now needed a hip replacement too:, Dr. X offered new options, "You've been through a lot of surgeries." She said, "If you're getting sick of it, I can take your hip. We can amputate." He considered his options and understood that prosthetic technology had become much more advanced than when he was 15, "I know in 17 years, there's been so much that's changed in prosthetics and even in knee replacements and joint replacements. I see it in P/T when I work and stuff." Quentin also felt that as an adult, he had a better understanding of the decision-making process:

> Now that I'm older, I was able to ask more questions. So, what will this be able to do? Will this give me more opportunities to do things and have less pain in my leg? She says, "Yeah." She said, "I didn't want to give you a hip before because you were still really young and I wanted it to last as long as it could." I said, "Doc, I've already done the knee replacement. We'll just do the hip and then, we'll be done."

With the new hip came new opportunities for activity, "She said I could run actually more than I could before with this because the force and the bone and everything too, and the trauma on it. She's like, 'No marathons, though.'"

Quentin recalled the cautions Dr. X had when he asked about being a police officer with the limb salvage; he had hopes that with the new hip, that might be a possibility, "After my hip replacement, when she said, 'Oh, you can run more. You can do this.' I was like, 'So, can I be a cop?' I still, even after college. She said, 'No.'"

Issue domain: Information seeking. Quentin was diagnosed before internet was popular and cell phones were rare:

> That was before you had iPhone where you could put whatever you wanted in and just be like, "Oh, wow. That's what bone cancer looks like. This is what limb salvage looks like." You know, you never even thought about that.

All of his information for making surgical decisions came directly from his surgeon. She used a variety to tools to explain surgeries to Quentin:

> She actually had a hip replacement in her office…. She had what a hip looked like, and she showed me how it would attach to my femur where the rod's already at and stuff. She wanted me to make sure that I knew what could potentially happen if she couldn't fix that bone with plate and screws. She made me well-informed. I think with her information that she gave me, I felt confident in the hip that she was going to give me. From day one, she was 100 percent honest with me with everything. When she told me about the amputation and showed me the video of the kid running around with the amputation, if I wanted to have it done, I could still do that stuff. She's like, "I wanted to make sure you have all your options." I think that's the biggest thing, when you have all your options and when you have the confidence of the surgeon.

He felt his surgeon was honest and positive what she could do for him:

> I think one of the biggest things that I really—I mean, I'm blessed to have the doctor that I had. She was always so straight forward. I think her positive attitude was a big part of it, too, like, "Hey, we're going to get this taken care of. We're going to get it fixed. You're going to be back to cruising again, you know." She

> was a big part of being calm about things. When you see a surgeon that's calm and confident in their work, that makes a big difference too.

His relationship with Dr. X continues to this day, "I have her on Facebook. She has an alpaca farm. She's like, 'You want to come down and come see them?' She's been a very special person to the family."

Quentin was a resilient individual; he made his mind up at 15 that he would adapt:

> I just learned to go with it. I think after that first decision at 15, not knowing really what to expect, I think I told my parents I'm going to roll with the punches and just whatever happens, happens. We'll deal with it as it comes.

His high-level outlook gives him purpose:

> Once you wrap your mind around it and you accept that, "Hey, I'm here. I'm alive. I've got my knee joint. I got that." Like, "Hey, I beat cancer and I'm a walking miracle for people to see that." I've helped a lot of people along the way, I feel like, with everything I've gone through.

He felt he was being given a new direction for his life goals:

> My whole life is changed, now. So, I went to a job career fair. They had somebody talk about athletic training and sports medicine, and how the jobs will be probably rising in the next years and stuff. Something just said, here you go.

Quentin's faith may have directed him to this career path when he realized he could not be a police officer:

> Maybe, then coming from our faith that sometimes there's a little bumper in your life that pushes you the direction that you're not supposed to be going. So, I think about maybe I wasn't supposed to be a cop, maybe I wasn't supposed to go to the military, maybe I would have been killed in service or on-duty as a police officer. That wasn't my calling, that wasn't my—that's not what was meant to happen. So, kind of holding onto some stuff like that.

Looking back, the limb salvage outcomes didn't affect him all that much, in spite of the many surgeries, "I think I ended up having more surgeries than she anticipated." Quentin does have some concern about implant wear and unexpected surgeries down the road, especially as he ages:

> I'm thirty-two years old and I've still got a long life to live. That's one thing I do think about—…I see some older people that are getting their hips and knees replaced at 60, 65. So, I'm thinking how am I going to feel at 60, 65 having a total knee and hip that will be replaced. Is it going to be good? Is it going to be bad?

One thing he can control that will help delay complications is maintaining a healthy weight, "That's the thing, the unknowing of that but I think that if I can control my weight. That's one of the things, too, doc told me. More weight on that joint, on that prosthetic, is not good for it." Quentin felt prepared for the unexpected by his surgeon with whom he maintained a relationship and trust. Not surprisingly and despite multiple complications and limitations, Quentin scored low on the decision regret scale.

Vignette 14: Ray & Ramona

Ray was 10 when he was diagnosed with osteosarcoma of the distal femur. He was an extremely active, athletic boy, "Every day during recess I would play soccer. I would play it constantly, at least eight nine hours a week. The travel team we played on was very competitive." Ray had a limb salvage surgery and after several years of complications, he had a rotationplasty at age 16. He was almost 17 at the time of his in-person interview with his mother, Ramona, at the family's home in Maryland.

Issue domain: No options offered. There were no surgical options offered according to Ramona, "'We'll do limb-sparing surgery.' It wasn't even presented as a decision." Ray had a Stanmore expandable implant as part of his LSS that could be lengthened as he grew. The family did seek a second opinion with another surgeon; Ramona recalled that meeting:

> At the time, all I'd heard was LSS. We asked about allograft. He's like, "No, no, no. The Stanmore is amazing. It's the newest, greatest thing. It's phenomenal." He did say there's this other procedure called rotationplasty. He described it to us. It sounded awful, so we immediately said no. We looked it up later and it was shocking.

So the family decided to go with LSS and the Stanmore expandable implant. Ramona said the risk of complications was never mentioned, "It was presented as a panacea."

Issue domain: LSS complications – drop foot, infection, pain. Soon after his limb salvage surgery, there were obvious complications, "He had drop foot. He had severe neuropathic pain… He wound up with a huge wound on his foot from swelling, and all of that contributed to poor rehab. The surgical care was not good at all." At this point, the family switched to different providers. A lengthening of the implant was scheduled but they discovered that there was an infection and the implant had failed. After four months of antibiotics, he had a second limb salvage with a new implant.

Ray's pain was very difficult to manage, and he was in agony – it affected the whole family. Ramona recalled the impact on her youngest son, in 2nd grade, "…finding him curled up in a ball in his room with his hands over his ears. That's the reminder to me of just how much pain Ray was in." In the new hospital, there was an outpatient pain clinic where Ray was switched to Lyrica, "Almost immediately, we saw a change. So, things got better….They also got him into aquatherapy as well as physical therapy. They really kind of took charge in managing his rehab which was really good."

Things seemed to be going well for a few months, then there were different complications, “They did a lengthening and basically, everything around his knee kind of seized up. He couldn’t straighten his leg all the way. He couldn’t bend it as much as he’d been able to.” Ray’s surgeon said the scar tissue had built up…Then he developed a big bubble on his scar that turned out to be another infection.

Issue domain: Another decision to be made. Ray’s surgeon suggested redoing the LSS again but said he might have to have removed every 2-3 years. She also gave the option of osseointegration (implant inserted into bone with stem outside the body), amputation, and rotationplasty. So at 16, Ray and his family were back where they started six years– minus LSS as a good option and plus osseointegration as a potential option. Looking back, Ramona said that it was Ray who decided not to be involved in the decision at age 10, “It was Ray. He was a pretty squeamish kid. So, I really think a lot of it was—the talk of surgery, he was not comfortable with at that time. He also, he trusted us, I think.”

Issue domain: Researching options. For the second decision, Ray wanted to participate in researching all of his options; he talked about what that was like for him:

> We talked to a Swedish doctor about osseointegration. He was like one of the top minds in it. Because Sweden, they don’t have the same regulations. They had more data about osseointegration. So, I was very much a part of that. I conferenced with everybody. I learned which each of the options, what they’re about.

He also knew from personal contacts what options would be best for the sports he wanted to continue:

> Two years ago I went to hockey development camp for disabled hockey. Ninety—eighty percent of the guys there are amputees, some of them double amputees, some of them single amputees. There are guys there who had rotationplasty. So, I seen all this and I have known that it was a viable option.

One of the factors around the decision-making now was accommodating for growth again; at 16, Ray was still growing. Previously with the first surgery, the doctors thought he was nearly done:

> Part of why they recommended LSS was, they said, “Oh, he’s so close to hitting puberty. He’s not going to get that much taller. We can accommodate for the height.” They missed the mark on that one as well...he’s still growing now. I don’t know if that would have impacted our decision too. You know, that was something—they really messed up on that, on how tall he would be. We had fused the growth plates on his left knee because the prosthetic was not going to be able to expand any further.

Issue domain: Complications with LSS. Meanwhile Ray was having issues participating in his adaptive sports; he was saying he wished he'd amputated the first time. Ramona recalled Ray talking about his hockey teammates:

> You know, when we get off the ice, these guys who are amputees, they get up, they snap on their leg, and they run. I get up and I have to find my crutches and have to straighten up my leg and I feel stiff.

They also had some concerns about weakness in Ray's quad muscles, especially since his physical therapist and the surgeons all said that Ray could run one day:

> Ray was struggling to walk down steps alternating feet. He just had poor control. When we asked (the second surgeon) that, she said, "Well, he lost half his quad muscle. Of course, he's going to struggle walking down the steps." When we went back to our physical therapist, she said, "Oh, well that explains a lot." Then, we realized he was never going to be able to run because he lost so much of the quad. We had already left (the first surgeon) at that point, so I never asked him if that was anticipated or not. I know that at diagnosis, they told us that it had spread outside of the bone. So, I have to assume they knew they were going to remove some of the quad.

It was never clear to the family if he missing quad muscle was due to the cancer or to some aspect of the surgery.

Issue domain: Second opinion/bias to save the leg. With so many factors to consider, their surgeon suggested they seek a second opinion. So they met with one doctor:

> He was very, very strongly pushing doing LSS again. He really did not want to entertain rotationplasty at all. He thought LSS was the way to go. We asked him about the patella and the scar tissue. He was very dismissive of those, which was disappointing.

They asked the Swedish doctor if Ray would be able to run with osseointegration, "He said they had no data on whether or not the legs can support that. So, the answer would be no." They went back to their original surgeon and said they decided on rotationplasty:

> She was kind of obnoxious. She really pushed Ray to make sure that he understood what it was going to be. She kind of played the bad cop. Even the last appointment before the surgery, she was still playing the bad cop, "Are you sure? Are you sure? Are you sure?"

Ramona felt that Dr. X was leaning toward the other two options:

> Dr. X, was very clear that she thought osseointegration or rotationplasty would be better because a straight amputation, the socket would be so high and it would be harder to get a functional knee. So, at the osseointegration, whatever it is that they do, the knee works better. Then, with the rotationplasty above the natural knee.

Issue domain: Information seeking. Ramona read through posts on the Osteosarcoma Facebook page and on the ACOR listserv. Together she and Ray watched YouTube videos of kids with rotationplasty. Ray emailed with a boy who had had rotationplasty after LSS, Ramona said both he and his mom were helpful. It was hard to find someone locally who had rotationplasty from osteosarcoma but Ramona pointed out that Ray was meeting others in his wide circle of friends in adaptive sports:

> You know, Ray's now in this world of adaptive sports where you get to know, you see people's disabilities more and you see how normalized they are. I think that's something, also. Before his cancer, we didn't know anyone who had a prosthetic. You know, we've never been around that. So, it was very startling, the idea.

As Ray accumulated knowledge about his options, he found himself closer to making a decision:

> I think I really had my mind set on rotationplasty. Osseointegration was just another option that we had heard about that we wanted to learn more about. So, we considered it. If it sounded better than rotationplasty, I probably would have gone with it. Rotationplasty did sound better. Also, since osseointegration is so new in the United States, you know, it only recently got approved to be used. The past three years, it was approved to be used. They've never done it on someone my age before, so they have absolutely no data for that. Yeah. I had no idea how that would turn out.

He felt that his risks for infection again with LSS were greater given his existing propensity to develop infections with implants:

> Also for rotationplasty as opposed to LSS, because of the infection appearing twice, there was potential that it could continue to appear and I would need to have surgery for the rest of my life. I did not want that. I knew that amputation would be a very finite
>
> decision. It was.

He understood that there were risks with rotationplasty but began weighing those risks against the risks of other options:

> Beyond rotationplasty, there was the complications that could go wrong from rotationplasty, I mean, obviously there were complications that can go wrong but it didn't seem as likely as the LSS possibilities. Especially since I was going through lots of problems with LSS.

She and her husband also had some concerns Ray's foot and ankle for this next decision:

> My husband and I didn't want him to have another LSS. We felt like that ship had sailed. We were both very curious about osseointegration. So, part of the complications from the first surgery are that his foot hasn't grown since that surgery. He has osteopenia in the ankle and in the whole lower leg actually.

Their concerns coupled with the experts shared knowledge helped them to keep it all in perspective; but in the end, they really wanted Ray to make the decision:

> He hasn't gotten his prosthetic leg, yet. I'm worried about overuse of the ankle and the health of that coming back. We felt like osseointegration would remove those issues. We really listened to Ray and we wanted this to be his decision. I think we kind of monitored his decision-making process. You know, we wanted

> to make sure he was making the decision the right way. That was what we cared more about.

Issue domain: Communication issues. Ramona compared the communication styles between the first surgeon who did limb salvage surgery and the second surgeon who did rotationplasty:

> So, Dr. Y is very much "I'm the great Dr. Y and I'm going to solve all of your problems." Dr. X has a kindness and a thoughtfulness. She sits and thinks while she's with us. She makes sure that she's covered everything before she leaves. She just takes that time. We've had some heated discussions with her. She pushes back. She also listens.

Communication and empathy differed considerably between the two surgeons:

> After the surgery when we had follow-up appointments with Dr. Y. it was difficult to get in touch with him. He didn't come to see Ray when he was inpatient. So, we had to make outpatient appointments. They were in a different building. It was just difficult. Dr. X, her scheduler, is phenomenal. Whenever we have questions, we get a call back from PA or a nurse within a day. Dr. X has called us at eight o'clock at night and talked at length. We never had that kind of access with Dr. Y. Dr. X, I get the sense, really knows Ray. Granted, she's been seeing him a lot longer, but I get the sense that she really understands who he is and his priorities and what's best for him. I feel like Dr. Y had the opportunities to get to know Ray and didn't take them.

Ramona explained how Dr. Y treated Ray after he had severe pain in post op after his LSS:

> After surgery, Ray was in the hospital for two weeks because they couldn't control his pain. They had him on a Dilaudid drip and oxycodone. He was screaming in pain. They could not figure it out. They had put him on the oncology floor because the nurses all knew him, but it meant the ortho residents weren't there. It's a big hospital. The pain team felt like their hands were tied as well… Dr. Y when we talked to him about Ray's pain, he said, "Well, he's going to be in pain. He's going to have to deal with it." Yeah. We were like, "He's screaming. What do you mean he has to deal with it? This is extreme pain. You can't expect him to deal with this." He said, "He's going to have to suck it up and deal with it."

Dealing with pain issues has always been a big concern for Ray and his family; they found out later that there were pre-op solutions to manage pain but it was after his first surgery:

> There is a study that Mayo Clinic did that said that Mayo Clinic now premedicates with Lyrica for all of their LSS procedures. That had been out before Ray's surgery. Of course, we didn't even know to look.

For the rotationplasty, Ramona and her husband were more prepared to advocate for Ray:

> I think because of everything that had gone wrong the first time with the LSS, my husband and I, that's been something we asked all along. What will the complications be? With the rotationplasty, we got connected with an anesthesiologist before the surgery and got Ray pre-medicated for nerve pain. We very much have in our minds now, you know, let's work to prevent problems.

She said she knew of some kids who did well with LSS but felt that rotationplasty had the highest level of satisfaction from reading about patient reported outcomes, “I’m on the rotationplasty group. I’m in both Facebook osteosarcoma groups. I don’t know anyone who has said, I wish I’d never had rotationplasty. I think that’s also significant, you know. I don’t know, maybe they are out there.”

Ramona questioned whether there might be a deliberate effort to avoid talking about potential complications for the first surgery; she remembers how overwhelmed she felt at that time:

> I would kind of be curious to know from a surgeon that I really respect, like Dr. X, how much she shares about potential complications with patients for their first surgery. Because when I think back to that time period, life had just stopped and we were so concerned about Ray’s immediate well-being. It was very difficult to think long-term….to be given all the potential complications with these surgeries, in such glaring terms, I don’t think you could really process it while your child is going through chemo.

Ray’s parents had to negotiate gathering information and data for managing the first series of limb salvage surgeries and then start over again with Ray when a new decision had to be made. This time they felt more prepared, in large part because they were not making a quick decision weeks after the cancer diagnosis. Finding a surgeon who was not authoritarian was a challenge. At 16, almost 17, Ray was able to participate more in making the decision, from researching to talking with peers to thinking deeply about his values. Ramona said Ray’s pain is gone and he’s able to do so much more, “This summer, there’s one point he said to me, You know, everyone acts like this is such a big deal, but my leg feels great.’”

Vignette 15: Shelley & Ted

Ted was 18 when he was diagnosed with osteosarcoma in the distal femur and had a limb salvage surgery. Now 33, Ted lives in a small town in rural Illinois with his mother, Shelley – his father passed away when he was 11. They invited me to their family home for an in-person interview where we gathered at their dining room table with coffee and friendly felines nearby.

Ted had no outward sign of a tumor; in fact, the tumor was found after he kicked a can and his femur broke. His diagnosis of cancer came soon after he lost a family member: “…my grandfather passed away from his cancer, so. Definitely a scary thought.” He wanted to save his leg but also worried about survival, “Terrifying at first, thinking about losing the leg. Then, just more talking about it, it was like if it comes down to the leg or my life. It’s like, obviously, lose the leg.” Ted started chemotherapy but the necrosis rate was not high and Shelley was worried.

Issue domain: Limited options due to location of tumor. Shelley said the doctors were concerned about it too, “It was a low necrosis rate which meant chemo wasn’t very effective.” So they added two more chemo agents to the mix “because he had broken the bone, it had contaminated so much of an area that they were concerned that it would spread because of that, too." The doctors felt that the contamination meant that Ted would have a very high amputation on the femur so this affected their options, "Rotationplasty was never an option because where they would have had to amputate would have been too high. There wouldn't have been enough femur left at all. That was gone."

The surgeon did talk about uncertain outcomes associated with limb salvage though there was no mention of comparable limitations with an above knee prosthesis – so it was hard to compare the two options:

> There was some talk about that, like which degrees of what difficulties and complications there would be going forward. Like, there would be possible breaks and injuries with the leg, risks of infections. Didn't at all talk about difficulties with adapting to a full-length prosthesis. So, I just weighed the options. Neither option was really good. It was kind of just horrible hand either way. So, it was like, "Okay. We'll just go with whatever hand we get dealt." I guess that we were just so prepared for either way, we didn't really get hit by it…

Issue domain: Height affects surgical option. At first, Ted's surgeons were saying they'd have to amputate, "That's what we were pretty much leaning towards. Because, I mean, if it was necessary, like yeah, go with that route first. If you can save the leg, do it, but don't compromise my life just to save the leg, kind of deal." The surgeon was considering limb salvage and called Shelley prior to surgery to discuss it, citing his height as a factor for consideration:

> That was actually the decision we made the night before surgery. Before that, it was a definite amputation. Because it would be so high up, because of the contamination. The night before surgery, the oncologist called me and said, you know, "Shelley, I don't know what to do." So, I said, "Well, if you can get clear margins." I said, "I mean, totally clear. I don't even want them close. You get clear margins, then save the leg. If you can't, take the leg." The thought process behind of saving the leg was because he is so tall. He's a big guy… So, he was concerned about that too. Dr. X tried so hard to keep the leg and do the limb salvage was because it would have been so high, fitting a prosthesis would have been very difficult and because of your size. It's your height…He did get clear margins and he saved the leg.

Issue domain: To know or not to know? Although her son was legally an adult at 18, Shelley did not share everything she learned, and Ted said that was okay with him, "Only as much as I kind of needed to know." Ted wanted someone else to be the decision-maker but he was interested, "He just did not want to know anything. He says, 'You make the decisions.' I mean, I didn't on my own. We talked about it for sure." He was also concerned about how that responsibility would affect his mother, "It wasn't just like completely kept out of the loop. Just enough that I needed to know without it just weighing down on you too much." Shelley also felt that survival information would have detrimentally affected her son:

> At first, the percentages scared the heck out of me. Because he had broken it, his odds were very low for survival….he never knew that. It's funny, when he went for his one-year check-up with both oncologists on the same day. The first one said, "Well, P__, you're lucky to be alive. We didn't think you'd make it." He's like, "Okay." The next one says pretty much the same thing. He looked at me, he says, "Mom, did you know that?" I said, "Yes, I did." He says, "Why didn't you tell me?" I said, "Because if I had told you, you would have died." He would have. He would have given up because I know my son.

Ted agreed, "Unfortunately, my brain's more analytical. I would have seen those numbers and been like, 'Well, I'm screwed.' I think I was smart to know, I was better not knowing."

Shelley said she was advised not to do research on her own, "I think she was the P.A. I think she told me to stay off the computer. I go, "Yeah, nope.'" She looked mostly to sources she knew were credible, "Back then, it was I tried to stay with more of the medical hospitals like Mayo and Sloan Kettering. You know, those, the big ones, instead of just going to Web Doc MD or something."

Issue domain: Advocacy requires knowledge. It seemed unusual that a surgeon would consult with the family on the phone the night before a scheduled surgery, but Shelley said, "All the orthopedic surgeons I've met have been a little quirky. You know?" She appreciated him sharing his thoughts: "He knew I had done a lot of research and I'd kept up on everything." Even though this was before internet was easily accessible, Shelley understood the importance of patient advocacy and was researching online, "I just knew from the start that the more I knew, the more they would share with me and the more they would consider me part of the team. That's what I wanted. It worked."

The family felt like they had a unique relationship with the surgeon. Shelley said she trusted him more after he called and said he didn't know what to do:

> I felt more confident because I think he was going to do what was best anyway. He just wanted to run it by me. Maybe somebody else would freak out about that. I felt very honored that he trusted my opinion. I think if we didn't have that relationship, I don't think he would have done that. I don't think he would have called me and said, "I don't know what to do." You know, it wasn't like, "Oh, my God. I don't know what to do." It was, you know, here are the options. What do you think?"

Even though they weren't sure what the outcome would actually be until the surgery was over, they felt informed about the reasons why and trusted Dr. X, "I guess we were just prepared for either direction. So, it was like, okay, this is the direction we've got now."

They learned later that the whole team was in on Shelley's preference for her son:

> One thing I do remember, is that one of the nurses in recovery took me aside and said, "I want you to know that every time they took another piece to see if he had clean margins, the whole operating room stopped and prayed." So, that made me

> feel pretty cool especially since I think I embarrassed the heck out of Dr. X when I said, "You're not going in there without praying with me first." He was kind of shy. Yeah, I don't remember. I know we were all excited in the waiting room, you know, when they came out. They called and said they were saving the leg.

Issue domain: Missing information – pain and quad muscle removal. Ted had an infection soon after his LSS and two years later a part broke, revealing yet another infection two years after that. Then he went about ten years before his full revision. Ted had some concerns about his limitations:

> It still aches a lot, especially in the cold weather. It gets a lot more stiff, kind of. Pops and cracks sometimes which is a little unnerving. I keep freaking out about something breaking. It's just kind of a dull ache, like the restless leg syndrome kind of thing, just constantly. You just have to learn to tune it out eventually.

He relies on an assistive device when he knows he's going to be out walking:

> I use the cane for long walks, mostly for balance—the leg gives out sometimes because there's no muscle holding it all together. So, it's not like a regular leg where it just kind of like locks and you're fine. I have to constantly just be holding it sort of, if that makes sense. So, when I'm walking, just in case it gives out, I need something to put all my weight on. Other than that, not too many horrible issues with it.

Shelley described an orthotic brace that an orthotist made for him:

> He custom fit a brace for him because his leg doesn't lock. He'll—even walking, he has to consciously think of moving his leg out to go forward. So, this one, the brace, he can lock if he's standing a long time, or he can unlock it and then he can walk. You don't use it very often, though.

Ted agreed that he didn't use it much, "No, it kind of just puts pressure on some parts. There's no muscle or anything there to like brace anything. So, everything just goes straight to the bone. It just kind of hurts. So, it just pushes on, like, the nerves are just painful."

He adds that it's not as bad as it sounds, "It's not like a nightmare thing. I really can't say if it would have been better or worse to have had the amputation. I have nothing to compare it to obviously." Shelley added, "We do have a wheelchair in case he needs it for long distance."

The lack of quad muscle contributes to the leg weakness though it did improve after his revision two years ago:

> It could be built up a little bit but it's never going to be back to normal level, at all, not even close. I think it was a month after physical therapy, like after the most recent surgery. Before then, I couldn't really even go upstairs easily. I just didn't have the strength to push my whole body up with that one leg. I had to go up on my good leg but one step at a time. It's gotten better. I can do that at least.

Shelley said Dr. X had to take more of the quad muscles to make sure they had clear margins and showed me, "…because it was contaminated when he broke the femur. You can see, actually, on his leg where his leg actually dips." She believes that retaining the quad muscle is what impacts limitation later:

> I see so many kids from the original osteo group, the ACOR group, you know, climbing mountains and hiking. I think of all these kids doing all these things. I think the key is if they don't have to take any part of the quad muscle or the tissue around it, you've got a much more successful surgery. Again, that's something I didn't know. You find out later. Would that have changed our decision? It might have if we had known.

Ted added, "The quality of life aspect."

Shelley also got information and support from other groups after Ted's surgery; she mentioned the America Cancer Online Resources listserv, "ACOR. I got a lot of information from them, a lot." Once the osteosarcoma Facebook groups started, she also joined those:

> I probably did get a lot of information from them. It was a good place to ask questions. You know, like, "Okay. He needs platelets again." "Don't worry. It's normal, it's normal." That type of thing. Probably after the initial diagnosis, I probably went more with ACOR and didn't start looking into things other places because I got all the information I needed there. The hospital gave me a lot of information, a lot of information, so that was good.

Shelley became so well-informed that healthcare professionals commented on her expertise, "Every doctor I have been to, new doctors with Ted…Every doctor has said, 'Where did you get your medical training?' I said, 'Nope. Just a mom-cologist.'" She even sent research studies directly to Ted's surgeon:

> I had sent the surgeon probably a couple—this is why he called. I had sent him a couple of studies. They were small because it's rare just to have osteo and then have a pathological fracture. I believe it was one at Mayo that had only 10 kids in the study but showed no difference between amputation and limb salvage in a pathological fracture. There were a couple other things, articles, I had read and sent it to him. Back then, the thought was save the leg. In hindsight, I'm not sure I would have fought so hard to save the leg.

Ted remarked, "Hindsight's 20/20." Shelley said, "Yeah, I know. Because just the pain and he's disabled now through Social Security, can't work. We just have to figure out some business you can start at home here."

Issue domain: Chronic pain management. They looked at different ways to treat Ted's pain, Ted said, "Use heating pads in the cold…those help a lot. Got one in my room all the time. Gabapentin, the nerve blocker. Tramadol, when it gets really bad. Other than that, basically just trying not to run marathons or anything."

Ted described what the pain was like for him, "The muscles start aching and tensing, like kind of like cramps and stuff. Not really spasms. It's weird. I just start kind of aching, mostly a flu-like ache where your joints just kind of ache soreness most like."

She wondered if saving the leg was the best decision 15 years later:

> Was it a best thing? I don’t know. You know, he can’t walk very much. We go out shopping, like we used to do weekly shopping at Target, he’s down for three, four days in pain. He can’t sit, stand, or walk very long at all. You know, you did the revision surgery. He had everything taken out except for that top part. It’s helped. You’re not in as much pain. It didn’t cure it.

I asked if they would have chosen amputation if they knew what they know now and Shelly said, “I would have, I think. Would you have?” Ted responded, “It’s hard to say… You really don’t know either way. You can only know one option…the limb salvage, we go that way and then, it doesn’t work out, you can go with the amputation. You can’t go the other way.”

For Shelley and Ted, the decision was impacted risks associated with Ted’s height for getting a good prosthesis fit vs. multiple surgeries; their options were further limited by the contamination of cancer cells in his upper femur. Current issues include pain and mobility issues that affect Ted’s quality of life, much of which they attribute to the missing quad tissue in his salvaged leg. Although both Ted and Shelley are committed to managing complications as they arise and Ted scored low on the Decision Regret scale, Shelley recalled missing information that might have factored into making the decision:

> I don’t remember reading anything about whole femur, knee, and half of his tibia, a third of his tibia, is all metal. How much those quads would have made a difference? Never knew that until years later that that was the difference. That kind of makes me angry, that I didn’t know that beforehand. Because I think those things needed to go into the decision-making. It would have been helpful. I don’t know. Would your life have been different if you had amputated? In what ways? Yes, it would have been different but in what ways? You wouldn’t have had pain. You would have more mobility issues. I think you could have overcome them more, maybe. Again, I don’t like to look at it because I can’t go back and change it. I mean, make a decision and make it right. That’s what we’ve done. Those coming up with that decision, yeah. You need to know some of the things that maybe are not the best. You know, maybe it’s not going to work so much for him.

Vignette 16: Uma & Vera

Vera was 11, almost 12, when she was diagnosed with osteosarcoma in her distal femur; she started her treatment on the day she was going to have her birthday party. She was an avid skier, played baseball, and enjoyed dancing. Vera’s surgeon decided that a limb salvage surgery was best for her. Vera, now 14, and her mother, Uma, invited me to their home in Illinois for an in-person interview. We chatted briefly while the coffee was brewing and I met Vera’s brother and her father before we made our way to the living room to begin.

Issue domain: No options offered. Vera and Uma expressed frustration with their medical team at the start of the interview. They made many attempts to meet with the surgeon for more than two months after the diagnosis, according to Uma:

> They kept making appointments for us and then canceling the appointments because something would happen with the doctor's schedule or whatever. We literally didn't see the doctor until two weeks before surgery. We'd been asking. I mean, it was March, March 21st. We'd been asking since the beginning of January.

After Vera was diagnosed, Uma started researching, "I wanted to know what our options were, right? So, I did a lot of research. I talked to a lot of people." One of Uma's friends is a naturopath; she had experience doing research after her own daughter was diagnosed with cancer, so she helped Uma get started:

> She went online and found the osteo groups and asked some questions in the osteo group about, "Is there anything else happening besides MAP? Is there a doctor that is doing anything different?" She did all that for me, and came back and said, "I don't know if you're ready to be on these forums, but here's where you can go when you're ready."

Uma joined all of the osteosarcoma Facebook groups and some oncology groups not exclusive to osteo in an effort to be informed about surgical options for Vera.

Issue domain: Information seeking. Uma's friend recommended that she attend the osteosarcoma conference, MIB FACTOR, in Miami even though it was soon after Vera's diagnosis:

> She said, "There's this organization and you need to go to this conference." That was one of the hardest decisions I'd ever had to make in my life was going to that conference, because she was pre-surgery. I mean, that was what—was that still in January? …, she was just a month in and she was having chemo that weekend, and I went to the conference because my friend told me that I needed to and I needed information.

Despite the challenges, Uma felt her time there was helpful in understanding osteosarcoma and treatment option, "I heard you talk about Zoe 10 years later riding her bike for the first time. I was like, 'I do not want that for my kid. I want them to be able to move.'" Listening to parents like me helped clarify her quality of life goals for Vera:

> I heard other parents saying all kinds of things about what their kid could do, or what they all couldn't do. Vera was a kid who was a very active athlete. I wasn't saying she couldn't have LSS. I was just saying, I want you to be open to these other ideas because I want you to be able to run if you want to run. I want you to play baseball if you want to play baseball.

They met another osteosarcoma family who had issues with LSS and vowed to avoid a similar situation with Vera:

> They kept doing surgeries and surgeries and surgeries. He's been in a wheelchair for two years, right? All kinds of problems and finally, they had the amputation this past year, the last few months ago, right? I was like, "We're not doing that, just know." I mean, that was afterwards. That was what I was afraid of. That that's what would happen to us. I just wanted it in her head that we were not going to stay that way if she didn't have mobility. I don't want her to have to live like that. We watched him do that. Three years, trying to save his leg, horrifying.

Uma developed friendships with other parents who served as expert patients for her:

> Both of them, were really great about coming out and saying, "Don't trust this. Don't trust this. Don't trust this. Ask questions, ask questions. Push it, push it." Both of them felt like they didn't do that enough. It didn't work out for them. So, they've been really strong advocates for us because when we would have questions about what they were offering us, I would go to them and be like, "What do you think?" They've been through it with the same people pretty much.

Parents she met at FACTOR also shared resources such as the revered ACOR listserv where the expert osteosarcoma physicians offer input, "I learned about that one at the conference because somebody was talking about it. I went up and said, "What's that, that you're talking about? ACOR?" So, we got on there after that."

Uma also described a parent-formed group at Vera's hospital that would meet for support and shared resources. It was hard to find the group initially:

> I think the thing had been around for a while, but she was really active in trying to get people on it and using it, and has made a big impact on a lot of people because they're people who are feeling—our hospital completely refuses to acknowledge any parent-to-parent anything. Arrogance. I think it's arrogance. I think it's insecurity.

Meanwhile unbeknownst to Uma, Vera went online to do her own research:

> Well, I made the terrible decision of the first day that I got diagnosed, my immediate reaction was to go on the computer and look it up. Terrible idea. I saw all these pictures of failed surgeries of all this awful stuff and of dead people. I was just like, "Oh, my God." So, yeah, that was not a good idea. I didn't try it again….I don't think I even got to the article part. I was just like, "Nope. Done with this."

Uma added that they also did research together and looked up YouTube videos of the options:

> We looked at videos of LSS. I googled rotationplasty and there are videos of people who are like, "Look what my kid can do!" Yeah, more than with LSS, actually. LSS was harder to find videos of people successful and happy with LSS, maybe because it doesn't look so dramatic.

Issue domain: No surgical counseling leads to lack of trust. But time clicked on while Vera was in chemotherapy and they still hadn't met with the surgeon past the initial appointment. Uma began to feel misled about the sequence of appointments, "Our experience with our doctors related to the surgery was just terrible because they sold themselves as a team and said that they were going to help us navigate this process."

Issue domain: Patriarchal decision. By the time Vera did meet with her surgeon two weeks before her surgery, she had made a decision:

> Well, my mom tried to convince me to do rotationplasty because of the mobility and how much more mobility rate it had. She eventually convinced me that I would do it. Then, we went into meet with my surgeon and he said, "No, you're doing limb salvage. There's nothing you can do about it."

After waiting for almost three months to talk to the surgeon, Uma was shocked at his responses to her questions about options:

> It was already in March when he finally saw us. Then, he wouldn't talk about anything else. He just said, "You're having LSS." I said, "Well, why can't we talk about what her options are?" He basically said, "You don't have any other options."

Vera tried to talk about options with Dr. A, "Well, he said that he done rotationplasty to, I think, about four different people and that he just refused to do it on me. That he wasn't going to do it and that it was not my choice." Uma felt like the family's preferences didn't matter in the surgical decision when they asked about rotationplasty:

> It wasn't an idea that he couldn't, wouldn't do, whatever. He really just didn't think it was any of our business. He got to decide what was going to happen to her. He didn't acknowledge those things, those as options.

Issue domain: Recurrence risks. Uma's understanding from her research was that recurrence rate could vary based on the surgeon's skill in getting clear margins; she pressed for information from Dr. A:

> Going to the FACTOR conference and listening to, might have been Dr. B, talking about his success rates of LSS versus amputation in terms of recurrence rates and all those kinds of things. I said, "What's your recurrence rate when you do amputation versus LSS?" and he refused to talk, to answer the question. He refused to acknowledge that there's any difference. He refused to acknowledge what his history was. He refused—I mean, he just, he basically said, I know everything. You don't need to know anything, essentially, right?... I know that they track that stuff. I mean, I've heard doctors talk about it. He refused to answer any of those questions.

When Uma expressed her frustration with the impending surgery, he told her to get a second opinion though it was with another surgeon in the same practice. Dr, B agreed with Dr. A but his bedside manner differed dramatically and he asked Vera about her long-term goals:

> We really liked that doctor, actually still her doctor, Dr. B, he agreed with Dr. A that LSS was probably a good fit for her and that she would—they did ask what did she want to be able to do? If she couldn't run, what was the other thing that she would want to do? She said she wanted to be able to dance and musical theatre. They both said that they thought that she could do that.

Uma compared the differences between Dr. A and Dr. B when they explained the surgery:

> Dr. B explained to us exactly what was going to happen. Like, our surgeon (Dr. A) wouldn't explain anything to us….Dr. B told us that they were going to take her knee and her own surgeon didn't tell us that….So, then I called Dr. A's office. I'm like, "Are you kidding me? Are you taking her knee and you didn't bother to tell us?" The response was, "Yes, her knee." No, "Hey, I'm sorry we didn't explain that better", or whatever. "Yes, her knee" was the answer, like three days before surgery. At no point did they tell us any of that. We had to have this other surgeon say that.

Uma said they would have changed surgeons but they worked in the same town, "Yeah. If he would have been able to get us in within a couple weeks, I would have changed surgeons but we didn't because we were afraid to get off of our MAP timeline." Retrospectively, Uma felt unprepared to advocate because she initially trusted Vera's team:

> I wish I had known that I could question and really push my team. I wish I'd known that I needed to question it. I needed to make sure that we saw the surgeons ahead of time, that we shouldn't wait for them to decide to do it. Not to trust the hospitals, I shouldn't have trusted them.

Issue domain: Pros, cons, and patient preferences. When Vera was making the decision, she not only thought about what she liked to do at the time but also what she'd most like to continue doing:

> Well, I really like musical theater. So, I wasn't sure if I was going to be able to dance and walk on stage or anything like that. I'd kind of given up on the fact that I was going to play baseball again. Even if I had amputation or rotationplasty, I wouldn't be able to run the same. Things would be very different and I didn't really want that. So, I just kind of looked at what I could do in each aspect and what the things about them were.

Thinking about what the next decision might look like impacted how Vera thought about the first decision for surgery:

> I kind of was leaning toward LSS just because it was my real leg, then I could also always go back and do something else. What was mostly appealing about it was that I had my real leg and it wouldn't be as big of a obstruction.

She understood her what her limitations would be like for activity with LSS:

> I technically can, but I'm not allowed to because the impact it makes since I have like high impact bones replaced. If I ran enough, then I would have to have surgery way sooner and it was just tear up my leg. So, I decide not—so, my motto is "If someone's chasing me, I will run. If not, I'm not gonna."

Issue domain: Leg length differential. Given the lack of time to explore options with other surgeons, Vera moved forward with her limb salvage surgery as planned by Dr. A. It was two and a half years after her initial surgery when we met and some issues presented during that time that continue at the time of the interview. One was a leg length differential that could have been addressed by adding more length at the time of the LSS:

> One other thing that Dr. B said that he would do when he explained to us what the the initial surgery would look like, was that he would try and make sure that he's stretched her out just a tiny bit so that she'd have a little bit of that cushion. Dr. A refused to do that. He refused to tell us why he refused to do that. I mean, I know it's probably—I know if you stretch too much people have nerve issues, so I do understand what it is. I did all the research. He refused to acknowledge that he wasn't willing to risk that and why that was important to him.

Knowing that a solution for this problem could have been addressed in surgery was problematic for Uma and Vera soon after treatment ended:

> Her differential is about a inch now because he wasn't willing. When chemo was done, instead of being like, "Okay. You'll be able to heal faster, let's get this done." He never did a bone study. He never did those things to see if she was still growing and should he do it. He just didn't bother. He just didn't do it. So, we waited a couple more months. I'm like, this is getting bad. We went to see Dr. B and he was like, "Oh, no. You've got to fix this."

Vera eventually moved to Dr. B after surgery, but Uma felt he family was affected by dishonesty from the beginning, "They had sold themselves as a team. Well, that was a lie. Her oncologist, her nurse practitioner, and her surgeon were sold to us as a team, but they weren't a team."

Issue domain: Pain and lack of quad muscle. Although she had done well in the first two years and was walking miles at a time, skiing, even riding a bike, Vera was starting to experience pain and issues with the hardware after working short distances, "My hip would start hurting and it would feel like the bones were grinding against each other. My knee would stop working and it would just click and just stop bending." In spite of efforts to regain that muscle, they were unsuccessful, "We've done a lot of PT. In addition to PT—last year, she wasn't getting stronger with the PT."

They started working with a new physical therapist and personal trainer to find answers and discovered that the pain is related to the lack of muscle, "She had this like, hole where her quad used to be." This turned out be more complicated than they thought and would affect Vera for a lifetime:

> It stopped clicking when her muscles got strong. So, the lack of muscle was—I mean, it was a self-fulfilling thing. She didn't walk very much, so it didn't get strong enough to overcome those things. You weren't building up the muscle enough. Because she had such good—we were making progress on PT, but then her doctors took her off of PT because she had good mobility and she had good range of motion and all those things. Compared to other people with what she has, she looked beautiful but she didn't have the strength yet. Then, she went way back and she was not able to do as much because we weren't doing any physical therapy or any real muscle building stuff. She was losing the muscle again. What, I mean, really what's come out of it is, she's got to continually make sure she's rebuilding that muscle so that it stays strong. When she goes for weeks and weeks without it, her back starts to hurt, her hip starts to hurt, her knee

> starts to click. Now, whenever she's clicking, we're like, "Oh, you haven't been working out." Right?

Uma started Vera on a training program to build strength, "I made her go to the gym with me to a trainer who specifically worked on her leg and back strength twice a week… She hated me, actually. After a few weeks, she's like, 'You're right. It's working.' Then Vera went to summer camp for six weeks and got busy with life but she noticed a difference in that short time without the rigorous training, "Now, I want to go back." Uma agreed, "It was huge. The quality of life is very different."

Issue domain: Missing long-term outcomes. Looking back, missing information from the surgeon about what to expect long-term was a regret for Vera:

> I wish I had been warned about how much physical activity that I needed to continue after getting the surgery. Afterwards, I kind of just laid off the physical activity. Then, I got super weak and then, it was harder to get back into walking and stuff like that. I wish I had been warned about that besides from my mother.

Uma echoed how the lack of information impacted the family, especially after they researched and received information elsewhere:

> We're scared anyway. If you're telling me not to worry about it, that doesn't make me not worry about it, right? If you're not giving me the information to make a decision, obviously we're not going to know everything. You know, my doctors were very much like, "We are in charge. We know everything and you don't need to go down that road." All I wanted was people to be open to hearing this other stuff that I was hearing and giving me feedback, why do you think that isn't the way to go?

There was a deep desire to know what the options were and potential outcomes might be for Vera, even if the medical team couldn't say conclusively:

> I understand that we're not going to know a hundred percent but the unwillingness to talk about it, the unknown, was a huge barrier to getting through this successfully without incredible mental strain. It would have made a huge difference just to acknowledge, "I don't know, but what we know so far, this is why I don't think you need it." They were unwilling to talk about any of that at all.

Not responding or acknowledging the family's concerns only raised more issues:

> That just leaves more unknown and more uncertainty. I don't think it decreases it. Acknowledging the uncertainty, I do not think decreases—I mean, not acknowledging that and kind of, only staying on that path which is what I think definitely our doctors did. So far, nothing's changed my mind and you don't need to know that. It doesn't make people feel more comfortable.

Although they could fix the leg length discrepancy now, they are choosing to wait. Not knowing when the next surgery might be is part of their current uncertainty though they have accepted it as part of the overall picture of not knowing exactly when the next piece will break:

> I think a lot of people when they have a differential like hers, I think there's a lot of pressure on the surgeon to go in and fix it. We're like, "Hell, no. If she's all right, we're not doing that." We'll do that when—you know. It's like a jigsaw puzzle, right, so you can add a little piece. When she has to have it, then we'll add a little piece. Until then, hell no. I'm like, but we're going to wait until it breaks. There are people who it breaks within a year. There are people, I mean, there are people on the osteo who have had the same prosthesis for 30 years. So, who knows?

Ultimately the lack of knowledge translation from the medical team was overall the issue that impacted this family the most – from dismissing their questions and concerns to figuring out how to manage the lifelong impact of limb salvage surgery. When framed against the overwhelmingly negative process of communication, the clinically successful short-term outcome is not what mattered most to this family:

> He didn't give a shit what her long-term prognosis was. All he cared was that his surgery was pretty, and it was. I mean, I am fabulously grateful for that but that's not the only thing that matters.

Vignette 17: Wilma

Wilma was 30 when she was diagnosed with osteosarcoma in her distal femur and had rotationplasty. Now 36, Wilma invited me to her home in the mountains of Virginia where she and her friendly house cat greeted me at the door. We sat at her kitchen table with fresh brewed coffee for her interview, surrounded by her children's artwork and an array of whimsical sculptures and mobiles.

Issue domain: No information provided. Wilma started her story by stating that her symptoms were limited to swelling in one leg with no pain. She had an ultrasound and the radiologist thought it looked like osteosarcoma. Since Wilma and her husband are both academics (neither are in health-related fields), they immediately started to research it – which was both edifying and frightening:

> We started doing osteosarcoma research at that point. My husband and I both have PhDs, which is dangerous. So, we had access to lots of medical journals, too. We can understand what they say, which is really exciting but also kind of terrifying….my PhD is in science and technology studies… my husband is in physics.

Issue domain: No options. Wilma saw her surgeon about a week later, "[She] told me I was just going to get an amputation, a high, above-knee amputation, that's what I was told. I wasn't given any other options because of the size of it." However, since she and her husband had prepared for this meeting by doing their own research online and were ready to discuss options:

> So, she was telling me, at that point, I was getting a high, above-knee amputation. I'd already read about rotationplasty. I was like, "What about rotationplasty?" My spouse was like, "What about limb salvage?" To which she was like, "It's too big." She didn't answer a lot of our follow-up questions. She

> was like, “It will all be answered along the way.”… She was just not helpful in explaining anything at all.

I asked Wilma if this surgeon knew she was involved in academic research. She said, “Yeah…she seemed annoyed that I knew about it.”

Issue domain: Unmet needs for information. Wilma’s relationship with this surgeon did not serve her knowledge translation needs for her surgical options. However, Wilma liked her oncologist and wanted to stay with him. She realized she needed to get information elsewhere; so, she turned to a friend who is a horse surgeon:

> I liked the medical oncologist. We would stay with him. What we did is, we—couple different things. One of my friends is a horse surgeon. She went through all of my imaging with me, piece by piece, even though she’s usually looking at horses. Like, we talked about—because she does amputate animals occasionally, we had weird conversations from her veterinary world… I was grateful that she was willing to walk me through every single picture on my CDs.

Issue domain: Misinformation. She also had input from a friend’s daughter who worked with a surgeon. Wilma sought a second opinion and realizes then that the information she received from her first surgeon was wrong:

> We also have a colleague whose daughter works at (major medical institution. Through her, we were able to send the imagining that I had gotten to (major medical institution) and had one of the doctors look at it. One of the doctors who does rotationplasty said, “You’re definitely a candidate for rotationplasty. You know, you could come here if you wanted to, but you should go (another surgeon at a different hospital). So, we went to (that surgeon), who said, “Yeah. You’re a candidate for rotationplasty.” The first thing you were told was just simply incorrect.

The communication differences between the first and second surgeon were considerably different:

> I mean, the first surgeon just told me I wasn’t a candidate and wouldn’t explain why except that I was maybe too old…they were busy and didn’t want to spend time with me. That’s what I got from that interaction. Like, I was not their focus because they wouldn’t bother explaining. She was like, “You’re getting an above-the-knee amputation.” No talk about what limb salvage would look like. No talk about what a rotationplasty would look like…it seemed like a threat to her.

Even though the second surgeon reassured Wilma that rotationplasty was an option, she still sought advice from a third surgeon at a major hospital in another state, “She walked me through it. Everything she said, (the second surgeon) echoed. So, I knew neither of them was an outlier at that point.”

Issue domain: Autonomy and preparation. Wilma and her husband had choices for where they would have the surgery and considered themselves fortunate to be able to afford options, “We are affluent enough that that would not have been a problem for us. Like, we were very lucky." Having multiple layers of choices for the care both before and after the surgical decision gave them a sense of control during the uncertainty of planning ahead:

> I mean, we were willing to travel and put in the time, the limited time, we had between hospital visits to get this right, same with the prosthetist. Like I was going to get that all set up. I wanted to plan ahead as much as possible. You know when your world feels like it's out of control, I mean, the one decision we do get to make really as osteosarcoma patients is this.

Issue domain: Preparing children. With the assurance from both the second and third surgeon that her preferred option was indeed a possibility, Wilma began to prepare her children for the amputation and appearance of rotationplasty:

> We enjoyed the YouTube videos from (member of the rotationplasty community). My kids watched them a million times. At one point, my daughter goes, "When am I getting my rotationplasty?" It says it's for kids 5 through 12, and she was 5. She's like, "I'm just like you and I'm the appropriate age." As a five-year-old, she asked me when she was getting hers. So, yeah, I mean, the videos were really helpful. They explained it to my kids.

Issue domain: Influential factors to the decision. Form was less important to Wilma than function when making the decision. She also brought up no phantom pain with rotationplasty and a better fitting prosthesis as factors that influenced her decision:

> I was a 30-year-old married woman… because my choice was between high above-knee amputation and rotationplasty, it was going to look weird no matter what. I'd rather have functional and weird, and no phantom pains and weird, especially because it was high. The likelihood that I would have had to wear a belt with my prosthesis on it and navigate bathrooms in new ways. Right now, I can sit on a toilet and it doesn't touch my business which is great.

I asked her if her younger self as an adolescent might have been more concerned about form over function, wondering if she thought age impacted priorities in her decision. She said, "Probably, but no matter which way you slice it - oh, puns! - it's never going to look the same. Like, it's never going to be the same."

Issue domain: Permanent decision. Even though Wilma seemed comfortable with her decision for rotationplasty, it was still a difficult decision to process, especially given the length of time she had to think about it. Wilma struggled with knowing that some aspects of her life would be forever changed by her surgery:

> Yeah. I knew about three months ahead that I was getting it, which was really hard. Right, so I thought about everything I did - This is my last time doing it with my leg. I danced at both of my kids' birthday parties in December. My rotationplasty was scheduled for January. Right? I did all of the things I could do. I thought I wouldn't dance again. I'm still just as bad of a dancer.

Issue domain: Visible disabilities vs. invisible disabilities. While it was hard to think about the last time she'd be doing things like dancing, she felt that in some ways, having a limp with rotationplasty that was visible would be different than having a limp with a bad limb salvage surgery:

> I think that the hardest move is getting used to the gaze of other people, and the invasive and judgmental gaze especially if you're young and disabled. I mean, what's nice is there's an explanation for the way my body moves that's obvious. I feel like a lot of people who perhaps have poor limb salvages or who have cerebral palsy or MS, the way in which they're interrogated by other people for the way they move, is very different than an amputee because I wear it out on my body.

We talked about what it was like to have strangers ask and sometimes comment on your body. I told her that my daughter, who is a private person by nature, is learning to negotiate curious questions from kids and intrusive questions from adults, "Yeah. That's a whole thing to learn how to manage. I mean, I feel like becoming disabled is like getting a new extra job where you have to justify your existence and explain your body to strangers."

Issue domain: Lack of outcomes research. Wilma talked about continuing to seek information during her chemotherapy about rotationplasty after she'd already had the surgery. She was not seeking knowledge to make the decision but rather to know what she could expect – she wanted that data:

> Yeah. I mean, I found some literature about the comparison of outcomes and children, between above-knee amputation and rotationplasty. A lot of it was like looking at self-esteem and body image. Then, a lot of it looks at sports things. A lot of the cases in the literature are male and like, teenagers. So, I wanted the data, I sort of had to think about. At that point, like at some point while I was getting treatment, there was the first study about people over the age of 50, like two rotationplasties done on 50-year olds, essentially, 50 or 60 years old which was really interesting to read about. Back then, I already had my rotationplasty. That wasn't really part of my decision process but it was good to start to see more adults in the literature, although they're still under—it should just be used a lot more on adults is what I would say. I went in and she just told me it wasn't an option. Most people would just say no because all the literature was about children.

Once Wilma decided she wanted rotationplasty, she started searching for information on what she could expect as an adult. The information was limited, mostly moms who had sites about their kids:

> …the one site we could find about rotationplasty is run by someone's mom. There were like two moms of sons that did sporty things. So many of those sites are just all about, "Look, my kid can do sports things." For me, that information was pretty useless. Like, my questions were about pain and function, and also maintenance, right. I feel like you can waste a lot of hours with physical therapists and prosthetists that don't necessarily make your life any better.

Wilma felt the process of making the decision was different for her because she was an adult making the decision for herself and speculated on how that might be different for parents making the decision for their child:

> So, there's a lot of managing my own records. That sort of decision tree probably looked a lot different than a lot of the kids and their families, because of course, making decisions about my own body isn't the same as making decisions about someone else's body. So, the stress of it was probably a lot lower. I committed to rotationplasty, like I had one conversation with (second surgeon).

Issue domain: Knowledge translation, options, and complications. The relationship with the second surgeon was very different from the first. She said he gave her a better understanding of the complications that might occur given the size and location of her tumor:

> What we did is, we played out each of the scenarios, given the size of my tumor, given the vascular involvement, what each of the surgeries would mean. My tumor was like, was over ten centimeters across, so it was large enough that it was starting—like, the reason my lower leg looked large was because it was cutting off some of my blood flow. Essentially, I did have a blood clot but it was a blood clot produced because there was a bony tumor that wasn't letting the vascular things happen… It was the distal end of my femur.

Given the tumor location and size, her surgeon didn't recommend limb salvage:

> I mean, it was just starting to go into the knee joint. It was enough into the knee joint that if I had gotten a limb salvage with something that large and in (the second opinion surgeon's) opinion, I would be unhappy with that limb salvage and would experience pain regularly from it.

She felt that most surgeons would recommend amputation given the location and size, "Most people would tell you, 'You're getting an above-knee amputation - I'll tell you, if we were to do the limb salvage, you'll want me to amputate later' because of blood flow." Wilma weighed all three options but given the potential outcomes, she felt she only had two good options – which she counted as a plus:

> So, the only good options were above-knee amputation and rotationplasty was in some ways makes it a lot easier, right? Limb salvage would have been so unsatisfactory given my tumor's width and the vascular involvement at that point. I would have had trouble getting the right circulation and there's just too much going on.

Issue domain: Setting realistic expectations. Wilma's needs were very clear but she feels rotationplasty gets marketed to parents for reasons that aren't realistic long-term:

> I'm not sporty but I live in a very hilly place. I live on a campus that has a million stairs, some of which I've protested at this point. I mean, for me, rotationplasty isn't about sports. I feel like it's often sold to parents that way. The fact is, most kids who play sports, aren't going to play those sports for the rest of their lives. That's highly unlikely. I feel like people put too much emphasis on sports in general for kids. All the injuries that kids have by the time that they get to college now from intensively playing sports are pretty serious.

Even though Wilma chose rotationplasty, it was still a challenge to get used to her new body. She described her first conversation with the prosthetist she later chose as he helped her to accept the commitment she'd need to have with a prosthesis:

> So, we had this conversation. (He) was very straight with me. He's like, "Your body's never going to be the same. You got to get ready for it. You got to get used to the idea. Even if you're highly successful as an amputee, it's not your real leg. Right? Sometimes it might feel like that but they're going to be times you're unhappy with it, too. You know, prosthetics have gotten better, maybe, but also a sort of mastery you have to do as a patient is pretty serious as well." So, (he) had this really like, very much helped me accept it. Right?

She felt she had more realistic expectations for what she could do with a prosthesis than most amputees:

> I think a lot of what we're told about prosthetics is, "Oh, they're getting better. Everything's going to be great. You should take up running." Running's terrible. Why would anyone do that anyway, even if they have to need legs? Anyway, okay, the sort of fantasy people have about prosthetics is one that, you know, for most people's life it doesn't look like that. If it does look like that, it looks like that on one day. That's your best day…

Wilma cited cost, low maintenance, her busy life, and the distance she'd have to drive to see her prosthetist as contributing factors to the componentry she selected:

> [My prosthetist] and I had a real conversation about like, "So, how's an above-knee amputee with all the swelling and shrinking you have, the number of prostheses you're going to need." It's going to be higher, right? My foot isn't going to change shape radically, like I'm a grown adult. Here's the foot. You know, we might need adjustments along the way, but in terms of durability and my body fitting a device, what it looks like in a rotationplasty is much more satisfying. So, the maintenance issues were part of it for me. Like, I have a life to live and I don't want to be in the prosthetist office all the time especially since there's not—I mean, the local, I know some people who see the local outfit and they may like it. They drive the hour and they're satisfied.

Issue domain: Identity and visible disability. Although Wilma is very happy overall with her rotationplasty decision, she makes it clear that it is not without challenges:

> Oh, I mean, sometimes my gait is off, too. Like, like, when the weather changes, I was using a cane yesterday. Like, I have enough metal in my body, like anyone with anything replaced. I'm an adult. When the weather changes, I'm like, "Oh." I have hip pain, right, because I got some screws in because they put a big rod in. So, like, I do have pain. I limp sometimes.

Wilma's identity before her amputation included having an invisible disability, one that is not obvious from looking at her; she already identified as a member of the disability community. However, part of her new identity as an amputee is a shift to having a visible disability; she talked about how this makes her more credible as a representative and gives her a new role:

> I'm, sort of, unimpeachably disabled as someone with a physical disability which means a lot of people who complain about their disability status are invisible,

> such that people say, “Well, you’re not really disabled”, and discount what they have to say about disability. Being visibly disabled lets me do this work and gives me the privilege to me able to do it.

Issue domain: Pain management. Wilma speculated that her pain experience might be different if she lived in a less rural area with more specialists trained to manage pain:

> Part of it is, I think if we lived in some place where physical therapists had specialized training that I would be better off. A lot of my pain is related to the hardware, which I would have in any of the scenarios except above-knee amputation, right? They had to screw in this rod that keeps all my business together.

Managing pain is a part of Wilma’s life but she doesn’t feel that’s exclusive to her surgical decision, “There’s pain in every scenario, right? Like, that’s also the other band-aid you’re going to have rip off. Your body’s always going to be different. There’s pain in every scenario.”

Wilma’s experiences around making the decision were punctuated by a professional background in research coupled by zero knowledge of osteosarcoma. Her diagnosis led her down the path of early research to find an option that was not offered to her. Acting as her own advocate, she sought a surgeon who would give her the option she wanted. She continued to have knowledge gaps that were sometimes met with her own information seeking online and sometimes with providers like her second surgeon and her prosthetist. Wilma and her second surgeon made the decision together that rotationplasty was the best option for her – he provided her with different potential outcomes with each scenario along with the risks and benefits of each while Wilma chose based on how she wanted to live her life moving forward in her roles as an academic, wife, mother, and independent woman.

Vignette 18: Xena

Xena was 33 when she was diagnosed with osteosarcoma in the distal tibia two years ago. She had a five-month-old son and a three-year-old daughter at the time and had been a pediatric nurse, though she was not practicing at the time that we met. Xena suggested that we meet in the quiet evening hours of a hospital lounge in the hills of North Carolina.

Xena’s first sign of cancer was pain in her ankle but her medical training prompted more serious concerns, “The night of August 24th, I couldn’t get comfortable. My leg was bothering me. It gets worse at night. My nursing history was like, shit, this is bone cancer.” She didn’t have any patients personally who had osteosarcoma but she knew about it, “So, in my pediatric stuff, in training, we learned about it because it’s generally in pediatrics.” Her connections in medicine led to personal recommendations from providers. A nurse called her:

> She’s like, “We have found a mass. There’s a mass in your tibia. It’s about five and a half centimeters.” I’m like, “It’s cancer.” She says, “We don’t know what it is but there’s something there.” She’s like, “I talked to Dr. X and they

> recommending…”. She talked to the radiologist, I guess. The radiologist was like, “You need to call Dr. Y who’s the orthopedic oncologist down in [city]. He’s the guy. You got to go to him. He’s the guy.” I was like, “Okay.”

In addition to colleagues in medicine, Xena also leaned on her father-in-law who was a general surgeon:

> So, my in-laws had my kids. We went there. My father-in-law being a surgeon, like he had been on some board of oncological surgery with Dr. Y. So, I had a call from Dr. Y on Monday morning. I had a MRI that afternoon, a bone scan the next day in his office on Wednesday, looking at all the stuff, biopsy on Friday. By the following Friday, I had a diagnosis of osteosarcoma. It is like a railroad, it’s a runaway train.

Issue domain: Tumor location limits options. On Monday, she met with her surgeon to discuss options. Despite her training, Xena did not know what to expect with osteosarcoma protocols:

> In that appointment, he started talking about it. He’s looking at it, he’s like, “Well, we can take it out and replace it with cadaver bone.” I was like, “Oh, so I don’t have to have my leg cut off?” He was like, “Well, that’s never our first plan.” He’s like, “But, we’ll discuss your options as we get closer to surgery.” At this point, I had no idea what the treatment entailed. I had no idea that surgery wasn’t happening immediately. I had no idea that I was going to be in the hospital forever. All I had known were breast cancer patients, who go to the little infusion center, get their infusion, and go home.

The timeline for surgery differed somewhat for Xena since her tumor was in an unusual location, “He kind of told me when I would need to decide because he needed to know about ordering the bone because tibias are not, like distal tibias, are not readily available.” The cadaver bone would have to be ordered from California so they needed time to plan and put in a request if she decided on limb salvage, thinking they could save her foot by using a cadaver bone to replace her tibia. A metal implant not an option for Xena:

> I asked about it. He said, because of the location of the tumor, they wouldn’t have enough bone to get another end of the rod into to mount. It just wouldn’t be able to mount correctly. So, it wasn’t an option.

Xena’s surgeon discussed limb salvage as an option with her and with her father-in-law too:

> He looked and talked extensively with my father-in-law, too. So, I had a lot of counseling and being able to bounce ideas off of him which was a tremendous resource just having someone who knows what they’re talking about. It was like—so, to get clean margins for the limb salvage, they needed at least two centimeters of the distal tibia to be able to bolt everything in for the caging and everything for the cadaver bone to attach. That would mean they would have to fuse my ankle. They were only going to get paper thin margins that way. So, I was always going to walk with a limp and never run, jump, any of that kind of stuff.

Issue domain: Weighing risks and benefits. Xena wanted to know about what amputation would be like, “So, then, I was like, ‘Well, what would an amputation look like?’ He was like, ‘You know, functionally, your limiting factor with a below-the-knee amputation, someone young and active, you can do most anything.’” She considered how the long-term outcomes would impact her daily living and quality of life, as well as potential complications as she aged:

> I was like, “I have kids. They run into traffic, I have to be able to take off and run after them.” I guess that was my biggest thing in the decision making, was that I couldn’t be limited to not having any kind of high-impact issue. Then, I was also worried with having the fixed ankle and a limp, how that was going to affect hips, back, shoulders, everything else over the course of my life.

When she was younger, she was more active. After she was diagnosed, that previous activity became more important to her, “In high school, I played soccer. I go through bouts of running and not running. So, then it’s like, once this came, I was so worried about my mobility long-term. Then, I wanted to run again.” Although Xena was moving away from the potential limitations of limb salvage, she was also concerned with how an amputation would look, “I mean, that was my number one hesitation was appearance.”

Issue domain: Lack of long-term outcomes research. Xena used a variety of resources to find more information while deciding between limb salvage and amputation:

> Yeah, like I would go on PubMed. I think I like got access however I could trying to get logins to download, you know, Google Scholar. Trying to look for outcomes and I was really interested in survival and what was going to be better. Survival or event-free survival. I couldn’t really find much. They seemed to be pretty equivalent.

She became increasingly concerned about the risk of recurrence if she chose limb salvage and makes a decision:

> So, I was like, “I don’t have a good feeling about this.” So, from the very beginning my whole thing was like, “Get this out. I want this gone. Cut it off. I don’t care. Get it gone.” So, of course, you know, my father-in-law is like, “You made a very man-type decision on this. Just the get it out, let’s go, cut it off, I don’t care, let’s move on instead of the ‘I really want to keep my foot’.”

Xena said that pain wasn’t really a factor in her decision, “My biggest thing was I knew I was going to have pain either way. I did ask about chronic pain and which would, because I didn’t want to have chronic pain.”

Issue domain: Parenting values. Xena reflected on her reasons for choosing amputation and the benefits she feels she has now two years after making the decision, especially as her children are more mobile:

> I wanted to be able to chase after the kids. Like, I don’t want to have a super noticeable limp forever. It was the chasing after the kids. My son is a bolter. So, I

> am so glad now. I could tell—he was so curious from early on, sort of moving early on. I was like, "He's going to be trouble." I have to tell you with my daughter, if they're going somewhere that's in trouble or if something's happening. I need to be able to run, even if it's not the most beautiful run, the most comfortable run. I need to be able to do it. My daughter loves going to the trampoline park. I want to be able to do those things with her, you know.

She felt she had some peer support when she reached out to a friend from high school who had an amputation later as an adult:

> I don't know her story or anything. I know she had cancer as an adult, in her twenties. So I sent her a message over Facebook. I was like, "Hey, I'm being faced with this decision." She was just like, "Oh, my gosh." She lost her leg to cancer.

Her friend counseled her on realistic expectations for a below the knee amputation, noting travel challenges and the need for different prosthetic devices:

> She was like, "You know, the biggest hassle with it is having to get my leg on to go tend to the kids in the middle of the night." She's like, "When we go travel, I have a separate suitcase of all my legs. I have my everyday leg that I like to look like a normal leg as much as possible. It has the ankle that can wear heels. I have a ski leg for going skiing. I have a water leg. I have a gym leg."

Her friend also gave her guidance on advocating for herself to get the prosthesis she needs:

> She was like, "The biggest thing is just having a prosthetist who is on your side and being the biggest advocate for yourself that you can be." She's like, "Don't settle for something that's not working for you." Don't be afraid to speak up and all of that. I can be somewhat shy and timid but not when it comes to those sorts of things.

Issue domain: Limited peer support. Xena's peer support for options was limited to amputation, despite joining the Osteosarcoma and Ewing's Sarcoma Support Group soon after diagnosis:

> I never met anybody with limb salvage. I mean, I didn't know anybody with it especially not willfully. I feel like when you're in this world with Facebook support groups and stuff. It feels like the community is really big but when you think about how spread out it is and what different stages people are in.

She trusted her oncologist with the information he gave her, "I felt like he was very well-informed and up-to-date on stuff. He kept giving me the most recent research, he knew I was a research person, so he would present me with the treatment arms and all of that." He gave her detailed information that she felt he might not have otherwise given to other patients since she had a nursing background:

> He's like, "The most recent COG study on that versus MAP-IE", or "necrosis rates, this is what it says," or "at this point, it doesn't look like there's any better outcomes compared to the toxicity that you get with MAP-IE". He was like, "The standard treatment is to continue MAP." I was like, "Even with poor necrosis?"

> He was like, “Yep. You never know it might kill that one cell that is still floating around.” He’s like, “It killed ten percent. It might kill the ones you need.”

Issue domain: The decision belongs to the patient. Xena reflected on her surgeon’s communication style when he shared the options:

> He was very clear not to influence my decision. I was like, “So, you’re saying amputation is the better outcome?” He’s like, “I’m not saying that. I’m saying, it depends on what you want. What do you want? What are your goals?” Then, he did tell me, he’s like, “You know, as a orthopedic surgeon, you know, our number one goal is always to save the limb.” He’s like, “That’s what I’m used to.” He’s like, “In my career, earlier, I would have said absolutely do the limb salvage.” He’s like, “Because that’s the goal and big bad surgeon, that’s what we do. We’re going to fix it.” He’s like, “I’ve been humbled many times and that’s not necessarily always the best answer for people.” So, he’s like, “Ultimately, it’s your decision.”

Her surgeon discussed other decisions that might have to be made during surgery as well as the unknown after surgery:

> He’s like, “We go in there with the intention of it being a limb salvage but if it’s too far spread or it’s in just the major vessels and major nerves, sometimes we have to make a decision at that point. They wouldn’t know until they went in. Also, he was like, “You know, in an ideal world you would be more functional with the amputation but there’s those random patients that can’t get a good fit with a prosthetic.”

She said she continued to follow up with him outside of regular appointments to ask about long term outcomes, “We had a couple of phone conversations after that, really. I asked him, ‘Can I do this? Can I do that? Would I be able to do this?’”

Xena and her husband explored her options together, discussing risks and benefits of each:

> I think if anybody pushed me to make a decision, it was him pushing for the amputation. He’s like, “You know you’re going to get good margins. You know you can be functional.” He’s pretty much like, “It makes sense. Why wouldn’t you pick this?” I’m like, “Well, if I have the other one and I don’t like it, I can always get it amputated later.” I asked [the surgeon] and he was like, “Yeah.” I mean, you have to have good reason for it, not working out for insurance to go, “Okay.” They don’t like to cut off people’s legs for no reason, if you have a functional leg.

Ultimately it was the risks associated with limb salvage that helped her decide on amputation:

> I did ask about revision surgeries and if I was going to, if this was going to constantly be an issue throughout my life. He was like, “Yeah. At some point, the hardware’s probably going to loosen and we’re going to have to go in there and fix things up or if things break.” I was like, I wanted it out, done, over with. I wanted to get through my treatment and move on, which is not how this works. At that point, that’s what I thought. I thought it worked that way.

After Xena made her amputation decision, her surgeon still followed up with her to make sure that was her final choice:

> You know, I had already told him that I was leaning towards amputation. Then, the next time we talked I was like, "Definitely doing the amputation." I saw him. I was going in for a visit with my oncologist and he stopped in. He was like, "You said you want to do amputation. Tomorrow is the absolute last day I can order the bone if you want to do the other. Are you one hundred percent sure?" I said, "Yeah, I am." I even called him the next day and was like, "I'm still sure." Yeah, I mean, he wanted to make sure he held the option open for me as long as possible.

She valued the decision-making experience she had despite the challenges she encounters living life as an amputee:

> I felt like I was really empowered in the whole process. Days when I hate the fact that I have an amputation or when I just want to throw my leg against a wall and scream and cry, I'm like, "You know, when it comes down to it. This was my choice. This was my choice." That's what I have to fall back on. I went through all that. I went through all the what-ifs. I did that. It's just the whole—whether you have limb salvage or you have an amputation or you have osteosarcoma or you have breast cancer, cancer changes you. You view the world differently when you're faced with your mortality, I think. So, the normalcy that I'm searching for with hating my amputation wouldn't have been there the other way.

Although Xena did not know much about osteosarcoma, she felt that her background as a health care professional helped her both with the accessibility and the understanding of medical decision-making. The lack of information on long-term outcomes and potential limitations of function vs. appearance was problematic for her. Her surgeon was open and honest with her about the unknown factors that might impact her surgery as well as uncertain outcomes of limb salvage surgery. He was also open about his training, stating that his "number one goal is always to save the limb" but admitted that it's not always the best decision for all patients. He engaged with Xena to think about her values and quality of life goals while supporting her need for information. Ultimately, she felt empowered and has no regrets about the decision she made, even when she gets frustrated with her prosthesis. She said she is comforted knowing it was her choice and explored all of the risks and benefits before making the decision that was best for her.

Vignette 19: Yates, Zora, & Arthur

Arthur was 11 when he was diagnosed with osteosarcoma and had a rotationplasty. He has three siblings and was active in sports, playing both baseball and basketball in addition to hiking and camping with his boy scout troop. His mother, Zora, is a cancer survivor and his father, Yates, is a philosophy professor. Now 17, Arthur and both of his parents joined me for a video interview from their home in California.

Issue domain: The options. Yates started Arthur's story with the diagnosis when they had a meeting with a surgeon very early in the decision-making process:

> The first surgeon, pretty much, was in favor of limb salvage surgery. We were concerned about amputation. He was like, "Don't worry about amputation. We can do a limb salvage surgery." So, we didn't really discuss a whole lot further than that. That was pretty early on in the process. He just encouraged us that amputation was not necessary. He wasn't so much pushing the limb salvage as he was saying that we can avoid an amputation. That's what his words were.

At this point, the family started researching osteosarcoma, including the surgical options. Zora felt that Yates' academic background helped them deciper the information they needed, "So, all these articles and all the stuff that he read, the studies, you know, he knows at least something about it, what a study is and how to do one and all that sort of thing."

Yates explained that they went to another hospital for a second oncology opinion, but ended up with more information on surgical options:

> That oncologist, in the course of our conversation, said, "Yes, you'll have to have surgery. We can go down right now and meet the surgeon. These are going to be the options: rotationplasty—" by that time, I knew what a rotationplasty was. I'd looked it up—"and amputation and limb salvage would be our options." So, the oncologist took us down and met the surgeon there. So, we weren't seeking out a new surgeon. We were seeking out a new oncologist. It was because of that oncologist that we met the second surgeon.

Zora thought the location of the tumor influenced the options:

> I think the limb salvage was the initial thought because it was just in the proximal tibia. By the end, by the time [Arthur] got his MRI, the big long one right before surgery, the cancer had gone into his growth plate of the knee. So, that's one of the reasons why rotationplasty does seem like the definite option because he was going to have to lose part of his knee or have knee replacement with limb salvage.

The second surgeon explained more about Arthur's options, including risks of limb salvage:

> He drew on a piece of paper about rotationplasty. He had a couple of probably, I don't know who they were, interns with him. We just sat in a little tiny room. It was like, the whole day we'd been there because we met the oncologist as well. He kind of swooped in on us and just said, "Hey, these are the options and here's why Arthur would do really well with rotationplasty." Because he was 11 and then he saw Yates was 6'6" and he had a tall brother as well. He said, "If you do limb salvage, we're going to have to keep doing it."

Issue domain: Age of patient. Zora described Arthur's reaction to talking with the second surgeon:

> Arthur's pretty stoic overall, so he just kind of sat and absorbed what was going on and would nod. You know, I would ask him, "Do you want to know about this?" He would say, yes or no, and get further information that way.

Arthur added, “I was a part of all the meetings…they did want me to take part in all of the discussions and everything like that…but my dad usually asked all the questions that needed to be asked.”

Yates felt that the communication style of the surgeon really helped Arthur understand his options better:

> Really, Dr. X was rather exceptional because he talked directly to Arthur and looked directly at Arthur, just like a children’s doctor should do. He did make sure it was okay, but I remember him drawing the picture for Arthur, letting Arthur see it, asking Arthur questions. Always, when he came into the room, he talked to Arthur first. He was exceptional in that regard, I think.

Yates said, “He was 11, but we decided that we wanted him involved in the decision” and he wanted to be sure that Arthur was fully informed about his rotationplasty option:

> One of the things that was important to us was the aesthetics of it, that it was going to be his leg and his foot on his leg. We wanted him to see what rotationplasty looked like so that he knew ahead of time and was ready to embrace that.

Yates said that Arthur’s access to the internet was limited, “So, we looked up the stuff and brought it to him.” Zora said they watched a video on rotationplasty, “We watched that and we all just went, “Okay. There’s three horrible choices. Which one will be the most functional for [Arthur]?” Arthur’s parents found ways to explore the risks and benefits of his options:

> We weren’t going to just step in and say, “This is what we’re going to do.” So, we did a sit-down with him and we looked at all of the pros and the cons after the surgeon had talked to him. We sat down again with him and said, “Here are the pros and the cons. We are going to have to make a decision.” We did want him to make the decision.

Yates included the reduced risks of pain and no phantom pain with rotationplasty.

Arthur’s parents had concerns about the options; his mother had many questions about long-term needs of care:

> This was a major surgery. How long is it going to take him to walk again? What are the risks? What about infection? What does he need to what kind of limitations will he have? Then, all of a sudden, you start realizing a prosthetist is for life. That’s a whole concern because that’s a relationship that you don’t even think about. You know, I never thought the whole thing about a prosthetist. It never crosses your mind. It’s like, will my child live, first of all. That’s the thing.

Zora felt overwhelmed with information but was mindful that the surgical goal was to remove the cancer:

> I was just overcome by the whole—there were lots of options and the surgery was big. Yates kept saying to me, “Zora, the point of the surgery is that the cancerous bone is removed.” That was the thing that just kept me taking my next step.

Yates shared that Zora’s personal bout with cancer likely heightened her concerns, "So, she's had experience with the threat of her life with the chemotherapy. So, I think she was looking at it probably just being overwhelmed by returning of that sort of experience. Yates also weighed in on his concerns for associations of recurrence with the different options:

> The things that I was reading on the internet said that the probability of relapse was slightly higher, but it was within the margin of error. We couldn't find anything that definitively said that limb salvage was a significant sort of risk. The risk was there but was pretty slight.

There were cysts indicated in Arthur's lungs that were more concerning at the time than the risks of recurrence to the leg:

> At that point, we had some issues with what appeared that it might have been lung metastases at the time, some cysts in his lungs. They disappeared. We don't know whether they were the start of cancer and then disappeared with his chemotherapy or they were just cysts because he had had a cold the week or two before. So, we really don't even to this day know. So, we were more concerned about his lungs than we were about a recurrence of the site. Felt like the goal of the surgery was to remove the cancer, if it was removed with clear margins. That factor didn't really come into the decision about which of the three surgeries to go to.

Yates used his training to search for information on recurrence risks:

> I just did Google Scholar and started looking in medical journals rather than just the regular Google studies for that. They did show, as I recall, about a five percent higher relapse with limb salvage surgery but the margin of error of the studies was close to that five percent. So, it was really hard to say. Nobody would say definitively, this has a higher probability of relapse. They would cite studies that seemed to show it but they weren't conclusive.

While statistics were important to Yates, he did not overburden Arthur with numbers:

> They're fairly important to me because if you read the studies carefully, you're able to see how many people were involved in the study and how accurate the study can be in terms of the conclusions that are being drawn. That seems like a significant part of that discussion. So, yeah, the percentages and the numbers were important to me. I don't know how they were important to Arthur. So, I always try to be careful with him to not burden him with the numbers, not burden him with the studies. You know, when I see a study that says, the probability of return of cancer, if you've got some lung metastases or some bone—what do I want to say? Bone—in his lung. Yeah. So, he has calcification. So, the probability that the calcification was due to the osteosarcoma. The probability that that would have in terms of return of cancer, we just never wanted to burden him with those things. When it came to numbers, they were important to me but I also felt that it was important not to dump numbers on him.

Arthur was glad for this, "I didn't really get numbers. I didn't really get the numbers, like actual numbers and percentages. I just heard that there was a better chance of me to do what I wanted with rotationplasty." Arthur weighed his options carefully and compared them against his quality of life goals, functionality ranking high on his list:

> Well, obviously, I heard about both the positives and the negatives. So, obviously, limb salvage you'd be able to start walking right away but then, you won't be able to do physical sports and stuff like that. You'd have a lot of other surgeries, obviously. I didn't want to go the amputation route because that wouldn't allow me to be as functional as rotationplasty with the ankle acting as a knee. So, I'd be in control of my own knee joint instead of just having a prosthetic be able to do that.

Appearance was less important to Arthur, "It didn't really matter as much to me about how it looked, you know. Looks a lot different than how it would normally. That's not a huge factor inthe long run, you know?" Factors that were important to Arthur were more mobility outcomes and decreased risk of additional surgeries:

> Just how long it would be—I don't know. I guess I wasn't at that stage where I was thinking about the long run. I was, what would be my best option for being able to walk better and run better and do activities like that? Which one would keep me out of the surgery room? Which option would help me walk better?

Ultimately, Arthur felt like he was primarily the decision-maker, "Mostly, I made this decision."

Issue domain: Psychosocial support. On the day of Arthur's rotationplasty, the family decided it would be best if Zora stayed home with the other three children. During this time, Zora searched for information on the internet. She found an online blog penned by a mother about her daughter's osteosarcoma experience that brought her comfort on this day:

> I found her blog, somehow. I read through her whole blog about how they decided, when she was diagnosed, what their surgery was. I just read through all of that stuff. That was really helpful to me, that whole day. I probably spent two or three hours just reading through all the things. That gave me a really good picture. It was nice to have access to that.

Issue domain: Not one and done. There were early infections that impacted the success of the surgery. Yates explained that while they thought they were making one decision for one surgery, there were actually several more:

> One of our goals was with rotationplasty that would just be the one surgery and we'd be done. In spite of that fact, we had infections and problems with fusion, so we had a revision surgery 11 months after the first surgery. Then, we also—there was an alignment issue. So, the ankle didn't align with the knee as he grew. So, we've had two revision surgeries beyond that. So, we ended up having three revision surgeries beyond the rotationplasty surgery. That was a little frustrating because you go into rotationplasty thinking, unlike the limb salvage surgery, it's one and done. It's not always.

He said that they knew there were risks but these complications significantly delayed rehabilitation and return to Arthur's expected quality of life:

> We knew that going in. We knew that there were the possibility of complications and problems. You know, it's frustrating but you realize no point of being frustrated, it's just real life. So, this most recent revision surgery was in May of

> this past year. He’s still doing physical therapy. He’s still rebuilding some strength.

Arthur recognized his limitations but felt he was doing as well as could be expected under the circumstances:

> It’s going all right. I mean, it’s not—obviously, it’s not going to be back to what it was before I had the surgery but it’s going well. I’m still kind of recovering in my muscles from the previous surgery that I had, the revision surgery that I had back in May, the end of May. I’m nottdoing much active stuff right now. I’m just focusing on school stuff, like that. Yeah, I am able to do what I would like to do.

Arthur commented on his mobility:

> It’s going all right. I mean, it’s not—obviously, it’s not going to be back to what it was before I had the surgery but it’s going well. I’m still kind of recovering in my muscles from the previous surgery that I had, the revision surgery that I had back in May, the end of May. I’m not doing much active stuff right now. I’m just focusing on school stuff, like that. Yeah, I am able to do what I would like to do.

Retrospectively, Yates summed up the challenges of getting information best when making the surgical decision:

> Owning the fact that none of these options is what any parent would ever want for their child, but all three of these options give you a response to what’s going to kill your child if you don’t deal with it. So, you know, I’m much more a frank person about that sort of thing. I want to know, okay, well I have to do one of these things. So, which is going to give my son the best opportunity to enjoy the things that he values the most. It was more of a value issue than it was the probability of failure. We weren’t told that. I had to go look that up. I wanted to own that in my own mind and recognize that this isn’t a guarantee. This is something that has a good probability.

Yates reiterated that there is no right decision, stressing that for surgeons to do so ignores patient preferences and individual values:

> When you look and you find out that limb salvage surgery has other issues and other problems, footdrop, addiction to pain medications, and these sorts of things, then you realize all these things have their strengths and weaknesses, and have their problems. Nobody should be promised, this is the right solution. I wouldn’t say the first surgeon did that, but I think that he was close to doing that by saying, “You know, we can just do so much with limb salvage, now. There’s no reason to have an amputation. You know, limb salvage is the way to go.” He didn’t mention rotationplasty, so there wasn’t even that consideration. I think, you know, he didn’t own that not everybody values the things that limb salvage has to offer. Not everybody—he didn’t own any of the potential problems of limb salvage.

Vignette 20: Brody

Brody was 28 when he was diagnosed with osteosarcoma in his proximal tibia. He had limb salvage as his first surgery, followed by an above knee amputation at age 30, 3 months prior to his interview. Brody was currently reviewing his treatment options for recurrence in his lungs.

He invited me to his cozy home under a magnolia tree in the rural Georgia where he greeted me at the door, walking with his new prosthesis and a cane, a dog and cat trailing behind him.

Issue domain: Adult patients have few options. Brody started his story with conversations he had with his surgeon at diagnosis when he was told his options were limited because of his age. The only option he was given was limb salvage surgery with the repeated assurance that he was not going to lose his leg. His surgeon said amputation wasn't necessary due to the size of the tumor located in his tibia:

> He said, "You know, in younger patients, there are some options, rotationplasty. In your case, limb salvage is our best bet." He said, "You're not going to lose your leg. You're not going to lose your leg. You're not going to lose your leg." He said it three times. I was like, "Okay. Well, this is good." So, that was the only option I was really given as far as removing the primary tumor just because he felt that I was too old for rotationplasty. Then, he didn't see the need for amputation at the time. It was a relatively small tumor. I think it was only about 10 centimeters in its largest part.

Brody assumed that his age contributed to not having any options other than amputation:

> So, I didn't have any options. I wasn't presented with any. I was told that there was some other things that you do with this diagnosis if you were younger. Being in my situation, there weren't really an option for rotationplasty.

My interview with Wilma had taken place just a few days before Brody's and her first surgeon told her the same thing – rotationplasty was just for kids. She went elsewhere for her rotationplasty and was very happy with her decision. I asked Brody if he knew why his surgeon said rotationplasty wasn't an option for him:

> He just said because the healing time would be so much longer and because he had done these surgeries multiple times and the quality of life would be better with the limb salvage. I was on board for that at the time. I was like, "Oh, yeah. That sounds better.

Issue domain: Recurrence. Brody had a limb salvage surgery; two years later he had a localized recurrence – this time in his femur - and lung metastases. He started chemotherapy that proved ineffective in shrinking the tumor, thus beginning the conversation for amputation as the tumor had not only grown but was moving up in his femur:

> It went from just one spot, about 5 millimeters, that spot had grown to like 7 millimeters. Then, there was another spot that just a little bit higher up. So, you know the chemo had not touched it. So, that was when they said, "Okay. We need to have an amputation." That was the last thing I wanted to do, you know?

Brody decided to get a second opinion with an osteosarcoma specialist who reviewed his scans as necessary. Rotationplasty was mentioned as an option, albeit briefly:

> …I'd send all my scans to him… he's been involved from the beginning. So, I sent them to him. Said, "They're recommending amputation. What are your

> thoughts?" He said, "Why don't we get you up here? I want you to meet with the team that does rotationplasty ." I'm not sure what you would call it. It would be radiology oncologists, maybe. "I want you to meet with the surgeon." So, I took my scans up there, went and met with Dr. X. Met with them, and basically the whole team… That was when they said, "We agree with (your home team), the best case for long-term survival is just to go ahead and do the amputation."

Issue domain: Communication. Brody wanted to have his amputation in his home hospital, but they were too busy. This delay was especially concerning to him because the tumor was moving up his leg:

> So, logistically it would have been easier to do it in (home hospital) but I couldn't get—they couldn't get me on the schedule until the end of August to even get a consult…That was the end of June… It's going to be six weeks before I can get a consult with the guy that had done the five surgeries prior to. That was terrifying for me because I mean, I was on chemo and it had grown, and it actually moved up. I'm worried at this point. Well, I want to save as much leg as I can just to be as functional as possible.

Communicating with his home surgeon was also a concern given the challenges he'd had so far. Brody's endoprosthesis broke two times in the year after his limb salvage:

> The first time my leg broke—no, it was the second time. I knew exactly what it was because it felt the exact same. So, I sent him a portal message and said, "Hey, my leg broke, exact same thing as last time. Can you order the parts and let me know what the plan is?" I didn't hear from him for over 24 hours. When it breaks, literally, the only thing holding my lower leg on is my skin. I mean, there's nothing. It's nasty. I could take my leg and move it all the way over here.

It became increasingly challenging for Brody to communicate with his surgeon. A family friend had originally referred him to his home surgeon, and he contacted his friend again:

> So, that was when I got mad. I called my family friend that referred me to (home surgeon). I said, "Hey, this happened again. Last time, I came to your office, got x-rays, sent him a picture of the x-rays. Can you reach out to him directly and tell him what's going on and tell him that I need to do something?" I said, "I can't get ahold of anybody at his office."

When he finally did talk to his home surgeon, he was blamed for doing something wrong to make the leg break a second time:

> Yeah. So, that was frustrating. You know, the first time, he's like, "I've never, ever seen this before." The second—the manufacturer had reps there during the first replacement surgery and all. The second time, he said the same thing. So, he actually called the rep on the phone, sent him a text message of that same break. He said, "You won't believe this but this is (Brody) again, same guy." He made it out like I was doing something wrong. He's like, "What are you doing?" I was like, "I was walking through my kitchen to get a glass of water before I went to bed." It just let go.

Communication and trust in your surgeon are important factors when making a big decision like amputation. Brody decided to consult with his second opinion surgeon and brought up his recurrence, wondering how the cancer could come back if the tumor had been removed with clear margins:

> I asked him, I said, "You know, if they get clear margins. They tell you they get clear margins with the resection. Then, how do you have a recurrence?" He's like, "Well, with resections like that", he said, "it's really not as uncommon to have a recurrence as you think." He said, "Really, it's more like one out of every five." So, I mean, he told me that. I was just like, "Wow, that's twenty percent." That's a pretty big number.

Brody remarked on the quality of the time spent with the second opinion surgeon in that time:

> So, I had never heard those statistics until I was sitting in the office making the decision to do the amputation. I spent more time with (the second opinion surgeon) face-to-face in that first meeting than I probably spent with my surgeon in three years. You know, we sat for an hour and he answered every question I had, at the time.

He felt that his later lung metastases were connected to the recurrence in his leg:

> That's where most of my recurrences were. The only time that I had any lung mets were when I had a recurrence in my leg. So, I mean, I've had—my right lung, I've had three surgeries on it. My left lung, I've had one. Every time it was one nodule that was less than a centimeter. So, they were tiny and there was just one at a time.

Issue domain: Unclear treatment plan. Brody reflected that maybe he should have had the amputation earlier, at the time of his first recurrence:

> Looking back, when I had that first recurrence, probably should have done it then, you know. That goes back to not really being able to communicate with my surgeon. So, right before he took me back to take the spot out, he said, "It doesn't look like osteosarcoma to me." He said, "If it is—"—you know, I'm getting ready to go back into surgery. He said, "If it is, we'll have to talk about amputation." Then, that's the last thing he says before I go into surgery. That's the first I had heard of amputation. After the surgery, it took like a week and a half to get pathology results back. It came back as osteosarcoma. When I went in to see the surgeon post-op to take stitches out or whatever it was that we did, he said, "Well, we got clear margins." So, he said, "I think (your home oncologist's) got a good plan. We're just going to monitor it right now. So, we'll do a PET scan in six weeks." That was when they found the other two spots, so six weeks post-op.

Brody's home surgeon had not revisited the possibility for amputation, but without knowing if the tumor was osteosarcoma, he probably wouldn't have chosen amputation at that point anyway:

> Well, that was what surprised me. You know, he said it before surgery, and it came back as osteosarcoma. Then, I met with him. Then, all of a sudden, the topic didn't come up. You know, I don't know that I would have done it then without knowing that there was a recurrence.

He never knew what the treatment plan would be if the new tumor was osteosarcoma, especially since the surgeon had said amputation would be discussed and then wasn't:

> The idea is always in the back of your head, “Well, maybe we got it all and that’s it. It’s the last time.” So, but it just didn’t—there was never a clear understanding of what the treatment option should have been. My oncologist, he wanted to save the leg, too. I mean, that was the direction he was going. He didn’t want to have to do an amputation. I never got any other information from the actual surgeon along those lines. He mentioned amputation once and then, I saw him again. He didn’t mention it.

Not having a clear understanding of the surgical plan was a missed opportunity for planning, according to Brody. He also speculated that he may have had some role in that for not bringing it up again but thought the medical team would be planning that for him if they thought an amputation was imminent:

> So, this was not a very clear plan on his part because it was always—there was some kind of lack of communication between him and my oncologist. I may have dropped the ball somewhere in there, too, but I felt like they should have been coming up with the plan and let me know. So, anyway, here we are.

Issue domain: Changing surgeons. The lack of information regarding the treatment plan led to a greater awareness of a communication divide between Brody and his surgeon. He compared that quality and quantity of time of that time spent with the second opinion surgeon to the time spent with the surgeon who did his limb salvage, citing communication as his biggest problem:

> So, that was my biggest problem with (home surgeon). Anytime he called, he called from a blocked number. The only—I could reach out to him patient portal but that may sit in there for two days without being opened. I could call his office and leave a message and maybe get a returned call in the next day or two.

Communication and trust were clearly waning in the relationship between Brody and his home surgeon. Given his level of frustration and the length of time he would have to wait, it’s not surprising that he decided to have the surgery where he had been getting second opinions. He saw this as a chance to start over:

> So, yeah. I was frustrated with him anyway, so it did not hurt my feelings to go to (the other surgeon) to do the amputation, and just kind of reset. I was just thinking, “I’ve had all these recurrences and just have not had a good positive experience at (my home hospital). I’m just going to clean the slate, go somewhere else and we’ll start this new journey through somebody else.” So, that was kind of my thinking, too, just from a mental standpoint. I was just so frustrated. I didn’t want to do the amputation. That was the last thing I wanted to do.

Brody started to negotiate with the surgeon who practiced several states away:

> So, I was up there. I said, “Listen, what kind of time frame should we be on because I can’t even get in to see my surgeon?” He said, “Well, you know if you want to schedule the surgery now, we can schedule it.” So, this was July 17th. I had the surgery in X on August 9th. So, I had been up there twice in the amount of time that it would have taken to see my doctor.

Communicating with the surgeon from the second opinion team proved to be radically different than what he had experienced with his home team:

> I got his cell phone number. I could call him right now. I text him pictures instead of emailing. I got his nurse—his nurse called me two weeks ago just to say, “Hey,” to see how I was doing. The patient care—or, I guess, the relationship between the doctor’s office and the patient is night and day from the experience I had here.

Brody’s new team communicated well with his home team although they had different processes:

> Everybody is kind of on the same page as far as the treatment plan. So, once we developed such a good relationship with (the second opinion oncologist), the virtual visit is just great for somebody like me that lives in Georgia. He’ll block an hour out to go over scans. He puts all this information in an email and a PowerPoint. You know, it’s in chronological order. I can go back and look at my last email from (him). It’s got information from my very first scan to my last scan. It’s got all of my dosages and everything from the chemo… he’ll take screenshots of my scans. Say, I’ve got a lung nodule. He’ll take a screenshot. He’ll put it on there, put the date, and then whatever the treatment plan is.

The poor communication was limited to his orthopedic surgeon on his home team. The ability to communicate with his oncologist was not problematic, making the decision easy to have his surgery with the other surgeon:

> Now, that’s with my surgeons. Now, with my oncologist, I could call him now. Actually, I just emailed him direct a minute ago. My lung surgeon is the same way. I’ve got their personal emails and personal cell phone numbers, but (the home team orthopedic surgeon), he would just not give that information out. So, that was also part of the reason. Once I met the surgeon in (the second opinion hospital) and had his contact information, he scheduled me, had me in with surgery within two weeks. I mean, it was a no-brainer.

Even though he didn’t have his amputation with his home team, he still valued the relationship he had with his oncologist and he appreciated that his home team communicated well with Dr. X. He expressed the relief he felt when he made the decision to amputate:

> My oncologist—I love him….He and Dr. X, they know each other. They communicate really well. So, I just told (my home surgeon) “Listen, I can’t wait.” Once I made the decision to go ahead with the amputation, it was like I couldn’t do it fast enough at that point.

Issue domain: Self-advocacy. Sometimes information about the patient is missing when there are multiple specialists, even when they’re in the same facility. Having the ability to communicate with the various members directly allowed Brody to eliminate a ‘middleman’ and advocate for himself:

> Even, there’s still some lapses in communication between my oncologist and my lung surgeon, but being able to talk to both of them directly, I can fill the gaps in. Where with my orthopedic surgeon and my oncologist, I can’t talk to my orthopedic surgeon, so all the questions have to go through my oncologist to reach out to him directly.

Like many patients, Brody didn’t want to be a pest; he was sensitive to that as well as the doctor’s busy schedule. Yet he felt he had to advocate for himself once he knew the providers in the same facility were not:

> You know, I don’t want to be bugging (my home surgeon) to ask a simple question that I should be able to get an answer to relatively quick, you know. I mean, he’s got enough patients to worry about than without having to advocate for me with somebody that’s in the same facility that he’s in.

Issue domain: Pain. Brody reflected on the chronic pain he had from the limb salvage and compared it to the short-lived pain he had following his amputation:

> …the only real pain I had was, I had some really intense phantom pains for a week. Finally, I was taking gabapentin and we had changed some doses and finally got on top of that. That subsided after about 10 days. I could not believe then, how much more energy I had, how much better I felt because I wasn’t in just chronic pain all the time. I didn’t realize how bad my leg was hurting. With the limb salvage.

He expresses some decisional regret with the first surgery, wishing he’d had amputation from the beginning. Retrospectively he didn’t realize how much pain he’d grown accustomed to until it was gone:

> So, looking back, I was like, “God, I wish I would have done this from the get-go. I would already have a prosthetic and probably know how to walk on it and would be able to get around. I mean, I had to walk up and down steps just like I do with this because my leg just didn’t work right. The swelling, every day, just the swelling and just the ache, the dull ache. This time of year, when it’s cold and rainy, I mean, I just had no idea how much pain I was in. I didn’t realize physically, and I guess, maybe even emotionally how draining it was. So, after the amputation, you know, after the 10 to 14 days post-op with the phantom pains and stuff, once that went away, I couldn’t sit here. I felt great. I felt the best I had felt in two years.

The difficulty of executing the decision was not mitigated by the relief of making the decision, “You know, looking back, I was like, “God, I wish I would have done this sooner.’ At the same time, it was the hardest thing I’ve ever done, walk into a hospital and get my leg cut off.”

Brody didn’t think he would have chosen amputation if he hadn’t relapsed, “I don’t know that I would have ever made the decision unless it was completely necessary, but now looking back, I wish I would have done it sooner.” He felt reassured knowing that even though he has a shorter femur than what he’d like, he has more than what is considered to be clean margin and his surgeon padded the limb below it with tissue:

> So, the surgeon, he was aiming for—he said, “I want to aim for at least 3.5, 4 centimeters of margin. We ended up with 13.5. So, he got well above it. So, hopefully that’s it for the leg. It’s about eight inches from the hip point, so it doesn’t seem like much, but I actually got a good bit more of tissue below it.

Issue domain: Amputation expectations. Now that the chronic pain has gone away, Brody feels like his discomfort is minor and temporary relating to the surgery, and his limitations are mostly around the fit of the prosthesis:

> The only complaints I've got right now is just, I guess, I'm so recent from the surgery, my leg just changes shape throughout the day. So, you know, getting this to stay on is my biggest hurdle. As long as I'm up and walking, it's fine. You know, if you sit for a little while, I'll lose suction.

He felt prepared for these challenges though and has a cane that he uses while he's learning to use his prosthesis:

> …the guys at my prosthetic company, they were like, "Listen, usually it's around three months before people are starting actually walking with this thing." So, they just said, "Don't get frustrated, hang with it." I've not lost enough volume yet for any sock liners or anything. You know, that's probably not too far out. You know, I can get around fine, especially here at the house I don't really use that cane that much but out somewhere that I'm not used to, new places or when I go up to the office. Stuff like that, I'll have it just in case. I've been getting around pretty great.

He was not prepared for the challenges of his limb salvage surgery, however:

> I was under the impression this was a normal knee replacement, that I would be able to run and full function. That may be the case for some people, I don't know, but it wasn't for me. I could not run at all. My leg just didn't work. I could walk around fine. You know, I had a slight limp. there wasn't a whole lot of explanation as to the whole surgery process. It was mainly you show up this day, we're going to do it.

Brody felt he was unprepared for the lack of function with LSS in spite of intense rehab, "In my mind, at that time, I was like, "Well, if anybody can rehab it and get it back, I can do that."… but I did a lot of time researching extensor lag and there's nothing. There's no way to fix it. I mean, there's really not.

Issue domain: Resources for credible information. Concerns about having too much information or bad information was a factor in Brody's decision not to do much research on his own. He talked about not wanting to know some statistics, such as those for survival:

> I've learned in this whole process that sometimes information is bad for the patient. So, I've kind of even let my sister and my wife just take off, you know, go down that rabbit hole. For me, I don't want to read statistics. I don't want to read into when you have a recurrence, what's your long-term survival statistic number is. You know, that doesn't matter to me.

The only time they researched for additional information was when a decision had to be made:

> So, as far as, digging for a ton of information, the only information that we look at is when we have to make a decision on treatment plans. They've been on the same page for so long that there's not really been a whole lot of questions in my case.

When the decision for different kinds of chemotherapy agents arose, he searched online:

> I have looked some up. On some of the chemos, Dr. X is wanting me to start with a low-dose methotrexate oral and Votrient and there's a third one. I have gone and tried to find something on those. They're used for so many different types of cancer that it's hard to find something specific to where I'm at. So, it's hard to I guess to see through the weeds on what is appropriate and what is not based on this clinical trial and how many patients they had, and these age groups. That's hard to make a decision based on reading a clinical trial. There's just so many variables that you don't know if it would work or not.

When he was first diagnosed, Brody did look to the Facebook support group for some information, "When I started the chemo, finding out what to expect from a side effects standpoint. I used that Facebook page, it's great. I haven't really looked lately…I just kind of searched for what other people were doing in it." Much of Brody's information came from Jack, a new osteosarcoma friend:

> I met a guy [Jack] in the hospital doing chemo for this. He was diagnosed a year ahead of me. He was 27 when he was diagnosed. We were really close in age. So, his wife and my wife and my sister made really good friends… his was actually in his lower femur. So, I leaned on him for what to expect. We were doing chemo. When you do the methotrexate, you have to stay in the hospital until your levels clear. He had given me a ton of information on ways to make that better. That was the worst part to me. The side effects from the chemo, was not that big of a deal compared to just the mental part of being in the hospital for seven days. So, we became really good friends

Jack was a great source of information but there were some differences in their diagnosis:

> I had a really great resource with Jack that had gone—you know, he did the same exact chemo treatment. He had 100 percent necrosis. I had 15 percent. He had a ton of bone mets. Fortunately, it sounds like I have had a lot, but I don't think that we got it all from the beginning in my leg.

Jack also recommended the osteosarcoma specialist out of state:

> Well, he had several recurrences and it was bone mets. So, they had reached out to Dr. X but once Jack went up there, he's like, "Man, you got to come up here. This place was unreal." He says, "You know, the level of care is just great." He's like, "Dr. (home oncologist) is awesome," he said, "but Dr. X has been doing this forever. He's the man."

Brody talked about using the specialist as a second opinion for all his tests and about the relationship between his home team and Dr. X:

> That was when I started doing the virtual visits and making sure every scan I got, I'd send up there. We would get a second opinion on everything. You know, the whole time, he and Dr. (home oncologist) agreed with the plan we were on. The only time they've ever not agreed was right now, whether to do more chemo. Right now, technically, there's not anything to monitor. So, Dr. (home oncologist) thinks that if we're going to do chemo, we should have something to shoot at. Dr. X wants to do it kind of preventative but there's not a whole lot of information out there that it prevents it from coming back. So, I told them just to work it out. You know, somebody tell me what to do and I'll do it.

Sadly, Jack passed away several months previously:

> …after they resected his primary tumor, I think he went six or eight months and was great. Then, it just he had one or two ribs and a spot on his back that showed up. Then, they couldn't ever catch up with it. It just bounced around. So, he passed away in April.

Brody also said he got a lot of information from his wife and his sister who were avid online researchers on his behalf, "My wife and my sister, they think they're oncologists, now. (My wife) actually, she's a stenographer. She does cardiovascular imaging and works in a hospital. So, between her and like I said, my sister, they're on it." Much of the knowledge for treatment came from medical professionals, sometimes at other hospitals if Brody didn't get enough information from his home team or his second opinion doctors, "Either I reach out to (home team) or I'll reach out to (second team), one of the two. Early on, my sister and my wife had gotten some other opinions in (third major hospital system)." When Brody was in the information seeking stage, he met with the surgeon who ultimately did his amputation. He was advised on advances in prosthetic technology and the likelihood that he would do better with an amputation than with a rotationplasty:

> He mentioned it but he told me that—he's like, "You're healthy. You're strong." He's like, "I think you would do so much better." He said, "The recovery time in getting you into a prosthetic is a lot faster than if we did rotationplasty." It was an option, but he suggested amputation. He's just, he's like, "You look just like these people that came back from Iraq." He said, "The technology has made such big strides in the last 10 years that you'll be fine. You'll be up and going." He said, "You'll probably have more mobility than what you had with the limb salvage." So, they gave me the option and definitely gave much more information than when I was originally diagnosed. So, there was other options there, but we were all kind of on the same page as the amputation.

Issue domain: Recurrence risks. Brody discussed his biggest concerns when making the amputation decision; his biggest concern was to be NED – No Evidence of Disease:

> So, I had, you know, I didn't want to miss a ton of work. Then, as this whole process has gone on from the chemo and having to stay in the hospital. That was my number one complaint was being, basically feeling trapped. It took a little while to get over the feeling of being trapped because I couldn't run. You know, that was a big deal to me. I couldn't—because that's what I liked to do. So, I guess, my biggest concern was, one, what is the biggest chance for success because I want to get out in front of this. I'm tired—I would like to be NED one day, you know. So, that was my number one concern. Amputation gave me the best chance of that.

Issue domain: Return to work. His second biggest concern was his keeping his rehabilitation time to a minimum so he can be active again; his values included returning to work:

> Number two was the downtime, how long would it take to get back out to where—I mean, I didn't go anywhere for two and a half months. We just didn't. We went to a couple friends' houses that are really close friends. I worked from

> home luckily. Yeah, fortunately, I've been able to work the whole time. We do environmental consulting. It has made it harder not being able to be out in the field, but I can still take care of my responsibilities. The guy I work for has been great. My biggest concern was best chance for long-term survival and second biggest concern was how fast can I get one of these and learn how to use it and go and do. So, those were my biggest concerns.

Issue domain: Phantom pain. When Brody was concerned about phantom limb pain from the amputation, a specialist came in to talk to him about the plans for TMR – targeted muscle reinnervation:

> He had basically a hand surgeon come in and rather than just cutting the nerves off when they do the amputation, the—the way he explained it was they reassigned those nerves to a muscle group and splice them and reattach them to muscles so they have something to grow onto. The way he described it was, a lot of times when they cut those nerves, they'll create a cluster at the end of them. That's what generates all the nerve pain or the phantom pains that people have. So, they had had really good success doing the TMR.

When it came to information seeking, Brody says he did look for information on TMR:

> I tried to read up on it after (amputation surgeon) mentioned it the first time we ever met. The oldest study I could find was from 2016. So, I think it's relatively new. That was the earliest I could find from an actual scholarly article. I think that they started it with upper-body amputations more than—I think that they were saying the phantom pain was a lot worse in the hand if you lost your arm, like in your fingers and stuff. They had started using that for all amputations, so.

He searched online for information, "Well, I was kind of just googling and searching what TMR was. So, you know, you got your Wikipedia, then you also can scroll and find some scholarly articles from universities. So, I was just googling it." For the most part though, Brody felt comfortable with the medical information he was receiving directly from professionals, "Once I established that relationship with (Dr. X) between Dr. (home oncologist) at (home hospital) and Dr. X at (second opinion hospital), I've not had the need to find any more information." He describes his ongoing relationship with his oncologist as positive but dreads going to visit him because he's in his home hospital:

> I love [my home oncologist]. I enjoy going to see my oncologist because he's fun to talk to and he's super smart. I wish we had more time just to talk outside of the reason I'm there. I tell him. I hate coming down here. I don't mind seeing you, but I hate coming here. I don't even want to go by the infusion center. I would never go back up on the seventh floor. That's where I had to stay for methotrexate. That was, that was awful, God.

As a young man ready to start his family with his wife, Brody's primary concerns were surviving recurrence, staying NED, returning to work, and managing pain. While he maintains positive relationships with most of his medical team, both at his home hospital and at the out-of-state hospital for his amputation, it was a long road to get there. Communication was one of the most prominent issue domains around surgical decisions, from what his options were to how a localized recurrence would change the treatment plan to waiting more than a month to discuss options for a fast-growing new tumor. The relationship domain was negatively impacted by his inability communicate effectively with his home surgeon, leading him to seek a second opinion and eventually amputate at a hospital several states away. He appreciated that his medical team would communicate together at times but was frustrated when he had to be the middle man and fill them in on his current status. In spite of credibility concerns for online resources, he did use Google, Facebook, scientific articles, and other web-based options to research on his own. Information was also provided by his friend Jack and by his family members in addition to knowledge translation directly from providers on both of his teams. He wanted research when he had to make a decision (risks for recurrence with a specific option) but not if it didn't contribute to the decision (risk for survival after the decision was made). Brody felt that his later lung mets were connected to the recurrence after limb salvage but stated that had he known he had a 20% risk of relapse, he still wouldn't have chosen to amputate at that time. He scored low on the decision regret scale for his LSS. While Brody valued some statistics - rate of recurrence when there are clear margins for example - he did not want to know the survival statistics after recurrence.

A brief summary of the vignettes. There were three main overlapping issue domains around information/communication, patient/parent concerns, and current complications.

Information/communication issues include information overload, misinformation, and missing information about options, what to expect with a prosthesis, long-term outcomes, peer support, limitations, potential complications, and uncertainties. Managing provider recommendations and patriarchal decision-making over patient preferences was a common theme. A bias for limb salvage emerged over other options as well as a gender bias against rotationplasty for girls due to the appearance.

Patient/parent concerns included the age of the child and how growth might affect the choice as well as function over time and psychosocial issues related to appearance. Surrogate decision-making for parents was an issue domain; parents were conflicted about planning for a child or for an older teen who did not want to participate in making the decision. The relationship between necrosis and risks for recurrence were also an issue domain as well as risks for additional surgeries, infections, and chronic pain.

Current complications are primarily centered around pain and negation of the pain experience such as dismissive remarks from providers. Desperation is also a theme as patients seek amputation or rotationplasty after limb salvage to escape the debilitating pain that accompanies everyday living. Complications that require additional surgeries are a theme mostly centered around limb salvage but not always; Arthur continued to require surgeries six years after his rotationplasty. Mobility issues and limitations were mentioned by all twenty of the participants, the degree of impact differed from minor inconveniences to the need for assistive devices. Many attributed functional limitations to missing quad muscles that could not be regained. These issues usually overlapped with pain as well. Most of the participants had either made or were in the process of making a new surgical decision or anticipated having to make one in the near future.

While not all the participants were involved in the first decision, they were all involved in the second decision or indicated that they would be if a second decision needed to be made. The lack of self-advocacy skills was another theme that emerged across the narratives; this was problematic due to the shock of diagnosis in addition to the lack of information needed to make the decision. Some participants indicated that the short amount of time to make the decision contributed to feeling overwhelmed while those felt they had more time felt they gained more knowledge to make the decision, although the patient preferences might still go unheeded.

Dissemination Domain

Recommendations for the decision aid ranged from the format to content suggestions. Uncertainty was frequently mentioned as hampering the ability to choose one option over another but not having enough information to make the decision remained the biggest issue. The gray area of not knowing the outcome of a decision is especially challenging for parents who are surrogate decision-makers for their children. Discussing uncertainty is an important part of the decision-making process and can impact the decision, according to Wilma:

> I think you have to make that information known, that there’s no perfect anything. Patients react differently. Adjustment periods vary. I feel like surgeons don’t want to talk about a lot of those things. I do think it’s important to be okay with it not being normal or to make it your normal. I assume most of this will be used by parents. Parents feel very bad for their children. That can get in the way of making decisions, making them in time to plan.

Yates suggested a values assessment component, “There’s a lot of footwork that you do as a parent to try and sort out what it is that you might be interested in. What are some of the values that you might need to start thinking about?”

There were three main themes for dissemination: format/structure, content, and psychosocial support.

Format/structure. The format includes suggestions for how the information would be presented, paper-based materials like brochures or booklets, online with a website, or some combination of both. One participant suggested an app that could be accessed from a smart phone. Structure includes units of information such as case studies, videos, images, and patient stories.

Paper. Given the short amount of time to make the decision, Opal thought a one-page document detailing the three options should be shared at diagnosis:

> Honestly, probably as you could fit it as neat and tidy on one sheet of paper. I mean, because you know, you're handed so much stuff. If it could just be something that's like, really just trying to show them the options and they can look at it from there. I really feel like with confidence I can say had somebody handed me something that said, "amputation, rotationplasty, limb salvage", from day one we would have chosen rotationplasty had it been offered. I know it's a huge surgery and that's there lots of complications and stuff.

She also recommended that resources be shared at diagnosis in paper format:

> Resources basically on a piece of paper. Not too many, because I know that I was so overwhelmed by just trying to process anything. If it could just be like, "Here are some options: amputation, rotationplasty versus this versus limb salvage whatever." Give three top, I'd say resources for each. To me, that would be the most helpful. Sometimes, just seeing it on paper and then being able to look into stuff on your own, is just enough to get your brain thinking that way, I think.

Phoebe thought reading might be good but not for younger patients, in part because chemo makes it challenging, "I think reading is more for the parents than young kids." Ingrid also thought that a paper format with appealing visuals would be best for young adults:

> For young adults, I think probably I wish there was a literature, like a pamphlet. If there could be an MIB packet or something, that explained each option, risks and benefits. So, something with nice info graphics, something that's eye-catching but something that really goes into depth about what the young adult might experience versus a child. A lot of the—what's the word? Literature, I guess, I've seen about osteosarcoma is kind of geared towards children and it's about children. So, I think having something specific for young adults or teenagers going through this that has info graphics along with information and that kind of thing. A little pamphlet that could be given out, would be probably the best way. That's how I would have like to have made my decision.

Shelley thought a booklet for each option would be useful if the hospital were to distribute it:

> I'm thinking like in the hospital, something to pass out. Probably a booklet with like, separate booklet for each option. You can have pros, cons, weigh each option equally and just see what kind of breaks and what type of situation they're starting with for each one. You discover the tumor before anything breaks, those are the options. After a break, these are the options.

Bert also recommended a paper format:

> A booklet or brochure is great. If it had recommendations for other sources you could see down for further information, that would be good. When I say that, I think that—at that time, especially looking back, you're so inundated with information and parents who are generally not equipped and not accustomed to making these kinds of decisions, have to make a hundred decisions a day, some

> bigger than others. So trying to find what you think is the best information but you can't help not to be inundated with stuff.

He felt that one resource was best:

> I think one big piece of information is better. I think people can generally take a manual or a booklet or a book and easily find the sections that are relevant to them. Having that kind of a resource, if that's something that's available to you, is great. I think if that's a doable thing, that would be very helpful.

Brody thought paper was not conducive to sustainability and used his own experience with patient education to illustrate his point:

> So, I've got stacks of papers where I've gotten on at-home care and tons of information from the doctors that I—it takes two weeks to get it out of my truck and then, I throw it on the table. Every once in a while, I'm like, "Where's that paper at?", because it said something about this or that. So, I don't know that as far as handing out a brochure or a book itself, I don't know if that would be as effective as it would having access at your fingertips all the time.

Paper and Online. Bert acknowledged that he may be partial to the written word so having two options for receiving the information would be best:

> I think the more ways that you can present the material, the better. I think people learn in different ways. Especially, even though I'm kind of a dinosaur, I understand that there are people who—my children do everything on their phones, so if you could marry published material with something that's reliable online, I think that would be the best, yeah.

Arthur (11 at diagnosis) recommended both a paper format and a webpage:

> Probably both. I mean, you probably have the website like with more immediate information that people would want to access right away. The book, would be the more in-depth thing where you would have all the statistics and stuff and people stories and stuff like that.

Arthur's mother, Zora, agreed that an online version with patient stories was best due to timing:

> I think the online because it's a timely issue. You know, you're told your child has cancer and in six weeks they're going to have to have a surgery and you have to make a decision right away. I think having it online, having some links to different people's stories, positive stories for each one of the surgeries, a clear list of what the prospects and concerns and problems. We sat down with Arthur and did a pro/con on a sheet of yellow legal paper. What are the things that are strong about this? What are the values? What are the downsides of each one?

Online. Opal suggested an online format, "I would think online if you can get some website, Facebook group, type of thing." Shelley also recommended an online format, "A website with the options would be great."

There's an app for that. Brody suggested a website but also an easy to access app:

> The most user-friendly thing is—I swear, this sounds crazy but everybody downloads an app for everything. So, if you had the information available in an app, even if it was just a broad information like, "You've just been diagnosed with osteosarcoma. Download this app, all this information is in here." That

> could be a website, too. The most user-friendly thing that I got is the MyChart where I can go and I can pull my labs up. Everybody's going to have a cell phone with them all the time. Nowadays, everybody's got a smart phone. I don't know that as far as handing out a brochure or a book itself, I don't know if that would be as effective as it would having access at your fingertips all the time.

Road map format. Levi recommended a format that is similar to a roadmap:

> I think it's going to be super beneficial going forward if you are able to put together—I don't want to say protocols. A program or a gateway or a path for kids and parents with their kids to start to establish, "Okay. Here is x amount of people who have had x, y, and z type of surgery from osteosarcoma of the blank." Being able to put out on a piece of paper or in a video or layout a roadmap, like literally a roadmap. All right, here you're diagnosed, chemo to here, you have the surgery here, and then, you could actually end up splitting here where this could happen or this. Right, I'm literally thinking of an actual roadmap. So, to be able to actually put that in a visualization form instead of just a conversation, I think that would be a lot more impactful to parents making decision and kids making decision for what they want to have done and what's going to best for them in the future. Obviously, every situation is different. You know, some have osteosarcoma of their entire femur, some are the parts, some in their knee, some in their tib, some in their tib and fib, some in there—right? Wherever it's going to be, they need to be able to look at. Okay, if I'm going to have this, that's going to lead to this which is going to lead to this, or B. Right? I'm kind of repeating myself here, but I think I'm just trying to make the point across for the recording, that's something super important for you guys to look into so it could be visualized for them

Flow chart format. Xena recommended a flow chart similar to Levi's roadmap, something she wishes she'd had when she was diagnosed:

> I'm really a big fan of flow charts and infographic-type things. I really liked the children's oncology group treatment arms. It's like, is there going to be a limitational function? Yes, no? Okay. Even just a flow chart of questions to ask your surgeon I think would be really helpful. Like, is this going to be limiting to me? How so? What capacity? Are there activities that I will be restricted from? Is this the same in all options? Just all these questions I had to come up with, it would be really nice if there was something to be like, "Here."

Decision tree format. Yates suggested a values-driven decision tree:

> Even a decision tree that looked at values and said, what are the things that you value? What are things that are important? Here is the way to achieve those values. Here's the costs and benefits and probabilities of success. Something where it was organized into a single place, I think would be helpful because you find out all of the information but you've got to go here for one thing and here for another thing and here for another thing. Then, some of the information is conflicting. So, which is the newer information? Which is the better information? There's a lot of footwork that you do as a parent to try and sort out what it is that you might be interested in. What are some of the values that you might need to start thinking about? You're faced with a tough decision. You're going to have to have a surgery.

Images. Quentin thought that including screened images would be good to include since people might be scared if they looked them up on their own:

> I think the biggest thing that kids look up now is Google images. You know, just typing in "what are the complications with a knee replacement." If you do that though, there's 30 different complications and that can really scare somebody off. If somebody took a mass production of everything and just put everything out there, which is nice but pharmacies do that, too, with certain meds. They might be treating one thing but you might have 20 different issues come along with it. You're like, "Why would I want that?" That's the thing is, when you type something online anymore, you don't know what you're going to get.

Vera(12 at diagnosis) had concerns about photos but thought they still should be included, "I feel like pictures of things that aren't too gruesome and won't scare a bunch of people would be good, but also just written out experiences and pros and cons and stuff like that." Mona thought including pictures with content would be useful:

> I mean, to me, just visual. Having pictures, whether you show the inside of what the leg's going to look like and then what it looks like on the outside. I think really explaining it with pictures and then pros and cons.

Animation. Levi, who was 10 at diagnosis, suggested animated videos for kids:

> Kids love cartoons, right? Putting something in a cartoon version…This way they can get that visual version of, "Okay, down the line, this is something that you might have to do, you know, even if you're 8, 10, 13 years old." I don't care what anybody says. Kids are freaking smart these days. Even if you're 10 years old or 12 years old, I still think them seeing that video of something saying, "Okay, we're taking this bad bone out, putting a good bone in that's not as strong but it's what you need. Down the road, when or if this does happen, you are going to have to do this to make you normal again type of thing." Right? That's like, I don't know, it's just how I envision it was explained to me.

His mother, Joy, added "Kids would more or less look at that than hearing the regular doctors talking and showing graphs." Levi agreed, "It just wouldn't make sense, you know."

Visual aids. Opal suggested visual aids to understand timelines for the options, "I think anything that you can look at, not a spreadsheet necessarily, but a simple spreadsheet. If you amputate, you're looking at three to four months of having a prosthetic and up and walking." There should be caveats for chemotherapy and complications:

> If you're limb salvage, you're looking at seven months to never of walking. That's very biased. I mean, the truth is that with the limb salvage when you're going through chemo is, you're not going to walk. You're not going to be weight-bearing before that six to nine-month mark. I guess, just even facts of that type of stuff… it was a huge concern of mine, is the risk of infection of having such a large implant in your leg. Then, personal stories of people.

Case studies. Aretha recommended a case study approach but also suggested content ideas for long-term outcomes and how to find a good prosthetist:

> I think best articles to read, case studies of people who have had each one and how long it's lasted and how many surgeries they've had." Would love videos of

> people describing what they did. I think, most importantly, I would want to see—what's the word, not case studies, not super-official, but seen, okay, this person had a limb salvage in this year and it lasted five years before surgery, and then 10 years. You know, I would want to know. This person had the rotationplasty. It's been 20 years, they're doing great. I'd want to see case studies of how long things have lasted. I think that's what's key with the limb salvage, is to be aware of all the things that you may be facing with it. Same with amputation. You know, you should be aware of the difficulty in getting the right prosthetic and rotationplasty, you know. Finding a good pro—I can't even say the word. What are they called? Prosthetist?

Quentin also recommended case studies, he was familiar with them as a healthcare professional too but he didn't think that patients would think to look them up on their own, "We've always based things a lot off of case studies and evidence based and stuff, too. Someone's just not going to go look for a case study."

Patient stories. Bert felt that patient stories were more important that statistics:

> Personally, I always weighed the patient stories more. I have a mistrust of statistics in general and it's such a rare disease that I don't think the statistics are particularly helpful when dealing with osteosarcoma. I'll tell you that one oncologist that we met at Sloan Kettering said to us, he said, your son is not going to survive seventy percent. Either he's going to survive, or he's not. So, if another doctor tells me it's a seventy percent survival rate, that's really not helpful to me.

He also felt that by having real stories in a decision aid that surgeons recommend would help families feel they are more credible:

> It would be very useful. I don't need them to be edited by the medical team. I just need to know that they're real patients and real parents, and they're not just somebody typing on the computer. The reason I say that is because Facebook, any other—our Facebook groups are good. Our administrators do a good job, but you still don't know where this information is coming from…I think if there was information included in materials that were given to me from the hospital and I knew the sources were reliable, that would be very helpful.

Vera (12 at diagnosis) recommended surgical recommendations followed by patient stories:

> I would have liked to have seen the doctor's point of view, like this is actually what's going to happen. These are the things that you can and cannot do, like specifically. Then, I would also want to see families opinions of "This is what happened. This is what happened to us. This does not mean it will happen to you but these are possible outcomes," because the uncertainty of what goes on is extremely nerve-racking but just knowing even some possible outcomes even if they're not great, you can prepare for that and be mentally ready for that when you go in for the surgery.

Gloria felt that the inclusion of patient stories would help to balance what the surgeon says:

> I feel like that would have to be something that's balanced. So, what the surgeon says isn't necessarily what you're going to experience. You need to get as much information from all the different sources that you could.

Cleo recommended including patient stories to hear the experiences of others and equates the patient's expertise with that of the provider:

> I feel like there's a lot of power in personal experience. I feel like if you google things, you don't have all the information. You don't have, you know, people's actual experience. I feel like your medical team is so important, not just because they went to school for it but also because they've dealt with it before with other people. I kind of feel like that's the same thing with the people in the Facebook groups. Everyone has different experiences but everyone's experience can offer insight. So, that has been really helpful to me before I had my revision. You know, people saying that they had dealt with the same thing or what had worked for them. It's just—you know, I don't have a lot of—I don't have any peers that have artificial joints in my personal life. So, it's nice to have people with similar experiences.

Bert was ambivalent about adding patient stories but said it would have been helpful for him to see people with various outcomes associated with different levels of limb salvage. Ramona highly recommended the inclusion of patient stories, "Patient stories, I think, are really, really important." Shelley and Ted also suggested patient stories:

> I'd probably say that personal experiences so it doesn't look all clinical. It's like, "Okay. Here's somebody who knows, who has been through it. This is why we chose it, why we thought it would work best. This is how it has worked best, our decision." Or, "This is how it didn't work." Like Z___, you know just, we tried it this way because that's the most conventional. That's what everybody kind of wants to do at first. Then, realizing no, that wasn't best for her.

Ted felt that this would be the best way to present long-term outcomes:

> You'd actually see like 10 years, like years out, time out from it. Just seeing how people adapted to it, like after surgery. The actual personal experiences, not just like, "Okay, this is how this works. This is how that works." Somebody actually saying, "Okay. This is how it worked for me."

Both Shelley and Ted thought all outcomes should be included, not just the successes:

> You got to show the other side, too, that it's not going to work for everybody. Yeah, I'd quite—yeah. You don't want to discourage people either, make them give up and go, "Oh, well this is ____", you know. I don't how you do that.

Ted added, "It's kind of just different for every person, really, so it's hard to say."

Zora pointed out that parents of younger children had a harder time since they had to make the decision for someone else:

> I think having parents talk about how they made the decision, especially parents of very young children, how they had to just make a decision for their child. We felt in some ways lucky that it wasn't us making a decision for Arthur. There's a lot of, not survivor guilt but guilt of having to choose something that's hard for your child. You have to choose something but the guilt of making that decision is really, I think, an issue. Having different parents talk about how and why they chose what they chose for their child. Then, with the older kids, some videos of how and why the older children chose what they chose.

Phoebe would like to see patient stories included:

> Mostly hearing people's stories and what happened, can kind of help you decide, 'That might work for me. That's not going to work for me.' I think just getting stories for each would be nice, different ages or where they're at in life. No one really talks about in treatment, about you surviving. So, I feel like they don't ever say future when you're in treatment. I never heard your future looks like this. I've never heard that.

Ted suggested videos or pictures would be the best way to show the different options and what they look like for everyday living:

> Probably pictures of the just getting around on the legs. Showing how much they limp, how much they walk, how difficult it is, how easy it is depending. Videos, probably. Going through physical therapy. Showing the leg in action, like working or not working, as it might be.

Aretha recommended videos:

> Would love videos of people describing what they did. I think, most importantly, I would want to see… this person had a limb salvage in this year and it lasted five years before surgery, and then 10 years. This person had the rotationplasty. It's been 20 years.

Opal also recommended videos, "I would say videos are good." Phoebe also though videos would be best, "I think now reading is good, but I know my 15-year-old self on chemo, I couldn't read very much. I just couldn't. Maybe, like videos and a web would be nice." Videos were helpful to Vera and Uma when they were researching options, "I think the videos were big for us. We looked at videos of LSS. We looked at videos of rotationplasty." Uma did express concern about the reliability of online videos, implying the need for videos in the decision aid:

> I think people are posting their—it does scare me a little bit. There are people who are posting a lot of my kid's playing baseball and riding with their LSS. People are like, "Ooh, you can do that." Well, yeah, you can, but should you? It's a choice, but I think people have all this hope that, "Oh, my doctor says I can't run but look at that kid. He's running." That makes me nervous because I think that that—I think that's a little bit of misinformation. It's wishful thinking that this is a great idea.

Xena would like videos that show people over time, "I think a video could have been nice, just hearing perspectives from people who chose different options. Seeing where they are, 'x' amount of years out." Videos on rotationplasty were helpful to Ramona and she recommended that similar ones be included in the decision aid:

> I think the videos of Gabi are extremely powerful. There's another video also of somebody swimming. What's great about that is you see her take off her prosthetic leg and then kneel walk to the edge of the pool. So, you get to actually see, "Oh, you have this half a leg but you can still do things with it." I must have watched Gabi's videos so many times. So, I think the videos are really powerful.

Wilma recommended videos but very specific types of videos:

> I feel like we have videos but they're all sports videos. They're often not people narrating their own experience. There's like some overlay of a person telling you a story of triumph. The story always has to be triumph and not "I've adapted to my body." They're always overcoming stories and not "human beings are adaptive creatures that can handle whatever is thrown at them." I want different stories to occupy this space because so much of what's in rotationplasty is kids with sports. Like, I'm not a kid, I don't play sports. The information is useless to me.

Yates discussed the values vs. the risks of negative patient testimonials given that all of the options have the potential to fail:

> I do think that somewhere in the tool, whether it's on the website or in a pamphlet, there needs to be you need a note to parents and to older patients that you need to be aware that none of these things are perfect, that all of these things are life-changing, all of these things have risk of failure. That's a part of the process, is saying, what does failure look like here? What does failure look like in other cases? So, you know, one of the things that we looked at was, well, if rotationplasty failed, then what? Then what's there seemed like it was going to be a higher amputation. Because his was in his tibia and not in his femur, there wasn't going to be as much of his femur removed in the first place. So, if the bones had not fused or they had not been able to deal with the infection, he would have ended up with an amputation. So, it was kind of like, well, that makes me feel a little bit more towards rotationplasty because I may end up with an amputation anyway. If that's the worst-case scenario, this looks like it has some prospects to be better. Just recognizing that all of these things have prospects of failure. Where do you go from there? I think that was important. It was very important to me. I don't know how to have testimonials of failures which would operate in an independent way to help people make decisions as opposed to causing fear and paralysis. All of these things have failure stories, so I don't know where to go.

Accessible to youth. Gloria felt that the older adolescents would benefit from having a decision aid since it's their body and often their decision:

> Sometimes, the parents do make the decision. In our case, we did not make the decision. She one hundred percent that was her. We had talked about it, but at the end of the day, I would have never felt okay making that decision with her age. There's no way, like none, because she had to live with that the rest of her life. She needed the tools to be able to make those decisions.

Content. The content recommended included everything from recovery expectations to what to expect in rehabilitation with the different options. Participants also suggested content in limitations and benefits of options, insurance, information on the prosthesis, how weight affects the prosthesis, statistics, long-term outcomes, and resources such as the osteosarcoma conference and patient portals to communicate with the doctor. Many participants such as Wilma and Brody talked about the need to understand how tumor location impacts the decision; Wilma said, "Our tumors are all different, in slightly different locations. Like, I think the factors are really hard to weigh at some point." Brody watched an osteosarcoma friend who had LSS play golf and sail through other physical activities while he struggled to walk; realizing that there are the location of the tumor impacts function with LSS was something he wished he'd known.

What to expect in recovery and rehabilitation. Mona recommended information on post-op and rehab, especially for the parents:

> Here's one thing that I would love, I'm going to be selfish here. As this evolves, the next step after you do all this, I would be really, really curious—I'm going to be blatantly honest because of what happened to Mandy. What is the rehab like for the patients that you talk with? I don't know if you've asked rehab questions. I'm very curious because Mandy had her amputation, I had to beg for therapy. I had to beg for it. Right when she got her amputation, we're back in the room and I have an aide and a nurse. I have no idea what to do with my kid who now doesn't have a leg. How do I get her to the bathroom? She doesn't know what to do. How do we move her from the bed to the bedside commode? I had aides and nurses who didn't really know what to do.

Several participants talked about how chemo affected their ability to participate in rehabilitation. Brody thought this should be included in the decision aid:

> It would have helped me mentally. So, I had a lot harder time mentally after my limb salvage when I was finishing chemo. I had this surgery. I'm trying to rehab. I'm trying to get back to where I was at, having all this pain that I don't really know why, if it's something I'm doing, if it's normal. On top of that, I'm doing chemo. From a mental, you know, mentally to get ready to do a week's worth of chemo, you have to kind of get in a place. I did, I had to get ready mentally to go and do that. You know, that's a battle in itself. I struggled a lot more just from spending so much energy and effort on trying to get my leg back to where I thought it could get to, while also doing chemo. So, I think it would have helped me having a little better expectation, or realistic expectation. Like I said, after the limb salvage, I struggled a lot more with chemo than I did prior to. Some of that's probably physical, too. That's a major surgery.

Gloria suggested content on the recovery process, "I think more about the process and what has to happen before, during, and after the surgeries. Knowing that would be helpful. For example, the nerve block and how they would plant them, probably."

Options: Limitations and benefits. Phoebe suggested clear descriptions of each option with the limitations and benefits of each, "I kind of think to is what you can do with each. I had no idea what I couldn't do with limb salvage, what limb salvage entailed." She recommended using plain language:

> All they said was cutting the bone and that was it. I think my mom had more medical lingo than what I did. I think sometimes doctors would talk in a more medical lingo. So, I didn't' understand fully what was going on. I think more in a layperson's terms would be nice, too. Like, "This is what limb salvage is." You can and can't do this. You could have multiple surgeries if your kid is growing. I know for some families, they didn't realize that. That was big for them. That's going under a lot and they didn't know that. Then, for prosthetics and stuff, I didn't know anything about prosthetics and stuff. It would have been nice, like I

> said, just terms and you know, "You'll be going to see a prosthetist sometimes more, sometimes not many times depending on how your leg fits." Then, rotationplasty, I see a lot of kids with those and they seem to do really well with them.

Quentin felt that information on recovery was missing for the limb salvage option, "I think another big thing is the knowledge in teaching those people, too, that when you do have a knee and hip joint, you have to be mentally ready for that, too." Mona suggested adding content around pros and cons of each and presenting them truthfully, especially related to long-term outcomes:

> I think there's a lot of negatives that don't always get presented with limb salvage. I think it's the reality. I don't know if you have to figure out what those percentages are, or of infection. So many people that I hear are like, "Oh, I have to go in and have this adjusted. It's been five years or seven years." So, then you're going back and having more surgery. That was, again, that whole risk of infection every time you go into have a surgery. So, I think just being realistic with it's not just—it is in the now, but it's also in the future.

She pointed out that there are negatives associated with all of the options:

> I think definitely being presented with the options. You know, with the amputation, what are the negatives? When you don't have a leg, you have to hop around. There are negatives to that. You have to hop around. You can end up messing up this leg. You hurt your good leg, then what's going to happen? Those are all things you have to think about. I had friends that said, you know, "You need to make sure you always have a wheelchair and crutches." "Why do I need a wheelchair for?" "Well, what happens if something happens?" Like, "Oh, I didn't think about that."

Vera would like to see pros and cons along with a visual for each one:

> I think what would have been really great, would be just a list of pros and cons of each one and an example of a successful of each one. Like, some kid riding their bike with an LSS. Some kid running with their amputation. Like, these are different things that you know, there are positives here. If you can get through this whole process, these can all be okay. These are the pros and cons of each one. Like, we kind of made that for ourselves. Right? We were like, "Okay. Here's the pros and cons of LSS here. Here's the risks of that. Here's the pros and cons of—you know?

Uma made suggestions for the pros and cons, "This one, you're not supposed to run and jump. The other one you got to deal with putting the prosthesis on, there's less chance of recurrence. There's less chance of more surgery." Xena suggested adding questions to ask the surgeon for your options, "There's some questions that you should think about asking when you're faced with limb salvage versus amputation."

Brody recommended including information on the location of the tumor since that affects the outcomes:

> So, there's a huge difference in it being in your proximal tibia and your distal femur just from the way that your knee works. I didn't know that either. I had talked to [an osteosarcoma friend] prior to surgery. He was getting around fine.

> So, that was the other expectation that I had was that, [my friend is] walking around playing golf, having a good time, and I'm having a hard time my knee not buckling, I'm falling down the steps.

Even though they both had limb salvage surgery, there were differences:

> That was frustrating to me but it was just a different type of surgery. Where the tumor is at, is a big deal. I had no idea that all of these ligaments that were in there, weren't going to be there anymore. You hear limb salvage but that's such a broad term. In regards to limb salvage option, you know, explaining the surgery based on tumor location a little more would be helpful.

Similarly, Shelley suggested that information about the potential removal of the quad muscle to get clear margins should be included in the decision aid:

> I don't remember reading anything about it being such a high [amputation] pretty much his whole femur, knee, and half of his tibia, a third of his tibia, is all metal. How much those quads would have made a difference? Never knew that until years later that that was the difference. That kind of makes me angry, that I didn't know that beforehand. Because I think those things needed to go into the decision-making. It would have been helpful.

Yates thought it was important to include information on where the tumor was located to understand the different levels of amputation:

> Understanding what the difference between a high amputation and a low amputation is and what the energy costs are for a mobility, I think is important information. Having it in a particular location, I think would be tremendously helpful.

He also said that knowing the risks and benefits ahead of time helped them make the decision:

> One of the things that one of his prosthetists asked us was, why did you choose a rotationplasty? One of the things that he said—why not just an amputation? One of the things that we'd said was, no phantom pain. Knowing, that was a factor in my talking to Arthur about what the costs and benefits would be. So, knowing that there's information that says, "Okay, so here's some of the things that you're going to have to expect. Here's some of the things that you would expect of a rotationplasty.

Insurance. Phoebe suggested information on insurance for RP and AMP: Insurance and knowing what I want and what I like, but it took a while. It just takes time. It'd be nice to have somewhere you can go and be educated on it.

Prosthesis. Phoebe highly recommended information on the prosthesis:

> I think a hundred percent because I spent a lot of time at prosthetists. Like, sometimes I spend a week there. Just knowing it's something that's going to be a long-term fixing and stuff, which I didn't know. I didn't know anything about cutting stuff to relieve medially and just some of the lingo, I had no idea. Once I became educated on that, I mean, I can take care of my limb pretty well without him until I seen it. So, I think if I would have known the process, it would have been a hundred percent more helpful because I feel like it took me three years to figure it out. It took me a while to become educated on prosthetics. Even with limb salvage or if people choose rotationplasty, I feel like you need to be

> educated on the long term… My biggest thing is I wish I would have gone and saw how a socket's made, how they make it, kind of like prosthetist shopped like you doctor shop. You've got to meet someone that you gel with.

Zora, mom to Arthur who was 11 when he had his rotationplasty, also said that information about the prosthesis was missing when they were making the decision:

> I wanted to add, as you were talking about what would be included. I think a good thing to include would be pictures of what the prosthetic looks like. That was a big mystery to me. We didn't know what it was going to look like, where the foot cup was going to be. [Another rotationplasty patient]'s looks real different because she has skin and whatever all over hers. Some of these other ones, will there be bleeds? Will there be feet? You know, what does the prosthetic look like for a rotationplasty patient?

Like Arthur, Wilma also had a rotationplasty and suggested information specific to their needs for finding a prosthetist who can fit them properly, "all prosthetists can make an above-knee amputee leg" but "if you get the wrong prosthetist then you're better off not having rotationplasty."

Weight: Phoebe would include information on long-term health care needs such as how weight affects the fit of your prosthesis:

> I think another thing too was being on your weight which I didn't realize with prosthetics. I know that we don't like to talk about weight in our culture but I'm really on my weight because if I gain weight my leg doesn't fit. It's easier for me to lose weight than to gain weight. I've lost over fifty pounds. So, that's hard to do with prosthetics. Like, I didn't know, I've gone through a lot of sockets if you lose weight. Right now, I'm losing a little bit of weight just from being in school. Me and my prosthetists have found ways to go around that with pads and stuff. I had no idea about having to maintain my weight the best of your ability, because then your socket doesn't fit.

Quentin also mentioned weight gain as a risk factor to include for LSS, "I think some people that have hip and knee replacements, you see them put on a lot of weight because they become less active."

Statistics. Generally participants did not think survival statistics should be included in the decision aid but if numbers were available for outcomes, they should be included. Xena would have liked to see the numbers associated with the diagnosis:

> Oh, I'm a numbers person.I was like, "I want to know the numbers of everything. I want to know.I wish I would have known more about genetic testing beforehand because I want to know. I want to know exactly what my odds are. No one every knows exactly what their odds are.

Nerisa would like to see statistics in a visual form included in the decision aid:

> I, personally, would have like to seen written statistics that I could visually see. Something that gave me information about children who have limb salvage versus amputation. Something like that rather than just him saying those things. If he had given us something to look at visually on a piece of paper, that would have been helpful.

Brody would like to see statistics on the successes, "The statistics on success rates with a limb salvage. I didn't know that until I was finding out that I needed an amputation. Just a success, you know, clear margins."

Long-term outcomes. Aretha suggested one focus one content focus on long-term outcomes:

> I'd want to see case studies of how long things have lasted. I think that's what's key with the limb salvage, is to be aware of all the things that you may be facing with it. Same with amputation. You know, you should be aware of the difficulty in getting the right prosthetic and rotationplasty, you know. Finding a good pro—I can't even say the word. What are they called? Prosthetist? Finding one of those, especially rotationplasty. So, you know, knowing what difficulties—basically, pros and cons of each one, I think. Seeing them, if there are any, long-term studies of how long this person has had it or how long people have had these different procedures and what has happened with it.

Brody didn't know what to expect with his limb salvage, so he suggested information about what to expect long-term:

> I would like to have had more information as to what to expect after the limb salvage surgery. Like I said, I was under the impression it was a normal knee replacement and I would be back to normal function in a couple months. Probably, that's the goal. I don't know if some people do that. Now, there's a little kid from my hometown that is 10 who's had Ewing's sarcoma. He was told from the get-go that there will be some other surgeries, some rod-lengthening processes, but playing high-activity is not in the picture. I was not aware of that, had no idea. So, that's one thing.

He felt that by setting realistic expectations for options would lead to less discouragement later:

> So, kind of setting some realistic expectations prior to the limb salvage. You know, where you're just not—in my mind, I was going to be back to a hundred percent. That's not the case. The only thing is, you know, with some surgeons I think there's—that's their goal, is to get you back to a hundred percent. So, I don't know that they would ever—you know, they're not going to go into it shooting for 80 percent, you know. They're going to shoot for doing the best that they're going to do. Sometimes, that's just not feasible.

For most of the participants, the first decision was not the last decision – many had to decide whether to continue to have surgery to save the leg years later or to decide on another option. Wilma felt there are other decisions that could be made after deciding on the surgical option and that these should be addressed in a decision aid too:

> So, there are some outcomes I just wasn't afraid of. I was afraid of surgery but you can cope with whatever body you get, no matter what you choose. You can always change your body later. It's not ideal. You can get a new prosthesis. Try on bodies until they fit. You're seeing more rotationplasty after limb salvage. There's been a huge uptick in that. It used to be people wouldn't do that at all. There are a couple places that will now. I don't

> think (my surgeon) will. There's some guy in Texas. [Interviewer: There's always some guy in Texas.] That's true, true. Embroider that in a pillow right now.

Resources. Bert felt that meeting people in person was helpful and would include the MIB FACTOR osteosarcoma conference as a resource:

> I don't know if this is helpful in your study but as a personal matter, when it came to the MIB conference and I met you and I met Zoey and I met others like you, that was the most help for me. I mean, meeting people have been, kind of, walked through the fire and been through exactly what—There was no—as far as I know, there was nothing like the MIB conference before. Whereas I go and I sit through all the lectures and I take away some of it. To me the best part of that is being with others in the community that have been through something similar.

Diana also suggested adding resources to the decision aid, bridging ways to connect with the importance of personal stories:

> Maybe people, phone numbers or contact information of people who are willing to answer questions for folks. I think having the personal connection and receiving information about personal experiences is very valuable. So, I think in some way, shape, or form, if there's any kind of contact method, I think that would be helpful.

Zora said that reading a blog was helpful to her and recommended adding other resources:

> I found her blog, somehow. I read through her whole blog about how they decided, when she was diagnosed, what their surgery was. I just read through all of that stuff. That was really helpful to me, that whole day. I probably spent two or three hours just reading through all the things. That gave me a really good picture. It was nice to have that access to that. Something like that, you know, a link to a journal, Facebook page, or a Caring Bridge on somebody's journey.

Brody said that a direct line of communication is what helped him the most so he suggested that we include a section on patient portals as one way to communicate directly with the doctor. However, he also said that these can vary greatly depending on the hospital system:

> There's a really big difference in the systems that the hospitals use. [Hospital] got MyChart. It is so user-friendly. All the information that you need, like you get your labs. As soon as your labs, as soon as the doctors get them, you get them. It's updated in that system. I've got an app on my phone where I can go in here and check that right now. Well, at [another hospital] they're on a different system. Their portal is not set up the same. I don't know that it would be something that you could fix with a broad brush. The problem I have is I'll go and I'll sit down with a doctor and I'll have a million questions when I leave that I just didn't ask, forgot to ask. You know, sometimes the discussion changes directions before you come up with that. So, I don't know what the best tool would be, but just having and being able to reach out directly to the physician is, in some way, it doesn't have to be a personal cell phone number.

Tumor location. Yates felt that understanding how the tumor location affects the decision would be good information to include in the decision aid:

> You're faced with a tough decision. You're going to have to have a surgery. For Arthur, when we very first started it wasn't a foregone conclusion that he's going to lose his knee. Very shortly thereafter, we knew, he would have to lose his knee. What are the options for that? They're beyond just simple rotationplasty. The tibia plasty that your daughter had, we know another amputee who did that. Just knowing what those options are. She actually did that initially, not because of the metal in her but because of the location and how far up her femur the cancer had gone. So, they said, you're either going to have a very high amputation. Understanding what the difference between a high amputation and a low amputation is and what the energy costs are for a mobility, I think is important information.

Psychosocial Support. Unmet needs for psychosocial support include mental health counseling for patients, peer support for both patients and parents, and decision support.

Mental health. Quentin felt that information on mental health should be included:

> I don't know if teaching, you know, like having resources of seeing a psychologist before that. I mean, you're already dealing with the cancer, but then you're going to be dealing with something that you're going to have to carry on for the rest of your life. You don't just do the chemo, but you still have your remembrance of having that knee and hip joint replacement because of the cancer. So, maybe having resources of a psychologist or to have somebody to talk to about it. Then, coming up with the decision if they're having a hard time making that decision, they don't have that support system.

Levi also thought mental health awareness should be a part of the decision aid, he hoped that people could find ways to match this unmet need with the what works best for the child:

> For me, being in a playroom and playing games and keeping my minds off of things was super imperative, right? That is what gave me my positive attitude and helped me get my chemo during the day and still be playing a video game or playing cards with the other kids or something. I think it's super imperative to find out what's worked for each individual, compare it all together to see what is the same, what's different. Start to kind of relay that out to parents of children so that parents can relay that to the children. So, this way it makes things a little bit easier as they're going through the chemo, the surgeries, the downtimes, the spiritual loss, right, which I see a lot of. I know that's a part of most oncology units but at the same time, I still feel like from what I've seen, it's not focused on enough.

Peer support. Aretha suggested a way to connect people with osteosarcoma should be included, "I think would be most key, just—also being able to link, I mean, linking people to support groups would be huge. I think it's invaluable being with other bone cancer people, because different experiences." Arthur (11 at diagnosis) says meeting other people would have been helpful to him:

> I kind of would have liked to—I would have like to have met someone that had had this surgery before, and asked them about their story and everything like that. It probably wouldn't have changed my decision at all. I just would have like to

> have known their thoughts and how they handled the situation. I mean, a phone call probably would have been better than just not talking to someone at all. I mean, video chat is obviously better but just talking with someone who's gone through that, you know.

Peer support was also recommended but in slightly different ways. Phoebe also recommended peer support as having a mentor, "Even mentoring, I think is huge. I love mentoring because I think it's a way for them to ask questions and get your side of what worked for you without imposing your beliefs onto them." Ramona talked about peer support as a way to hear patient stories, "The best form, in my opinion, are people living with it in person, not videos or pictures, In person meeting somebody especially older or at your age with it. Around your age, is probably the best." Opal suggested including information for online support groups specific to osteosarcoma, in particular, addressing rotationplasty as a valid option:

> If they can get on some of the support groups and talk to other parents, I think is invaluable. I know it was for me. I'm like, "I need to talk to somebody who has had a limb salvage." Someone who is having a rotationplasty because of a birth deformity is not helpful to me and to us in our situation. There's so many people that had them after limb salvage, I feel like it's super-beneficial. I'm just putting it out there that there is that option. I think rotationplasty, while people are becoming more aware of it, it's still not the first thing you think of.

Quentin also thought peer support online would be good to include and likens it to having an in-person support person to get expert patient stories:

> I guess, it's hard to really come up with one thing that would help me but I think with cell phones these days maybe having the osteosarcoma page on Facebook. That's a huge reach-out. We never had that when I was going through it. I think having somebody as a support system that has gone through it, is a very big thing, too. That's why I think the social media aspect of Facebook and having that group, seeing that, I was like, "Wow. This is awesome." I wish I had something like this when I was younger, because I could go on and be like, "Hey, is there any survivors out here that had this? What was your ideas?" I think first-hand knowledge in dealing with that is some of the best knowledge.

Bert felt that personal stories directly from a peer would be good for hearing experiences:

> I don't know if it's part of your study but we did talk about having personal stories in whatever materials result from this. Personally, for me, it's helpful talking to people. You touched upon the idea that we did not meet with any amputees or people who have had rotationplasty. If that were available and I was able to meet with either the patients or the parents or family and talk to them, that would have been very helpful to me. I find the personal interactions very helpful. Face-to-face is always better, but cyberspace is second. I mean, just reading you can gather a lot of statistics, but you can tell a lot about how somebody feels by actually speaking with them and seeing them.

Decision support. While social workers, therapists, and patient advocates as an entity couldn't be included in a decision aid, some participants mentioned them when discussing it. Ramona would have liked someone to help her talk through the decision:

> It also would have been great if there was either a social worker or a psychologist to work with us on the decision-making process other than just a surgeon. Ray had a wonderful psychologist who he worked with in the hospital, who was phenomenal. I think that if Ray had been invested in making the decision, he would have helped Ray to work through the decision. We had a very abysmal social worker working with my husband and I who knew next to nothing about osteosarcoma. So, it would be great if there was a social worker who was well-educated in the choices to help us think about things.

Nerisa also recommended an advocate who was trained in advocacy:

> The surgeon had this PA who was a completely different person than him. She was very kind and connected with us more than he did, I feel like. She was more understanding and could see our reactions, whereas he was not seeing anything. I don't know if this is what you're thinking about but it was nice to have her there. I feel like that was beneficial. Having someone else, not just him, to give feedback about what was going. Then, like you said, we didn't record anything. You're not allowed to, to my understanding. Maybe, if they had a patient advocate that's with you who could go over, "Okay. You just met with the surgeon. Make sure you understand everything. Did you have—" I think patient advocacy, there's a lack of that. I think in something as serious as what we went through, you should have someone there. I always felt like we were out there just hanging on and hoping we got all the information and tell everybody that's going to ask us a thousand questions.

Wilma suggested a more systemic approach to decision support in terms of how we think about disability. She was concerned that parents were choosing the option that makes the child look the most normal rather than the most functional and that reinforced the surgeon's preferred position to save the leg:

> …there's a sense in which they're not open in hearing your child is going to be disabled. They want the option that makes them the least disabled or the most able to blend in, whether that's blending in in terms of sports participation or in terms of how things look.

Ray said he wished he had known more about adaptive sports for example when he was making his decision for rotationplasty after limb salvage complications. Changing the way we talk about limitations in the decision aid is also an opportunity to talk about what people can do with a prosthesis. When thinking about the decision aid, Wilma had concerns about parental fear of disability for their child when she felt that having a disability didn't prevent anyone from living a good life:

> I think about how so many messages in our society tell us that disability is something to fear and that this is built into so much of our decision making around surgical choices like this for instance. I think that's a real shame because it doesn't allow us to imagine our futures as disabled. They all will be, through one thing or another. It increases fear and policing

> about who can be in a space when, you know, a good life is doing things. It’s not how you do them.

These results indicate some clear content areas by participants.

Discussion of Dissemination Domain

The integrated model of KT and the dissertation domain was a major strength for this study. Survivors and families expressed interest in participating because they knew their experiences would serve a useful purpose – to aid future families with making this difficult decision. The role of the dissemination domain in the theoretical framework supported not only the development of the decision aid but also fulfilled the benevolent needs of the community; they valued the opportunity to share their stories to help others. The validity of narrative research is inherent by empowering participants to contribute to social change and to inform future studies (Kohler Reissman, 2008).

Recommendations were made in three broad areas: format, content, and psychosocial support. In addition to these three areas, recommendations were made for resources and patient stories. Format recommendations ranged from brochure/booklet to a webpage; the paper version was sometimes mentioned as a first introduction to the options with the option of going to the decision aid for more information. Content recommendations for the decision aid merge with the issue domains in the vignettes - the need for more information about long-term outcomes and potential complications was indicated by most participants.

The need for psychosocial support was also discussed during the questions for the dissemination domain. In a review of original studies on parent decision-making, researchers recommended prospective methods to better understand how to support parents when making difficult treatment decisions for their child (Lipstein, Brinkman, & Britto, 2012). Although this is a retrospective study for the first decision, it is prospective for some of the participants who are making another surgical decision. Furthermore, it is a recommendation to include resources in the decision aid to address support needs for future families. While parents and youth often looked online for information, they were concerned about the medical knowledge needed to decipher articles and websites found on the internet. In a study that evaluated 56 websites on osteosarcoma (excluding academic sites). more than 60% received over 50,000 in the month of study, 86% failed readability standards and only 12% included content aimed at adolescents/teenagers, the primary OS population (Lam, Roter, & Cohen, 2013). This further reinforces the need for a standardized, patient-friendly decision aid for both patients and parents.

Patient stories were recommended by most participants, whether those are video-based or told through images. Research indicates that there is insufficient evidence that adding personal stories to decision aids will help to support the user's informed decision-making (Bekker, Winterbottom, Butow, Dillard, Feldman-Stewart, Fowler, Jibaja-Weiss, Shaffer, & Volk, 2013). Bekker et al recommend additional research about the type of story that "encourages people to make more reasoned decisions, discourages people from making choices based on another's values, and motivates people equally to engage with healthcare resources" (p. 1).

On the other hand, participants come to the expert patients because they want to hear the stories of real life outcomes, including the so-called failure stories. Reframing "failure" as something other than broken might be one way to present patient-reported outcomes. Wilma talked about the need to see real people adapting to their disability in the patient stories, not stories of triumph. She felt stories about how people handle their disability are more valuable than stories where they overcome their disability, stressing resilience while finding new ways to live well. As many other participants pointed out, it will never be the same again; as Brody and others pointed out, it may even be misleading to let people expect that with dedication, they can return to their previous level of activity.

Some participants indicated that if they had known about the significant complications they or their child would face, they might not have made that decision. However, many said they would not have changed their decision if they had more information in this area. This is congruent with the Leinweber et al (2019) study on decision aids:

> Interestingly, most of the decision aids were not associated with a significant effect on the actual treatment decision made by the patient. While it seems evident that decision aids may provide knowledge to patients, it still remains unclear exactly what influences decisions for patients facing surgery. (p. 361)

There are many different types of patient narratives such as the Yale Persuasion Model and the Elaboration Likelihood Model but most models are used for persuasion and focused on attitude or behavior change (Shaffer, Focella, Hathaway, Scherer, & Zikmund-Fisher, 2018).. While patient stories as process narratives and experience narratives do not "bias" decisions the same way as outcomes narratives (Shaffer, Hulsey, & Zikmund-Fisher, 2013), it is outcomes narratives that are sought online by patients and families when making the surgical decision for osteosarcoma.

The intended development of the decision aid is similar to one used to inform the design of a decision aid for CF - Cystic Fibrosis (Basile, Andrews, Wang, Hadjiliadis, Henthorne, Fields, Kozikowski, Huamantla, & Hajizadeh, 2019). In-depth interviews were used to explore participants' knowledge, conversations with providers, and medical decisions to determine the information needs of adults with CF and their families with the intent of designing a patient decision aid. Similarities between the osteosarcoma study and the CF study include the number of participants (24) who felt it was important to hear patients' real-life experiences and desired more peer interaction with other CF adults. The direct result of the CF research is a decision aid that offers two types of information – biomedical and personal-level/experiential (Basile et al, 2019). The results of the Basile et al study in many ways mirror the dissemination recommendations from the osteosarcoma study – information about surgical options and patient stories, further supporting the development of the tool.

Overall, these robust recommendations for the decision aid are invaluable as formative research. In conjunction with the issue domains identified by the 29 participants as well as those revealed in the Facebook content analysis, this study reveals the foundational needs of the community.

CHAPTER FIVE

DISCUSSION AND CONCLUSION

Discussion

In this chapter, my goal is to synthesize and integrate key findings from the vignettes and Facebook posts, pulling examples from across both data sets, and using those to illustrate my conclusions. Paramount to understanding the lived experience of decision-making is the uninterrupted preservation of the patients' voices as much as possible. Presenting the results in a narrative vignette offers a 'slice of life' from those whose lives are most affected by the decision.

Here I discuss the overlapping themes between the two studies and the three models of decision making as they were described by patients, survivors, and families. A section in this chapter includes osteosarcoma-specific patient decision-making with a discussion of the results for both parents and youth. The decision aid recommendations are discussed as well. Theoretical implications, medical education implications, and decision aid implications are discussed. Strengths, limitations, and future directions conclude the dissertation.

Overlapping themes. The overlapping themes from study 1 and study 2 are information knowledge gaps (information overload, misinformation, missing information, biased information), concerns about risks (limitations, recurrence), the unknown (complications such as foot drop, missing quad muscles, additional surgeries, nerve damage, chronic pain) and psychosocial issues such as concerns about self-esteem, appearance, and peer support. These concerns were nearly identical to those identified as areas of concern in clinical research when comparing limb salvage and amputation: survival, short-term and long-term complications, function, and psychosocial issues (Simon, 1991). Hearing about patient-reported outcomes overall is an unmet need according to participants like Bert, "In terms of statistics of revision surgeries and people being happy, nobody every discussed that with us."

The expression of strong emotions and feelings were shared in both groups. While those expressed in the Facebook posts emerged as fear and feeling overwhelmed; those in the interviews were more reflections of frustration and regret when looking back on the first decision-making experience.

These themes are important identifiers for future content areas in the decision aid.

Models of decision-making for osteosarcoma surgery. The purpose of this study is to understand the specific needs of patients and parents who are making this difficult decision. A common theme across the posts and interviews is confusion about who is ultimately the decision-maker, the patient/parent/child, the surgeon, or a combination of the two? All three models of medical decision-making were found in both study 1 and study 2 and are guided by the surgeon's preference for decision-making. Parents and patients who are not comfortable with the prescribed role, often seek second opinions if they have time. Communication is fundamental to all three decision-making models; one cannot not communicate. As Zora observed about silence, “By not telling me anything else, you’ve led me to believe something else.”

Patriarchal model in osteosarcoma surgery. Several interview participants described the model in which the surgeon decides on the option yet there were fewer instances of the patriarchal model of decision-making in the Facebook posts. This may be due to the intent behind the post; most posts seek information, implying that they have some efficacy in making the decision, which may or may not be accurate. That said, some posts indicated that surgeons were making the surgical decision for the patient without discussion for patient preferences. One family met with the surgeon only once; after two months of researching, she later questions the surgeon’s decision two weeks before her 9-year-old son’s surgery:

> The orthopedic surgeon hasn't saw him since she did the biopsy I have questions I need answers to the hem/onc doctors can't give me the answers I want. The questions I have is why can't he get LSS? Why is his only option amputation? (6)

Others indicate that there is a bias for limb salvage surgery, with the surgeon stating that there is more than one option but pushing for one over another, “The surgeon feels that saving the limb is what’s best for her” (8) and “The Drs have been pretty forcefully promoting limb-sparing surgery” (4). Ultimately, we don’t know what the decision-making model is until after the surgery is performed; just because someone posts for information, that doesn’t mean that their preferences will be heeded. To understand how preferred roles are received, we have to know outcome of the decision.

Study 2 provides a deeper understanding of how decision-making roles are communicated by both patients and providers, at least from the patient’s point of view. Some participants defaulted to the surgeon’s preference even though they asked about other options. Opal describes these roles when discussing treatment options for her daughter, Olivia, “We had asked about amputation and the doctors were all like, ‘No, no, no. You don’t want to do that.’ So, yeah, I was very overwhelmed and just kind of, ‘Okay. Well, I guess you probably know what’s best.’” In this case, Opal was asking for information about an option, but told that’s not what she’d want; she accepts that the doctor knows best in large part because she’s overwhelmed. According to Charles et al (1997), this model clearly places the patient in a passive, dependent role with the surgeon as the expert.

While some interview participants were engaged in the decision-making, many were not; most surgeons decided on the best treatment and that treatment was LSS. Like the mother of the 9-year-old son from the Facebook study, Uma and her daughter, Vera, did not meet with the surgeon a second time until they were two weeks from surgery. When they finally met with him and were prepared to discuss options, they were dismissed, “He just said, ‘You’re having LSS.’” Gloria and her daughter, Holly, had a similar experience. Their surgeon said, “‘Okay. This is what we’re doing.’ There was no options presented.” Aretha’s said the same, “It was presented as, ‘This is what you do. We’re going to do the limb salvage.’” The assumption that the doctor knows best is grounded in the patriarchal model:

> The model assumes the patient and physician share the same goals, values, and preferences. Decisions made by the physician promote patient well-being but are independent of patient preferences and values, which is the primary weakness of this decision-making model (Slover et al, 2011, p. 123).

That said, this model can be useful when the patient’s decision-making preference is for the surgeon to make the decision about what they think is best.

Informed model in osteosarcoma surgery. In the informed model, the patient and family are the sole decision-makers for the surgical treatment. The process of this model places the responsibility of the decision on the shoulders of the patient and family: “After the patient evaluates the effectiveness, benefits, and risks of each option, the decision is presumed aligned with the patient’s values because the patient makes the decision completely independently of the physician” (Slover et al, 2011, p. 1048). Many of the Facebook posts in Study 1 indicated that patients were gathering information about outcomes in the expertise/experience theme to make a decision though it was unclear whether it was an informed model of decision-making or an SDM model. Some posts imply that the decision is shared but it’s difficult to tell due to the brevity of the post. Many also sound as if the patient is making the decision, such as this one, “I had another consultation with my surgeon today and am facing an impossible decision” (12) and “I understand of course that if amputation is the only life saver then we will go with it but I need to understand if we have the opportunity to choose is it the norm that LSS comes with much trouble later?” (10). In the absence of communicating directly with the individual posting the query, there is no way to know if they are seeking information to make the decision on their own or in partnership with their surgeon. This is useful for designing the decision aid as we won’t know if the user has found it on their own or at the recommendation of their surgeon. A section should be included early on decision-making roles.

When Study 2 participants indicated that the decision followed the informed model, it was usually when they were either in the process of or had already made a second decision. Two were preparing for second surgeries at the time of their interviews, Diana and Ingrid. Diana was in the process of choosing amputation after her limb salvage left her with chronic pain and limited mobility:

> I did the research and then I brought it up to my surgeon when I had my most recent scans which were in July. He was able to tell me a little bit more and tell me how much stump I would have left and all that. Then, he scheduled a amputee clinic appointment for me. That was a couple weeks ago. That was when I think I became more comfortable with the idea. That's when I was just like, okay. I think this is going to be good for me. The amputee clinic appointment was with a prosthetician and a orthopedic surgeon who wasn't my original surgeon.

Ingrid also made the decision for rotationplasty independently of her surgeon after her limb salvage caused extreme, debilitating pain. Interestingly, although her surgeon did not have the skillset to perform that surgery himself, he supported her second decision:

> My doctor said, "Look, if this is what you need to do, if this is what's going to help you, I'm going to come watch the surgery because I've never seen one before. You need to do what's going to be best for your life and your lifestyle."

Five others (Aretha, Bert, Nerisa, Ted, and Vera) were exploring options for a second surgery after LSS, either in the near future or if additional complications compromised their quality of life to the point that they would choose something else.

Four participants had made an informed decision and moved forward with a second surgery. Phoebe made the decision for amputation after LSS after spending three years trying to save her leg:

> I decided to amputate. Doctors in (city) weren't too happy about it. So, I kind of doctor shopped and found the best doctor that really understood what was going on. I was in so much pain and couldn't do anything. So, I was over it and wanted to move on with my life.

Olivia chose RP at age 13 after three years of complications post LSS. Holly chose to amputate at age 14 after a year of complications post LSS. Ray chose RP after six years of complications post LSS. These proactive efforts to get the surgical decision they want through the informed model of decision-making often came out of profound dissatisfaction with the patriarchal model that preceded it.

One exception is Wilma who chose the informed model of decision-making for her first surgery, making the decision for rotationplasty as an adult after her first surgeon told her she wasn't a candidate for RP. With a history of research, a PhD, and other health issues, Wilma was comfortable doing the research necessary to feel fully informed with online searches using PubMed and personal stories in blogs, YouTube videos, and expert patients on Facebook. This research coupled with information she received from other surgeons helped her feel confident that rotationplasty was the best choice for her and she searched for a surgeon who would agree to perform one.

Frank and Ernie's story stands out as remarkably different from those in which the patient desires to make the decision. For this family, it was Frank's surgeon who set the stage for informed decision-making. Frank was 14 at the time of his diagnosis and the family had no medical background; the surgeon's treatment recommendations for the patient were not part of the decision-making process (Emanual & Emanuel, 1992). The knowledge translation was limited to strictly information provision by Frank's surgeon, according to Ernie:

> I really felt like they wanted us to make the decision on our own. It was really weird. I'm like, hey, we're totally dependent on what you're asking us to do here. We would want to know what are the limitations. The only thing I really feel like I got from the surgeon was, if we do the surgery, given where the tumor is, if we do limb salvage, over time the endoprosthetic could break down. If we do an Allograft or an endoprosthetic, your leg's never going to be as strong as it was. If you do the rotationplasty, with time, you'll be able to do whatever you want to, sometimes not as good as you could before but maybe only a step behind. That's really the way that the negatives were portrayed to us.

Ernie tried to understand why the surgeon wouldn't make any recommendations for treatment:

> So, the doctors themselves, I really feel like they were trying to stay away from convincing us or influencing us too much. They were just saying, hey, if something were to go wrong here. We want it to be your choice, not our choice. That was hard, because I was like, wait. We're in a new world here. We need you guys to talk to us about the pros and cons.

Retrospectively, Ernie rationalized that the lack of guidance from the surgeon was due in part to litigation concerns, especially as the surgeon became more interactive once the family had made the decision for rotationplasty:

> Once [Frank] had made his decision, then the oncologist was like, "Hey, [Frank], that's what I would have chosen." All the sudden, he would let us know, "I'd seen patients in here for years. They do a lot better. If I had to make that decision, that's what I would have made." I'm thinking, okay. One, I get it. You don't want to influence. You can't, like if something bad happened in a lawsuit, they can't say, yeah, we told them that's what you should do or something. I don't know. I'm guessing that's what they're thinking. Then all of a sudden, for him to drop his guard a little bit and say, "This is what I would have done." The surgeon was very—he was very guarded, too. Once the surgery actually happened, he was a lot more jovial around [Frank]. He would come in and check on him a lot more than he did before. So, we saw him more post-op than we did pre-op. Again, I can only attribute that to they don't want to be seen as the influencer or let us do whatever the choice. Then, once we've made the choice, then they're more open to talking.

This family struggled with the limited amount of time further compounded the issue of gathering and processing information to make the decision:

> You're just forced to say, especially with sarcoma where they do the neoadjuvant therapy and you have two months to make the decision, like very rushed, very compressed. It's all the sudden, and you've got to make an informed decision. It's tough.

With the shock of the diagnosis, many families would feel uncomfortable without some recommendations from their surgeon. Indeed, according to Slover et al (2011), this is a distinctive disadvantage to the informed model of decision-making:

> The challenge with this model is that the decisions are often difficult, requiring the analysis of complex information and concepts, and may be beyond the capacity and comfort level of some patients. Ensuring complete delivery of the necessary information to make the decision is difficult. (p. 1048)

This aligns with the findings from both the Facebook posts and for the personal interviews as patients and families describe the overwhelming task of getting the knowledge they need to make the first decision. The amount of time they have to make the decision further complicates an already complex process of understanding the disease and managing the side effects of aggressive chemotherapy. However, the findings from the personal interviews indicate that many patients prefer this informed model of decision-making for the second decision, particularly if the first decision was patriarchal. This has implications in medical education for orthopedic surgeons that patient preferences matter for patient satisfaction and long-term outcomes. As surgeon Carol Morris was quoted in the rationale, “if the patient doesn't want the option, then it doesn't work at all.”

A recent study conducted by Tate & Rimel (2020) analyzed differences in decision-making approaches and suggests that the treatment stage impacts how oncologists present treatment options. The researchers analyzed transcribed interactions between patients and oncologists in which “option-listing” (OL) was used to facilitate shared decision-making by listing more than one option for treatment. What the investigators found was that in the early stages of cancer, OL was constrained “through expression of physician bias.” However, this constraint disappears when cancer is more advanced and “OL is presented without physician preference and demonstrates rescission of medical authority” (p. 71). They concluded that “when physicians do OL for patients in initial treatment, they are skewing more toward one preferred treatment route by mobilizing their medical expertise” (p. 75). While the context of this study is the progression of disease (early vs. late while in treatment), it is consistent with the findings in osteosarcoma patients. The initial consult usually has a physician preference of varying degrees while the informed model is preferred by patients for later treatment options, regardless of whether or not the osteosarcoma has advanced. For the patient seeking amputation or rotationplasty after attempts at reconstruction have failed, their condition may equal that of disease progression due to chronic pain and functional limitations.

Shared decision-making model for osteosarcoma surgery. The differences between the models are vast compared to the SDM model where doctors and patients bring together their respective expertise and patient preferences are a priority. Recent studies demonstrate the importance of shared decision-making for these types of surgeries, “Orthopedic surgeons must be knowledgeable on the concept, practice, and current outcome data of SDM practices to improve care delivery" (Klifto et al, 2017). It has been recommended that shared decision-making begin early with families to be effective (Swota & Bradfield, 2015, p. 34):

> Several considerations need to be taken into account in order to maximize the likelihood of a successful family/ provider interaction. For instance, the conversations with the family need to start sooner rather than later. The earlier these conversations start, the more time there is to build relationships with families, establish trust between parties, elucidate parental values and preferences, and have ample opportunities to revisit these values and preferences as the condition of the patient changes.

When Chief of Orthopedic Oncology, Valerae Lewis, MD at MD Anderson is asked how she begins her osteosarcoma counseling conversations with parents, she says she starts early with long-term quality of life considerations: "I start talking about the different options early, so they can explore what's right for them, because a prosthesis for one child may not be the right option for another child and another family" (MD Anderson Cancer Center, 2011). Thinking about the long-term outcomes of the decision is an important part of shared decision-making; this is most noticeable during the values clarification exercises recommended in the International Patient Decision Aid Standards. Both prospective concerns and retrospective regret about long-term outcomes were in both studies, the SDM that did take place in Study 1 usually included potential limitations and thinking about the patient's future quality of life.

Parents and providers both made efforts to include the child or adolescent in the decision-making process. Joy and Kent described the SDM model when they made the decision for limb salvage surgery for their 10 year old son, Levi. As the family leaned more toward LSS, their surgeon shared stories, pictures, and videos of other patients who had the same surgery. All three options were presented and they discussed them together; Joy said, "I looked into it. I did my research talking to Dr. X." Levi was involved in the decision-making process at 10; Kent felt it was important that he be included even at a young age, "You have to say, they have to have some say. It's their body." Levi felt that younger children may need parental guidance for decisions but felt strongly that adolescents should be involved in the SDM process, "If your kid is 10 or 12 or 14 years old, I think it's very imperative that they allow them to have some type of say and make sure everything explained in detail what's going on."

Quentin also described a shared decision-making model when he was 15 and deciding whether to do amputation or LSS, "Dr. X said, 'We can give you a total knee replacement or if you don't want to deal with the multiple surgeries that may be ahead,' she said, 'we can also amputate.'" His surgeon brought in the values Quentin had expressed to her about his vision for his future as she described different possible outcomes:

> My doc told me, she's like, "If you play sports, if you want to continue playing football, if you want to continue playing basketball, if you want to be a cop," she said, "choose that, I can amputate. Because then if you break that prosthetic, you can fix that.

As part of the SDM process, Quentin's surgeon also discussed possible complications, "I think the most she said was you know, there could be possible other surgeries down the road. Didn't really know how many, didn't know what possible surgeries." As a result of setting these realistic expectations about the unknown outcomes during SDM, Quentin, age 32, felt better prepared when he needed several surgeries (8 before he was 18) and even a hip replacement. Interestingly, Levi and Quentin were the only interview participants who were very satisfied with their limb salvage surgery and counseled using the SDM model of decision-making; they were also long-term survivors at 10 years and 17 years post op respectively, with many surgeries to repair the limb salvage behind them. Cleo was also a long-term survivor (19 years) who described SDM discussions with her surgeon and chose LSS for her first surgery. She was mostly satisfied but had considered amputation due to pain.

Pieterse & Finset (2019) speculate about the ramifications of uncertainty when framed by the shared decision-making model:

> Possibly, feeling better informed about the options and their pos and cons, and knowing better what matters to oneself, are key to limiting regret about the decision in the long term, more so than feeling certain about the decision per se. (p. 1947)

Both Levi and Quentin described themselves as very satisfied with the long-term outcomes of their decision for limb salvage surgery according to the decision regret scale (Brehaut et al, 2003; O'Connor, 2003). The five statements read to the participants often elicited further comments on their decision. Of the five statements, Levi and Quentin both strongly agreed that it was the right decision, they would go for the same choice if they had to do it over again, and that the decision was a wise one; they both strongly disagreed that the decision did them a lot of harm. Levi strongly disagreed with the statement – I regret the choice that was made – while Quentin rated it as neither, stating:

> I'd probably say neither, because it's one of those ones where I'm glad that I did it but still when you mentally know you can do it but physically it won't let you, sometimes that's where you get frustrated. I say neither nor because I mean, just having it done I don't have any issue.

Several participants commented that responding to the scale was problematic because they have no way of knowing what the outcomes might have been with a different decision. Brehaut et al (2003) discussed this possibility that low levels of rated regret may be a result when participants had no knowledge of what their outcomes might be if they had made a different decision:

> For most medical treatment, decisions, knowledge of the outcomes for alternative choices is not known (e.g.,if one has chosen surgery, one cannot know what the outcome would have been had an alternative treatment been chosen). (p. 289)

Furthermore, the researchers suggest that knowledge of regret when comparing the chosen option may be one way to address regret:

> Whatever regret does exist may stem from people assuming that another treatment choice would have yielded a better outcome. If this interpretation is correct, it suggests that one way to reduce regret after a decision might be to remind patients that bad (perhaps worse) outcomes may have occurred had the alternative been chosen. (p. 289)

As illustrated in Levi and Quentin's experiences, they had no decisional regret and they were counseled on the potential complications and limitations of the treatment options. Therefore, by including the unknowns of risks during SDM, osteosarcoma patients may have less regret because they were informed of the potential limitations and uncertainties for all options. In Quentin's case, he had never heard of rotationplasty – even at the time of the interview – which may contribute to his lack of regret; except for his "neither" response, he had zero decisional regret. At their screening, Quentin said he was "very satisfied" and Levi said he was "1000% satisfied" with the outcome.

Both Study 1 and Study 2 indicate that SDM for osteosarcoma surgeries is an opportunity to discuss potential limitations and common complications, an unmet need in both studies. Surgeons posited limb salvage surgery as the best option to save the leg, glossing over and often avoiding discussion of functional limitations, pain, and the need for additional surgeries. Brody felt that patient expectations should be addressed both on the provider side and in the decision aid:

> So, kind of setting some realistic expectations prior to the limb salvage. You know, in my mind, I was going to be back to a hundred percent. That's not the case. The only thing is, you know, with some surgeons I think that's their goal, is to get you back to a hundred percent… they're not going to go into it shooting for 80 percent, you know. They're going to shoot for doing the best that they're going to do. Sometimes, that's just not feasible.

Orthopedic surgeon Michael Simon (1991, p. 268) asserts the following about patient expectations for limb salvage and surgeon's obligations:

> Young adults often imagine that limb salvage, especially with a mobile knee, will enable them to continue their normal activities with no change in performance. The patients and their families must be made to realize that none of the reconstructions will enable them to have a normal limb, and that all will be partially disabled.

It is noteworthy that this was penned nearly three decades ago yet limb salvage is still the preferable procedure by most surgeons. In fact, although this dissertation study represents the patient/parent voice exclusively, what osteosarcoma patients say is quite the opposite. In 2020, patients report that they are encouraged to choose limb salvage because they can 1) save the leg and 2) have a normal-looking leg; their perception of the surgeon's position is that it is presented as "the gold standard." There is often little discussion about resuming their activity level post-operatively, leaving some patients to assume they can resume high level activities. It is possible that patients hear "normal" and extend that to normal function, expecting that as an outcome. Even so, the evidence in this study suggests otherwise; Aretha sums up her 26-year experience with limb salvage since age 15:

> I feel like surgeons, they don't think past the surgery. They think my success is getting this leg replaced and haha, it's a success. They don't think about how it's going to actually function for your life. I don't think they think of other options. They think, well, an amputation or a rotationplasty would be like a failure. No, I'm going to fix the leg. I just don't even feel like they know or are aware that they should be thinking about quality of life, not just what your leg's going to look like. I wish I would have known, because I had no concept that I was signing myself up for a life of surgeries.

Some patients and parents are unsure of their role in decision-making and may follow the surgeon's lead for who will make the decision. Others are sure of their role and disagree with surgeon's preferred model of decision-making. This is important information to understand for the decision aid as information to include in the content, particularly since patients who have a sense of efficacy in making the decision are usually more satisfied with the outcome.

Osteosarcoma patient decision-making. According to the American Cancer Society (2020), about half of all osteosarcoma cases are in children and teens, making parents the surrogate decision-maker for their child under the age of 18. Parents in both Study 1 struggled with making a "life-changing decision" for their child, often choosing dialectical language to describe the tension such as "I know that this is the right decision for him" (3), "I don't want to make the wrong choice for him" (9) and "What if you make the wrong choice?" (2). Parents were conflicted about how much information to share with their child and how to involve the adolescent who preferred to be passive. A father from Study 1 is debating between rotationplasty and limb salvage for his 14-year-old son, "When I try to talk to my son about this he shuts me down and doesn't what to talk about it" (11). Similarly, Bert, a father from Study 2, wrestles with making a new decision for his 14-year-old son who does not want to discuss his options for further reconstructive surgeries or amputation. Retrospectively adults in the study who were adolescents at the time of the surgery expressed regret about not being more active in the decision-making process. Phoebe was 15 when she was diagnosed; now 27, she said, "I wish, at that time, I knew to voice my opinion more." Aretha was also 15 at diagnosis; now 41, she said:

> Honestly, I was kind of the clueless teenager… So, I really didn't ask a lot of questions. I didn't know a lot of details. I kind of just ignored everything and just waited for it to happen to me. I didn't know a lot about what was happening. I remember years later being embarrassed that I didn't even know details about what they did or what was in there or anything. I think it was willful ignorance and my parents went along with it. I don't know.

In a study on surrogate decision-making, factors that influence parent treatment decisions for their child include their child's health status, other members of the community, prior knowledge, and personal factors, such as emotions and faith (Lipstein, Brinkman, & Britto, 2012); parents also "struggle to balance these influences as well as to know when to include their child in decision-making" (p. 246). Findings in both studies expressed a desire for more support in terms of both patient reported outcomes (PROs) and clinical research when making the surgical decision, indicating an unmet need for support with survivors and decision support from their medical team. This need for PROs is consistent with the recommendation of Swota and Bradford (2015) to provide parents of children who have cancer with data on factors identified by parents as important in their decision-making process. Information seeking on patient experiences and decision support is the driving force behind joining the osteosarcoma Facebook support group.

While appearance was a concern for many participants at the time of decision-making, this later function was so lacking that many of them had or were seeking a second type of surgery. A study that looked at psychosocial and function outcomes in long-term survivors of osteosarcoma who had LSS or AMP found that those with more functional lower limbs had a better quality of life than those who had less functional lower limbs regardless of their surgery (Robert, Ottaviani, Huh, Palla, & Jaffe, 2010). Patients looked back on their early diagnosis and noted a surgical bias for LSS, saving the leg, and looking normal as opposed to function. In fact, many said the limitations for LSS were never discussed.

The lack of muscle needed for rehabilitation was one of the leading factors that limited their function and mobility and led to decision regret. The goal for removing the tumor is to achieve clear margins, noted in both studies as concerns. However, the definition of what constitutes a wide margin has changed over time (Anderson, 2016) but the goal remains to preserve as much normal tissue as possible. According to the interview participants, more of the muscle and tissue were removed than expected, rendering them less functional than they were led to believe. The American Cancer Society (2018) makes this statement about LSS and limitations:

> It takes about a year, on average, for patients to learn to walk after limb-salvage surgery on a leg. Physical rehabilitation after limb-salvage surgery is more intense than after amputation, but it's extremely important. If the patient doesn't actively take part in the rehabilitation program, the salvaged arm or leg can become useless.

However, the implication is that the patient is choosing not to take part in rehabilitation, not that they can't or are physically unable to participate. In a study looking at the Repiphysis implant, there were 37 complications and 13 revisions with 10 patients over a 6 year period, prompting researchers remark on the psychological toll of repeated surgeries, "The negative impact of extended treatment on quality of life should not be underestimated, especially given its potential to interfere with the school and social activities of this patient population" (Cipriano et al, 2015, p. 837). My daughter had a Repiphysis in 2007 and a revision in 2008. No amount of effort on the part of patients with a Repiphysis would help them regain their function. Brody expressed similar frustration with his efforts at rehabilitation, which he attributed to an inability to rebuild his missing quad muscle.

Conclusion: Patient Preferences Matter

The findings from Study 1 and Study 2 demonstrate that patient preferences for surgery matter for patients and parents, both before and after the surgical decision is made. Their choices matter for supporting their quality of life goals, self-efficacy and decision satisfaction. Survivors and parents shared powerful stories of a life-threatening diagnosis that impacts their identity and mobility for a lifetime. Patients and families who participate in making the decision with their surgeon and are aware of the potential complications are better prepared to manage the uncertainty of long-term outcomes. In a decision fraught with uncertainty, patients need more than just information. They need decision-making support from other families and survivors. In addition to clinical information, patients want to know about both short-term and long-term outcomes from survivors who have been through this traumatic experience. Patients and families need to know that there is no right or wrong decision and that there will likely be more decisions with each option. Yates stressed the need for realistic preparation about outcomes, "…that none of these things are perfect, that all of these things are life-changing, all of these things have risk of failure. That's a part of the process, is saying, what does failure look like here?" The process of making the decision as well as additional decisions to continue efforts to save the leg or choose another surgical option resulted in powerful personal testimonies in Study 2.

The power of the narrative. In the screening for the interviews, potential participants were eager to share their surgical decision-making experience and to provide feedback for a tool that would help future families make this difficult decision. They wanted their story to be heard (even if they were dissatisfied with the outcomes) and they wanted to help future families making this difficult decision. Patient stories like these are referred to as illness narratives in the field of medical humanities and in the practice of narrative medicine. The value of illness narratives has become increasingly beneficial to both patients and providers for the understanding of the human experience of illness and how illness shapes identity (Kleinman, 1988; Brody, 1987; Charon, 2006; Frank, 1995).

The diverse experiences described in the vignettes were complex and messy at times, with unique circumstances for each participant. Laura Ellingson, PhD, is a health communication scholar and also an osteosarcoma survivor; she describes her use of narratives, “Rather than seeking to eliminate the messiness of real experience by defining and quantifying it, I embraced the mess as a source of meaning” (p. 501). Ellingson includes lived experience as an “organic process of knowledge production grounded in (not abstracted from) human experience” (p. 511). Although the patient stories for the length of the interview were necessarily condensed, the power of these narratives is evident even in the shortened vignettes. The resulting knowledge gleaned from these stories can help clinicians empathize with patients as human beings. Research with illness narratives suggest that these stories are important for building relationships between providers and patients:

> The divides in health care need to be bridged in order for effective treatment to proceed. I have proposed that narrative means might help to bridge these chasms, because narrative ways of knowing and experiencing the world and self are held in common by health care professionals and patients. (Charon, 2006, p. 39)

Physician and noted author in narrative medicine Arthur Frank posits that illness narratives are dominated by the quest narrative, beginning with accepting the disease as the beginning of a journey, the course of trials that includes “the sufferings of surgery and stigma” (1994, p. 7), and the return to living and survival. When asked to tell me their story at the beginning of the interview, survivors and parents both talked about a normal life interrupted. In her book, The Year of Magical Thinking (2006), author Joan Didion described how life can change in an ordinary instant when unexpected illness occurs. Ted, who was 18 when he found out he had osteosarcoma, shared the incredulity of that kicking a can could result in a broken femur; 15 years later, he still has the can. Other participants also started their stories with normal activities: “So, he was in basketball at the time. It was the end of seventh grade” (Nerisa), “It was my first day in college…I was playing volleyball” (Diana), “I slipped and fell on ice in Ohio at my first year of college” (Ingrid), “So, he was ten and a half. He was playing football (Joy), and “I had gotten into doing some long-distance running…I was actually training for a 5K…” (Brody). According to Frank (1994):

> The boon granted at the end of the trials of illness is generally an enhanced subjectivity, extending toward others specifically and toward life generally. Most illness narratives culminate in some expression of how the self has been changed by the experience of suffering. (p. 8)

Quentin was glad for the opportunity to share his story in the interview, “It’s kind of neat that I get to tell my story to someone other than students.” Telling their own story is one way that illness narratives allow people to retain their individuality and not be like everyone else (Frank, 1998). With the power of the narrative comes the sharing of human experience, extracting some sense of meaning out what is otherwise a chaotic event. For the participants, sharing their story with me as a member of the community gave their traumatic experience meaning knowing that their stories would be part of a research study and in the development of a tool to help future families.

More than information is needed. Providing patients with information or research alone isn’t enough to support those who are making decisions (Hargraves et al., 2016), particularly with a decision as life-changing the osteosarcoma surgery. What the patient or parents decide is best for themselves or their child is more important than what the surgeon feels is best for them; patient satisfaction is greater when they are fully informed about the potential complications and they have better satisfaction when they have positive relationships with their providers. To feel fully educated when making the decision, more than information is needed; patients and families need to know what to expect long-term with each of the options. They need the support of their surgeon in a shared decision-making model through conversation and not just information exchange to choose the option that best fits their values and quality of life goals for the future.

In addition to information from providers, patients and families searched for information online, seeking other options or patient expertise. The inability to schedule follow-up appointments with surgeons to discuss options further impacts the relationship. By the time they do meet with surgeons, there isn’t enough time to change what the surgeon had planned without answering questions or considering the patient’s preference. Research indicates that anxiety over making the “right” decision may put patients in a passive position of “surrendering” decision-making to the surgeon (Frosch & Kaplan, 1999), as these last-minute decisions seem to indicate. Parents and patients both succumbed to giving in to the surgeon’s recommended surgery when their preference was challenged, and they were concerned about making the right decision.

Quality of life is impacted by complications, not by the decision. Ultimately, a survivor’s quality of life is not impacted by the surgical decision but by the complications. Patients need to know the potential risks and common complications associated with each choice. Uncertainties should be included so they have realistic expectations and feel prepared for what might happen after the surgery. This includes the lifelong need for a prosthesis and other assistive devices for rotationplasty and amputation. Patients who feel prepared for potential outcomes expressed greater satisfaction even when they had many surgeries and complications.

In the absence of receiving information directly from the surgeon, parents and patients alike seek information and support from other families online and through social media. The information they receive can either support their relationship with providers or it can impede it. Both studies indicate a reluctance on the part of surgeons to listen to patient preferences; in fact, some were annoyed or dismissive when patients voiced their preferences or asked questions. This is a missed opportunity for providers as online health information seeking can improve the patient-provider relationship if they have a positive relationship and the patient discusses the information with the provider (Tan & Goonawardene, 2017).

There is more than one decision. As participants discussed in the interviews, there isn't just one decision. Of the 20 interviews, every single person had additional decisions to make after the initial decision for surgery. Even with amputation or rotationplasty, it's not "one and done," both may require revision surgeries. The first decision may or may not be made by patient; that decision, usually limb salvage surgery, was made by the surgeon. Subsequent decisions are usually made by the patient or family, such as whether to have additional surgeries to save the leg or try amputation or rotationplasty. This is consistent with another study that demonstrated the evolution of patient involvement, "Decision making is often an ongoing process in which patient participation may change over time" (Fraenkel & McGraw, 2007, p. 533). For some families, thinking ahead to what the next decision might be helped them to make the initial decision.

Patient preferences matter and parents and patients will go to great lengths to get the surgical option best for them. Two orthopedic surgeons and a bioethicist presented the case study of 17-year old boy with osteosarcoma and a failed limb salvage (Betsy et al., 2005). His surgeon only offered above knee amputation although the boy and his parents requested rotationplasty. They were told rotationplasty was no longer being performed so the family traveled 850 miles to have the procedure done elsewhere. The boy is now thriving and very satisfied with his rotationplasty. The authors deliberate the ethics of letting the patient choose the option that best fits their lifestyle, values, and preferences; they deduced, "In the end, it is the patient who lives with the outcome. This is why the choice belongs to the patient" (p. 1655). Findings indicate the same from both Study 1 and Study 2; the decision affects the patient and family for their whole lives and the choice belongs to them.

Implications

Theoretical implications. Knowledge translation uses evidence to guide medical decision-making. This study adapts the interactive model of knowledge translation to explore the issues for decision-making in osteosarcoma surgery. The interactive model of knowledge translation stresses that information is not linear and includes multiple inputs (Estabrooks et al, 2006). In the absence of receiving sufficient knowledge from providers, osteosarcoma patients seek information from survivors in online support groups and social media. One of the unmet needs for information in both studies was the need to know what the uncertainties are for success and failure of the options. In the absence of evidence for the risk of complications, providers often provide no information on potential complications at all. According to researchers, surgeons have been trying to find good ways to predict function and quality of life to find the best surgical option, but the data is almost always retrospective (Anderson, 2016). Consequently, they have no way to prospectively predict who might do well with limb salvage and who might not. Also, there may be contributing factors during surgery that will affect functional outcomes, such as the unanticipated removal of additional muscle to achieve clear margins. Interview participants clearly stated that not knowing is still information; Uma talked about the need to know about these risks, "I understand that we're not going to know a hundred percent but the unwillingness to talk about it, the unknown, was a huge barrier to getting through this successfully without incredible mental strain."

The implications for the interactive model include 'not knowing' as a construct of knowledge; in other words, the absence of knowledge is still knowledge. Bert described the problems and challenges of information not shared, "They told us that it might fail, generally, but we never knew about the more common problems and challenges of limb salvage surgery survivors experience." In this case, the lack of information left Bert and his family feeling unprepared for the complications that ensued, "We never really had those conversations. I don't think that it would have changed our decision in any way, but it's certainly something that I would have like to have known." Similarly, Zora describes the implied meaning in not discussing the unknown:

> If you don't tell me now what the downsides are, what the probability is that there would be of complications, then because you're not confident about that probability then you've told me something. By not telling me anything else, you've led me to believe something else.

While the absence of knowledge may or may not impact the decision, it does affect patient satisfaction and quality of life when they encounter complications later, Nerisa said, "Had I really had an understanding of the pain that Nate would have to deal with on a daily basis, I may have made a different decision." In cases like Nerisa's, not knowing that pain might be a daily issue might have impacted her decision for limb salvage surgery and for Bert, it wouldn't have changed the decision, but it would have made him feel more prepared. If nothing else, knowing that it's complicated and it depends may be a good way to start a discussion and acknowledge that there are unknowns. According to Uma, not talking about it causes more distress, "That just leaves more unknown and more uncertainty. I don't think it decreases it. It doesn't make people feel more comfortable."

The four domains of user, issue, research, and relationship gave structure to the telling of the participant's story using narrative analysis. The isolation of the issue domain helped to identify knowledge gaps and unmet needs while exploring the lived experience. The fifth domain, dissemination, will guide and inform a tool to address the gaps in knowledge translation for osteosarcoma surgical decision-making. This application of the interactive model will be valuable for future studies using the framework, especially qualitative inquires exploring patient experiences with the intent to identify a problem and develop a resource to solve the problem.

Medical education implications. Surgeons in both studies were described as having biases for limb salvage surgery – saving the leg, keeping the leg, looking normal – and in steering requests for rotationplasty (not for girls) and amputation back to the gold standard of saving the leg. Results also indicated that families felt options were available to them but not discussed, leaving them to seek information and sometimes surgery elsewhere or to give in and move forward with the surgeon's choice.

The lack of shared decision-making to elicit the family's preferences and input in the decision has implications for medical education. The bias for limb salvage surgery points to changes needed in medical education to provide all options as equally valid. It has been suggested that changes in health profession education shift from a biomedical positioning to one that includes the social sciences like communication and these be taught with challenging perspectives and dialogue with the inclusion of patients as partners (Thomas, Kuper, hin-Yee, & Park, 2020). Thomas et al conclude in their study that "without a major transformation in what, how, and with whom we teach, our future clinicians may be unprepared to enact SDM in a manner that does justice to the patient voice" (p. 8). The development of a tool that includes values elicitation exercises at the time of diagnosis may help surgeons and patients alike to choose an option with the best prognosis for the individual patient.

Future efforts should be focused on education in orthopedic surgery on the long-term outcomes of limb salvage surgery, rotationplasty, and amputation to eliminate patriarchal decision-making. Borrowing from the conclusions of Thomas et al (2020), I suggest having patients as partners in the orthopedic curriculum for a system change to promote values elicitation, shared decision-making, and rethinking the save-the-leg-at-all-costs mentality:

> We propose that academic programs include and recognize persons with patient experiences—as official members of the pedagogical team and include them in all levels of the educational continuum, from admissions, to curriculum design, teaching, assessment, and program evaluation (p. 8).

Findings from this study can be adapted as role-playing scripts in which students in orthopedic oncology can practice real-life communication scenarios while also playing the role of patients and parents.

The results of this study also has implications for obstetrical care as Cleo mentioned that as problematic during her two pregnancies with LSS:

> I don't think many OBs are used to dealing with people of child-bearing age having artificial joints or limbs Especially, the amount of survivors across the board, you know, childhood cancer survivors are growing up now. They're having kids and I don't feel like anyone is that educated in how to handle long-term effects because there haven't really been survivors of this length of time that were treated as children before.

The surgical decisions for the treatment of osteosarcoma in the lower extremity are not limited to osteosarcoma specifically and may be options for other types of sarcoma and cancerous tumors. I suggest the development of a resource to be distributed to the obstetrical community to better prepare clinicians for the unique challenges associated with pregnancy for cancer survivors who have rotationplasty, amputation, or limb salvage surgery.

Implications for the patient decision aid. Osteosarcoma is predominately a rare pediatric cancer. Resources are limited and adults diagnosed with the condition are left feeling further isolated as a rare member in an already rare community. While the survival rate for older patients is identical to the pediatric population (Aljubran, Griffin, Pintilie, & Blackstein, 2009), the four adult patients who were 21 and over at diagnosis felt that they were in the minority as adults for what was already a rare pediatric cancer. Brody (28 at diagnosis) said his surgeon told him, "You're 15 years older than you should be to get diagnosed with osteosarcoma." Xena (33 at diagnosis) said her surgeon told her, "You know, the chances of bone cancer are so slim, in general, and you are not in the age group to be diagnosed." Wilma (30 at diagnosis) felt fortunate to diagnosed quickly, "Most people spend a long time waiting for the diagnosis in some way, especially as adults because it's not common in adults." Ingrid went to the osteosarcoma conference at 21 but felt she was expected to be with the kids, "Nice to be around people who understood, but it was also hard because I didn't want to hang out with the kids. Like, I didn't want to do kid's activities. I wanted to go listen to the scientific conference." The recommendations for including patient stories in the decision aid provides an opportunity to address the challenges associated with adults seeking information about surgical options.

An additional implication for the decision aid is the potential inclusion of other reconstruction options such as Levi's vascularized fibular allograft and the osseointegration aa surgical option for Ray's second surgery. My daughter had a procedure called a tibia turn up plasty for her second surgery, 11 years after her limb salvage surgery. This surgery used her existing tibia to give her a longer residual limb in an above knee amputation; without this procedure, she would have had a very short limb, adding to the challenges of the prosthesis. A subset of options for secondary surgeries if reconstruction efforts fail should be considered as unit in the decision aid. This inclusion would support the conclusion that more decisions will be made after the first decision and better prepare patients and families to understand what options may be available to them in the future as they make their first decision. For example, amputation after limb salvage may be an option but it might not be a good option if the residual limb is too short to effectively use a prosthetic device. Further discussion with orthopedic surgeons on the Osteosearcoma Decision Aid Medical Advisory Team is advised.

Lastly, the decision aid should prepare patients and families for a realistic expectation of decision-making both during treatment and as part of survivorship. The decision-making process does not end with the surgical decision; for each of the options discussed, there will be a lifetime of decisions related to additional surgeries to save the leg, prosthetic options, and even to choose another surgery entirely. The implications for communication skills training for a lifetime of self-advocacy will empower survivors to participate as an active partner with health care providers.

Strengths. A strength of this study is the inclusion of interview participants who are actively making a surgical decision at the time of the interview. Although these are decisions for the second decision and therefore limited, this data captures concerns as they are shaping. Some participants, like Athena and Bert, were in the stage of researching options while others, like Diana and Ingrid, were actively planning a future surgery.

The inclusion of the adolescents and young adults is often lacking in pediatric cancer research, is a strength of this study. The concerns of survivors as young as 14 at the time of the interview and 10 at the time of diagnosis is important in understanding the needs of the participant who is most directly affected by the surgery.

The involvement of the researcher having personal knowledge of the condition can also be a perceived strength. The ability to both step back from the content of the interview and step in when appropriate is considered part of being a “reflexive interviewer” (Attia & Edge, 2017). For example, during Opal and Olivia’s interview, Opal said there were no good options. Her daughter had rotationplasty after limb salvage and she knew my daughter’s story of amputation after limb salvage. When I agreed with her and said, “There’s no good choice,” Opal responded with, “No, I mean, with you guys too. By the point you got to have Zoe’s surgery and where we were at, it’s like we didn’t have anything to lose.” Quentin alluded more to my researcher role in his interview, “I think what you’re doing is awesome, going through and finding out what the best course of action so everybody knows the issues that come along with it, you know.” This stepping in and out of the parent/researcher role connected me to the survivors and parents and prompted participants to share more since I had an intimate understanding of the lived experience.

Limitations. This study is limited by several factors. First, both study #1 and study #2 reflect only the subjective account of the patient or family member who either posted on Facebook or who participated in a personal interview. These accounts are rich in the descriptions of the decision-making experience, but they are influenced by the passage of time and memory, especially for those who are many years past their surgical procedure. They do not include the perspective of surgeons nor do they include the lived experience of patients and families making the decision prospectively for the first decision.

The recruitment method from the Facebook osteosarcoma page is a limitation due to the digital divide; patients and parents in very rural areas or who are impacted by socioeconomic constraints may not have access to the internet. Knowledge of online support groups or Facebook specifically is also a limitation. These participants may not be representative of the patient population.

Third, participants for study #2 were self-selected so they wanted to share their experiences; it is unknown what concerns other parents and patients might have if they had concerns about participating in a research study or if they choose not to respond.

Fourth, selective sampling was used to collect a range of surgeries and geographical locations, over 60 people responded to the recruitment notices. It wasn’t until after the interviews started that five of the participants who said they had RP or AMP, had LSS first. In addition to those, four other participants were either actively making a new decision or looking at options. While those who had surgeries other than LSS for the first surgery were included, the range of RP or AMP surgeries was not as broad as anticipated. Further limitations include English speakers only, eliminating the perspective of other cultures and languages. Another limitation is the lack of diversity; all the participants identified as white.

Lastly my own role as a member of the community affects my ability to subjectively report on this study. Steps taken to address the reflexive nature of parent/researcher include letting the data in the interview speak for itself by using narrative analysis in the patient’s own words and refraining for interjecting my own story and perspective in the interviews. Furthermore, the requirements of dissertation studies do not allow for second coders.

Future Directions

This study paves the way for future directions in pediatric cancer and osteosarcoma-specific research in decision-making, trauma, social media, telemedicine, and communication theory.

The results of this study have ramifications for future research in the complex web of decision-making and how people deal with the trauma of making medical decisions for children. Parents who make life-altering decisions are at increased risk for PTSD, especially if their child relapses (Jurbergs, et al., 2009). Pediatric cancer survivors (Marusak et al., 2019), particularly those with osteosarcoma (Fauske Bondevik, et al., 2015) and their siblings (Alderfer, 2003; Kaplan et al., 2013) are all deeply affected by childhood cancer. Further studies on how complex decision-making affects the family over time may help future families prepare for additional decisions.

The use of social media for pediatric cancer information-seeking contributes to the understanding of the informational and support needs as well as patient-provider communication. Research with Internet-informed patients recommends that traditional models of patient-provider communication be revisited to adapt to this changing landscape of knowledge translation (Tan & Goonawardene, 2017). Additionally, in the aftermath of the COVID-19 pandemic, there will likely be nontraditional changes to shared decision making between patients and providers, particularly in oncology where immediate chemotherapy weakens the immune system. In a recent study investigating patient portals and the responses, over 90% of the messages were aimed at addressing needs and care with varying levels of risk and complexity (Robinson et al., 2017). The use of patient portals and other modes of telehealth will be paramount for information-sharing and shared decision making between patients and providers.

While knowledge translation and specifically, the interactive model, was selected as the best theoretical approach to understanding the communication needs in the osteosarcoma community, there are communication theories that could be applied to this study as well. The application of uncertainty reduction theory (Berger & Calabrese, 1975) to Study 1 and Study 2 would be useful in understanding how patients and families manage uncertainties and what events lead them to feel uncertainty when communicating with providers. Rapid and unequal communication with providers can violate the rules of social penetration theory and impact the necessary building of the relationship needed for trust between surgeons and patients/parents (Nazione et al., 2019). Indeed, many of the participants stated that they liked their surgeons because they shared information with them about their personal lives or said what they would do if they had a child in a similar situation. Problematic integration (PI) theory (Babrow, 1992) would be a useful theoretic lens to view how patients and parents integrate the threat of a life-threatening disease with a life-altering surgery. PI through communication with providers and expert patients includes the consideration of probabilities while balancing uncertainties to choose the best surgical option for themselves or their child.

Next Steps

This two-study dissertation examined the knowledge gaps for osteosarcoma surgical decision-making through a content analysis of Facebook posts (Study 1) and a narrative analysis of lived experience as shared in personal interviews with survivors and parents (Study 2). The purpose of this study was to understand the information needs of patients and families as formative research for the Amputation Patient Decision Aid for Osteosarcoma in the Lower Extremity, part of a five-year cooperative agreement between the Administration for Community Living and the Amputee Coalition.

The decision aid is informed by this study, providing the information necessary to understand the what patients and families need most when the decision for surgery is being made. This information will help fill the knowledge gaps identified for future families who are making this difficult decision. The use of integrated knowledge translation as participatory research with patients will aid in the transfer of knowledge to the implementation (Gagliardi et al., 2017).

A formal report of the results and recommendations will be shared with Members of the Osteosarcoma Patient Decision Aid Medical Advisory Team (O-DAMAT). O-DAMAT members include orthopedic surgeons, pediatric oncologists, social scientists, and decision aid experts as well as other health care professionals such as physiatrists, physical therapists, and prosthetists.

All 29 participants indicated on the last question of the interview guide that they would be interested in participating in future research; from these, the O-DAMAT Patient Partners will be formed. O-DAMAT Patient Partners will include youth, parents, and adult survivors in quarterly meetings to advise on the design and content of materials through 2024. Liaisons from the Patient Partners will also attend O-DAMAT meetings. Opportunities for co-authorship of publications throughout the development and co-presentation for dissemination at national conferences will be provided to patient partners.

The content areas will be outlined by the overlapping themes between the two studies. Specific content will be informed by the issue domains from the vignettes and the concerns from the posts. Recommendations for patient stories from the interviews are also supported by the experiences/expertise of survivors in the Facebook posts.

Due to the variances of decision-making models revealed in both studies, a section early in the decision aid on decision-making roles makes sense. While shared decision-making is a preferred style of communication, some patients and families prefer that they make the decision independent of the surgeon's recommendation or that the surgeon make the decision based on what the surgeon thinks is best. All three are valid models of decision-making. However, as an issue domain in the interviews included the need to advocate for patient preferences, self-advocacy skills will be promoted throughout the decision aid. Suggesting ways to talk about the decision with a health professional and ways for patients to share what matters most to them is also a component of the original 74-item checklist for patient decision aids with the International Decision Aid Standards (IPDAS, n.d.).

The accumulation of knowledge about options, limitations, and complications differs logistically in these groups due to the amount of time since diagnosis. Those on Facebook may have been diagnosed for days or weeks before posting while all the interview participants were at least one-year post op from their first surgery. The added dimension of survivors who had unexpected complications adds to this knowledge gap at the front end when making the decision. Together, the two data sets provide a nuanced picture of the knowledge gaps and unmet needs osteosarcoma patients face in surgical decision-making. The findings from these two studies set the stage for a comprehensive patient decision aid that includes long-term survivorship needs associated with limb loss or limb salvage.

APPENDIX A

Study 2 Recruitment Notice

Research Study for Bone Cancer

My name is Janet Panoch and I am a PhD student in Health Communication at IUPUI; my daughter is an osteosarcoma survivor.

I am conducting a research study for my PhD dissertation exploring surgical decision-making for bone cancer in the lower limb (amputation/limb salvaage surgery/rotationplasty). This study requires a 10 minute phone call to determine eligibility, a one hour interview, and a 10 minute follow up survey. Adult survivors, partners, parents. and children 14-18 with a parent are eligible to participate.

All participants will receive a gift card as a thank you for your time.

This study is IRB approved.

If you are interested, please contact me by email at jpanoch@iu.edu or by text/phone at (xxx)xxx-xxxx

APPENDIX B

Study 2 Eligibility Screening

Screening for Interview

Good morning/afternoon and thank you for reaching out to me for the research study on osteosarcoma decision making. I'd like to ask you some questions to confirm that you are eligible for the study; this will take about ten minutes of your time. Your responses are solely used solely to determine that you qualify for this research study; the information you provide to me now not be used as data. Would you like to continue?

Before we begin, I need to ask your age (if between 14-17, ask if a parent can be present on the call for the eligibility screening).

To start, my daughter is an osteosarcoma survivor so I understand what it's like to make a surgical treatment decision. I am interested in learning what it was like for other people. The results of this study will be used to develop a tool for surgeons and patients to use together when making this difficult decision.

Do you have any questions for me so far?

First I'd like to ask you who in your family was diagnosed with osteosarcoma.

How old were you/was the patient at diagnosis?

May I ask the date of the diagnosis?

And where are you/is the patient now in the treatment protocol?

Was a surgical treatment decision made?

What was your role in making the decision?

Would you say you are satisfied or dissatisfied with the decision right now?

In which city/state do you reside where a face to face interview might take place?

And lastly, do you/the patient have access to the internet for a video interview if a face to face interview is not possible?

Would you/the patient be able to conduct the interview in privacy?

Thank you so much for taking the time to talk to me about this study.

I will be in touch with you by email or by phone to schedule an interview to let you know if you are selected as a participant. May I collect that information now?

APPENDIX C

Study 2 Interview Guide

Intro	Good morning/afternoon and thank you for agreeing to be part of this research study to about making surgical decisions for the treatment of bone cancer in the lower leg. As I mentioned in the Study Information Sheet, this interview is being audio recorded but any names, dates, or locations will be removed Do you have any questions for me before we begin?
Personal story	**Thank you for your responses. Before I begin talking with you, I'd like to first ask you to share your story with me and you may start wherever you like.**
Part I **User Domain**	Thank you for sharing your story. I'd like to start the interview now; there are five sections where I'll be asking specific questions. First, I'd like to learn a little more about your background with doctors and your expectations for making the decision.
	What other kinds of medical decisions have you made in the past?
	What was your attitude about patients making a surgical decision?
	Who was involved in making the final decision?
	What surgical options were offered to you?
	How much time did you have to make this decision?
	Where were you going to find the information you needed to make a decision?
	What did you expect to learn from your sources (internet, other families, doctors)?
	What was most important/least important to you when making this decision?
Part II **Issue Domain**	I'd like to shift gears just a little bit now and talk about what it was like to make the decision itself.
	Do you feel like you had the expertise necessary order to understand the decision?
	Did things change with the decision making? (infection, complications, outcome of chemo)
	Can you talk about the role (if any) of uncertainty when you made this decision?
	What conflict did you experience when making the decision? Were friends and family members supportive or did they disagree about the decision?
	What was the hardest part for you when you made this decision? What were the risks of being the decision maker?
	Please tell me how you are feeling now about the decision you made.

Part III **Research Domain**	Now we’ll talk about the information you received for making the decision. I’m interested in learning more about how patients learn and where they get their information to feel knowledgeable about making a surgical decision.
	What research was available to you when you were making the decision?
	Were the resources you used clear and easy to understand?
	Was the information for options consistent from different sources?
	How did you feel about the quality of the information you received?
	How was the research presented? Did you learn about numbers/statistics or patient stories?
	Was the research that you found detailed or was it broader?
	Did the research state things you could do?
	How relevant did the research feel to you or your child?
	In what ways were the options in your research incompatible with your expectations or priorities?
	Would those incompatibilities have disrupted your (your child’s) quality of life?
	What quality of life changes were mentioned in the research that you felt would work – or not work – for you/your child?
Part IV **Researcher Relationship Domain**	I’d like to ask you about the relationship you have with your medical team. This might include your oncologist, surgeon, or anyone who comes to mind when you were thinking about making the surgical decision.
	How would you describe the trust and/or communication between you and the medical team?
	Did you know any of your medical team prior to diagnosis?
	Were there other things happening in your life that might have affected your ability to decide? (chronic illness, moving, stress, etc)
	Was there a specific member of the medical team with whom you interacted with more than others?
	How often did you meet with that person? Did you meet in person or talk by email or phone?
	Did you and the medical professional agree on what the goals were when making the decision?
	Did you and the medical professional agree about who would be making the final decision?

Part V **Dissemination Domain**	We're almost done. The last section I'd like to explore with you is recommendations for future materials that will help families like yours feel better prepared to make a decision and more satisfied with the outcome.
	Was it better for you to seek information about options elsewhere or to have the information provided to you by the medical team?
	What would be the most appropriate way to receive the information: written or oral, formal or informal? (follow up with video, audio, interactive module, etc)
	For written materials, which format would have been best for you? Brochures, booklets, images, illustrations, statistics, etc. What are your preferences for length?
	How much detail would you have wanted to see? (surgical details, outcomes, etc)
	Would the research be more vivid with patient case examples?
	What would be the optimal size or scope of information for you? (ie basic info or more details)
	How much information do you think you could absorb at the time each time you used it?
	Would it have been helpful to have more than one option available for you?
	Would it have been useful to have a medical professional check in with you while making the decision?
	To what extent and in what ways should the doctor continue to be available to patients/parents after presenting the options?
	Often the results of any surgical option for osteosarcoma can vary from person to person for reasons that can't be predicted statistically or as a number. When you think about a tool to help future families make these kinds of decisions, how important do you think it is to communicate this kind of uncertainty?
	Other than what we've talked about so far, as you look back to when you were making the decision, is there anything you wish you had known then that you know now?
	Thank you again for agreeing to participate in this research study. Before we end, I have a few demographic questions to ask you: **(Age) Which category below includes your age? (for all interview participants)** **17 or younger** **18-20** **21-29** **30-39** **40-49** **50-59** **60 or older**

	(Race) Do you identify as White, Black or African American, American Indian or Alaskan Native, Asian, Native Hawaiian or other Pacific Islander, or some other race? **(Gender) What is your gender?** **Female** **Male** **Non-binary** **Other** **(Marital status) Are you now married/partnered, widowed, divorced, separated, or never married?** **(Education) What is the highest level of school you have completed or the highest degree you have received?** **Less than high school degree** **High school degree or equivalent (e.g., GED)** **Some college but no degree** **Associate degree** **Bachelor's degree** **Graduate degree** **(Household income) How much total combined money did all members of your household earn in 2010?** **\$0 – \$9,999** **\$10,000 – \$19,999** **\$20,000 – \$29,999** **\$30,000 – \$39,999** **\$40,000 – \$49,999** **\$50,000 – \$59,999** **\$60,000 – \$69,999** **\$70,000 – \$79,999** **\$80,000 – \$89,999** **\$90,000 – \$99,999** **\$100,000 or more** **Finally, I'd like to ask you about the level of satisfaction with the decision that was made. I will read five statements and ask you how much you agree or disagree with each. The answers will be strongly agree, agree, neither agree nor disagree, disagree, strongly disagree.** **1. It was the right decision.** **2. I regret the choice that was made.** **3. I would go for the same choice if I had to do it over again.** **4. The choice did me a lot of harm.** **5. The decision was a wise one.**
Conclusion	We are at the end of our interview. Is there anything I did not ask that you think I should have asked? I will be emailing you a follow up survey with your gift card so if you do think of anything, please let me know. Would you be willing to participate in additional research for the development of a surgical decision-making tool?

	And finally, would you like to be notified about the results of this study? Thank you again – I will now turn the recorders off.

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Sallinen, M., & Mengshoel, A. M. (2017). "I just want my life back!" - Men's narratives about living with fibromyalgia. *Disability and Rehabilitation*, *41*(4), 422–429. doi: 10.1080/09638288.2017.1395085

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Shaffer, V. A., Focella, E. S., Hathaway, A., Scherer, L. D., & Zikmund-Fisher, B. J. (2018). On the usefulness of narratives: An interdisciplinary review and theoretical model. *Annals of Behavioral Medicine*, *52*(5), 429–442. https://doi.org/10.1093/abm/kax008

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Stevenson, J., Tsagkozis, P., & Grimer, R. (2016). Functional and quality of life outcomes in bone sarcoma following amputation, rotationplasty or limb-salvage. *Expert Review of Quality of Life in Cancer Care*, *1*(4), 303–312. doi: 10.1080/23809000.2016.1203725

Street, R. L., & Voigt, B. (1997). Patient participation in deciding breast cancer treatment and subsequent quality of life. *Medical Decision-making*, *17*(3), 298–306. doi: 10.1177/0272989x9701700306

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Tate, A., & Rimel, B. (2020). The duality of option-listing in cancer care. *Patient Education and Counseling*, *103*(1), 71–76. doi: 10.1016/j.pec.2019.07.025

Thomas, A., Kuper, A., Chin-Yee, B., & Park, M. (2020). What is "shared" in shared decision-making? Philosophical perspectives, epistemic justice, and implications for health professions education. *Journal of Evaluation in Clinical Practice*. doi: 10.1111/jep.13370

Thoren, E. M., Metze, B., Bührer, C., & Garten, L. (2013). Online support for parents of preterm infants: a qualitative and content analysis of Facebook 'preemie' groups. *Archives of Disease in Childhood - Fetal and Neonatal Edition*, *98*(6). doi: 10.1136/archdischild-2012-303572

Torre-Díez, I. D. L., Díaz-Pernas, F. J., & Antón-Rodríguez, M. (2012). A content analysis of chronic diseases social groups on Facebook and Twitter. *Telemedicine and e-Health*, *18*(6), 404–408. doi: 10.1089/tmj.2011.0227

Tozer, L., Stedmon, J., & Dallos, R. (2018). 'It is that bad but it isn't that bad': Exploring children's experiences of their mother's non-terminal cancer with a focus on attachment, resilience and trauma. *Clinical Child Psychology and Psychiatry*, *24*(1), 53–68. doi: 10.1177/1359104518781922

Vaismoradi, M., Turunen, H., & Bondas, T. (2013). Content analysis and thematic analysis: Implications for conducting a qualitative descriptive study. *Nursing & Health Sciences*, *15*(3), 398–405. doi: 10.1111/nhs.12048

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Woodgate, R. L. (2005). A different way of being: Adolescents' experiences with cancer. *Cancer Nursing*, *28*(1), 8–15. doi: 10.1097/00002820-200501000-00002

Zhang, Y., Yang, J., Zhao, N., Wang, C., Kamar, S., Zhou, Y., He, Z., Yang, J., Sun, B., Shi, X., Han, L., & Yang, Z. (2018). Progress in the chemotherapeutic treatment of osteosarcoma. *Oncology letters*, *16*(5), 6228–6237. https://doi.org/10.3892/ol.2018.9434

Curriculum Vitae

Janet Elizabeth Panoch

Education

PhD	Health Communication Minor, Medical Humanities Indiana University-Purdue University, Indianapolis	2020
MA	Communication Indiana University-Purdue University at Fort Wayne	1997
BA	English Indiana University-Purdue University at Fort Wayne	1993

Current Positions

2018	Amputee Coalition, Washington, DC Resource Development Manager & Co-PI for Administration for Community Living, Dept. of Health and Human Resources, for the National Limb Loss Resource Center,
2017	IU School of Medicine, Indianapolis, IN Co-PI, IU Health Values Grant for Education, *PACE-talk Patient Education: An Interactive Simulation Game for High School Health Classes*

Previous Positions

2017-2018	Research Assistant, IU School of Medicine, Pediatric Endocrinology, Tamara Hannon, MD 2017-2018
2017-2018	Research Assistant, IU School of Medicine, Pediatric Urology, Katharine Chan, MD
2017-2018	Research Assistant, IU School of Medicine, Children's Health Services Research, Nerisa Bauer, MD
2017-2018	Research Assistant, IUPUI, Communication Studies, Graduate Director Marianne Matthias, PhD
2013- 2018	Research Assistant, IU School of Medicine, Dept. of Obstetrics, The Center for Pediatric and Adolescent Comparative Effectiveness Research, Brownsyne Tucker Edmonds, MD
2013-2018	Graduate Student Mentor, IU School of Informatics, Josette Jones, PhD, Health Informatics
2014-2016	Co-PI, IU School of Medicine, IU Health Values Grant for Education, *Patient Communication Skills Training Module for High School Health and Wellness Classes*

2012-2015 Adjunct Faculty, Department of Communication, Ivy Tech Community College Courses taught: Interpersonal Communication, Introduction to Public Speaking

1997-2000 Adjunct Faculty, Indiana University-Purdue University at Fort Wayne. Courses taught: Interpersonal Communication, Public Speaking, Nonverbal Communication

1994-1997 Teaching Assistant, Indiana University-Purdue University, Fort Wayne, IN

Invitations

2017- present Formal advisor, Pediatric Oncology, invited appointment with annual honorarium, *K12 Child and Family Patient Centered Outcomes Research Scholars Program*, Katie Greenzang, M.D., Dana-Farber/Boston Children's Cancer and Blood Disorders Center

2020 MIB FACTOR, invited speaker, *"How do you make this life changing decision?": A Qualitative Inquiry on Patient Decision-Making for the Surgical Treatment of Osteosarcoma in the Lower Extremity*, Year One Formative Research for the Osteosarcoma Decision Aid, Phoenix, AZ

2019 Patient Engagement Working Group, session moderator, International Shared Decision-Making conference, Quebec City, Canada

2017 MIB FACTOR, invited speaker, *Self-Advocacy: The Patient's Perspective*, Osteosarcoma conference, February 23-25, Miami, FL

2016 *High School Meets Medical School: Teaching Teens to Actively Engage with Healthcare Professionals.* Invited keynote speaker for the Indiana Society for Public Health Education Conference, Indianapolis, IN

2013 *Addressing the Long-term Health Care Needs of Children Who Survive Pediatric Cancer*, in cooperation with Senator Dianne Feinstein and Safer Chemicals Healthy Families, briefing at the Dirksen Senate Office Building, Washington, D.C.

Awards

2020 IU School of Liberal Arts PhD nominee, Queener Graduate Student Excellence Award

2019 Patient Scholarship award for travel, accommodations, workshops, and registration, International Shared Decision-making conference, Quebec City, Canada

2018 $600 acceptance award to present advocacy research, *PACE-talk Patient Communication Skill Training*, Role/Play: Collaborative Creativity and Creative Collaborations Student Fellows Symposium, National Academy of Sciences, Washington, D.C.

2018 Patient-Centered Primary Care Collaborative, Key Policies to Elevate Primary Care Conference, Washington, DC. Transforming Clinical Practice Initiative (TCPI), funded by the

Centers for Medicare and Medicaid Services, full scholarship award of $1250 for travel expenses, hotel, and registration.

2018 British International School of Chicago, Lincoln Park, ComSciCon-Chicago, a two-day workshop to communicate the complex concepts arising in science, engineering, and other technical fields to diverse audiences, full scholarship award.

2017 *Elite 50* Top 50 Graduate Students, IUPUI. Award recognizes academic achievement, university service, and community benefit. Received 2017 Best in School award, School of Liberal Arts.

2017 3 Minute Thesis Competition for Doctoral Students, *Research Presentation on Self-Advocacy,* $350 Runner Up Award and $150 People's Choice Award, IUPUI Graduate School.

2017 Departmental Grant for General Dissemination and Travel of Advocacy Research, Communication Studies, $600 award.

2017-2019 Co-PI, IU Health Values Grant for Education, *PACE-talk Patient Education: An Interactive Simulation Game for High School Health Classes with Yale's Center for Health and Learning Games*, $94,569 award.

2016 Service Learning Assistant Scholarship, *Patient Communication Skills for High School Health and Wellness Classes*, $4800 award.

2016 Departmental Grant for General Dissemination and Travel of *PACE-talk Advocacy Research*, $600 award.

2016 Best Patient-Centered Poster Award for *Patient Communication Skills Training for High School Health and Wellness Classes Year 2: Partnering with Herron High School for Implementation of PACE Module,* Partnering with Patients in Decision-making Conference, Johns Hopkins Medical Research, Baltimore, MD.

2016 National Communication Association Student Travel Grant for *PACE-talk Advocacy Research*, 102st NCA, Philadelphia, $200 award.

2016 National Communication Association, Top Panel Award for the Training and Development Panel: *Turning the tables on patient advocacy: Responsible patients informing responsive providers.* Philadelphia, PA.

2016 Dissemination Grant, IUPUI Center for Service and Learning for *Patient Communication Skills Training for High School Health and Wellness Classes Year 2: Partnering with Herron High School for Implementation of PACE Module*, Johns Hopkins Medical Research, Baltimore, $500 award.

2016 IUPUI Graduate - Professional Educational Grant for Applied Interest Group Panel Submission, *[R]evolutions in Patient Advocacy: Effects of Online Engagement in Patient Health Initiatives*, Eastern Communication Association Conference, Baltimore, $500 award.

2016 IUPUI Communication Studies Travel Grant for *Patient Education in High School Health and Wellness Classes: Teens Review a Video Prototype of PACE-talk Communication Training*, Kentucky Conference on Health Communication, $250 award.

2016 Humanities Intensive Learning and Teaching scholarship and sponsorship, IUPUI Arts and Humanities Institute for the 2016 HILT conference course: Working with Scalar, $500 award.

2015 Humanities Intensive Learning and Teaching scholarship and sponsorship, IUPUI Arts and Humanities Institute for the 2015 HILT conference course: Digital Storytelling, $500 award.

2015 National Communication Association Film Festival, Honorable Mention, Las Vegas, NV.

2015 Honorable Mention Award, Film Festival, 101st NCA, Las Vegas. *The PACE-talk Module: Patient Communication Skills Training for High School Health and Wellness Classes.*

2015 NCA Student Travel Grant for Dissemination of Advocacy Research, 101st NCA, Las Vegas, $275 award.

2015 Top Student Presentation Award, 3rd Biennial D. C. Health Communication Conference, Communication Competence and Health Promotion, *Patient Communication Training for High School Health and Wellness Classes.*

2015 IUPUI Graduate - Professional Educational Grant for *PACE-talk Patient Training* Top Student Presentation Award Acceptance, DC Health Communication Conference, $500 award.

2015 IUPUI, Communication Studies Grant for Dissemination of Self-Advocacy Research, Shared Decision-making-International Society for Evidence-based Health Care conference, Sydney, Australia, $1600 award.

2014-2016 Co-PI, IU Health Values Grant for Education, *Patient Communication Skills Training Module for High School Health and Wellness Classes*, $62,000 award.

2014 CTSI (Clinical and Translational Sciences Institute) Researcher Award, 6th Annual CTSI Meeting, *Patient Communication Skills Training Module for High School Health and Wellness Classes*, $1000 award.

Publications

Chan, K. H., **Panoch, J.E.,** Carroll, A., Wiehe, S., Cain, M. P., & Frankel, R. (2020). Knowledge gaps and information seeking by parents about hypospadias. *Journal of Pediatric Urology*. doi: 10.1016/j.jpurol.2020.01.008

Salama, A., **Panoch, J.E.,** Bandali, E., Carroll, A., Wiehe, S., Downs, S., Cain, M., Frankel, R., & Chan, K. H. (2019). Consulting “Dr. YouTube”: an objective evaluation of hypospadias videos on a popular video-sharing website. *Journal of Pediatric Urology*. doi: 10.1016/j.jpurol.2019.11.011

Chan, K., **Panoch, J.E.,** Carroll, A., Wiehe, S., Downs, S., Cain, M., & Frankel, R. (2019). Parental perspectives on decision-making about hypospadias surgery. *Journal of Pediatric Urology*. doi: 10.1016/j.jpurol.2019.04.017

Chan, K., **Panoch, J. E.,** Carroll, A., Downs, S., Cain, M., Frankel, R., Cockrum, B., Moore, C., & Wiehe, S. (2019). Community engagement of adolescents in the development of a patient-centered outcome tool for adolescents with a history of hypospadias repair. *Journal of Pediatric Urology.* doi:10.1016/j.jpurol.2019.04.011

Tucker Edmonds, B., Mckenzie, F., **Panoch, J. E.,** Litwiller, A., & Dicorcia, M. J. (2019). Evaluating shared decision-making in periviable counseling using objective structured clinical examinations. *Journal of Perinatology, 39*(6), 857-865. doi:10.1038/s41372-019-0366-1

Tucker Edmonds, B., Mckenzie, F., **Panoch, J. E.,** White, D. B., & Barnato, A. E. (2016). A pilot study of neonatologists decision-making roles in delivery room resuscitation counseling for periviable births. *AJOB Empirical Bioethics, 7*(3), 175-182. doi:10.1080/23294515.2015.1085460

Tucker Edmonds, B., Mckenzie, F., **Panoch, J. E**., Wocial, L. D., Barnato, A. E., & Frankel, R. M. (2015). “Doctor, what would you do?”: Physicians’ responses to patient inquiries about periviable delivery. *Patient Education and Counseling,98*(1), 49-54. doi:10.1016/j.pec.2014.09.014

Tucker Edmonds, B., Mckenzie, F., **Panoch, J. E**., & Frankel, R. M. (2015). Comparing neonatal morbidity and mortality estimates across specialty in periviable counseling. *The Journal of Maternal-Fetal & Neonatal Medicine, 28*(18), 2145-2149. doi:10.3109/14767058.2014.981807

Tucker Edmonds, B., Mckenzie, F., **Panoch, J. E.,** Barnato, A. E., & Frankel, R. M. (2014). Comparing obstetricians’ and neonatologists’ approaches to periviable counseling. *Journal of Perinatology, 35*(5), 344-348. doi:10.1038/jp.2014.213

Mentor Experience

2020, IU School of Informatics, Health Informatics. Mentor for two health informatics graduate students at the MIB FACTOR to conduct the Osteosarcoma Decision Aid Survey, Scottsdale, AZ.

2019, IU School of Informatics, Health Informatics. Mentor for SP/2018 INFO-I535_NURS-I631: Clinical Information Systems project, PACE-talk Patient Communication Training for High School Health Classes: Branching Dialogue Development/Input & Evaluation Tools for Assessment.

2018, IU School of Informatics, Health Informatics. Mentor for SP/2018 INFO-I535_NURS-I631: Clinical Information Systems project – *Metaphors Matter: Establishing a Digital Collection of Doctor-Patient Interactions from TV/Film for Medical Education.*

2018, IU School of Informatics, Health Informatics. Mentor for SP/2018 INFO-I535_NURS-I631: Clinical Information Systems project – *YouTube Videos & Surgical Decision-Making: Evaluation and Collection of Credible YouTube Vides to Help Parents make Decisions for Hypospadias Repair.*

2017, IU School of Medicine, Children's Health Services Research, mentor for full time summer intern on *PACE-talk Patient Communication Skills Training.*

2017, IU School of Informatics, Health Informatics. Capstone advisor for four graduate students in their final project developing a Game Playbook in collaboration with Yale University for *Patient Self-Advocacy.*

2016, mentor for Health Informatics IUPUI graduate students, SP/2016 INFO-I535_NURS-I631: Clinical Information Systems - *Interactive Advocacy Game for High School Health and Wellness Classes.*

2016, mentor for Health Informatics IUPUI graduate students, SP/2016 INFO-I535_NURS-I631: Clinical Information Systems, *Adapting the PACE-talk Patient Communication Training Skills Module for High School Health Classes to the VA Audience.*

2015, mentor for Health Informatics IUPUI graduate students, FA/2015 INFO-I535_NURS-I631: Clinical Information Systems, *Patient Communication Skills Education Module for the Veteran Audience.*

2015, mentor for Health Informatics IUPUI graduate students, SP/2015 INFO-I535_NURS-I631: Clinical Information Systems, *PACE Project: Creating Educational Questionnaires for Healthcare Decision Support in High School Health and Wellness Classes.*

2014, mentor for Health Informatics IUPUI graduate students, SP/2014 INFO-I535_NURS-I631: Clinical Information Systems, *Adolescent Patient Communication Training Skills.*

2013, mentor for Health Informatics IUPUI graduate students, FA/2013 INFO-I535_NURS-I631: Clinical Information Systems, *Teaching Patient Centered Communication Skills to Adolescents using PACE.*

Volunteer Experience

2020, The Patient - Patient-Centered Outcomes Research, Adis Journals, reviewer.

2019, Certified Peer Visitor, Amputee Coalition.

2019, Certified Peer Visitor, MIB Ambassador Agents for Osteosarcoma Support.

2017-2018, International Shared Decision-making 2019 Conference committee member.

2017, Judge for the Case Study Student Competition at the Indiana Society for Public Health Education Conference, Ball State University, Indianapolis, IN.

2017, IU School of Medicine, *Metaphors Matter: Learning to Listen for and Mirror Metaphors for Heath Care Professionals,* workshops conducting usability tests at IUSM-Terre Haute and University of Pittsburgh – Johnstown.

2016, National Communication Association Reviewer, Health Communication Division, reviewer.

2016, International Society for Medical Decision-making, Decision Psychology and Shared Decision-making (DEC) Division, reviewer.

2016, Pennsylvania Communication Association, Interpersonal Communication Interest Group, reviewer.

2015, National Communication Association Reviewer, Health Communication Division, reviewer.

2015, Central States Communication Association, Heath Communication, reviewer.

2015, Indiana Medical Reserve Corps volunteer for Marion County and Hamilton County.

Conferences and Advocacy Research (Presenting Author)

Panoch, J. (2020, October). *How Many Experts can you Fit in a Decision Aid? The Challenges of Diverse Contributors in a Decision-Making Tool.* Symposium, 75-minute presentation, Society for Medical Decision Making annual conference, Chicago, IL

Panoch, J. (2020, March). *Challenges to Success and Support in the Pediatric Limb Loss/Limb Difference Population: An Interdisciplinary Research Collaboration.* Oral presentation at the Association of Children's Prosthetic Orthotic Clinics conference, Portland, OR

Panoch, J. (2020, March). *Amputation Patient Decision Aid for Osteosarcoma of the Lower Extremity: Which Option is Best for My Child - Amputation, Rotationplasty, or Limb Salvage Surgery?* Oral presentation at *the* Association of Children's Prosthetic-Orthotic Clinics conference, Portland, OR

Panoch, J. (2020, February). *An Overview of Online Patient Decision Aids: Developing a Decision Aid with the Osteosarcoma Community* (poster & survey), Surgical Decision

Aid for the Treatment of Osteosarcoma in the Lower Extremity (podium presentation & panelist), MIB FACTOR Osteosarcoma Conference, Scottsdale, AZ

Panoch, J. (2019, July). *Amputation Patient Decision Aid for Malignant Tumors of the Lower Extremity: Which Option is Best for My Child - Amputation, Rotationplasty, or Limb Salvage Surgery?* Poster presentation at the 10th International Shared Decision-Making conference, Quebec City, Quebec, CAN

Panoch, J., Wiehe, S., Cain, M., Carroll, A., & Chan, K. (2018, October). *Parental Information Seeking and Decision-Making in Pediatric Urology: Evaluating Youtube Videos Using the Patient Education Materials Assessment Tool for Audio-Video.* Poster presentation at the 39th Society for Medical Decision-Making conference, Montreal, QC, CAN

Panoch, J. (2018, October). *High School Health Classes and Lifelong Self-Advocacy: Patient Communication Skills Training in a Video Game Design*, 30-minute roundtable presentation, American School Health Association conference, Indianapolis, IN

Panoch, J., Wiehe, S., Cain, M., Carroll, A., & Chan, K. (2018, September). *Community Engagement of Stakeholders in the Development of a Patient-Centered Outcomes Tool for Adolescents with a History of Hypospadias Repair*, moderated poster presentation, Pediatric Urology Fall Congress, Atlanta, GA

Panoch, J. & Hecklinski, T. (2018, April). Invited to present a two-hour workshop at the University of Pittsburgh –Johnston on *Metaphors Matter*, Healthy U for a Healthy Community: Scholarship for the Real World conference. Johnstown, PA

Panoch, J. & Hecklinski, T. (2018, March). *Metaphors Matter*. Poster presentation, 15th Biennial Kentucky Conference on Health Communication, Lexington, KY

Panoch, J. (2018, March). *Translating Videos to Interactive Role-Play Video Simulations for High School Health Classes: Teens Inform PACE-talk – The Game.* Oral presentation, Role/Play: Collaborative Creativity and Creative Collaborations Student Fellows Symposium, National Academy of Sciences, Washington, D.C.

Panoch, J. (2017, December). *Patient Communication Skills Training for High School Health & Wellness Classes: Adapting Video to Video Game.* Poster presentation, 9th Annual Clinical and Translational Sciences Institute Meeting, Indianapolis, IN.

Panoch, J. (2017, November). *Developing Interdisciplinary Collaboration and Stakeholder Relationships: The Many Faces of Public Health.* Invited speaker and panelist for the Indiana Society for Public Health Education Conference, Ball State University, Indianapolis, IN

Panoch, J. (2017, October). *Patient Communication Skills Training for High School Health & Wellness Classes: Adapting Video to Video Game.* Poster presentation at the 39th Society for Medical Decision-Making conference, Pittsburgh, PA

Panoch, J., Cegala, D., Anderson, P., Wieczorek, S., & Warner, M. (2017, June). *Partnering with Patients: Advocacy Advances in Technology and Education.* Oral presentation/invited panelist at the 15th International Communication/Medicine/Ethics (COMET) conference, Indianapolis, IN

Panoch, J., Bauer, N., Parrish Sprowl, J., & Nafiseh, A. (2017, June). *Translating Advocacy Videos to Interactive Video Games for High School Health Classes: Teens Inform PACE-talk – The Game.* Poster presentation at the 15th International Communication/Medicine/Ethics (COMET) conference, Indianapolis, IN

Panoch, J. (2017, February). *Patient Advocacy.* Oral presentation/invited keynote speaker at the Osteosarcoma Alliance Conference, Miami, FL

Panoch, J., Cegala, D., Aldrich, J., Jerry, C., Tonsing, B., Warner, M., Wharton-Michael, P. & Wieczorek, S. (2016, November). *Turning the tables on patient advocacy: Responsible patients informing responsive providers*. Training and Development Panel: National Communication Association, Philadelphia, PA

Panoch, J. (2016, October). *High School Meets Medical School: Teaching Teens to Actively Engage with Healthcare Professionals*. Invited speaker for the Indiana Society for Public Health Education Conference, University of Indianapolis, Indianapolis, IN

Panoch, J. (2016, October). *Patient Communication Skills Training in High School Health and Wellness Classes*. Invited vender/information booth for the Comprehensive Medical Update for Primary Care Allopathic and Osteopathic Physicians, Seven Springs, PA

Panoch, J., Lazarus,K. & Leverenz, A. (2016, September). *High School Meets Medical School: Herron High and IUSM Partner in Patient Education Pilot Study.* Poster presented at the 8th Annual Clinical and Translational Sciences Institute Meeting, Indianapolis, IN

Panoch, J. & Lazarus, K. (2016, June). *Patient Communication Skills Training for High School Health and Wellness Classes Year 2: Partnering with Herron High School for Implementation of PACE Module.* Poster presented at Partnering with Patients in Decision-making, Johns Hopkins Medical Research, Baltimore, MD

Panoch, J. (2016, July). *Introducing PACE-talk: An eHealth Intervention for High School Health and Wellness Classes.* Keynote Community speaker, Humanities Intensive Learning and Teaching Conference, Indianapolis, IN

Panoch, J., Tonsing, B., Warner, M., Wharton, A., Wharton-Michael, P., & Wieczorek, S. (2016, April). Panel presentation at *[R]evolutions in Patient Advocacy: Effects of Online Engagement in Patient Health Initiatives*, 107th Eastern Communication Association Conference, Baltimore, MD.

Panoch, J., Lazarus, K., & McKenzie, F. (2016, May). *Patient Communication Skills for High School Health and Wellness Classes.* Poster presented at IUPUI Research Day, Indianapolis, IN

Panoch, J., Lazarus, K., & McKenzie, F. (2016, April). *Patient Education in High School Health and Wellness Classes: Teens Review a Video Prototype of PACE-talk Communication Training.* Poster presented at 14th Biennial Kentucky Conference on Health Communication, Lexington, KY

Panoch, J., Cegala, D., Lazarus, L, Bute, J., Anderson, P., Di Corcia, M., McKenzie, F., & Wieczorek, S. (2015, November). *The PACE-talk Module: Patient Communication Skills Training for High School Health and Wellness Classes.* Oral presentation for the Film Festival award at the National Communication Association conference, Las Vegas, NV

Panoch, J., Cegala, D., Lazarus, L, Bute, J., Anderson, P., Di Corcia, M., McKenzie, F., & Riche, S. (2015, September). *Patient Communication Skills Training for High School Health and Wellness Classes – Year 2: Partnering with Herron High School for Implementation of PACE Module.* Poster presentation at the 7th Annual Clinical and Translational Sciences Institute Meeting, Indianapolis, IN

Panoch, J., Cegala, D., Lazarus, L, Bute, J., Anderson, P., Di Corcia, M., & McKenzie, F. (2015, July). *Patient Communication Skills Training Module for High School Health and Wellness Classes*. Oral presentation at the 8th International Shared Decision-making-International Society for Evidence-Based Health Care (ISDM-ISEHC), Sydney, AUS

Panoch, J., McCallum, R., Ward, M., & Whitis, N. (2015, July). *Nicole's Story: A Case History to Better Manage the Illness Experience of Gastroparesis with Patients Using ISDM*. Poster presentation at the Informed Shared Decision-making-International Society for Evidence Based Health Care), Sydney, AUS

Panoch, J., Ward, M., Whitis, N., & Babrow, A. (2015, April). *Nicole's Illness Experience: Problematic Integration in Adolescence for Contested Symptoms*. Poster presentation at the D.C. Health Communication Conference, Fairfax, VA

Panoch, J., Stoltzfus, K., Wieczorek, S., & Lee, Y. (2015, April). *Using Big Data to Study Patient-Physician Interaction and Online Support Groups.* Panel presentation with chair: Maria Brann at the D.C. Health Communication Conference, Fairfax, VA

Panoch, J., Cegala, D., Lazarus, L, Bute, J., Anderson, P., Di Corcia, M., & McKenzie, F. (2015, April). *Patient Communication Training for High School Health and Wellness Classes.* Poster presentation at the 2015, D.C. Health Communication Conference, Fairfax, VA

Panoch, J. (2015, April). Junior Investigator Presentation: *PACE-talk for High School Health and Wellness Classes.* Invited oral presentation at the Annual International Children's Health Services Research Symposium, Indianapolis, IN

Panoch, J., Cegala, D., Lazarus, L, Bute, J., Anderson, P., Di Corcia, M., McKenzie, F., & Riche, S. (2014, September). *Patient Communication Training Skills for High School Health and Wellness Classes.* Poster presentation at the 6th Annual Clinical and Translational Sciences Institute Meeting, Indianapolis, IN

Panoch, J. (2014, November). Roundtable Research in Progress (RRIP): *Patient-Provider Communication and the High School Student: Adapting the PACE Model to the Adolescent Audience.* National Communication Association conference, Chicago, IL

Panoch, J., Young-Johnson, C., Young, L., & McCully, W. (2014, November). Panel: *Retraining in the Rustbelt – Stories of Success and Struggle with Students Pursuing Mid-life Career Changes.* Oral presentation at the National Communication Association conference, Chicago, IL

Panoch, J., McKenzie, F., Bute, J., & Tucker Edmonds, B. (2014, April). *Missed Opportunities for Partnership: An Analysis of Obstetricians' and Neonatologists' Steroids Counseling for Women at Risk for Extreme Preterm Delivery.* Poster presentation at the 13th Biennial Kentucky Conference on Health Communication, Lexington, KY

Panoch, J. (1997, November). *Judging the Book by its Cover: Dominance and Submission in Young Adult Romance Book Covers.* Oral presentation at the National Communication Association conference, Chicago, IL

Panoch, J. (1997, April). *Lipstick and Lollipops: A Critical Analysis of the Paradoxical Imagery of Adulthood and Childhood in Advertisements Intended for Adolescent Girls.* Oral presentation at the Central States Communication Association Convention, St. Louis, MO