

Dear parents and guardians and caregivers of boys born with hypospadias (and epispadias- I use the word hypospadias throughout)

I wrote this thesis for you.

You might be feeling pressured to begin the process of agreeing to genital surgeries to repair your son's hypospadias. The case for agreeing to surgeries is clearly based in the wish to normalize your son's penis, and there are many surgeons who offer that hope.

I want to offer you an alternative; the case for not doing surgery until your son grows up and is at least over 18 and can participate fully in these medical decisions. I offer this material because there are many cases when the risks of early surgeries involve lifelong complications and lifelong problems. These lifelong problems complicate everyday urological functioning and sexual functioning.

Sometimes these medical problems get worse as the man ages. Often, surgeons cannot accurately predict or calculate how to get a good outcome. For many boys the risks of surgeries may be greater than the benefits over the years.

In order to make these decisions, you need a lot of information. It is my hope that this paper contains much of the information that you need, or at least points you in the direction of getting that information through a process called shared decision making.

The idea of shared decision making is an ideal that is rarely available to parents making these important decisions. Indeed the words shared decision making actually mean taking more time and getting a wider variety of information than might be currently available. It is my feeling that you need much more time and much more information in order to get informed so that if you give your consent to surgeries, your informed consent follows as much education as you can get, including time to adapt to your new baby, time to question basic assumptions, and time to consider all the options- the option of doing nothing, waiting, or the option of surgeries.

The model of shared decision making is described in an article by Karkazis, Tamar-Mattis, and Kon which is on this website. That article includes many questions that you should be asking. I add many questions as well. Those questions are listed on pages 23-24 of my paper, which is also on this website.

This thesis is long, but the decisions will have lifelong consequences. In order to orient you here are some guiding ideas that begin the discussion.

In general surgeons distinguish between distal hypospadias which they consider easier to correct, and proximal hypospadias, which is much more complicated to correct.

One: Dr. Douglas Canning, a pediatric urologist, represents a small minority of physicians who are questioning the medical wisdom of doing surgeries on young babies and children (see page 34). Responding to an article by Stanasel et al which openly discusses the fact that complications that require reoperations are “higher than previously reported,”- especially with proximal hypospadias- Canning notes that “we urologists routinely underestimated the problems our patients have experienced following hypospadias surgery... we have misled our patients, their families and our colleagues...”

Two: Parents and professionals are often concerned about sexual functioning. But I feel that everyday urological functioning is not discussed enough in the medical reports. After Dr. Canning’s questions about long-range problems that go unreported, how do we ask the right questions to plan for the lifelong best interests of our boys? (see pages 23, 24). Should the question about healthy urination come before the question about sexual functioning? I quote an experienced hypospadias surgeon who said (see pages 18 and 26) that a natural urethra works well- the urethra that is made in the operating room is not going to work so well. It is likely that your baby has a well-functioning urethra and meatus, so should we remove that and reconstruct one that is not as natural and well-functioning without his full adult consent?

Note the following private conversation I had with one 32 year old, born with severe proximal hypospadias, who is now a survivor of six trips to the operating room for reconstruction and repair of those reconstructions. He lives with urological problems, and may well need more surgeries. He said, “I know that my medical future is hell, but I still think that parents should be offered the possibility of doing surgery.” Perhaps he has integrated all the experiences and complications into his identity in such a way as to see them as normal. But his quote sums up the messy dilemma that parents face- what is truly in the long range best interests of our boys born with hypospadias; should we leave a natural

urethra and meatus intact? or try to reconstruct them to conform to our ideas of what a normal penis should be? or should we change our ideas so that our sons have the opportunity to make their own decisions about their bodies when they grow to adulthood?

Critical questions about the safety, impact, possible trauma, and the medical outcomes of these surgeries abound. Psychosocial issues abound. If you decide to reject surgeries or if you decide to have surgeries, there might not be a lot of guidance, mentoring, or ongoing support for parents and growing boys. Supportive mentoring and guidance would be ideal. I think that the goal is to consider deep acceptance- to accept that a boy has a meatus, urethra, and penis that are different but healthy in their natural state, and that doing nothing while giving them all the love and respect that we can, might be in their best interests, at least until they are adults and can participate in or take control of the decision. We might do well to consider that we need to accept our sons as nature made them so that we can grow in our love and respect for our sons.

I need to make a note about language- how do we talk about these differences? Leaders in interACT and the Hypospadias/Epispadias support groups have been looking for language that respects these congenital differences, language that takes away the idea that these birth differences are pathologies. I support these efforts and look forward to a day when phrases like “hypospadias cripple” are terms from the past. However, most practitioners (and most people in general) will use the language of pathology to describe your son’s hypospadias, so I have not gone the extra mile to change all the language.

If you would like to contact me, please do so through this website or at bonniesteinberg2@gmail.com or call me at 516 428 0933. Leave a clear message - with your contact information –saying that you want to talk about hypospadias alternatives.

Bonnie Steinberg

**Hypospadias Treatment Decisions:
Resources for parents and professionals to use
for shared decision making**

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Chapter One: Introduction

When a baby boy is born with different genitals (the most common example is hypospadias) there is often a rush to make an early decision regarding genital reconstructive surgeries with the goal of making the genitals look and function like the majority of male genitals. Sometimes the babies have a medical problem that is life threatening and needs immediate medical diagnosis and management. But in the majority of cases, the babies have no medical problems, they just look different. The urethral opening and meatus are not at the tip of the penis- they may be just below the tip, to anywhere on the penis, to between the testicles. In the past, hypospadias has been considered a medical emergency, and parents are counseled to consent to surgeries to repair or reconstruct the penis within the first two years of the baby's life. More current thinking has questioned these early surgeries as the standard of care, and there is a controversy over whether or not the surgeries are necessary or elective, and whether or not early surgeries are in the best long-range interest of the boy.

Most parents may be unprepared for this kind of genital difference and for these kinds of decisions. Often they are asked to make serious decisions about surgeries without the time to adapt, without the time to get wide varieties of information and other opinions, without the time to meet adults who have grown up with these differences, or compare information and advice from different sources. Parents have numerous concerns about the impact of these genital differences on their babies as they grow, and about what the psychosocial issues might be. Parents and professionals wonder how the child will fit in with his family, school and friendship communities, and whether the child will feel confident about his body in social and intimate situations. They are concerned about the reactions of others, and aware that their boys might be teased and stigmatized. All of these are legitimate concerns.

Most often, parents are offered the option of surgeries in the hope that cosmetic, reparative or reconstructive surgeries will make the genitals look and function normally, so that the child can develop without most of the psychosocial problems that the parents fear.

Recent thinking has questioned the assumptions that genital surgeries can fix the differences and set the baby on a normal course of development. Recent thinking has questioned the idea that the surgeries will fix the genitals so the problem of having different genitals will actually go away. There are many adults who had the surgeries, as young babies and as children, who are reporting that the surgeries caused more harm than good, the risks outweighed the benefits. Some physicians are beginning to write about the long range complications that have resulted from the surgeries- fistulas, diverticulae, urological problems that cannot be addressed- and are beginning to question the wisdom of early surgeries. Many of the adults who had hypospadias repairs as babies and young children are asking for the involved doctors to reflect deeply on their practice of doing early surgeries, and to stop doing them. They are asking the involved doctors to turn their attention to their adult needs and the follow up that they need over the long-range. These vocal adults are assuming that if parents were given time and the guidance of multidisciplinary teams to gather a lot of information and to adapt, then the numbers of surgeries would go down. There is a small but growing number of physicians who are beginning to question the results of the surgeries themselves. This small number of surgeons and professionals are also beginning to wonder if the risks outweigh the benefits. They are beginning to wonder if parents have been given too much hope that the surgeries would correct their babies' genitals so that they could lead lives with normal genitals. This paper will focus on the example of hypospadias, one of the most common genital differences that is addressed through surgeries. I will review many of the writings that are available that outline the major points that are being made by bioethicists, by adults who have had the surgeries, and by surgeons who do the surgeries. I will also review some writings by legal scholars, and review internet sources.

A note about nomenclature and the language that we might use to describe hypospadias; is it a Disorder of Sex Development or an Intersex condition? Adults who are leaders in the Hypospadias Epispadias support groups do include themselves in these categories and ally themselves all together as a group. Internally, they are looking for language to describe themselves that takes away the stigma, or that describes without pathologizing, and they

debate the language. However, when it comes to advocacy for their health, they group themselves together with Intersex and DSD's and when they read the books and articles about intersex conditions and DSD's they see themselves together with the entire spectrum of differences – from mild or distal hypospadias to severe or proximal hypospadias. In personal emails with the leaders of the group, their identity is reflected in the email I received from Dr Arlene Baratz on April 10, 2016. Baratz writes,

“I think of DSD as variations of reproductive development that are differences rather than disorders, and I include hypospadias in that category. We know that some anatomic variations, such as cloacal and bladder exstrophy, which involve the urinary and GI systems, require urgent surgery because they are life-threatening. Other variations (which often have gotten treated with early surgeries in order to prevent urinary infections are questionable because) there is no proof of an increased rate of infection. ...Urologists are starting to reevaluate recommendations for hypospadias repair... (finding) high complication rates that were worse than expected...there have also been unexpected findings at the other end of the spectrum, for distal hypospadias repair. When they evaluated parents a year after surgery, Canadian urologists were surprised to find that although all parents had chosen surgery, half had significant decisional regret...they suggested better (preop) education (for parents). In an Italian study ... (they) found a similar rate of decisional regret, ¾ of the parents wished they had received more information preoperatively. At the very least, these studies suggest to me that consent is far from ‘informed’.”

Later I will refer to some of the books that discuss DSD's in general or intersex conditions in general. Leaders in the Hypospadias and Epispadias Association, while they understand the different natures of the wide variety of conditions and the differences in treatment decisions, see themselves as part of that whole group.

Recent thinking has suggested that parents should be offered the opportunity to go through a process called shared decision-making to give them information, time, and exposure to a wide variety of approaches before they make the decision to go ahead with surgeries for their babies and young children. Usually hypospadias has not been addressed through a team approach, but the adults who had hypospadias surgeries are advocating that new parents should be offered a similar multifaceted team to guide them through the shared decision making process before deciding on surgeries. I hope that this project will contain much information to aid in the process of shared decision making for parents and professionals. My goal is to present the references, ideas and sources so that parents and professionals can get as much information as possible to use in the process of shared decision making. My best hope would be that this paper could begin to give parents and professionals some of the information they need in order to decide whether or not to agree to have surgeries for their boys with hypospadias.

Shared Decision Making and Informed Consent

There are three sources that I have chosen that indicate that shared decision making is an ideal goal for parents going through the process of deciding about surgeries. Indeed, one gets the

feeling that shared decision making should be the process of getting all the information that parents need to make treatment decisions. It is not the goal of this paper to analyze or deal with all the issues in the area of informed consent, but one gets the impression that the process of shared decision making would be the ideal way to handle the whole process of informed consent for hypospadias (and other decisions about genital surgeries for babies and young children) because parents need time and information before they make their decision to consent to surgeries or decline the option of surgeries. In chronological order, the resources are the 2006 Consensus on Management of Intersex Disorders by an international committee of 50 experts (Lee, Houk et al), the 2010 article called Genital Surgery for Disorders of Sex Development: Implementing a Shared Decision-Making Approach by Karkazis, Tamar-Mattis, and Kon, and the 2011 edition of Greenspan's Basic and Clinical Endocrinology text book edited by Gardner and Shoback.

Parents of boys with hypospadias are not usually offered the process of shared decision making with a diverse team that is recommended for parents of babies with DSDs. But the leaders of the Hypospadias Epispadias Association would like the parents of boys with hypospadias to be offered that same process of shared decision making. They look to the consensus statement, described next, to outline their hope that parents of boys with hypospadias will be offered that same team process. The approach of the adults with Hypospadias is also discussed in the section on their responses below. In brief, the adults who had surgeries for hypospadias as babies have such a high rate of life-long complications and problems resulting from the surgeries, that they feel that parents make the decisions to agree to surgeries before they have the time to get all the information that they need. Their claim is that the only way parents can get the information in order to understand the risks and benefits of surgeries is to have a team guide them through the shared decision making process before any surgeries are done.

The Consensus statement starts by discussing disorders of sex development in general, but also includes guidelines for hypospadias repair. They call for "an experienced multidisciplinary team" to "work with the family to reach the best possible set of decisions" with "ample time and opportunity for continued discussion with review of information previously provided" (Lee, Houk et al 2006, page e490). Support groups can also have an important role in this care Lee, Houk et al 2006, page e490). Regarding the specific case of hypospadias, they note that "patients must not be given unrealistic expectations about penile reconstruction..." (Lee, Houk, et al 2006, page e492). Over time, "authorization is given in stages to allow time for the parents to come to terms with their child's condition" (Lee, Houk et al 2006, page e497). Some physicians and many adults who had the surgeries as babies and young children agree that the risks of the surgeries and the psychosocial issues raised by having different genitals mean that parents need this team approach with education over time in order to make a fully informed decision about treatments.

The 2011 edition of Greenspan's endocrinology textbook also suggests that parents and caregivers need time to get a wide variety of information.

“The parents need to be presented with the whole picture... possible medical and surgical therapies, risks, complications and unknowns. It is critical that they have the knowledge and time to make an informed decision about any surgical procedures” (Gardner and Shoback 2011, page 507). This textbook notes that “the timing of any surgery remains controversial. This is a decision that needs to be made after careful discussion with the parents.”

This textbook does not use the words “shared decision making,” but it would seem that they are suggesting the process without reference to the article above.

The article by Karkazis, Tamar- Mattis and Kon is most helpful in describing the process of shared decision making. Because of the controversial nature of the surgeries, and the debate about the risks and benefits, they outline a six-step process for shared decision making that can be used for any difficult medical decisions (Karkazis, Tamar-Mattis, and Kon 2010, page 790). They write,

“the process of thoroughly examining alternatives and the encouragement of transparency and questioning... can help to ensure that the best interests of the child and family are served, patient care and the doctor-patient relationship are improved, satisfaction with the decision-making process is increased for both physician and parents, decisional conflict and regret are minimized, and ethical and legal requirements for informed permission (and assent when appropriate) are met” (Karkazis, Tamar-Mattis and Kon 2010, page 790).

This process “...necessarily requires clinical caregivers to reveal their reasoning, values, and biases, and to similarly explore the understandings, values, and reasoning of patients or their surrogates... This process can also help caregivers meet legal and ethical standards for informed consent (Karkazis, Tamar-Mattis and Kon 2010, page 790) and “allows caregivers and parents to engage in a thoughtful discussion of the pros and cons of treatment options” as it helps elicit the “best option” for care (Karkazis, Tamar-Mattis and Kon 2010, page 791). The article includes a long list of questions that parents need to review with the professionals on the care team. It includes a section of questions that are specific to hypospadias repair (Karkazis, Tamar-Mattis and Kon 2010, page 802) and later in this paper I add my own questions to that list. Note that these questions are centered around the risk of multiple surgeries that increase the risk of becoming what is sometimes called a “hypospadias cripple,” a problematic term describing a problematic situation that is discussed below in the chapter on articles by surgeons. The six-step process of shared decision making that is described by Karkazis et al, along with the questions at the end of the article, seems the ideal process for parents and professionals.

A parent might look on the internet and find both a site from the Centers for Disease Control (CDC) and the Hypospadias and Epispadias Association (HEA). I reviewed both sites on March 7, 2016 and found both clarity but also contradiction. The CDC site leads to the HEA site but with the note that the views of the HEA are its own and do not reflect the official position of the CDC. It is unclear with which views the CDC would agree or disagree. Perhaps the CDC recommends surgeries for most forms of hypospadias, not just the most severe. The HEA website seems to

make some suggestions for surgeries and then some suggestions against surgeries. In the section near the CDC material, the HEA suggests that the most severe forms of hypospadias require surgery to allow basic sexual function and normal urinary function. The HEA suggests that for the lesser forms no surgery is often chosen. However,

“...the most severe cases of penoscrotal hypospadias with severe chordee [curvature of the penis] require creation of a complete urethra in multiple staged surgeries. No one would argue that the child with the most severe case of hypospadias will require surgery, often to allow basic sexual function and normal urinary function where none existed previously. For this child the risk of surgery is clearly a sensible undertaking compared to the benefit of undergoing procedures... Surgery in the mildest cases is cosmetic in benefit and will not confer much functional improvement to the patient, if at all. In this case the risk is relatively higher, since the benefit of the process is not as great as it was for the first patient with severe impairment of function” (Hypospadias and Epispadias Association website, accessed March 7, 2016).

Parents researching options would read in both the CDC and the HEA websites that if their son has a severe case of hypospadias, surgery is recommended. Yet, reading further, this recommendation advocating surgery for the most severe form of hypospadias (called penoscrotal or proximal) is contradicted elsewhere within the HEA website (<http://heainfo.org/index.php/category/hypospadias>). The recommendation for surgeries is contradicted in multiple ways, including recommendations that the parent not rush into a decision, but rather gather information, consider delaying surgery until the boy is old enough to participate in the decision, and consider doing nothing because of severe side effects. These side effects often include fistulas, scarring, stricture, re-repair, or loss of function. The HEA website includes many personal stories of the lifelong difficulties of addressing these risks and of finding good care over the course of the man’s life. See also page 21 below for a review of the comments about hypospadias on the Intersex Society of North America website.

Fausto-Sterling (2000) notes that

“studies of hypospadias surgery reveal good news, bad news, and news of uncertain valence. The good news is that adult men who have undergone hypospadias surgery reached important sexual milestones—for example age of first intercourse—at the same age as men in control groups (who had undergone inguinal, but not genital, surgery as children). Nor did they differ from control groups in sexual behavior or functioning. The bad news is that these men were more timid about seeking sexual contact, possibly because they had more negative feelings about their genital appearance. Furthermore, the greater the number of operations the men had, the higher their level of sexual inhibition. Surgery was least successful for men with severe hypospadias, who could often have normal erections but found that problems such as spraying during urination and ejaculation persisted” (Fausto-Sterling 2000, page 87)

She goes on to write that although the medical literature writes confidently about the genital makeovers, “the procedures are complicated and risky. From 30-80 percent of children

receiving genital surgery undergo more than one operation. It is not uncommon for a child to endure from three to five such procedures” (Fausto-Sterling 2000, page 86).

One also needs to ask not only what the surgeries and recoveries are like for the boys, but what the medical exams are like for the children and how success might be measured. Fausto-Sterling notes, “ An intersexual man pointed out to me that one method of measuring penile growth and function in intersex boys involved the doctor masturbating the boy to achieve erection” (Fausto-Sterling 2000, page 86). Preves (2003) also reports that when she interviewed people about their experiences, “participants regularly reported that their genital exams routinely involved doctors’ stimulation of their genitalia to assess genital responsiveness and size” (Preves, 2003, page 73).

A parent who has read the website from the Centers for Disease Control and Prevention and then compared it to the Hypospadias and Epispadias Association website might notice the following contradictory things. One, the CDC leads to the HEA information but does not seem to endorse it, saying that it does not reflect the official position of the CDC. Second, the CDC site recommends surgeries for many types of hypospadias- from severe to moderate- while the HEA website suggests that parents should wait, gain lots of information, consider the side effects, and let the boy grow up in order to participate in the decision making. The parent will read many personal stories of the difficulties that the boys and men face as they grow up or need corrective surgeries for the surgeries that were done when they were children. Some of the personal stories contain assertions that the boys or men were glad to have had the surgeries. But as one goes on to read about their chronic problems, one might wonder how they assess satisfaction because these everyday difficulties with urinary and sexual function seem difficult to live with and in some ways contradict that report of satisfaction.

The parent who reads these reports sees that there is a conflict over treatment, that there are difficult decisions to be made and that there is a conflict between the suggestions of the CDC and the Hypospadias Epispadias Association. These contradictions and the questions that they raise will be echoed in the articles by surgeons and physicians when they report the challenges of hypospadias repairs.

In order to get oriented for the conversation, a parent might need to consider the sources in a deeper way. There will be sources that are from medicine and research, articles by surgeons and physicians. There will be articles and books by ethicists, researchers from sociology, psychology, biology, philosophy, and the personal anecdotes from the adults who have had the surgeries. There are a couple of legal experts to consider. Perhaps the parent needs to consider the hierarchy of values in the conversation, or the political nature of the sides, or the power that doctors might have over their thinking and over the decision making process. Parents and professionals might want to consider those power assumptions. Does the parent assume that the doctor is the more informed and therefore most appropriate source for information, and that the patients are just unhappy? Parents and professionals might need to reflect on their personal biases and responses to these differing sources of information. In bioethical matters, the ideal is that all voices are heard in the conversation. But power and hierarchy of education

often dominate our thinking and can influence the outcome. One might automatically assume that sources from medicine are more valuable than the personal anecdotes. One might assume that sources from medicine have statistics on their side. One might assume that too much passion or anger from some of the adults who had the surgeries negate the seriousness of the information. In this conversation there is much passion on each side, and much questioning of the value of the anecdotes and personal testimonies of the adults who have had the surgeries. Parents and professionals need to think through how they are responding and note these values and ideas as they engage in the conversation. In general many who value sources from medicine or science will discount the “soft sciences,” and people who value the anecdotes and personal histories. In turn, these “soft or social sciences” might criticize the hard scientists for ignoring the personal stories that the adults have.

Parents and professionals need to take all the information seriously. For example, parents naturally value having a child that fits the physical norms and has normal genitals. Facing the difference that a baby with hypospadias has will take some adjustments. The wish to repair might be so compelling that a parent might not be able to assimilate the controversies over whether or not the risks are worth the difficulties of surgeries. The parent might be unsure of which information has more value or weight- the narratives of the adult patients who are speaking out or the assurances of the surgeons. The parents need to be given time to see if their values or opinions change after getting more information. The following is an introduction to the conversation about how parents and professionals might value the hard sciences versus the softer sciences.

Chapter Two- Science versus Anecdote, Quantity vs Quality

How should parents and professionals evaluate the sources of information? The first thing that parents need to consider is the controversial nature of this decision, how hotly it is debated. There are several sources of information for parents. Physicians and researchers consider their information scientific and quantitative. Scientific information and information from medical sources generally get preferential treatment. The adults who have lived with the results of the early surgeries, on the other hand, tell personal narratives. The bioethical and legal advocates write long narratives that include the references to all the sources from medical writings as well as the personal narratives of the adults who have had the surgeries. These qualitative writings sometimes question the science of the quantitative writings. Parents might need to reflect on the inherent authority and power that our society gives to doctors. Parents might want to consider that if they give the medical sources more power and weight, then when they hear the personal stories and read the bioethical, qualitative and social science material will they be able to give each source their full attention and respect. The writers from the medical perspective generally do not reference these qualitative reports. One wonders if the medical sources have read the social science sources, or if they discount the social science sources as simply reports from a minority, therefore they are too personal or too anecdotal. It is interesting to note that the majority of the medical writers are men, the majority of the bioethics and social science writers are women. Given what we do know about the marginalization of women's voices in

science in general, one wonders if the gender of the source also affects the value people place on the material presented.

Additionally, in today's world a parent would search the internet for sites about hypospadias. Those sites are often weighted heavily against surgeries. How should the different kinds of information be measured for value? Which should get preference? Accounts from adult survivors are obviously subjective and anecdotal. A parent has no way of knowing what percentage of the affected community the adults represent. Are they a small minority or are they half of the group? Where are the testimonies from satisfied adults? Should a parent only listen to reports from the medical, quantitative science sources and discount anecdotal qualitative information? What counts as the most reliable information?

In general scientists might agree that the gold standard is a randomized clinical trial, but Karkazis (2008) notes that a randomized clinical trial to evaluate genital surgery in children would be ethically unacceptable (Karkazis 2008, page 282). Adult advocacy groups have been asking for deeper dialogue with the physicians who care for them, and have been disappointed in the lack of serious access to that dialogue. Johnston (2012) tries to describe this lack of serious dialogue between the medical sources and the social science sources. According to her, the emotions of the adults who had the surgeries drive the scientists apart from the adult survivors of the surgeries. Perhaps the dialogue between medical practitioners and the adult community and their legal and bioethics advocates is hampered by this science versus anecdote divide. As Karkazis (2008) notes, "...it is not uncommon for researchers more accustomed to quantitative approaches to view qualitative research as a marginal methodology (Karkazis 2008, page 293, note 9)." It is very hard to find any quantitative studies that help parents understand the long-range outcomes (see the chapter below on writings from medical sources). It is important to note that both physicians writing articles and the non-physicians agree that real statistics and numbers and outcome studies are hard to come by in the United States. The United States has no national registry, and we do not have long range outcome studies that fit the ideals of statistical analysis. Karkazis describes her method as just talking to everyone involved (Karkazis 2008, page 16), like an anthropologist. I think that her approach and her material should be counted as good enough to be taken seriously. Surgeons Moneer Hanna and Gina Cambareri (Cambareri and Hanna, 2015) note that the long range studies are not good. Writing about hypospadias, they note that "follow up studies after sexual maturity... are very limited (page 35), "long-term outcomes ...are scarce"(page 40), and "progress in hypospadias surgery will require long-term patient-reported outcome studies to better determine how the patients fare in adolescence and adult life" (page 41). Indeed in order to get real outcome studies, after appropriate review by Institutional Review Boards for compliance with ethical guidelines, we would need many more surgeons to open their files and follow up with all patients who are available.

I report the following in some detail because I believe that it might help parents and professionals think through the research science versus social science qualitative research divide. A pediatric psychologist and clinical researcher at a prominent university, is an expert on research with DSDs. I called him on August 8, 2014 and the following is a summary of what I

took away from that phone interview. This researcher's comments in that phone conversation highlight the tension between science and anecdote, between hard data and the personal recounting of difficult experiences. It seemed clear from our phone conversation that he gives more weight to science. Indeed I would characterize him after the phone conversation as data hungry. Research driven, he wants quantitative outcome studies and almost discounts personal qualitative narratives. I later looked into his wide ranging work and noted that he collaborates with the qualitative researchers. Since he works in both worlds – data and human testimony - I report the conversation and then in a later chapter I look at some of his writings closely. Since adults with hypospadias want the same consideration given to their treatment decisions, I report this conversation as relevant to issues that parents face with hypospadias.

He quoted Daniel Patrick Moynihan saying, "You are entitled to your own opinion, you are not entitled to your own facts," perhaps in order to support his belief that anecdotes do not make good science. Regarding the claims of the adult survivors that their surgeries did damage, he questioned the validity and value of hindsight. He is asking whether the adults really experience more sexual dysfunction than the general population, and whether one can blame their sexual dysfunction on the surgeries? What is the effect of subjecting a child to repeated harsh exams over time? He notes that 20% of the general population reports experiencing sexual distress, so we need to ask if distress among adults who had genital surgery can really be entirely attributed to the surgeries. When he said, "distress is common as dirt and it's bad scholarship," I believe that he is again discounting anecdotal evidence (qualitative reports) in favor of more measurable (quantitative) outcome studies.

This researcher's comments are representative of a preferential treatment given to medical opinion based in science that is measurable—he wants hard evidence. That is not a bad thing, but what does a parent do while the research continues? The scientific community cannot yet present parents with hard data on long-range outcomes. His position also raises several concerns. First, there was a deep discounting of the trauma of the surgeries, their frequent complications which require follow-up surgeries and the medical exams. The difficulties of those surgeries is a central fact for anyone who has undergone them, witnessed the surgeries or accompanied someone during the recoveries. Some of the recoveries are lengthy and compounded by medical complications that are difficult to address and become increasingly problematic (see the section below on adult survivors and their advocates). Similarly, by questioning hindsight, he discounts the seriousness of the personal narratives and memories from the patients. He also discounts distress as relevant to scholarship. Are the testimonies and memories of adults merely interesting but not really relevant for parents or professionals? Are they vague mis-rememberings, are they simply suspect because they are a looking back, or are they just some bad memories? Memory is often not literal but that does not mean that it is worthless. If adults are reporting trauma, then that should be taken seriously. By asking if this is just like living with any complex medical condition, this researcher seems to be including cosmetic surgeries that reconstruct genitals in a similar category along with surgeries that save lives and have many medical advantages. As a parent, I would want to know if adults who had surgeries as babies for other congenital differences are working together to ask the doctors to change their practices. Comparing the work and long range outcomes and problems of any

other advocacy groups would be very fruitful to see if there are parallels, but beyond the scope of this project.

More generally, how does a parent weigh the subjective memories and experiences with the hard data that this researcher is working to get? How do we humanize the data that we do have on outcomes, so that a parent can balance the stories that they hear with the material written by surgeons about their outcomes? Karkazis (2008) notes that there are important differences between scientific/quantitative and narrative/qualitative work:

“Unlike quantitative studies that focus on the measurement and the analysis of causal relationships between variables, qualitative research explores processes and values in an effort to explain how social experience is created and given meaning... The inductive nature of qualitative approaches makes them especially suitable when the phenomena under study are emerging or not fully understood... (We gain) depth, expanse and novelty of the findings...” However, “it is not uncommon for researchers more accustomed to quantitative approaches to view qualitative research as a marginal methodology” (Karkazis, 2008, pages 292- 293, note 9).

Parents and professionals need both science and anecdote in order to have access to the best information they can get in order to make the right decision for their child. Indeed through the shared decision making process, all information has to be taken seriously. It seems to me that parents need to know that the pain their child will endure will be worth it over time. Parents need to know if the pain will be in the normal range, or if the pain will be an undue burden.

Parents’ most important questions include the following: Will these surgeries get us the results that we hope for? Will my child be spared the psychosocial problems of having these genital differences? Is the pain and trauma of the surgeries worth it? In order to consider these questions, I believe that parents need both scientific information and the personal narratives of the adults who had the surgeries.

The goal of the shared decision making process is to expose parents and professionals to a wide variety of information, and give them time to absorb the material with as much care and reflection as possible. The goal is to give people the time to examine if their initial gut reactions stand the test of time and more reflection- or should they change their values and thinking after careful consideration. The goal of a bioethical approach to shared decision making would be to include many voices and sources as information is gathered. The narratives of the adult survivors need to inform the medical reports in a serious way.

Chapter Three—bioethical points of view

1. Introductory ethical issues. The Consensus Statement of 2006 is considered a watershed in the treatment of genital anomalies including hypospadias, but does not address several ethical considerations. Dreger and Sandberg (2010) write that the ideal team for shared decision making does not really exist in most institutions, so that the collaboration of the ideal team with parents is rare. I suggest that Dreger and Sandberg might agree that the ideal team facilitates ethical decision making and should be the standard of care. “The multidisciplinary team...

statement includes specialists in pediatric endocrinology, pediatric surgery or urology (or both), psychology and/or psychiatry, gynecology, genetics, neonatology, and (if possible) social work, nursing, and medical ethics... To allow such children to be treated only by, say, pediatric endocrinologists or urologists is to practice in a way that is ethically questionable. (Dreger and Sandberg, 2010, page 152). This article could be seen as one in a small group of articles that advocates a long process of shared decision making with a highly trained team, even for hypospadias, and with an emphasis on psychosocial issues. My recommendation is that this team approach be applied both for distal and proximal hypospadias, even though surgeries for distal hypospadias are currently considered simple and not complicated.

The authors know that the consensus statement describes an ideal, and that most medical centers do not create or support the development of these teams with financial resources. "We would argue that the failure of health care systems to financially and institutionally support optimal care is in and of itself an ethical issue" (Dreger and Sandberg, 2010, page 153). They write that "two of the thorniest ethical issues are the open disclosure to patients of their medical history, and the use of surgical "normalization" procedures on children too young to consent for themselves" (Dreger and Sandberg, 2010, page 153). They also note that full disclosure is in the best interests of the well-being of the child and the family (Dreger and Sandberg 2010, page 154). Further, in saying that the team approach of shared decision making is so important for fully educating and informing parents and professionals, Dreger and Sandberg seem to be suggesting that when the team is absent for educating and informing parents, then informed consent is compromised. Dreger and Sandberg note that the consensus statement supports the consideration of early surgeries, but also notes that if there is no team to guide parents through the whole ideal shared decision making process, then parents might not have enough information to make a fully informed decision about genital surgeries.

Dreger and Sandberg also note that the consensus statement does not ask about the ethics of whether parents and professionals should be allowed to pursue genital normalization (page 157). The consensus statement does not make a critical distinction between what is medically necessary and what is elective (page 157). Referring to the anger of the adults who have had those surgeries (page 158), Dreger and Sandberg note that parents need to know about that anger over the surgeries, and about the difficulties of the treatment around these procedures which contributed to shame, psychological trauma and problems that they believed were a result of the care (page 158). In short, Sandberg and Dreger know that there are very few ideal teams to orient parents over time so that they can get enough information from that diverse team of experts. The implications for informed consent might be that if a parent is not part of this more rigorous process of getting information, informed consent might be compromised.

Questions about the validity of informed consent are raised in an article by Kishka-Kamari Ford which outlines the requirements for valid legal consent, concluding that the current model of treatment for genital surgeries "...fails the test for legal informed consent at every step" (Ford 2001, page 13). Parents must be fully informed, feel that the decision is voluntary and that they have real competence to make the decision. These three conditions are compromised by lack of information about the long range outcomes, vulnerabilities that parents feel, and the very

strong influence of the physicians. This all compromises the parents' ability to come to a decision that is fully informed (Ford 2001, pages 12-13). Ford also concludes that because these elements of informed consent are not usually met:

“In light of the questionable scientific basis behind [the use of surgeries], the lack of follow-up data on its benefits, and the overwhelming evidence of its negative physical and psychological results for many intersexuals [I would also apply her argument to hypospadias], a moratorium should be declared on the use of defenseless infants as the experimental subjects of genital-normalizing surgery” (Ford 2001, page 14).

In the same vein, Tamar-Mattis (2006) finds that too often, the parent is not given time and the wide range of information needed to make an informed decision, so informed consent is illusory (Tamar-Mattis 2006, page 87). I examine her arguments in more detail below (Chapter 7, Legal Issues).

The issue of the parental authority to make medical decisions on behalf of their babies is clearly a charged one. In most cases of medical necessity one would never question parents' rights to consent to medical care on behalf of their babies to do what is in their best interest. Given the history of treatment for hypospadias in particular and genital differences in general, parents and professionals need the time to consider this authority more deeply. Doing irreversible surgery for cosmetic and psychosocial goals when the risks are high and the information is conflicting, may mean that parental authority in these cases is questionable.

Bioethical principles. I now turn to specific principles for making bioethical decisions, and relate these to decisions regarding surgery for hypospadias. Beauchamp and Childress (2012) outline the following principles and concepts for making ethical decisions: the principles of autonomy, beneficence and fidelity, non-maleficence, and justice, and the practice of informed consent.

1. Autonomy. “The autonomous person acts in accordance with a freely self-chosen and informed plan...” (Beauchamp and Childress, 1989, page 68). Obviously babies are voiceless, and parents (or guardians) in collaboration with doctors are usually given the authority to make health care decisions for them. For most medical procedures, parents are the appropriate surrogates and decision makers for their babies, and the standard argument is that parents must be permitted to exercise autonomy on behalf of their minor children, handing over decision making authority to the child once she or he reaches an age of decisional capacity. In the past, hypospadias was considered a medical emergency, and parents were advised to give consent for surgeries while the babies were infants. There are many people who still consider this a medical emergency. But there are now many people who think that this is elective surgery that has many risks, and many today would agree that the early surgeries are controversial. Since hypospadias is not life threatening, since surgical repair might be seen as elective, because the urethra and meatus will be changed irreversibly, it makes sense to grant the boy autonomy over this decision and wait for the boy to grow up so that he can evaluate the pros and cons of surgery. Given the vulnerabilities of parents and babies, waiting would honor the value of autonomy and protect vulnerable parents from making a decision which they might regret. One looks to the process of shared decision making which leads to informed

consent in order to insure the medical necessity of the surgeries, and to insure that the best interests of the baby are protected.

The principle of autonomy would mean that we need to let the baby grow up with genitals in their natural state so that when the genitals are fully developed the boy can make his own decisions and evaluations about surgeries. Since the surgeries we are considering are irreversible; since they are cosmetic reconstructions that can permanently damage the nerves and muscles and cause scarring of the genitals; and since they alter the genitals, the surgeries often limit and sometimes close off future possibilities for the boy's bodily integrity and autonomy as the babies grow into their own. If the child grows up to have different feelings about what the surgeries did, or if the child wants to make his own decisions about his genitals, the early surgeries close off most possibilities. If the child grows up to have difficult complications from the surgeries, his options are limited and his quality of life is adversely affected by those complications. On the face of it the autonomy of the babies is violated.

2. Nonmaleficence . "Above all do no harm" is one popular version of a statement in the Hippocratic oath; another version is "I will... abstain from whatever is deleterious and mischievous." But according to Beauchamp and Childress (1989), "...the Hippocratic oath does express a duty of nonmaleficence together with a duty of beneficence: 'I will use treatment to help the sick according to my ability and judgment, but I will never use it to injure or wrong them'" (page 120).

Since many of the adults who have lived with the consequences of hypospadias surgeries over decades are saying that the surgeries with all the attendant medical attention have harmed them, we need to consider that this surgery is controversial. On the one hand, the standard of care has been early genital surgery, as the American Academy of Pediatrics (2000) has recommended until recently (Baskin, Wilcox and Kim 2015). On the other hand, the 2006 consensus statements from the U.S. and European endocrinological societies (Lee et al, 2006) do state that what the children experienced at the hands of the doctors was trauma (Feder 2014, page 88-89). Physicians might do well to consider that they have held on to the surgical standard of care for too long. Who sets the standard of care and what is the doctors' responsibility if the outcome is poor? Doctors set the standard of care, and if doctors do what their teachers taught them to do, they have been understood to be doing the right thing (Beh and Diamond, 2000, pages 14-17). Beh and Diamond also note that since physicians are not guarantors of positive outcomes, there is no presumption of malpractice from the mere fact of injury.

But what is the doctor's ethical role? Given that unique skills, great experience and being at a center of excellence are all required for good outcomes (see the consensus statement just cited, and the endocrinology textbook by Gardner and Shoback (2011), the doctors have a special responsibility to get the extra experience and training required and to be forthcoming about their experiences and results. Doctors need to be open about the options of refusing surgical interventions, and they need to be open about the limited and conflicting data on long range

outcomes. Physicians need to be open to examining the possibilities that they have held on too long to surgery as the standard of care.

3. Beneficence and fidelity. “Morality requires not only that we treat persons autonomously and refrain from harming them but also that we contribute to their welfare” (Beauchamp and Childress, 1989, page 195). How do parents and physicians decide if the surgeries will contribute to the welfare of the babies? Regarding all genital differences (not just hypospadias), Tamar-Mattis (2006) notes that “national intersex leaders claim to be unaware of any intersex person who is satisfied with surgical intervention, and no intersex person has stepped forward publicly to advocate for surgery” (page 62). On the one hand, it is true that a post-op person will have trouble stepping forward if he does not know or recall that he had surgery and did not need much additional follow-up. He may also prefer to live as an anonymous “normal” person, rather than as someone treated for a highly stigmatized condition. Yet the lack of public statements of satisfaction needs to be considered as an important fact regarding the lack of evidence for beneficence. Regarding hypospadias in particular, there is insufficient evidence (see chapter below on Medical Sources) comparing outcomes and satisfaction among boys who had surgery for proximal vs. distal hypospadias in their later adult years. Given the lack of such evidence, standard arguments for surgery in infancy (such as the value of a boy being able to urinate standing up, not being teased in the locker room, etc.) cannot be evaluated.

One way of demonstrating beneficence is through maintaining fidelity between physician and patient (Beauchamp and Childress, 1989, pages 341-342 ff). “Once having undertaken a case, the physician should not neglect the patient, nor withdraw from the case without giving notice... Abandonment would be a breach of faith, as well as a failure to discharge the obligation of beneficence” (Beauchamp and Childress 1989, page 342). Many adults report in personal conversation that they have increasing lifelong sequelae of the surgeries and that skilled and compassionate medical care to address these outcomes is hard to find. There may not have been any plans for continuity of care. Often this is a problem for several kinds of pediatric surgeries, but professionals and parents need to address this when they consider early surgeries. Note that Douglas Canning (2015) has suggested that this long-range follow through should be a part of the continued care. “Our handoff must be meticulous, because in pediatrics we are cursed with our inability to follow our reconstructed children throughout their adult lives” (Canning 2015, page 285). This topic is beyond the scope of this capstone project, but parents and professionals might do well to note that care over the lifetime might become an increasing need and concern. Yes, this is a genuine concern with many major pediatric surgeries. For those with pediatric cardiac repairs and many other issues it is challenging to find suitable care in adulthood.

4. Justice. Over the recent decades in the United States, minorities and people who were seen as “different” have taught us about the social, legal, and medical injustices they have endured. Through their efforts to gain more of their civil and human rights, our society has been challenged to redress those wrongs. Hypospadias and other genital differences have been stigmatized, and it has taken great courage for people with a history of surgery for such genital

differences to speak out. Our medical practices should incorporate those efforts to reexamine the automatic practice of surgeries and silence as the answer to different genitals.

5. Informed Consent. This is perhaps the critical ethical issue. What is the process of informed consent? Beauchamp and Childress (2009) offer a definition of informed consent in which “one gives an informed consent to an intervention if (and perhaps only if) one is competent to act, receives a thorough disclosure, comprehends the disclosure, acts voluntarily, and consents to the intervention” (page 120). How do parents get their information, what is the conversation between parents and doctors, and who else is invited into the conversation? What are parents offered by way of time to adjust, time to get an education, and how do parents get access to the resources to make this important decision about genital reconstructive surgeries? There are no reports in the materials written by medically oriented writers about the process of giving information to parents.

The consensus statement of 2006, already described, calls for team decision making at centers of excellence. One might note that if there is a center of excellence that has one of these teams, perhaps parents and professionals might be encouraged to consult with them and make use of their expertise through the use of the computer and technology to live stream consults. Otherwise facilities without such teams should be urged to refer patients and families to centers with the appropriate expertise.

The process of informed consent for genital surgeries has been criticized. Writers have described it as too fast and limited in scope. Parents feel pressured to make a decision without getting second opinions etc. Karkazis (2008) also provides examples of the difficulties for parents of getting fully informed as she describes the varying ability and inclination of physicians to involve the parents in decision making (pages 128-129).

Summary. In conclusion, what Ellen Feder (2014) writes about general genital differences (which she refers to with the term intersex) can also be applied to hypospadias. She notes a history of moral failure (page 111), saying

“Two decades after the beginning of intersex activism, physicians readily grant that the means by which children’s bodies have been normalized has effected harm. And yet, because the care of those with atypical anatomies may call into question the good of the goal of medical treatment in this case... the act of ethical reflection has been taken to be at odds, or somehow in tension, with medical practice...It is not only a matter of recognizing that sound arguments laying out the ethical violation that the standard of care entails have not resulted in the fundamental changes one might expect; it is also necessary to try to understand why the history of the medical management of atypical sex anatomies looks increasingly like a history of moral failure” (Feder 2014, page 111).

Given the absence of good research determining whether or not the long-term outcome of hypospadias surgery is acceptable for either distal or proximal situations (see chapter 4 below on assessing medical sources), I believe that Feder’s conclusions apply to surgery for hypospadias as well.

Chapter 4: Finding and assessing medical sources for parents and professionals regarding treatment options

1. Where are the reports of successes? Why are there so many reports of complications and challenges? How possible is the worst case scenario? The question for parents and professionals of whether the surgeries will be successful is clouded by the reports of complications and challenges of the surgeries which might result in some successes, but also result in long term challenges and increasing difficulties for the young man, adult, and the surgeons. What percentage are the successes? What percentage are the surgical complications that might increase over the years? Reports and outcomes differ widely from different surgeons and medical centers. It seems that surgeons' articles are concerned with the challenges that they face getting good results. For a baby with hypospadias, the worst case scenario might be that the surgeries would result in varying degrees of failure. Often surgeons use the phrase "hypospadias cripple" to describe this worst possible outcome. After the disabilities rights movements and after everything that people living with chronic disabilities have taught us, this phrase is offensive. However, it is still used by several surgeons, and parents and professionals need to know that and face the problems that the surgeries might create. Feder (2014) notes that since this term was introduced in 1970 by urologists Horton and Devine, "'Hypospadias cripples' has remained a term of art in the medical literature...". Feder suggests that the continued use of this phrase may mean that practitioners cannot face these surgeries as mistakes. "But if most physicians who continue to recommend and perform normalizing surgeries see these [failed] results as 'regrettable,' they see the problems in the bodies of those affected" (page 204). I think what Feder is suggesting is that there might be the view by surgeons that the condition of hypospadias cripples the baby anyway, and that if the surgeries further cripple the boy, this is all regrettable, but we tried through the surgeries to fix the crippling condition, and sometimes we fail. But regrettably, being a hypospadias cripple is the way the boy was born.

The question for parents and professionals of whether the surgeries will be successful is clouded by the reports of these long term failures. Assertions of success are in the articles by surgeons, but concerns about complications and repeat surgeries which sometimes make the complications worse are there too. The percentages are unclear. The ability to predict who will do well and who will not seems missing from several surgeons' reports.

"The comparatively robust evidence of the success of hypospadias repair has meant that criticism of this intervention has not had as much effect as criticism of other normalizing interventions for atypical sex anatomies. Despite what has become regarded as the routine character of these surgeries, among those who have undergone repair there are also a number of patients who have had repeated and unsuccessful repair, leaving them with significant functional problems" (Feder 2014, pages 203-204).

Karkazis (2008) also deals with the subject of complications from hypospadias repair. Even with all the technical advances, After describing the many complications (page 145, "...some patients suffer from multiple failed surgeries, thus being referred to as hypospadias 'disasters', or worse,

'hypospadias cripples)'), Karkazis goes on to note that "some suggest that surgery creates more problems than it corrects" (page 145). There are adults who need to catheterize themselves throughout their adult lives (Dreger 1999, pages 201-210). "Given that a hypospadiac penis enables pleasurable sexual sensation and orgasm, while surgery risks damaging erotic sensation and creating life-long complications, one may easily wonder why the risks and harms of such surgery may be outweighed by the harms of having a slightly different penis--which from what epidemiological studies tell us, is actually quite common" (Karkazis 2008, pages 145-146).

Preves (2005) also tells the story of a man who had 16 surgeries just while he was growing up. They didn't work, and left him with many urological problems as well as "... scars all over my body from places they've taken skin to plant in to my penis... [My penis] is something that looks like a bunch of skin sewn together with scars all over it that doesn't feel like much and doesn't really work very well... From my point of view, I wish they would have left me alone." Preves also writes, "...chronic complications resulting from surgeries to 'correct' the position of the urethra are so common... that doctors coined the term 'hypospadias cripples' to describe patients who experience ongoing and debilitating iatrogenic, or medically induced, complications as a result of surgery on the urinary tract" (page 31).

In considering hypospadias repairs, parents and professionals need to address those worst case scenarios. It will become clear from the writings of some surgeons that statistics on whether or not a boy will become a hypospadias cripple or live with several complications are hard to come by, but the concerns and the risks are not minimal (see below).

What are the original urethra and meatus like and how do we know if they are functionally optimal? How does a functionally optimal congenital urethra and meatus compare to the reconstructed urethra and meatus? This question was not addressed clearly in any of the articles that I read. Yet it seems to me the heart of the medical question. Parents and professionals also need to ask if the original congenital urethra and meatus function well. I have no medical training but I want to offer a guess. My best guess is that, for the majority of hypospadias cases, the congenital urethra and meatus of a baby boy born with hypospadias do function well. If the majority of the congenital urethras did not function well, perhaps urological problems would develop during the first months of the babies' development. None of the surgeons' articles mentions this problem. When the surgical plan includes closing that congenital urethra and meatus off in order to create a new one, complications that may arise are often significant and lasting. So for a hypospadias boy with a urethra between the testicles, the question is this: if that urethra is working well, and if the risks of later fistulas, urethral diverticula, and meatal stricture are high, then isn't it better to leave a good, working urethra in place even if it is between the testicles?

Perhaps one might characterize this penoscrotal urethra as in the female position (or feminine in style), and perhaps there is deep resistance to leaving a female-type of urethra on a boy. Feder refers to philosopher Martha Nussbaum who writes about disgust as a factor in driving decisions about treatment (Feder, 2014, pages 75-76). Therefore, parents and professionals need to think through how they feel about having a urethra that is in the female position on a

boy. They need to consider that perhaps it is medically safer in the long run to leave the natural urethra there, accepting the idea that the boy will need to sit down to urinate (see page 31 below for the assertion by an experienced hypospadias surgeon who said that the natural urethra works better than the man made urethra). With some deep thought and counseling or mentoring from adults with hypospadias, parents and professionals might need to change their attitudes and deep reactions toward the male culture of standing to urinate, or find strategies that help the growing boy to deal with the culture of the men's rooms, rather than run the risk of lifelong urological problems or catheterization in adulthood if the surgery causes urological problems. I grant that this kind of change is not easily accomplished but the current evidence does not support the assumption that surgery is preferable.

People who exhibit differences are certainly vulnerable, but in considering their lifelong urinary health, perhaps that anxiety over the psychosocial consequences of this physical difference should not drive a medical decision. Perhaps in the case of hypospadias it is better to work around the difference, balancing the need for privacy and dignity, until the young man is better equipped to deal with the social consequences.

The Intersex Society of North America has a site on their website called Hypospadias: Parent's Guide to Surgery (<http://www.isna.org/hypospadias.html>). The site presents a clear presentation of the issues that parents and professionals need to know, including a definition of hypospadias and how it will affect boys and men. Their approach is that hypospadias is a cosmetic difference, and that even though doctors recommend surgery,

“our discussions with men who have had hypospadias surgery lead us to believe that the physical damage and emotional trauma of genital surgery are frequently far worse than the hypospadias itself... A hypospadias penis is entirely capable of pleasurable sexual sensation and orgasm. Plastic surgery on the genitals damages erotic sensation; it cannot improve it. There are some conditions however, which may require surgery to save your child from pain or illness, such as chordee... exposed mucous membrane, or adhesions.” This site notes the many complications from surgeries that damage sexual function, and addresses the embarrassment of young men who discover this. This site notes that the embarrassment often prevents them from discussing this problem or seeking help. This site also describes the difficulties of creating a new urethra, the advantages of having a natural urethra that works, and the reality that it may be preferable to accept sitting in order to urinate, and to get counseling, even for the whole family, in order to deal with the emotional side of having a different penis. This site does suggest waiting for the boy to finish with puberty, when his penis has grown to its mature size, before considering the risks and benefits of surgeries: “...the surgical prospects are improved when working with a mature, full grown organ...”

This will be a helpful source for parents and professionals.

2. Current lack of consensus regarding standard of care. What are the surgeons writing in their articles? I will now look at several of the writings that come from medical sources. Obviously, parents need this information to make a fully informed decision, to educate themselves about

their child's congenital conditions, and to understand what the surgical options might be. These writings inform their medical team, and help parents understand and weigh the risks, benefits and long range outcomes. Perhaps through getting all this information, they also go through a process that helps them to adjust to the child that they have. Yet these writings are mostly in medical journals, and are written for physicians. Parents might need extra help to access these articles.

The surgeons are consistently concerned with their complications, and seem unable to predict which cases will do well and which will need multiple re-surgeries. Articles might start with the goals of surgery, but they often don't report clearly if the goals of surgery are mostly achieved. There are often internal contradictions and often clear differences in approach, so that parents might be left with more questions than answers. I wish I could report that some surgeons report clear successes, but I find that even if a surgeon reports some success, it is unclear from these articles if the surgeries will achieve their goals, or if the psychosocial concerns of parents will be addressed by surgery. My goal here is to review the selected articles that I have that are about hypospadias, see if there are clear guidelines, and note the internal contradictions and red flags of concern about long-range outcomes, in order to see if the surgeons tell a clear story about how the surgeries will address or solve the long-range concerns that parents have.

I begin with two sources concerning hypospadias, namely, Greenspan's 2011 textbook on endocrinology and an UpToDate for physicians about the management of hypospadias (Baskin, Wilcox and Kim, 2015, accessed on line November 13, 2015). A comparison between these two sources perhaps illustrates clearly the reality of the fact that there are differences in approach to the management of hypospadias. Greenspan's text book (Gardner and Shoback, 2011) was written in 2011, before the UpToDate material. The endocrinology textbook suggests that the standard of care for managing hypospadias demands a multifaceted team in order to provide the ideal practice as outlined in the 2006 Consensus statement that has already been discussed. The endocrinology text suggests that the parents go through a process like the shared decision making process that I have already described. The Greenspan textbook suggests treating hypospadias as a DSD (disorder of sex development) and talks about the complexity of a DSD (page 519). Given this complexity, management is "...best undertaken by a team consisting of a pediatric endocrinologist, psychiatrist or psychologist, pediatric surgeon or urologist, social worker, religious counselor if appropriate, and an informed primary care physician at a center of excellence" (page 519). Introducing the subject of the management of DSDs, the textbook goes on,

"The parents need to be presented with the whole picture diagnosis... possible medical and surgical therapies, risks, complications, and unknowns. It is critical that they have the knowledge and time to make an informed decision about any surgical procedures on their child... surgical reconstruction done in infancy may need refinement at puberty.

Surgery, if done must emphasize function rather than cosmesis. It should be performed by a surgeon who is experienced in the operative procedure and is cognizant of the importance of preserving the functional integrity of the genital area... The timing of any surgery remains controversial. This is a decision that needs to be made after careful

discussion with the parents. It should be noted that there are no outcome data for delaying functionally cosmetic surgery on an infant with ambiguous genitalia until an age of consent as suggested by some psychologists and patient advocates” (page 507).

From this endocrinology textbook, a parent would learn that there should be a multifaceted team approach to giving them information, as well as counseling and time to learn and adapt and consider all the options. Parents learn that surgery done while their child is an infant might need refinement during puberty, and that surgery has controversial aspects regarding timing (that is, how old the child should be) and preservation of function. They learn that they need an experienced surgeon, and that if they want to wait until the child grows up to consider surgery, there are no long range data, and that even without this long range outcome date, patient advocates and some psychologists suggest waiting.

The UpToDate (2015) material, on the other hand, which was written 4 years later and seems like a practical guide for physicians and surgeons, makes no mention of using a multifaceted team to work with parents. The UpToDate material assumes that surgery is the standard of care. UpToDate does not mention a team approach, nor does it mention giving parents time to consider risks and options. The authors write, “...a pediatric urologist should be consulted to determine the severity of the hypospadias, and review with the parents the clinical consequences of uncorrected hypospadias, and the need and potential timing for surgical correction (page 4). UpToDate continues,

“[patients with uncorrected mild hypospadias] may not require surgical correction... Patients with uncorrected hypospadias may develop anxiety regarding sexual performance and dissatisfaction in the appearance of their penis... The American Academy of Pediatrics recommends that genital surgery be performed before 18 months of age... General consensus within the pediatric urology community... is to perform surgery at six months of age in full-term healthy infants” (page 5).

Further, UpToDate reports that there is no consensus on what the best surgical approach is for correcting severe hypospadias, that the complication rates remain high regardless of the surgical techniques used, that further studies are needed to address consistency within the pediatric urology community, and that the postoperative care is complicated, with parents often left caring for their child on their own as they are discharged quickly (pages 5-7). The complications are also listed (pages 7-8) and after listing these complications, the authors conclude that “surgical correction generally results in an acceptable cosmetic and functional repair of the penis, and satisfactory genital self-perception, sexual performance, and fertility” (page 10). Parents learn here that they need to consider surgery before 18 months, and often during the time between 6-18 months; and that uncorrected hypospadias will cause their child anxiety about sexual function and appearance. Further, according to these authors, even though surgeons cannot agree on best techniques, and are accepting of complications, they are pleased with the results of their surgeries, and they find the results are acceptable.

UpToDate supports the use of surgery within the first 18 months of life as the standard of care that urologists use and that is advocated by the American Academy of Pediatrics that has

prevailed for decades for hypospadias treatment (and for treatment of DSDs in general). Does this reflect the approach of the majority of surgeons and physicians? I have to assume that it does represent the majority approach to treatment. In contrast, the endocrinology textbook (Greenspan 2011) reflects a new standard of care as proposed by patient advocate groups and the Consensus Statement of 2006. Parents might not know that they are not getting the best standard of care if surgeons don't know about the recommendation for delay and for team decision. Parents need to get this information in the first place so that they can evaluate these two approaches and decide whether to look for the shared decision making process.

3. Medical guides for parents and clinicians to use together. There are two guides that I found for parents making this decision. One is a book called *Disorders of Sex Development: A Guide for Parents and Physicians* (Johns Hopkins, 2012), co-authored by two physicians and one research psychologist. It is an accessible read and a basic introduction for parents, but it does not give in-depth information about pros and cons of surgeries. The authors readily note that surgeries are controversial (Wisniewski, Chernausek, and Kropp 2012, pages ix-x). In eight chapters, one gets a basic education in DSD's and their possible treatments. The footnotes are not extensive at all. For the chapters on treatment options and educating children (Wisniewski, Chernausek, Kropp 2012, chapters 5 and 6), there are no footnotes at all. Regarding genital surgery, this guide writes that "it is not required to protect a child from serious health risks. However, many parents choose to proceed with early genitoplasty because they want to spare their children the teasing or embarrassment that they might experience due to their genital appearance or for technical reasons related to the planned surgery" (Wisniewski et al 2012, page 66). On the other hand, "some parents... decide to forgo early genitoplasty for their child, allowing the child at an older age to make his or her own decision about whether to proceed with these types of surgeries. It may not always be in the best interest of your child to have early genitoplasty." When talking about early surgeries for boys, they note that parents need to be aware that "severe hypospadias often requires multiple surgeries... and may be associated with problems such as scar tissue formation" (page 71). In the summary they write that "sometimes no treatment is in the best interest of the child, other times it is warranted. Use this book as a guide to work with your health care team" (page 71).

These statements are good introductory statements. These statements hint at the controversy surrounding surgeries as treatment, and point the way to alerting a parent to the complications that might necessitate follow-up surgeries in the teen years. In my opinion, these statements are too neutral to give clear helpful guidance—yet they open the door to ask the questions about long range outcome studies, and about whether or not the surgeries are elective or medically necessary. In this same book, indicating that it might be hard to talk about all the issues that parents have thinking through medical care with many of the diverse experts that they need on a health care team, the writers note that there should be "at least one person on the team of caregivers who is good at interacting with parents, and this person will be the one you turn to most often when you need information" (page 48). With a diverse team of experts, one still needs one member of the team who is good at communicating. Are the writers suggesting that the team members don't have time to communicate fully or that they won't be equally available for questions and equally responsive to the patient's ongoing needs?

One of several reasons for not doing early surgeries is that communication and collaboration with doctors is sometimes hard for parents and their growing children, and that if a boy becomes needy due to the complications that are reported as routine, it does become harder for the physicians and nurses to address parental concerns on behalf of their children. We have also noted the embarrassment that many boys and young men feel in dealing openly with their genital differences. In later articles, the surgeons themselves will note that there are routine long-range complications, and that despite successive surgeries it is hard to solve those problems. Through reviewing articles below it becomes clear that there are frequently long-range issues of concern. Canning (2015), discussing hypospadias surgery, suggests that continuity of care over the adult lifespan is a significant concern. For example, when men need special follow-up care, they often find few surgeons and medical providers who can help.

Chapter 7 of *Disorders of Sex Development: A Guide for Parents and Physicians* is about the long term health of people with genital anomalies. There is no mention of the re-surgeries that are common when there are complications from the original surgeries. I cannot imagine that repeat surgeries don't have an influence on overall medical health for the boys and young men. Yet this risk is not addressed at all. Some assertions in the chapter about medical treatments simply say that long-term health outcomes could be good or could be bad, and recommend further discussion with the health care provider. In general this book is a good starting place, but contains many internal contradictions and questions that need addressing, and contains no footnotes to back up the most important assertions and ideas about surgeries in particular.

The second guide for parents facing decision making about genital surgery for their children is about the ideal process of shared decision making (Karkazis, Tamar-Mattis and Kon, 2010). It describes the process of shared decision making and contains an appendix which includes all the questions that parents and health care professionals need to use and answer in order to go through the process of getting clear information and understanding the controversies surrounding surgeries and other treatments. It reflects the work of the bioethics and legal advocates. Because it is in a medical journal I include it in this section. Perhaps its inclusion in the medical journal gives the process of shared decision making more support and exposure to the clinicians who might want to develop the process and increase the implementation of shared decision making. Yet because it is in a medical journal, it may not be easily accessible for parents. The questions presented are excellent, including issues around diagnosis, prognosis, rationale for proposed procedures, risks and benefits, alternatives to proposed treatment, post-surgical care, clinician experience, referrals for future support, special cases, and pitfalls to avoid.

I would add the following questions for parents to consider. Is the hypospadias surgery elective or medically necessary? What are the medicines that will be given, and what are their side effects? What are the effects of anesthesia (see Rappaport et al, 2015, on anesthetic neurotoxicity), antibiotics and other medicines on young children and their developing bodies and brains? Will any tests be given to determine sexual responsiveness? When and how are they given, what is their purpose and can they determine long range sexual responsiveness? Will any teaching be done or will photography be taken at any time during this process? Who is in the operating room who assists during surgery, and is this a teaching opportunity? Will there

be grafts? Where are the donor sites? Are there any side effects or long term effects on the donor sites? How do the donor sites heal and are there any long range outcome studies about the safety and effectiveness of those grafts? During the recoveries, what support will the patients and parents be given? When there are complications, how are they addressed? Will parents be given all medical records as soon as each treatment takes place? Will those records be complete? What if parents live far from a center of excellence? How will follow-up issues be addressed? Regarding urination, does the boy have a natural, congenital urethra and meatus that work well? If the hypospadias is penoscrotal, how do the involved parties feel about boys having a urethra that is in the girls' location? Will the urethra and meatus be changed or altered in any way? Does the boy have sphincter control now, before surgeries? If reconstructive surgery is planned that affects the urethra and meatus, will sphincter control be as good as the natural urethra or meatus? Looking very long range, given the widespread problems adult men have with prostate issues, if they change the urethra, how will that relate to long range needs and prostate issues that so many adult men have? When the boy grows up, and the original team has changed perhaps even many times, how will continuity of care be arranged over the boy's lifespan? In the event that the family or later the grownup son moves to other locations, can he access that care easily at any time he chooses? It may be unreasonable to ask such long range questions, but given the stories of the adults who have increasing difficulties in their adult years and find it difficult to get medical treatments, parents might want to explore this long range view before taking the risks of surgeries.

This article gives the impression that the special multifaceted team that gives parents time to get information, collaborates with them in that process and believes in the shared decision making process, might be seen as the current standard of care; and indeed should be seen as the new standard of care. Yet shared decision making as the new standard of care is an ideal that is very rare in reality. This becomes an ethical lapse in care (Dreger and Sandberg, 2010, page 153). The first thing a family needs to know is that if they are not getting input from a wide team, if they are being pressured to make decisions before they are ready, then they are not getting the ideal standard of care. The first thing that doctors need to explain to a family is that the issue of early surgeries is very controversial. I hope that physicians would work to create this ideal team to work extensively with new parents, and that parents and patients and health care teams could access this team from any distance as needed through technologies that the computer and the internet make possible.

4. From hypospadias cripples to the challenges of hypospadias repair—surgical controversies. Even though articles by surgeons are not written for parents, some parents might want to know what surgeons are telling each other about hypospadias repair. I was hoping to find some clear explanations of the benefits of surgery. Though there were statements that there were successes, as I read the articles it seemed to me that there were significant risks. One might even say that there is confusion about the benefits and risks.

In this section I review selected articles written by surgeons and physicians about hypospadias repair from 1981 to the present. The focus of the articles seems to be the challenges and complications of the surgeries. The surgeries often create so many problems that they end up

with what several articles describe as “hypospadias cripples.” This term was used in an article entitled “Hypospadias Cripples” (Stecker, Horton, Devine, and McCraw, 1981). The authors, a team of physicians, write, “The term ‘hypospadias cripple’ certainly applies to these patients who are trapped in this surgical maelstrom in which every operation may in fact, make matters worse” (page 539). In the editorial comment, “Avoidance of the creation of a ‘hypospadias cripple’ should be foremost in the minds of those surgeons who perform hypospadias operations only occasionally. How nice it is to have a place like Norfolk to which to send the disasters. I am certain the patients wish they had come sooner” (page 544). There is no suggestion in this article that these problems only occur in a small number of patients, or that there are a majority of success stories and that therefore the benefits outweigh the risks. There are no suggestions about what to do if you do not live near Norfolk, Virginia. Parents might wonder if their surgeon was trained in Norfolk, and what to do if their surgeon was trained somewhere else.

Articles often suggest that there have been many advances in techniques over the decades. But a parent wants to know the risk of creating a “hypospadias cripple”. Are the risks significant? Are there predictors? Have new techniques solved these problems? In 2015, a book was published which includes two articles by surgeons Gina Cambareri and Moneer Hanna (Cambareri and Hanna, 2015; Hanna and Cambareri, 2015) called “Approach to the Adult Hypospadias Patient” and “The Adult Hypospadias Patient: Technical Challenges in Adulthood.” These chapters by Cambareri and Hanna could also be said to be referring to hypospadias cripples. They express a certain hope for a good short-term outcome, but then they report on several long term problems.

“Modern techniques report an almost 90% success rate but often have short-term follow-up... The high success rates often cited for various techniques are virtually impossible to report with certainty considering the fact that late stage failures are well documented and reported in the literature... follow-up studies after sexual maturity has occurred are very limited and criteria for ‘success’ have yet to be defined... It is far more challenging to work with proximal hypospadias than with distal” (Cambareri and Hanna 2015, page 35).

The long term prognosis for complications is not good, should a parent again wonder if the risk of creating a hypospadias cripple is high or low. “Failed multiple attempts at hypospadias repair often leave the patient with a penis that is scarred, hypovascular, and shortened” (page 36).

“Long-term outcomes in patients with a history of proximal hypospadias are scarce... It is important to note that surgeon perception may be different than patient perception regarding successful outcomes” (page 40). They cite another author who notes that “overall patients were less satisfied compared to the surgeon with overall genital perception...” (page 40). Further, “This study highlights the importance of patient-reported outcomes in long-term studies related to penile surgery... For primary hypospadias repair, a near-perfect functional and aesthetic result represents a successful outcome and is indeed achievable for many using modern techniques. However, for the minority who require multiple surgeries, which compromised the quality of the genital tissues, the outcome can be severely disabling and the patient is required

to accept a lower standard for success... Progress in hypospadias surgery will require long-term patient-reported outcome studies to better determine how the patients fare in adolescence and adult life” (page 41).

Might parents wait until their son grows up, for techniques to continue to improve, and the young man can participate in the decisions regarding surgical treatments and interventions? (also see discussion of Hensle et al, page 31 below). This may be a good idea for some, yet there are still many technical challenges for the surgeon of adults with hypospadias. Hanna and Cambareri (2015) might help in understanding this option, in this article two urological surgeons discuss the technical challenges that surgeons and patients face when the surgeries take place on adults. They looked at adults who have not had surgeries who might want to try to normalize their genitals. They also looked at adults who had surgeries who need repairs of the original surgeries. The title of this chapter, “The Adult Hypospadias Patient: Technical Challenges in Adulthood” (Hanna and Cambareri, 2015, pages 77ff) does not suggest that there are successes as well as challenges. After concluding that better long term outcome studies are needed to make better progress in hypospadias surgeries, they summarize this chapter by saying

“Primary hypospadias repair after adolescence is feasible, but subject to higher complication rates compared with repairs in childhood... 10-30%... Repair of secondary complications in adulthood present a special technical challenge and are subject to complication rates ranging from 35-75%, particularly for the ‘hypospadias cripple,’ who presents with penile scarring and a dearth of tissue for repair” (page 94).

There are no descriptions of the successes, no sentences suggesting reports from patients of their satisfactions at having had the surgeries. Should we attribute this to the way that medical articles are often written about surgical outcomes? For a parent these are obvious questions. What are the normal complications following surgeries? If the complications resolve themselves, how do they resolve themselves? What is it like to fall into the trap of the surgical maelstrom mentioned both in the 1981 article about hypospadias cripples and suggested by the 2015 article in Wood and Wood? Should a parent try to repair distal hypospadias because it is less challenging for the surgeon? Are there complications following distal repairs that are acceptable and easy to live with? For proximal hypospadias, the more severe form, are the complication rates of 10% or 35% an acceptable risk? How often are the complications so great—30% or 75%—that there are a large number of ‘cripples’? Where will a child fall on the spectrum, and is it worth it to try to normalize the genitals? Or is the operative word “try”—is it worth trying to get a good result? If this is elective surgery, are there other examples of elective surgeries that create as many problems but are still worth the risks? What is it like for a man to live with these complications? Would men with hypospadias prefer their original urethra to the neourethra? Indeed, in a private conversation in the spring of 2015, an experienced hypospadias surgeon said that the urethra that nature made works well, and that the manmade urethra is not going to work so well. If you have long range problems voiding, isn’t that a significant health risk? If fistulas and diverticula are common complications, are both voiding and sexual function compromised? How does a parent evaluate the long range risks and benefits?

The article from 1981 seems to be echoed in the articles from 2015. What articles have been written between those years, and do they address any of the issues that usually concern parents? Here is a selection of articles. Is there long range information about the successes? Is there long range information concerning living with the complications of hypospadias repair?

For parents wondering how their child will fare in their adult years, there is little information from articles written by surgeons. Indeed none of the studies follow a significant number of adults who are ages 40 and over (for example, Bubanj, Perovic et al, 2004, page 1876, had an age range of early 20s to early 40s, with the mean age of 27). In conversations at a hypospadias support group (July 2015), adults suggested that many affected adults do not begin to grapple with the issues raised by the early surgeries until they are in their 40s and older. Also, the internet has become a way for many to begin to relate to others with similar conditions, and the internet has become a source for medical knowledge. But these adults grew up before the internet, and vary in their adaptation to use of the internet as a source for information and connection.

There could be many reasons for dealing with their medical issues later in life. Perhaps they were not given the full information about their medical histories, perhaps they were satisfied with their bodies, perhaps they just understood their differences or struggles as an inescapable part of their lives and felt that they could not share this information with others.

Embarrassment, shame, privacy and secrecy must be considered as reasons for not dealing openly with their medical histories and needs. Perhaps they did not know that many others shared their medical histories. Perhaps all the medical interventions made them either hardened to the emotional and/or physical sides of the issues, or perhaps they have been so sensitive that they do not want to be public about their personal medical histories. Even though our society seems to deal with sexual issues openly, it takes unusual courage to discuss genital differences openly. Perhaps after repeated attempts to address any concerns with doctors or health care providers, patients became frustrated by the lack of solutions or the difficulties of finding effective and compassionate care. All these possibilities are described in the literature by the adults who had the surgeries and their advocates. Their books and articles are discussed in the chapter later in this paper about their voices, and are also listed in the bibliography.

Many of the doctors' articles do not seem to explicitly ask some of the other general questions that a parent would be interested in. What do patients remember about their surgeries and medical care? How were the boys prepared for the surgeries? Did the boys need follow-up care, and how was that addressed? Do the boys and their families have a team of specialists that they can turn to when they wish? Would they recommend surgeries to others? Would the adults have surgeries done on their own child?

Research suggests that there is a difference in people's attitudes regarding doing surgery on themselves, compared to doing surgery on their children. For example, Suzanne Kessler, a professor of psychology at Purchase College, did an experiment with her students (Kessler, 1998). After explaining the risks and benefits of genital surgeries to college or graduate students, and then asking if they would have wanted their parents to agree to give them

surgeries, the overwhelming majority said that they would not have wanted surgeries for themselves. When asked if they would give their children surgeries, the overwhelming majority said that they would give their children the surgeries (Kessler 1998, pages 100-103; also in Feder, 2014, pages 44-45).

I now review a series of articles addressing hypospadias repair, going in chronological order. In the approach of Mureau, Slipjier et al (1996), a group from the Netherlands, correcting hypospadias is necessary to ensure voiding in a standing position, unhampered sexual functioning as an adult, and a cosmetic appearance that is as normal as possible (page 703).

But there is no agreement about how to assess satisfactory results, and the surgeons are more satisfied than the patients (page 704). Surgeons often judged the cosmetic results as excellent, yet patients may often judge the cosmetic results as poor (page 705). Their data looks at up to nine years from the original surgeries and notes that several patients had repeat surgeries (page 705). They note that the patients need to be followed into adolescence, but that complications often become more apparent many years after the surgeries (page 705). So for a parent who wants to know how these surgeries last over the lifetime, these surgeons are saying that they don't have the long-range data. A parent might note that there are questions about long range complications that are not addressed.

In 2001, a group from Columbia-Presbyterian Medical Center (Hensle, Tennenbaum, Reiley and Pollard) wrote an article about hypospadias repair in adults, calling the article "Hypospadias repair in adults: adventures and misadventures." They reviewed the records of 42 men, ages 18-47 (mean age 22.5), who had repairs for hypospadias between 1979 and 1999. Looking at a twenty year period, they reviewed charts and did urine cultures and other medical assessments in preparation for surgeries, including creating "artificial erections" (unexplained) for the majority. They do not report conducting any interviews. There were several surgical techniques used for the original repairs and subsequent repairs. They divided the 42 men into 3 groups: 8 had never had surgeries to repair the hypospadias, 12 had one or more procedures as children and were easy cases with relatively intact tissue, and 22 had histories of multiple unsuccessful hypospadias repairs. They report a significant number of complications for each group. For the 8 that never had surgeries and had surgeries as adults, there was a 37.5% complication rate.

For the 12 that had one or more surgeries in childhood but had tissue relatively intact, they had a complication rate of 41.67% after follow up surgeries. For the 22 who had multiple unsuccessful hypospadias repairs, there was a 63.6% complication rate after follow up surgeries (Hensle, Tennenbaum et al 2001, page 77). Many of the children (22 out of these 42 cases) had unsuccessful multiple surgeries as they grew up, yet Hensle et al conclude that "Hypospadias repair is best done in early childhood, usually at one operation and with few if any complications."

A parent needs to question how the surgeons can continue to assert that hypospadias repair is best done for young children when the number of complications and even failed attempts is so high. There are many surgical techniques to use, and outcome numbers seem inconsistent. For example, if the repair was a tube graft, complications developed in 13 of 53 cases, with failure in 16 of 53 cases (Hensle et al 2001, page 78). Regarding tube graft, "These less than perfect

results from a group of highly skilled and extremely experienced hypospadias surgeons raise some concern and provide some insight into the problems we face when treating the adult hypospadias population... Despite our overall 88.1 % success rate, a significant number of complications developed [in each group]" (page 78). With all these complications and multiple failures, how can they conclude an 88% success rate in any of these groups? They are not talking about simple complications. I think that the surgeons who write these articles understand that the complications for hypospadias repair are routine but serious enough to need re-do surgeries and the risk of increasing the seriousness of the complications grows with each surgery. See Bubanj et al below. I do not believe that the many reports by surgeons and adults who had hypospadias surgery in childhood, that reported complications and re-surgeries to address fistulas, diverticula, and dribbling, can be considered short-term post-op complications, nor can such surgeries be considered a good final resolution of the initial problem.

Looking at this list of complications, differing techniques, and the admission of the technical challenges for excellent surgeons, how is a parent to decide to try to get a good outcome when the risks of surgeries are so significant? Parents need more elaborate and thought-out explanations of the advantages of early surgeries when so many are unsuccessful. There are even more challenges for the adults who did not have early surgeries and try to address their hypospadias after growing up. In general, "Adults undergoing hypospadias repair must be counseled on all of these variables to avoid unreasonable expectations" (Hensle et al 2001, pages 77 and 79). As a parent, one needs to know if the surgeons can predict who will do well and who will not, and what the complications will mean over the decades for urological, sexual and psychosocial functioning. This article cautions parents to not have unreasonable expectations, but does not spell out how the complications will affect their children over their lives.

In the 2004 study by Bubanj, Perovic et al from Serbia and Montenegro (cited above in a different context), the authors looked at sexual behavior in 37 men who had surgery for hypospadias, compared to 39 men without hypospadias. Thirteen of the 37 men who had hypospadias surgeries had ejaculation difficulties, yet the authors concluded that sexual function of patients who underwent surgery for hypospadias in general was not affected. They suggest follow-up and counseling extending into adult life for hypospadias men (page 1876). Specifically, "Ejaculation difficulties (spraying or dribbling of ejaculate) were reported by a third of patients. This problem is common after hypospadias surgery as reported by others. Unfortunately, there may be little room for improving this inevitable consequence of reconstructing more proximal forms of hypospadias..." Yet, in the same paragraph, "We concluded that the sexual function of patients with hypospadias in general is not affected" (page 1878).

Any thoughtful reader would be confused. One third have ejaculation difficulties after surgeries for proximal hypospadias, those problems are inevitable, yet sexual function is not generally affected? This contradiction is left unexplained. Also, they are comparing men who had hypospadias surgeries with men who do not have hypospadias. It would be more helpful to find

the adults who had hypospadias who have not had repairs and compare their issues with both the non-hypospadias group, and with the groups that had hypospadias repairs. A significant number of the adult advocates said that they wished they had never had surgeries. How would that have served them over their lives? Parents need to know the answers to that if at all possible. Bubanj et al (2004) conclude that

“Only half of the patients with hypospadias were completely satisfied with their sexual life. This finding contrasts with the results of earlier investigations indicating that the majority of men with hypospadias lead an adult sex life that does not differ greatly from men with normal genitalia... There is room for surgeons to further improve surgery so that patients with hypospadias are closer to normal... In our series we did not find that multiple operations and age at first and final surgery showed significant impact on sexual behavior, but this finding may be the result of the relatively small number of participants in our series. Additional investigation is warranted” (page 1879).

However, in this study they excluded some of the complicated cases because “long-term outcomes of certain techniques are never as good as would be expected... [some procedures are no longer applicable due to changes in techniques]. Long-term sexual outcome of new techniques for hypospadias reconstruction is anxiously awaited” (page 1879).

The parent reading this learns that there are many surgical techniques that change, that these researchers do not want to include the difficult cases in their research, that surgeons still need to learn better techniques, that long-term outcomes of certain techniques are not as good as expected, that studies differ in their conclusions about the patients’ long term satisfaction, and that for proximal hypospadias (the more serious kind of hypospadias) one third have problems with ejaculation and those problems are inevitable. A parent might wonder, if their child has a procedure that seems like an excellent idea today, whether that procedure will be determined unsuccessful in the future? This is crucial information that a parent needs to know. Since the techniques keep improving, might parents do better to postpone surgeries until techniques develop even further? A parent might conclude that waiting until the child grows up has distinct advantages, because there is time for the surgeons to get more experience and hopefully new techniques which will address the complications and challenges of doing the surgeries on adults.

A parent might also wonder why sexual functioning is often studied, but general urological functioning is often not questioned, perhaps rarely studied. I found one article that does ask this question, notes the sparse data on urological outcomes (Moriya, Kakizaki et al 2007).

Someone who has problems with voiding has ongoing problems which could affect medical health. Someone who has sexual distress has a problem which affects quality of life, but might not be as significant a health risk as general urological problems, from the medical perspective.

Nuininga, DeGier et al in the Netherlands wrote an article in 2005 about “Long-term Outcomes of 1-stage hypospadias repair.” Again this study clearly did not deal with the difficult cases. They limited the severity of the cases for this study, noting that “Recurrent complications were not counted” (page 1545) and certain techniques were not studied. “No free graft, bladder or buccal

mucosa was used in our series and, thus, the severe crippled cases requiring multi-stage procedures were not included in our study” (page 1546). They list the complications—necrosis, wound dehiscence, meatal stenosis, urethral stenosis and fistulas—that need reoperation (pages 1546-7). They note “that there can be a long interval between the operation and presentation of the first complication” (page 1547) and that the long-term complication rate was 54%. The complication rate that they note seems higher than in other studies, suggesting that this might be because they study patients for up to ten years when others study patients for up to five years (page 1547). Perhaps these authors include small complications in their count, complications that other studies might ignore, leading to the higher long-term statistics. However, this possibility is unknown. They note that the more complicated the procedure, the more complications will occur (page 1547), that these results might only be the tip of the iceberg, and that as surgeons get more experience, they might expect better cosmetic results (page 1547). They conclude that “Even in the hands of experienced pediatric urologists the complication rate of hypospadias repair remains high, with an average of 54% at long-term follow up in our series. The results show that additional complications can present after a long postoperative interval. Therefore, final outcome of hypospadias surgery can only be evaluated once the patient reaches adulthood” (page 1547). These writers follow up their patients until the age of 5, asking their patients to come back at ages 10 and 15 (page 1547). This article does try to compare technique of repair with outcomes. But with the suggestions that as techniques change, several techniques were excluded from the study because the patients became severe hypospadias cripples requiring multi-stage procedures (page 1546).

A parent reading the above article sees no studies on adult men in their 40’s and older, a long list of complications, a high percentage of complications, reluctance to study the very complicated cases, several procedures that might cause severe crippling, and the note that even in the hands of very experienced surgeons, there is an uncertain outcome long range. If so many techniques cause severe crippling, how do the surgeons address the needs of the patients over the life span?

Schober’s (1996) article raises some important concepts that are applicable to all genital surgeries, including hypospadias. She refers to truth telling, referring to the fact that often there was a history of concealment between doctors, parents and patients. To raise this in 2006 means that perhaps concealment is still an issue in genital repair. Are there doctors who are not fully disclosing all the information that parents need? The idea that early surgeries make doctors and parents more comfortable is a critical one to discuss more fully. Surgeries are stressful, so perhaps the parents need to be interviewed to see if they are getting the comfortable feeling that Schober writes about here. Such an interview would elicit parents’ expectations and hopes. Follow-up interviews should also take place to see how parents respond over time and to see if their responses change or are consistent. It would also be valuable to interview doctors in order to learn whether the physicians see the high complication rate as routine, as indicated by several articles cited here. Since Schober writes that early surgeries make doctors more comfortable, another valuable question that can be put to surgeons in such interviews would concern reasons for their comfort, given the rate of complications. Physicians may indeed see the complications as routine, and that living with

them becomes normal for the patients. Are the complications addressed and solved by more interventions or by more surgeries? Schober observes that parents need counseling even if they are not going to plan surgery.

To review, the question about impaired sexual functioning in adulthood needs much more exploration. Schober notes that the wish for patients to choose their surgeries themselves may not favorably affect the outcome. Is she suggesting that complications are so frequent that even if a patient is older and participates in the decision making, the outcomes might not be more favorable? Further, is she suggesting that instead of parents shouldering the responsibility for making the decision, the older patient is now shouldering all the responsibility? Is she saying that the outcomes will be the same no matter when the decision for surgery is made and who makes the decision? Doctors often counsel that the baby will not remember the surgeries, but with all the complications and revisions, there is trauma for the babies and for the families. When there are revisions in later life, these early traumas can be reactivated. Schober is open about the high likelihood of revisions, and the lack of evidence that early surgeries provide a better outcome. The technical challenges are great at any stage for these surgeries, and it is clear that babies heal better than adults, but is it unwise to mistake this short range idea of healing with the long range issues? And what are the effects of anesthesia (see Rappaport et al, 2015), antibiotics, pain medications, and prolonged recoveries that need specialized care and attention?

Here I want to describe the Consensus Statement in more detail. In 2006 *Pediatrics*, the official journal of the American Medical Association, published a Consensus Statement on the Management of Intersex Disorders. With a committee of 50 international experts it included only one intersex activist, Cheryl Chase. The International Consensus Conference on Intersex- referred to as the Consensus Statement- was seen as a watershed for the treatments of DSDs. In line with the Greenspan textbook (see page 22 above), I include hypospadias as a DSD.

Regarding the guidance that a parent might get for decisions relating to surgeries, they wrote that “the pattern of surgical practice in DSD is changing with respect to the timing of surgery and the techniques used. It is essential to evaluate the effects of early versus late surgery in a holistic manner, recognizing the difficulties posed by an ever-evolving clinical practice. The consensus has clearly identified a major shortfall in information about long-range outcome” (Lee, Houk et al 2006, page e496). The Consensus Statement advocates a team including pediatric subspecialists in endocrinology, surgery, and/or urology, psychology/ psychiatry, gynecology, genetics, neonatology, and if available, social work, nursing, and medical ethics. “Ideally the discussions with the family are conducted by one member of the team who has good communication skills (page e490).

I might also add that surgery of any sort increases parental stress. This opens up the discussion of parental bonding and stress over having a baby with genital differences, the effect of the stress of surgery on parental bonding and attachment, and the balance between the stress of surgery, the hope of a good cosmetic and functional outcome, and outcomes regarding parental bonding and comfort.

Regarding hypospadias, the authors of the Consensus Statement write,

“...standard techniques for surgical repair such as chordee correction, urethral reconstruction, and the judicious use of testosterone supplementation apply. The magnitude and complexity of phalloplasty in adulthood should be taken into consideration... Patients must not be given unrealistic expectations about penile reconstruction... For the male who has a successful neophalloplasty in adulthood, an erectile prosthesis may be inserted but has a high morbidity” (page e492).

So the Consensus Statement implies that surgery is still the standard of care, but that parents and patients are undergoing a complex procedure and should not have unrealistic expectations because of the high complication rate. If they wait until adulthood for these surgeries, the surgeries are complex, more complex than with babies. However, both babies and adults have high complication rates.

From all of this, parents might see that if they have distress and need to improve their attachment to their child, then they might place a lot of hope on the surgeries. But if there are complications they might discover that the surgeries might make them more distressed. If the surgeries for babies are challenging for the surgeons, and waiting until adulthood is also challenging for the surgeons, and the definitions of success are unclear, then how is a parent to weigh all this information? For example, why would a physician call a neophalloplasty successful if the patient would still need a prosthesis that might fail? The Consensus Statement points the way to addressing some of the cautions regarding the decision about surgery. It emphasizes that there should be a diverse team to help parents. It also notes that there is a psychosocial screening tool to identify parents that are “...at risk for maladaptive coping with a child’s medical condition” (page e492). I suggest that complications that require many surgeries and still not solve the problems would contribute greatly to poor adaptations. A parent might note that if there is not a fully functioning team of experts with one person as the best communicator, then they might not be getting the standard of care. And if they are pressured into an early decision without the process that the team offers, then they are not getting the standard of care.

In chronological order I now review three recent articles. The first is an article called “Anesthetic Neurotoxicity- Clinical Implications of Animal Models” (Rappaport et al, 2015). Although data are not complete, concerns have been mounting for two decades over the use of anesthesia in children under the age of three. In 2009 the Food and Drug Administration established a partnership with the International Anesthesia Research Society called SmartTots from the name Strategies for Mitigating Neurotoxicity in Tots (Rappaport et al, 2015, page 796). There is not conclusive data on babies, but there are many concerns about the toxicities in animals. As the conclusion notes, “...mounting evidence that anesthetic agents cause neurotoxic effects in the developing brains of laboratory animals increases the urgency of the need for large-scale clinical studies” to see what the effects are on babies (page 797). Therefore this group recommends “... that surgical procedures performed under anesthesia be avoided in children under 3 years of age unless the situation is urgent or potentially harmful if not

attended to.” The statement also emphasizes the need to determine whether anesthetic and sedative drugs cause brain damage in infants, toddlers and children (www.smarttots.org/resources/consensus.html). This kind of information may shift the balance of judgment regarding risks and benefits for elective surgery before age 3.

Stanasel et al (2015) reviewed 56 patients who underwent repairs for proximal hypospadias repairs, the more severe form of hypospadias. It is beyond my expertise to evaluate the procedure that they used, but the follow-up was not long-range. That is, the repairs were done between 2002-2013, meaning that none of the patients are in late puberty yet. They conclude that the complications causing the need for reoperations is “higher than previously reported” (page 512). One of the limitations of this study is that “...it is a single institution, retrospective review of hypospadias involving multiple surgeons. The technique varied somewhat from surgeon to surgeon as well as from patient to patient” (page 515). They note the difficulty of getting good data and the need for studying patients and surgeons across different institutions (page 515-516).

In the same journal there is an editorial response to the article by Stanasel et al from Douglas Canning called “Can We Correct Hypospadias with a Staged Operation? If Not, Are We Bold Enough to Report it?” Canning calls the Stanasel et al (2015) article honest and courageous (Canning 2015, page 284), perhaps suggesting that this honesty might be unusual. The complication rate when repairing proximal hypospadias (the more severe form) “...is much higher than what most of us would quote to our prospective clients.” Canning writes more honestly and directly about the possibility or fact that follow up studies are too short-term, and that “Complications become more evident the longer we follow the boys. For years we urologists routinely underestimated the problems our patients have experienced following hypospadias surgery... we have misled our patients, their families and our colleagues” (page 284). He calls for much more follow-up and a commitment to understanding that the surgeon’s “...impressions do not always agree with those of the families” (page 285). He writes that surgeons should be following their patients throughout their careers, and that serious arrangements need to be made for their care throughout adulthood. “Our handoff to the next generation must be meticulous, because in pediatrics we are cursed with our inability to follow our reconstructed children throughout their adult lives” (page 285).

A parent or physician might want to know which kinds of hypospadias will do well with which kinds of procedures. It is beyond my skill set to fine tune the discussion to that level. But it is obvious that the risks with any kind of hypospadias repair in childhood are great. Since the voices of the satisfied adults who have had the surgeries are missing, we might conclude that the risks of surgical repairs may be greater than the benefits. Since the reports from surgeons are difficult to evaluate, and they often emphasize their complications and challenges, we can also wonder if there is a need for a moratorium on surgeries in childhood. It would seem to me that physicians who have great expertise in hypospadias should turn their attention to follow-up care for adults who may be underserved.

Chapter 5: Adult survivors and their advocates

I now turn to the writings of adults who had surgeries and their advocates. I think that these are the voices and writings that should get some extra attention by parents and professionals, as the adults' experiences may be underreported. The adults who had genital surgeries as babies and young children group themselves together as people who were born with a disorder of sex development, or as people who are intersex, even though they know that there are critical distinctions between DSDs and between surgeries for DSDs. Many of these adults who identify as one group are still looking for terms and nomenclature with which they feel comfortable. For example, leaders in the Hypospadias and Epispadias Association ally themselves with others who have had genital surgery, whether to change their gender or to normalize anomalies ranging from minor to major. While many students of genital surgery prefer to separate the outcomes for different kinds of genital surgery, the adult survivors and social scientists studying genital surgery tend to group outcomes together. Perhaps this grouping together of hypospadias with other DSDs creates a tension between adults who have had genital surgery, on the one hand, and doctors who have treated them, on the other hand. The physicians might want to keep definitions and treatments and outcomes separate for the sake of clinical practice and study.

The books that social scientists have written on their behalf also tend to treat surgical interventions for the wide variety of DSDs or genital anomalies as a group. These are influential books that parents and professionals should read, and need to be included in this review. These books parallel the development of the intersex self-help and advocacy movements that are evolving as the adults find each other, often through the internet, forming support groups, sharing information, and planning advocacy. The books might be considered the academic voices of their growing movement. The books include data from science and medicine, accounts of adults with DSDs, and deep discussions of cultural issues, bioethical, philosophical, and cross-cultural issues. Legal issues are raised as well. The books are long, extensively footnoted, and have a passionate quality. While I recognize that these books tend to include hypospadias as one of the DSDs, I review them briefly here because parents are likely to come across them and their points of view should be part of the shared decision making process.

These books document long-range harm from genital surgery, including hypospadias surgery, which I will focus on. Hypospadias surgery is often found to leave lasting scarring, fistulas, diverticula, diminished sexual sensation, and problems with urological functioning (these are also documented in surgeons' reports, cited in the previous chapter of this capstone project). Documenting the long-range harm reported by the adults, they are asking doctors to at least reflect more deeply on their practices and perhaps to stop doing early surgeries. They are the appropriate complement to articles and writings by the medical and scientifically-oriented writers. These writings are qualitative and also address the quantitative when possible. They address the needs and pain articulated by the communities of adults who had the surgeries as babies and young children. While the majority of the medical/scientific writers are men, the majority of the bioethicists/advocates are women. In general the physicians do not list these books and articles in their writings; that omission might reflect the lack of dialogue between the bioethicists, the adult survivors and the medical sources, and perhaps a resistance to that dialogue. I wonder if part of the barriers to dialogue reflects the continued marginalization of

women in science, and the prejudice that social science and emotions are not good science (as discussed above).

The earliest in the group of advocates is Suzanne Kessler's book, *Lessons From the Intersexed*, published in 1998. When she wrote the book, she was professor of psychology at State University of New York (SUNY) at Purchase. She wrote, "A follow up study of boys who underwent surgery for hypospadias concluded that there was a high incidence of fistulas with almost half requiring secondary operations... It is rare that boys are followed into adulthood, so claims about long term cosmetic and functional excellence need to be considered with caution" (page 69). The conflict over how to understand and interpret the reports of the results of early genital surgeries (including phalloplasty) is summarized by the following (Kessler 1998, pages 73-74): "A noncritical reading of the medical literature would lead to the conclusion that genital surgery... is routine, postsurgical genitals can pass inspection, and medical complications are few... A careful analysis of follow-up studies, however, argues for a moratorium on infant intersex surgeries." Throughout her book, she notes that adults who are critical of the surgeries and the results are told that doctors keep perfecting the techniques, or need better follow up studies. The adults begin to feel that they are contributing to doctors' pursuits of the better technique. Yet the medical viewpoint is that genital surgeries on infants, while not problem free, are preferable and worthwhile (Kessler 1998, page 75). "Parenting a male with a scarred and insensitive penis is seen as preferable to parenting a male with a normally functional [but small] one or one that does not permit a direct urinary stream. Nowhere in the medical literature is there an acknowledgment that these are value judgments" (Kessler 1998, page 76). Crucial point; this is the famous fact/value distinction in bioethics.

The second writer who has contributed a large body of work to this effort is Alice Dreger. In 1999 she published a collection of essays called *Intersex in the Age of Ethics*. At that time Dreger was an Assistant Professor of Science and Technology Studies in the Lyman Briggs School, and adjunct Faculty in the Center for Ethics and Humanities in the Life Sciences at Michigan State University in East Lansing. Her book includes two essays by men who had surgery for hypospadias in their infancy or childhood, "Growing up in the surgical maelstrom" by Howard Devore, and "Take charge! A guide to home auto-catheterization" by Sven Nicholson. Devore describes having 16 surgeries while growing up, each surgery breaking down within a year... If they had just left my urinary meatus where it was, at the base of the penis right by the scrotum, I could have avoided at least 12 of those surgeries" (page 80-81). Nicholson writes that from age 11 and into his adolescence, multiple painful recatheterizations allowed improved urine flow for only a week before urinary functioning broke down. The surgeon, a professor at a research hospital, never varied his technique (page 202-203). By his early 20's he had to learn to catheterize himself because his urinary flow did not work without it.

Dreger outlines the general problem clearly: "Some surgeons have argued that we ought not to hastily throw out the older Age of Surgery model of intersex management for a new model. They say we first need evidence that the older model has failed and that a different model would work better" (Dreger 1999, page 15). While the surgeons whom she cites do not distinguish, in this statement, between various kinds of genital surgery, Dreger continues to

note that there is no evidence that the surgical model worked well, the long range outcome studies are severely lacking, the personal reports are reports of failures and increased difficulties because of the surgeries, and while we wait for long range outcome studies, more and more children will be harmed by the surgical interventions (Dreger 1999, pages 15-16).

According to Tia Powell (personal communication, 2016), one could say that the ethical issue is a kind of conflict of interest. The surgeons argue for maintaining the status quo until evidence emerges that the surgical approach is problematic; but the surgeons are the ones who must generate that data and they are not motivated to collect it.

One article by physicians Wilson and Reiner notes that “Early uro-genital surgery should be strictly limited to that necessary to preserve the child’s health...” (Dreger 1999 page 128). I think that leaders in the Hypospadias and Epispadias Association would agree. It might be relevant to note how difficult it is to change a prevailing medical paradigm.

“As with many clinical paradigm shifts, in the absence of data, adherents of each protocol become increasingly dogmatic that their preferred approach is better for the patient, and that it would be unethical to subject the patient to the other, ‘less acceptable’ treatment. Individual clinicians’ attachments to specific treatment regimes result in the ongoing polarization of paradigms” (Dreger 1999, quoting Wilson and Reiner, page 131).

The next major contribution I review is that of Anne Fausto-Sterling, Professor of Biology and Women’s Studies at Brown University, in her book called *Sexing the Body: Gender Politics and the Construction of Sexuality*, published in 2000. Like other writers reviewed in this chapter, she groups together a critique of all the early genital surgeries for the wide variety of intersex conditions. Passionate about feminist thinking, Fausto-Sterling is open about the fact that she weaves both politics and scientific evidence together. The footnotes and bibliography include references from science and medical literature, and take up 192 pages (Fausto-Sterling 2000, pages 257-449). Her footnotes are almost as long as the text of the book itself. Some of the dialogue is in these footnotes. For example, Fausto-Sterling’s colleagues often ignore her work because of the fact that she both asserts that we need to stop doing genital surgeries on babies, and addresses the politics of gender and sexual orientation (Fausto-Sterling 2000, pages 79-81).

Aware of this charge, she refers to the fact that some criticize her for mixing politics with the well-being of intersex babies. Responding to that criticism, she notes that “The medical ‘cure’ for intersexuality frequently does more damage than good... strong evidence that early genital surgery doesn’t work: it causes extensive scarring, requires multiple surgeries, and often obliterates the possibility of orgasm.” Leaders of the hypospadias support groups agree with these observations (personal conversations summer 2015).

In 2008 Katrina Karkazis, Senior Research Scholar in the Center for Biomedical Ethics at Stanford University, wrote a book called *Fixing Sex: Intersex, Medical Authority, and Lived Experience*. She writes, “hundreds of surgical techniques for hypospadias have been described, and many technical advances have taken place.” However,

“no consensus exists about which techniques produce better outcomes, and whether surgery on the whole is more helpful than harmful... There are surprising few long-term follow up studies demonstrating the efficacy of these procedures, which raises concerns about unsatisfactory results, complications, and the need for subsequent surgical repair. Even less understood are the psychological, psychosocial, and psychosexual effects of hypospadias and its surgical treatment” (page 144-145).

Karkazis writes that there is a paradox--clinicians and researchers criticize the advocacy groups because their information is just anecdotal, and they will not change their practice without hard data. But since there is very little hard data, the clinicians keep practicing based on their own experience and the anecdotes of colleagues, trusting their own clinical anecdotes more than the stories coming from support groups (Karkazis 2008, pages 268-270).

Increasingly some clinicians have been influenced by the adult advocates, as seen in the consensus statement and the endocrinology text book (see the chapter on medical sources above). Feder (2014) notes that changes are taking place because the work of the Intersex Society of North America and the work of the Consensus Statement in 2006 prompted some physicians to question the standard of care. “But as marked as these changes have been, the majority of specialists have continued to take normalizing surgery to be in the best interests of children and their families” (page 195). The central argument that these books make is that the parents and physicians should adapt to the congenital bodies of the boys with hypospadias and reevaluate their gut reactions. The only way to begin the process of adapting to the child’s body is through a process like shared decision making. The goal of the shared decision making process would be to help the parents and the children flourish at each stage of their development together, as they learn to first do no harm.

Chapter 6—Internet sources and legal issues

Internet sources. Parents today would search the internet for advice on hypospadias (see page 4ff above) and perhaps on genital surgery in general in order to get information or add to their education about what to do about their children’s DSDs. On the one hand, an internet search using “hypospadias” as a key word obtains 676,000 websites, and so a sampling of these sites cannot be systematic. On the other hand, parents get information and support from these sites and so attention to internet sources is relevant to this capstone project. I have already referred to some useful internet sites in this paper on page 4 and following.

The selections that I have found in the internet seem to be against early surgeries. Many would debate the uses of the internet in making serious decisions about surgeries. Many might question the responsibility of the sources or the legitimacy of the sources. Is the internet a good source for parents? It is ironic that the Hypospadias and Epispadias Association could grow because of the internet, helping many to find each other when they had lived with secrecy and isolation. Parents need to see what is being written on the internet and evaluate it in the context it is meant. Given the reports from articles from the surgeons themselves, given the reports from the adults with a history of hypospadias repair, the internet is one of several

voices. The internet raises the questions that parents need to deal with, even if there is a question about the sources.

Until the advent of the internet, many men with hypospadias felt isolated. They began to use the internet to find each other, exchange information, and form support groups. This has many advantages. Alexander Springer at the Medical University in Vienna became a member of a German-speaking Internet based support group for hypospadias, and monitored the activity on that site between September 2002 and December 2008 (Springer, Reck, Huber and Horcher, 2010, page 250). Springer noted the importance of the online support group for the sharing of information and support, concluding that online support groups for hypospadias “play an important role in how parents and patients cope with the condition. Surgeons dealing with hypospadias should be aware of the importance of Internet-based information in the field of hypospadias. They may benefit from the valuable feedback information from patients and parents to improve their practice and outcome” (Springer et al 2010, page 520).

Legal Issues. Legal advocacy for individuals with DSDs has proceeded more quickly than legal advocacy for men with a specific history of hypospadias repair. Advocates for Informed Choice (xx), which does legal advocacy for patients with DSDs, has been approached by adults with a history of hypospadias surgery on a number of occasions to apply their legal expertise in this area for the legal needs of these adults. The legal efforts for these adults have been evolving (personal conversation, spring 2015, with the legal team working with Advocates for Informed Choice, Ann Tamar-Mattis and Kimberley Zieselman).

Most legal scholars who deal with genital-normalizing surgeries on infants “have called for higher standards of informed consent, suggesting that parents would decline to authorize surgery if they knew the long-term problems faced by intersex adults who [had the surgeries], if they also understood the questionable theory behind the surgeries, and if they knew the lack of evidence that there is benefit to the child” (Tamar-Mattis 2006, page 87).

A Colombian court also ruled on this question, concluding “that genital-normalizing surgery on infants should be treated differently from other medical decisions for children, with special attention to properly informing the parents through a “super-informed consent standard” giving the parents a long time to get informed and make the decisions (Tamar-Mattis 2006, page 87).

Tamar-Mattis goes even further, suggesting that even this higher standard of informed consent is flawed because there is a lack of long-range outcome studies and the cultural bias underpinning the decision-making process create conflicts of interest (Tamar-Mattis 2006, page 88). Doctors and parents are so concerned about genital appearance and conforming to the societal norms of boy or girl, that the conflict of interest is deeper than in other cases. “Radical surgery” is not the answer; counseling is the answer. Addressing parental anxiety and distress are important using a more neutral decision-maker and counseling to determine whether surgery is in the best interests of the child (Tamar-Mattis 2006, page 90).

I note that there are very few people who are neutral on this subject. Most people have been deeply influenced by the cultural considerations of wanting people to fit in to the boy or girl body pattern, and cannot imagine living in a different kind of body. The adults, who have had

the surgeries and want to stop the doctors and parents from doing the surgeries, are not neutral. The professionals involved are profoundly influenced by the experiences that they have had watching this process take place. But with so much medical and anecdotal evidence that there is great harm from the surgeries, the case against surgeries is easily made.

Karkazis (2008) reports the following regarding physicians' concerns over potential lawsuits. There is a controversy over how to find and study adult patients who had early surgeries. Some physicians are reluctant to give out the names of their patients for a variety of reasons. Patient privacy and HIPAA regulations are a primary concern. Karkazis notes that concern for patient confidentiality might cover the larger concern--the exposure of poor outcomes, the fact that the patients got treatments that are out of date, treatments that are not used currently. Tia Powell notes (personal communication, 2016) that there are standard ways of addressing these concerns. One may contact patients and seek consent to participate in a survey or registry of those who had the surgery in the past; this leaves the patient free to say yes or no, and only those who say yes can be contacted.

I also want to note that legal advocates might want to try to change the statutes of limitations on these kinds of surgeries. Since the adults often need care spanning their adult years, and since there is so much evidence that medical and urological problems are caused by the early surgeries, perhaps the institutions that supported the early surgeries should be challenged in the courts to support the best and most compassionate follow-up care that they can give over the life span.

Chapter 7: Conclusions

There are several voices that are missing from this conversation, such as the voices of adults--especially adults ages 40 and over-- who are satisfied with their early surgeries, the voices of physicians who have the evidence that their older adult patients are satisfied, and the voices of men who grew up with hypospadias that was not treated. Karkazis (2008) notes that "Although a few of the adults that I contacted expressed satisfaction with their treatment and quality of life, the majority reported past and continuing suffering, both physical and psychological" (page 219). It is hard to answer with confidence the question of whether we should advocate for surgical repair of distal hypospadias (the "easy" cases). One study (Lorenzo et al, 2013) notes that mild decisional regret was found in 41% of parents and moderate to severe decisional regret was found in 8% of parents within the first year after surgery of their sons' distal hypospadias.

Another concern is the lack of attention in articles to urological functioning over the adult years for men with hypospadias. There are a number of articles that concern sexual function, but urological functioning seems a neglected topic. It would seem to me that urological problems would be more medically serious than sexual dysfunction. Sexual problems might cause distress and psychological concern and these can affect quality of life. However adults in the support groups report urological problems that need medical attention. This seems underreported in the articles, which seem more concerned with sexual function.

I note that there is a lot of criticism of techniques, and constant searching for upgraded techniques and more surgeons with more experience and skill, and so this can be another reason to postpone any early surgeries. If the techniques are better now, why not wait twenty years and get the even better techniques with the involvement of the now young adult with hypospadias or a DSD?

The studies in process about the neurotoxicity of anesthesia in animals have caused great concern about the potential neurotoxicity for babies and young children. The new recommendation is that elective surgeries for children under 3 should be postponed.

Until we have those voices of the adults who are satisfied, we have to give parents the information that we have. Parents need to give equal weight to the narratives of the adults, the writings of the legal and bioethics writers who have become their advocates. Parents would also do well to pay close attention to what the surgeons and medical sources are writing.

Further, parents and professionals need to examine their personal reactions to physical differences, and consider whether their instinctive negative responses concerning differences in genitals should drive treatment decisions, or whether they should reexamine those reactions in order to accept these physical differences. Crucial point -- as you noted earlier, these are value judgments, not simply clinical choices. I would note that physicians (as well as parents) of babies whose genitals are different need time and perhaps counseling in order to adapt and adjust and accept the differences in their children's and patients' bodies before making serious decisions about irreversible surgeries. Physicians and parents who have an automatic dislike or disgust for what they see as they look at different genitals, need to examine what the implications are. Does that reaction merit serious surgeries which often cause so many adverse lifelong problems for the babies and the adults? Feder (2014) would argue that the core of ethical responses and action toward babies with different genitals is how to help them flourish, indeed how to help all concerned flourish (page 210).

Given the extent of concerns that surgeons have about the challenges of doing early genital surgery on boys with hypospadias, I am suggesting a moratorium on early surgeries. I suggest that the medical efforts on behalf of babies with hypospadias and genital differences should be dedicated to creating teams of excellence who will provide all the information above for parents and professionals, to guide them in an extended process of shared decision making which will lead to the best informed consent possible. Parents and children as they grow should be offered access to counseling and mentoring from well-trained members of the teams that conduct the shared decision making process. That mentoring should include adults from the support groups who are also trained to counsel parents and children. The mentoring should be open ended and long range.

Physicians and health care providers should turn their attention to long range care, following up on the babies who had the surgeries already throughout their lives. Outreach, medical care, and support should be improved and enlarged to expand the possibilities for access to compassionate care at any stage of life. It is my hope that the physicians and health care providers who have been involved in hypospadias care will enter into real dialogue with the

adult activists and their advocates, opening up their discussions in order to re-evaluate the impact of their practice. It is my hope that centers of excellence will create the diverse teams of professionals advocated by the consensus statement and the article on shared decision making, in order to widen the availability of shared decision making for parents, using technology to connect parents who live far from those centers to offer them access to the process. The multi-disciplinary teams are already advocated for intersex children, but I believe that they should also be available to all boys with hypospadias, whether distal or proximal.

I hope the legal advocates can not only continue to take more cases, expand their medical apology program, and also find a way to extend, do away with, or work around the statute of limitations around legal action. Perhaps through the legal process, the adults who had hypospadias surgeries and still need more medical care can request that the health care institutions that supported giving them surgeries when they were babies actively support their efforts to find compassionate ongoing care as they age; admit that their medical and psychosocial problems were often made worse through the surgeries that they had; and work to address and redress these harms. A precedent should be set that when the surgeries did harm, the institutions have a responsibility to follow through with any of the medical and psychosocial help that is necessary.

The wish and the hope to fix hypospadias is completely understandable and the efforts of the skilled surgeons is often remarkable. But with surgeons and adults who have had the surgeries reporting so many risks, repeat surgeries, and serious complications that compound over the years, one needs to stop and reexamine some deeply held wishes and fears and assumptions. The adjustment to differences and the ability to accept these differences will vary with the skills and temperaments of the parents and the professionals, but the rush to surgeries should not be clouded by the anxieties over what might happen if we do not do surgeries.

There needs to be a balance between maintaining privacy for the babies, without the stigma of secrecy and shame, and negotiating the inevitable difficulties of being different in settings outside the safety of a supportive home. Indeed the mentoring should address the issues that are inevitable with some intelligence.

An anecdote might help parents to think through some of their gut reactions. First, when confronted with different genitals we need to examine our feelings and wishes. The following points up the parent's wish to justify the surgery that they just had for their baby even in light of watching adults lobby Parliament to stop doing the surgeries. Sanders, Carter, and Goodacre (2011), in their article about the parental search for harmony in making medical decisions, note the following reaction of one pair of parents. They write that

“the controversies and dilemmas of early surgical decisions raised strong emotions in some parents: There's no way in my mind that we've done the wrong thing and that I ever think 'I wish we'd left him to make his own decision when he was older.' Definitely not. I watched a documentary [on TV] just after Neil was born... They were lobbying Parliament to stop making children have genital surgery and we just said 'how ridiculous' (Sanders, Carter and Goodacre, 2011, page 2225, quoting Anne).

Leaders of the Hypospadias and Epispadias Association, usually volunteers who are survivors of the early surgeries, are aware that they are in a vulnerable position. They cannot afford to alienate the physicians who are experienced enough to help them, yet they are hoping that an extended process of shared decision making will help new parents of boys with hypospadias decide to postpone any surgeries until the boys grow up to adulthood and can participate in the decision making process. Their hope is that when the parents get all of the information above, the numbers of early surgeries will go down. Their hope is that they can mentor parents to address their anxieties, help them see clearly that it might be better for the boys to have a healthfully functioning congenital urethra, even if it is in the female style, than to risk becoming a hypospadias cripple. Their hope is to offer mentoring and support to boys and men with hypospadias and epispadias. Their hope is that the physicians who are experienced in hypospadias surgeries can join them in giving the lifelong care that men with hypospadias might need with compassion and understanding. In order to redress decades of ethically questionable treatment, physicians and all the health care professionals involved should consider the bravery of the adult survivors' testimonies, read the writings of their bioethics advocates and enter in to the process of shared decision making in order to reexamine their practices.

Table of Selected Articles from Surgeons and Physicians

Article	Findings	Suggestions or conclusions
Bubanj et al, 2004, sexual function after hypospadias repair	Contradictory findings: "sexual functioning not affected" vs. major reported negative outcomes	Additional investigation is needed
Cambareri and Hanna, 2015, adult hypospadias patients	Secondary surgery rate: distal 9%, proximal 32%; many postpuberty problems (sexual, urinary, psychosocial)	Need long-term patient reported outcome studies
Canning, 2015, commenting on Stanasel's (2015) study	Parents may get misled into holding false hopes about success of surgery	Surgeons should warn parents of the high long-range complication rate
Hanna and Cambareri, 2015, surgical challenges in adult hypospadias	35% to 75% complication rate	Technical recommendations, especially considerations of availability of native tissue
Hensle et al, 2001, on outcome of hypospadias repair in early adulthood	Significant number of complications	Pre-operative counseling to avoid unreasonable expectations

Lorenzo et al, 2014, on decisional regret after distal hypospadias repair	50% of parents regret after one year	More pre-operative counseling
Moriya, K. et al, 2007, on urinary symptoms after hypospadias surgery	Few studies but reports are contradictory: “mostly good urinary control” but 32% moderate to severe dribbling	Despite advances in surgical techniques, terminal dribbling not uncommon
Mureau et al, 1996, satisfaction with penile appearance after hypospadias surgery	Surgeons more satisfied than patients, complications years after surgery	Follow-up needed into adolescence
Nuininga et al, 2005, on long-term outcomes of hypospadias repair	54% complication rate at long term follow-up and also later	Evaluate final outcome only after patient has reached adulthood
Rappaport et al, 2015, studies on animals re anesthetic neurotoxicity	Question raised regarding negative consequences for neurocognitive development in babies	Stop elective surgeries on babies under age 3
Springer et al, 2011, on online support groups for hypospadias patients	Groups help parents and adults exchange information and concerns	Surgeons should learn patients’ and parents’ concerns from these groups
Stanasel et al, 2015, on complications after proximal hypospadias repair	Additional surgery 68%, fistulas 57%, diverticula 14%, urethral stricture 14%	Complications higher than previously reported
Stecker et al, 1981, on the prevention of “hypospadias cripples”	Surgeon must know over 150 methods even though no one procedure is accepted by every surgeon	Without great care taken, likely consequence is hypospadias cripple

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