

# Should Surgery for Hypospadias Be Performed Before an Age of Consent?

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## Abstract

Hypospadias is a relatively common genital condition in which the urethral opening forms on the underside of the penis, as opposed to at the tip of the glans. Patients with hypospadias are typically referred for surgery during infancy or early childhood. Recent evidence, however, indicates that many individuals with hypospadias do not experience the functional or psychosocial difficulties that are commonly attributed to the condition, and that surgical intervention for hypospadias carries substantial risk of adverse outcomes. In this article, we review published outcomes data and conduct an in-depth analysis of the typical rationales for hypospadias surgery, taking into consideration both the potential benefits and harms of the procedure, as well as the existence of non-surgical alternatives. We argue, firstly, that most childhood surgeries for hypospadias are performed for anticipated future problems concerning function and cosmesis, rather than extant problems that serve to undermine the child's well-being. Secondly, we contend that the surgery can be safely performed after an age of consent without increasing the absolute risk of surgical complications to an ethically meaningful degree. We conclude that surgery for hypospadias should typically be performed only if requested by the affected individual, under conditions of informed consent.

**Key words: hypospadias, elective surgery, pediatric ethics, informed consent**

## **Introduction**

Hypospadias is a congenital condition in which the urethral opening forms on the underside of the penis or scrotum, rather than on the tip of the glans. It is estimated to occur in as many as one in 250 boys (Elder, 2011). Hypospadias presents along a spectrum, with individuals being affected to varying degrees. Individuals with hypospadias of all degrees, however, commonly undergo surgery for the condition during infancy or early childhood. While it has generally been assumed that the potential harms to the child of failing to perform hypospadias surgery are sufficient to warrant early intervention, recent reports suggest that many individuals with hypospadias do not experience the functional and/or psychosocial difficulties that are commonly attributed to the condition, nor desire surgery for it (Dodds et al., 2008; Schlomer, Breyer, Copp, Baskin, & DiSandro, 2014). In addition, existing outcomes data on hypospadias surgery highlight the substantial risk of significantly poor cosmetic and functional outcomes.

In this article, we present these emerging data to place the implications of a hypospadias diagnosis into a proper context, and then critically analyze the purported rationales for hypospadias surgery. Among other issues, we challenge the commonly held notion that children do better with hypospadias surgeries performed before the advent of conscious memory by reviewing the relevant data. We take into consideration both the potential benefits and potential harms of hypospadias surgery, alternatives to surgery, and the ethical principles involved in deciding whether to perform hypospadias surgery during childhood. Although we draw on evidence that is at times specific to varying degrees of hypospadias, we ultimately offer a set of ethical principles that we believe applies to all individuals with this condition. Our key suggestion is that the choice for parents (and patients) to make is not between an inevitable surgery that must be performed either early in life or later, but rather between no surgery, early surgery, or surgery performed with the consent of the affected individual. The primary task is to determine if any medically necessary interventions (i.e., interventions intended to preserve or restore a straightforwardly physical function, such as the ability to pass urine) must be made before the

child can be involved in the decision-making process. If not, then the choice should be reserved for the (future) individual either to refrain from surgery (if his hypospadias does not bother him, or if he sees the risks associated with the surgery as outweighing the potential benefits), or to elect the specific component(s) of a typical hypospadias surgery that would best suit his preferences and values. Along the way, we anticipate various objections to our position as well as offer our replies.

### **Hypospadias – a ‘spectrum’ condition**

Hypospadias is defined as a urethral meatus (opening) located proximal to the tip of the glans penis. It is classified based on the location of the meatus [see Figure], as glanular (on the glans penis), coronal (at the coronal ridge), subcoronal (the top 3 positions shown in the image), midpenile (the position shown between the dashed lines), penoscrotal, scrotal, or perineal (the positions shown under the lower dashed line) (Elder, 2011). It is often, but not always, associated with chordee of the penis, in which the penis curves to varying degrees, as well as with a lack of foreskin tissue on the underside of the penis. Other variations of genital development, such as a midline scrotal cleft, are sometimes present (Snodgrass, 2012).

Hypospadias may be associated with a downward angle or splaying of the urinary stream requiring sitting to void, difficulty with sexual penetration due to penile curvature (however, this difficulty only applies if chordee is present, and only then to sexually active individuals), and, more rarely, meatal stenosis (narrowing of the urethral opening) – though the more common scenario is that the opening is actually larger in individuals with hypospadias than in those without. Hypospadias is also alleged to cause infertility if the location of the urethral meatus prevents semen being deposited in the optimal position for conception during unassisted heterosexual intercourse. For example, in the ninth edition of *Campbell-Walsh Urology*, Borer and Retik stated that, “[t]here is no question that more severe degrees of hypospadias require repair to provide the ability to...achieve sexual intercourse, and effectively inseminate” (2007, p. 3710), and Schlomer et al. (2014) have noted that “[r]easons given to

parents for treating hypospadias include...difficulty with sperm deposition leading to infertility, [and] ventral curvature leading to difficulty with intercourse” (p. 2). Some authors also claim that a hypospadiac penis constitutes a cosmetic ‘deformity’ (Elder, 2011).

### **Hypospadias surgery**

A variety of techniques for hypospadias surgery exist, and surgery may involve one or a combination of these. Urethroplasty is used to bring the urethra to the tip of the penis, while meatoplasty and glanuloplasty are used to create a patent urethral opening and reapproximate the edges of the glans to resemble that of an unaffected penis. In some cases, skin flaps or grafts are used to substitute or supplement the urethral tissue (Snodgrass, 2012). Other aspects of the surgery can include release of chordee to straighten the penis; plastic surgery to create skin cover for the phallus; and scrotoplasty in cases involving bifid (split) scrotum. Surgery for hypospadias is most often performed as a single surgery, although individuals with more severe cases sometimes undergo a planned two-stage procedure. After surgery, a catheter is typically left in the urethra for several days to drain urine from the bladder and allow healing. Because of the pain of the genital surgery and irritation as well as infection risk from the catheter, patients undergoing hypospadias surgery are usually prescribed medication for pain, bladder spasms, and infection prevention (Snodgrass, 2012).

### **Historical intentions behind hypospadias surgery and more recent outcome data**

In 1968, Culp and McRoberts stated, “it is the inalienable right of every boy to be a ‘pointer’ instead of a ‘sitter’ by the time he starts to school ... [and] to write his name in the snow and have it legible” (pp. 312, 315). In addition to the ability to urinate in a standing position and with a strong, straight stream, surgery for hypospadias is typically performed with the goals of achieving a straight penis in erection, a meatus near the tip of the glans, allowing ‘normal’ sexual intercourse and fertility, and otherwise creating a functionally and cosmetically ‘normal’ penis (Schlomer et al., 2014).

Hypospadias surgery was not always seen as advisable. In a urology textbook published in 1917, before surgery for hypospadias was common, author and urologist Keyes stated, “Balanitic [glanular] hypospadias is unimportant; many patients have it without being aware of the fact, while the greatest inconvenience it produces is a slight imperfection in erection and a dribbling at the end of urination” (p. 546). It is possible that the shift in recent decades toward regarding hypospadias as being ‘important’ enough to warrant surgical intervention may at least partially reflect improvements in surgical methods and technology (and thus a greater ease of intervening, with comparatively less risk). However, it seems that a broader *intervention bias* may also play a role: the tendency of doctors to favor ‘taking action’, including surgical action, when non-intervention may be equally well justified on strictly medical grounds (see Foy & Filippone, 2013).

Surgical intervention can sometimes lead to regret. In 2014, a study of parental regret after elective hypospadias surgery (for their son) showed that 58 of 116 parents (50%) felt mild (48/116, or 41.4%) or moderate to severe (10/116, or 8.6%) regret one year after the operation (Lorenzo et al., 2014). In this study, parents were administered the Digital Regret Scale, a well-studied and validated measure that registers distress or remorse experienced after a health care decision. All of these parents had provided permission for their child to undergo the surgery, but later came to regret their decision. Although parental regret was statistically associated with the experience of postsurgical complications, parental regret also occurred in many parents of children who did not experience complications. The authors concluded that aiming to minimize surgical complications and providing parents with better counseling about presurgical expectations might be effective ways of reducing parental regret. However, they did not discuss whether the surgery was actually necessary in the first place, nor whether choosing *not* to intervene via surgery could reduce or even eliminate the potential for future parental regret.

Two recent studies of adults with hypospadias raise further questions about the advisability of early hypospadias surgery. One study, conducted by Dodds et al. (2008), surveyed 56 men who were

identified as having hypospadias by a urologist during an appointment for another urological issue. Of the 56 men, 9 (16%) had previously undergone surgery for their hypospadias, and all of them still had a hypospadiac meatus. Eighteen (32%) stated that they were unaware of their genital difference, and 6 (11%) reported that they only became aware of their genital difference after a healthcare professional brought it to their attention. Although 20 of the 56 (36%) reported spraying or a downward angling of the urinary stream during urination, only 3 of the 56 (5%) sat to urinate. Fifty-three of the 56 (95%) reported no difficulty engaging in sexual activities, and 41 of the 43 who had attempted to impregnate a partner (95%) were successful (the infertility of the remaining two men was believed to be due to low sperm count, not hypospadias). The vast majority of the total sample (95%) reported being satisfied with their penile appearance, and only one male expressed an interest in hypospadias surgery when asked (but eventually chose not to pursue this) (Dodds et al., 2008). The authors noted that adults, compared to adolescents, may be less likely to be bothered by their penile appearance, possibly because they have become accustomed to the appearance of their genitals, as well as have had the opportunity to form long-term relationships.

In the second study, Schlomer et al. (2014) conducted an online survey of 736 adult men (over the age of 18 years) who were recruited through a targeted advertising campaign via social media. All of the men completed a series of self-reported anatomy questions to provide an indication of the appearance of their penis and the severity of hypospadias (if any). The men were shown six diagrams showing different locations of the urethral opening, five diagrams showing different appearances of the foreskin, and five diagrams showing different degrees of ventral penile curvature during erection, and were asked to select the three diagrams that best resembled their penile appearance in each of these three domains. If the men selected a diagram that showed a urethral opening located at the proximal one-third of the glans or more proximal, and/or chose a diagram of a foreskin that aligned with the typical appearance of the foreskin in males with hypospadias, they were regarded as having possible untreated hypospadias by the researchers.

Of the 52 men who self-identified as having untreated hypospadias (7.1% of the total sample), 37 identified themselves (through selecting the equivalent diagram) as having a ‘mild’ (glanular or coronal) form of the condition and 15 reported having a ‘severe’ (subcoronal or midpenile) form. To validate the self-reported anatomy questions, the researchers pretested these questions on 26 participants without hypospadias (including 22 males and four females). These participants were shown a series of images depicting penises with differing degrees of hypospadias (including no hypospadias), and were asked to select the diagrams that best represented the penis shown in the image. In addition, 52 (7.1%) of the 736 men who participated in the study agreed to upload an image of their penis, and these images were reviewed by two pediatric urologists (who were unaware of the men’s self-reports) in an attempt to validate the men’s self-reports. Of the 49 men who reported having a penis that was “normal” in appearance, 38 were deemed to be “normal”, 11 were deemed to be “inconclusive”, and none were deemed to have hypospadias by the urologists. Of the three men who reported having untreated hypospadias, all three were deemed to be “inconclusive” by the urologists.

As part of the survey, the men were asked to indicate how often they sat to urinate on a 5-point scale (1 = almost never/never, 2 = sometimes [less than half the time], 3 = sometimes [approximately half the time], 4 = most of the time [more than half the time], 5 = almost always/always). The authors found no statistically significant difference ( $p = 0.8$ ) between the men in the ‘mild’ hypospadias group (mean score on scale = 2.0,  $SD = 1.3$ ) and a control group of men who did not self-identify as having hypospadias (mean score on scale = 1.9,  $SD = 1.2$ ). However, they did find a statistically significant difference ( $p < 0.003$ ) between the men in the control group and the men in the ‘severe’ hypospadias group (mean score on scale = 3.1,  $SD = 1.7$ ). The men who reported sitting to urinate completed an additional 5-point scale indicating the extent to which this bothered them (1 = not at all, 2 = very little, 3 = moderately, 4 = significantly, 5 = extremely). Interestingly, no statistically significant difference ( $p = 0.7$ ) was found between the scores of the men in the control group (mean = 1.4,  $SD = 0.7$ ) and the men in the ‘mild’ hypospadias group (mean = 1.3,  $SD = 0.6$ ), nor between the results of the men in the

control group and the men in the ‘severe’ hypospadias group (mean = 1.7,  $SD = 0.8$ ). This suggests (a) that some men sit to urinate, whether they have hypospadias or not, and (b) that sitting to urinate does not seem, on balance, to bother the men who do this (again, whether they have hypospadias or not).

With regards to fertility, more men in the hypospadias (combined ‘mild’ and ‘severe’) group reported successfully impregnating a partner than men in the control group (29/51 or 57% vs. 317/674 or 47%); however, this counterintuitive difference was not statistically significant ( $p = 0.2$ ). No statistically significant difference was observed with respect to diagnosis of infertility between the men in the control and hypospadias groups (22/677 or 3.2% vs. 2/52 or 3.8%,  $p = 0.8$ ). The men in the hypospadias group (both ‘mild’ and ‘severe’) reported lower mean scores on the Sexual Health Inventory for Men ( $p < 0.001$ ), more dissatisfaction regarding the shape and location of their urethral opening ( $p = 0.011$ ) and penile curvature ( $p = 0.048$ ), and more penile curvature ( $p = 0.003$ ) resulting in difficulty with sexual intercourse ( $p < 0.001$ ) than controls. Overall, the men in the ‘severe’ hypospadias group reported more negative functional and cosmetic outcomes than the men in the ‘mild’ hypospadias group, and also reported having more “unhealthy” days with respect to physical ( $p = 0.003$ ) and mental ( $p = 0.017$ ) issues than controls.

In another study conducted by Fichtner, Filipas, Mottrie, Voges and Hohenfellner (1995), the authors raised doubt as to whether anterior hypospadias can even be considered “abnormal,” rather than simply part of the developmental spectrum. The authors analyzed the urethral meatus location in 500 “normal” men and concluded that there is wide natural variation in the location of the meatus. They found that only 275 (55%) of the men had a meatus located at the very tip (distal third) of the glans. Of the remaining 225 men, 160 (32% of the overall sample) had a urethral meatus located in the mid-third of the glans. Sixty-five of the men (13% of the overall sample) had anterior hypospadias (49 glanular, 15 coronal, 1 subcoronal). Of the 16 men with coronal and subcoronal hypospadias, all but 6 were unaware of their genital difference, all but 1 (a homosexual man) had fathered children and only 1 (the man with subcoronal hypospadias) had associated curvature of the penis. All of these men were able to

urinate standing with a single stream and engage in sexual intercourse without difficulty. Based on these findings, the authors questioned “the need for meatal advancement in cases of anterior hypospadias without associated penile curvature” (Fichtner et al., 1995, p. 833). These data suggest that not all children with hypospadias will go on to suffer from the problems typically attributed to the condition, and thus cast doubt on the advisability of undertaking surgery to prevent such problems from occurring.

**Rationale for performing hypospadias surgery in childhood: What is the recommended window of timing?**

The influential *Nelson Textbook of Pediatrics* recommends surgery for all cases of midpenile and proximal hypospadias, but states that surgery is not necessary for boys with distal hypospadias (i.e., hypospadias with the meatus closer to the tip of the glans) who have no functional issues (Elder, 2011). The recommended timing of hypospadias surgery is between 6 and 12 months of age (Elder, 2011; Snodgrass, 2012). According to the hypospadias chapter in *Campbell-Walsh Urology*, this recommendation is based on “expert opinion” and is not supported by any specific data (Snodgrass, 2012). Elder (2011), however, offered four reasons to support this recommendation. Firstly, younger children (i.e., less than 12 months) are asserted to require less postsurgical pain relief compared to older children. Elder does not provide any references to substantiate this assertion. In fact, it is now recognized that even very young infants have a well-developed capacity to experience pain, and that developmental differences in *expression* of pain, including an infant’s lack of ability to use language, seriously complicate such an assessment (see, e.g., Fitzgerald, 1998; Franck, Greenberg, & Stevens, 2000; Frisch & Simonsen, 2015; Johnston, Stevens, Craig, & Grunau, 1993). Indeed, Fitzgerald (1998) argued that “an infant’s spinal sensory nerve cells are more excitable than an adult’s. This makes their spinal reflex response to a harmful stimulus much greater and more prolonged” (p. 21). Of course, it must be remembered that, since adults can self-report pain, they are able to procure post-surgical pain

control as needed; and, moreover, the *least* painful intervention is the one that does not occur at all. Arguably, these points speak in favor of avoiding medically unnecessary surgery in the first place—unless it is deemed to be desirable by the individual who will have to undergo the painful experience.

The second reason offered by Elder (2011) in support of the 6 to 12 month recommended window is that the risk of complications from general anesthesia is lower after 6 months of age compared to earlier in infancy. This sets the lower limit for surgery, on Elder’s analysis. Thirdly, penile growth is slower at this age (i.e., between 6 and 12 months) compared to later on, during puberty. This is presumed to make healing more reliable. However, it should be noted that penile growth between 6 and 12 months of age is not as slow as penile growth after puberty, when the penis has reached its full size (and when the patient is in a better position to provide informed consent for any genital operations; see discussion below). Finally, Elder noted that the child will not remember the surgery later in life if it is performed earlier, which appears to inform the “upper limit” of 12 months (although note that retrospectively reportable memories do not seem to form up to 5 years of age; see Jones, O’Brien, Chase, Southwell, & Hutson, 2009).

This final rationale for early surgery, namely the lack of a conscious memory, is also given by Jones et al. (2009). These authors surveyed 52 adolescents (aged between 13 and 15 years) who underwent hypospadias surgery in early childhood (the median age of first hypospadias surgery was 1.96 years). Of the total sample, 90% (47/52) of the adolescents reported being satisfied with the overall appearance of their body, and 81% (42/52) reported being satisfied with the overall appearance of their genitals in particular. The authors found that the adolescents who did not recall the surgery (as was the case if the surgery was performed before the age of 5) were more likely to have a positive body image and to be satisfied with their overall body appearance than those who did recall the surgery. As the authors did not report the mean satisfaction scores, but rather an “association” between these variables (Fisher’s test  $p < 0.031$ ), the real-world significance of the association is difficult to determine. Moreover, the authors did not collect appearance satisfaction scores from adolescents with

hypospadias who had *not* undergone surgery at all. Therefore, for the purposes of the present analysis, this study lacked the crucial control. As the authors stated, the “retrospective nature [of the study] and lack of controls mean that the results must be interpreted cautiously and the memory of events may well be modified by caregivers and other factors” (Jones et al., 2009, p. 1748).

We note that even if hypospadias surgery can be performed without a child’s (consciously) being able to remember it later in life, (a) more research is needed to determine the unconscious effects of early surgery on the developing nervous system (i.e., effects that are unavailable to verbal reporting; see Fitzgerald, 1995; Frisch & Simonsen, 2015), and (b) the potential harms of surgery for those who may not need it in the first place also need to be considered.

### **Risks of hypospadias surgery**

There are risks associated with all surgeries, and hypospadias surgery is no exception. Potential complications include wound infection, poor wound healing, edema, urethral diverticuli, urethrocutaneous fistula, iatrogenic meatal stenosis and urethral stricture, glans dehiscence, breakdown of the repair, functional obstruction of urinary flow, residual chordee, iatrogenic spraying of the urinary stream or spraying from several openings due to fistulae, hardening of the skin, and poor cosmetic result (Duckett, 1998; Hadidi, 2013). The term “hypospadias cripple” was coined in 1970 to describe a patient “who has undergone multiple unsuccessful hypospadias repair attempts, with significant resultant penile deformity” (Borer & Retik, 2007, p. 3740). For many of these complications, the rates of which are discussed below, revision surgeries are required; however, there is a possibility that even further surgery may not adequately address these issues. In addition to the specific risks associated with hypospadias surgery, there are also general risks that apply to all surgeries. These include adverse reactions to anesthesia, infection, pain, loss or change of sensation at the site of surgery, and, rarely, death (Jenkins & Baker, 2003; Marsh, 2006; Mouradian, 2006).

The degree of risk for each of these adverse outcomes is not known with certainty. It depends

upon numerous factors, including the specific anatomy of the child patient, the conditions under which the surgery is performed, the surgical technique used, and the surgeon's experience. In *Nelson Textbook of Pediatrics*, it is stated that the complication rate is "low" at 5% for distal hypospadias, 10% for midpenile hypospadias, and 15-20% for proximal hypospadias (Elder, 2011). However, the available long-term follow-up data call into question these reported rates, and suggest that complications may be generally underreported because of inadequate follow-up (see later discussion). In a 2013 review of 690 cases (over 12 studies) of primary hypospadias surgery with a follow-up range of 1 to 120 months, the authors reported up to an 11.5% rate of fistula formation, 7.67% rate of meatal stenosis, and 2.56% rate of wound breakdown (Wang, Xu, & Zhong, 2013). A review of 126 patients who had at least a 10-year follow-up after hypospadias surgery by experienced pediatric urologists found that 50.8% (64/126) of the patients reviewed experienced at least one complication. In this study, all patients with a fistula (23%, 29/126 patients) or fistula recurrence (11%, 14/126 patients) required additional surgery. Fifteen patients with meatal stenosis (12%) and 10 with urethral strictures (8%) also required reoperation; 6 of the patients with meatal stenosis (5%), and 3 of the patients with strictures (2%) required a second reoperation (three surgeries including the initial operation) (Nuininga, De Gier, Verschuren, & Feitz, 2005). Even if the numbers quoted by Elder were correct, however, that these percentages should be considered 'low'—especially for a surgery that may not be medically necessary—is debatable. Finally, it is worth noting that although some of these risks (e.g., of becoming a "hypospadias cripple") may be less likely to occur, their magnitude is so severe as to have a potentially catastrophic effect on the male's quality of life.

On the issue of underreporting, Springer (2014) identified numerous barriers to complete reporting of surgical, functional, cosmetic, and quality of life outcomes. These barriers include: numerous techniques in use; most publications reflecting single-center and single-surgeon retrospective case series with limited follow-up periods and small numbers of patients undergoing follow-up; transition of care from pediatric to adult specialists, thus limiting follow-up into the period when sexual

activity is likely to occur; assessment of cosmetic outcomes by surgeons who may be biased to approve of their own ‘work’; assessing urinary function in non-toilet-trained boys; and difficulty assessing sexual function and behavior in young adults (Springer, 2014). One report on adult patients who underwent hypospadias surgery as children suggested that, although these surgeries are often deemed to be successful initially (i.e., no apparent postsurgical complications), patients tend to experience postsurgical complications years later as their penis grows and they become sexually active. The authors of this report drew attention to this “natural evolution” of the results of hypospadias surgery and suggested that it is only with adequate, long-term follow-up—i.e., greater than 20 years—that these complications will be accurately identified and reported (Barbagli, Perovic, Djinovic, Sansalone, & Lazzeri, 2010). Further, a review article of the quality of outcome reporting of surgery for hypospadias showed that the reporting of complication rates is incomplete and has declined over the past three decades (Pfistermuller & Cuckow, 2012). The only conclusion that can safely be drawn at this time is that the complication rates are *at least* those reported in the literature, and may very well be higher, given all of the barriers to collecting accurate long-term follow-up data.

### **The importance of adequate long-term follow-up: Sexual dysfunction and hypospadias repair**

The effects of early hypospadias surgery are not limited to childhood or adolescence, i.e., the period in which data on complications are typically collected. Instead, the consequences of genital surgeries performed during childhood are far-reaching, affecting the future adult the child will become. Adults requiring further surgery to repair the complications described above that are known to occur after childhood hypospadias surgery must often undergo extensive reconstructive procedures and more than one operation. Myers, McAninch, Erickson and Breyer (2012) evaluated 50 adult men undergoing major reconstructive surgery to repair complications arising from childhood hypospadias surgery. The authors deemed the surgery to be ‘successful’ if it resulted in no urethral complications; only 50% (25) of the men met this criterion after the first reconstructive surgery. Of these 25 men, 18 underwent

additional surgeries. Thirteen of them ultimately achieved a successful outcome, resulting in an overall success rate of 76% (38/50). The authors noted that, although some complications following hypospadias surgery occur early on, others only become apparent with genital maturity. Based on these findings, the authors concluded that “Managing problems from previous hypospadias surgery is difficult with a high initial failure rate. Additional procedures are commonly needed ... patients must be prepared and committed to the possibility of several surgeries to manage these problems” (Myers et al., 2012, pp. 459, 463).

The results of a questionnaire study by Chertin et al. (2013) provide further evidence of the need for adequate long-term follow-up. The authors found that only 11% of adults with proximal hypospadias who underwent hypospadias surgery as children were satisfied with the final results of the surgery. Depending on the degree of hypospadias, 8.9% to 72% reported mild erectile dysfunction as adults, and 83.2% complained of premature ejaculation. These rates are far higher than those in the unoperated general adult population and present cause for serious concern (see also Selvin, Burnett, & Platz, 2007; Stein et al., 2014).

A more comprehensive study was undertaken by Herbst, Ferrer and Makari (2013). Their review of the Pediatric Health Information System (PHIS) database involved the evaluation of data from 1,679 children who underwent surgery for proximal hypospadias at 37 different US-based pediatric hospitals. The median age at the time of the surgery was 10 months. The researchers discovered that 440 (26.2%) of the patients who underwent prior proximal hypospadias surgery that was believed to be definitive required one or more additional hypospadias-related surgical interventions later in life (Herbst et al., 2013). Similarly, a long-term outcome study of 474 patients who underwent primary hypospadias surgery between 4 and 134 months of age (mean age: 22.6 months) at a single tertiary care center in Belgium reported a 24.1% reoperation rate. Of the 114 patients who required further hypospadias surgery, more than half (60/114 or 52.6%) did not present with complications requiring reoperation until more than one year after the surgery (Spinoit, Poelaert, Groen, Laecke, &

Hoebeke, 2013). It has also been reported that 50% of all patients undergoing hypospadias surgery will require further surgery during their lifetime (Mouriquand, 2006).

### **Alternatives to Hypospadias Surgery During Childhood**

Given the numerous complications associated with hypospadias surgery, it seems reasonable to ask whether there may be less invasive and less risky alternatives. This question is especially pertinent in light of the studies we cited earlier suggesting that hypospadias is quite common and does not result in serious adverse effects on health or functioning in a substantial percentage of those with the condition.

One alternative is to refrain from surgery—in childhood—altogether. The option here would be to adopt a conservative approach and wait to see whether the child does indeed experience problems related to his hypospadias that (a) have a negative impact on his well-being and (b) cannot be addressed more effectively, and less invasively, using non-surgical means. Children with hypospadias could be counseled about their genital anatomy, if necessary, and could eventually decide for themselves what problems, if any, hypospadias creates for them (and whether the risk of surgical complications is “worth it” to them, all things considered).

It is important to note that, even if a boy does undergo surgery for his hypospadias, this may not necessarily eliminate the need for psychosocial interventions later in life. As we described earlier, many males who undergo hypospadias surgery require one or more follow-up surgeries to address complications arising from the initial surgeries, and this may be experienced as distressing.

Hypospadias surgery may also result in unsatisfactory cosmetic outcomes and a penis that is atypical in appearance (more atypical than the original appearance), and this may also be a source of psychosocial distress for many males who undergo the surgery. Some males who undergo surgery for their hypospadias during early childhood may also experience feelings of resentment toward their parents for choosing this surgery for them before they could participate in the decision-making process, and

require assistance in managing these feelings. There is a large body of literature describing similar feelings of resentment experienced by some individuals who underwent ‘corrective’ genital surgery during early childhood, including individuals with hypospadias (see, for example, the numerous patient narratives reported in Dreger, 1999 and Intersex Society of North America, 2006).

If surgery is judged to be desirable by the male later in life, he may be able to select only certain portions of a typical hypospadias surgery. For example, if the patient has curvature of the penis making penetration difficult, he may choose to have a chordee release. However, if he must sit to urinate and is not bothered by this, he may prefer not to undergo urethroplasty and therefore avoid the specific surgical complications associated with this procedure, such as fistula, urethral dehiscence, and urethral stricture. If the patient is experiencing problems with infertility, he may prefer non-surgical methods of addressing the issue, such as the use of artificial insemination. There are numerous reports in the literature of men with hypospadias having children by this method. In fact, the first documented case of successful artificial insemination took place in 1785 after the Scottish surgeon John Hunter instructed the wife of a man with severe hypospadias to inject her husband’s semen into her vagina (Agarwal & Allamaneni, 2007).

There is significant evidence to suggest that performing hypospadias surgery during childhood does not necessarily lead to better surgical outcomes compared to operating in adulthood. A recent study by Snodgrass, Villanueva and Bush (2014) investigated surgical outcomes in 1,140 males with hypospadias (age range at time of surgery: 3 months – 62 years) who underwent urethroplasty. The surgery was primary in 883 (77%) of the patients (i.e. this was their first hypospadias surgery) and reoperative in the remaining 257 (23%) (i.e. these males had undergone at least one other surgery for their hypospadias in the past, some as many as 20). Sixty-nine of the males underwent urethroplasty (8 primary, 61 reoperative) during adulthood (mean age at time of surgery: 23 years), and the remainder during childhood (871 primary, 196 reoperative). Importantly, all of the urethroplasties were performed by the same surgeon using the same surgical techniques, allowing for a more meaningful comparison

than is typically available in the literature. The authors found no statistically significant difference in complication rates between children and adults. One (12.5%) of the 8 males who underwent primary urethroplasty during adulthood experienced complications, compared to 123 (14%) of the 871 males who underwent primary urethroplasty during childhood. Sixteen (26%) of the 61 males who underwent reoperative urethroplasty during adulthood experienced complications, compared to 69 (35%) of the 196 males who underwent reoperative urethroplasty during childhood. Although the authors found no correlation between the age of the patient at the time of the surgery and risk of surgical complications, they did find that reoperations were associated with a greater risk of complications than primary urethroplasties. Based on these results, the authors concluded that, “In contrast to popular belief, our data do not indicate a greater risk for urethroplasty complications after hypospadias surgery performed in adulthood. Repair in adults can be done using the same techniques as in children with the same goal of [creating] a neomeatus at the tip of the glans” (Snodgrass et al., 2014, p. 1730). Consistent with this view, the only available long-term survey (by Jones et al., 2009) performed on those individuals with hypospadias who were operated on as infants shows a comparable complication rate (38%) to studies of hypospadias surgery performed during adulthood (38.7%) (Hensle, Tennenbaum, Reiley, & Pollard, 2001).

### **Ethical considerations in contemplating hypospadias surgery for children**

Parents are generally considered entitled to make healthcare decisions in the best interests of their children (Buchanan & Brock, 1989; Kopelman, 1997). In the case of bodily differences that may adversely affect a child’s physical-functional capacities, such as a severe cleft lip (Marsh, 1990; Mouradian, 2006), and where the potential harms associated with not performing the surgery are sufficiently great, it may be ethically acceptable for the parents to give permission for a surgical intervention while the patient is still a minor, and for the surgeon to perform the surgery. When the bodily difference has a low risk of interfering with physical function, and is thus closer to being purely

“cosmetic”—such as in the case of a minor cleft lip, and indeed many cases of hypospadias—the need for ethical scrutiny is greater, as may be the prudence of refraining from surgical intervention until the individual who must deal with the consequences of the surgery is able to make an informed decision (see Earp, 2015a, 2015b, in press; Maslen, Earp, Cohen Kadosh, & Savulescu, 2014).

As stated earlier, the usual recommended timing for performing hypospadias surgery is 6 to 12 months; that is, on children who are not yet able to meaningfully participate in the decision-making process regarding surgery. Is this in the child’s best interests? While there is no clear consensus in the pediatric ethics literature on the kinds of factors that should be considered when ascertaining whether a proposed medical intervention is in a child’s best interests, factors that have been put forth include:

- whether the proposed intervention can be refrained from altogether until the child is able to meaningfully participate in the decision-making process (Kitipornchai & Then, 2011)
- whether the potential benefits of the proposed intervention to the child outweigh the associated risks (Hodges, Svoboda, & Van Howe, 2002; Van Howe, 2013)
- whether the physical and psychosocial risks of not performing the intervention outweigh the risks of the proposed intervention (Kitipornchai & Then, 2011)
- whether the proposed intervention is the most conservative and least invasive option (Hodges et al., 2002)
- the child’s wishes (if known) (Eekelaar, 1994; Kitipornchai & Then, 2011)

In the case of surgery for hypospadias, the potential harms of not operating, and the consequent ‘need’ for surgery, are not straightforward. It is important to note that most hypospadias surgeries performed during childhood are prophylactic in nature. The boy is not currently experiencing any problems related to his hypospadiac penis; rather, the aim of the surgery is to *prevent* such (potential) problems from occurring in the future. As previously noted, the main ‘problems’ hypospadias surgery seeks to prevent relate to urination and fertility. In most cases, however, these ‘problems’ are

psychosocial ones. In the case of fertility, the issue at stake is not whether the man can produce and ejaculate sperm, but rather whether he can impregnate a woman in the manner society deems to be ‘normal’, i.e. through unassisted heterosexual intercourse. Similarly, in the case of urination, most males with hypospadias do not have any difficulty passing urine, although some may need to do so in a sitting position. Sitting down to urinate is not *in itself* an issue, however; after all, biological females do this all the time without any problems (and indeed most men who sit to urinate—as noted above—do not seem to mind very much, regardless of whether they have hypospadias). Rather, hypospadias surgery is aimed at allowing urination in a position society deems ‘normal’ for males (Kessler, 1998). It is therefore complicit with the questionable social norm that males cannot be “real men” unless they stand to urinate (Dreger, 2014; Little, 1998). Moreover, it is worth noting that this social norm does not uniformly apply to all sociocultural communities. There are some communities in which it is considered normal for men to sit or squat to urinate, such that some authors have questioned the ‘need’ for hypospadias surgery in these communities. For example, responding to Culp and McRoberts’ (1968) claim that it is the “inalienable right” (p. 312) of every boy to urinate in a standing position, Anikwe et al. (2000) noted that “since urination in a sitting/squatting position is the rule in [our] community [i.e., of Saudi Arabians] ... [i]t may well be equally valid to state ... that it is the inalienable right of boys in communities such as ours to be sitters or squatters instead of pointers” (pp. 366, 367).

More generally, the justifications put forth in the literature for performing pediatric hypospadias surgery also neglect to consider the appropriateness of using surgery to achieve psychosocial goals (see Earp, 2014 and Little, 1998, for further discussion). For example, if the concern is that a boy with hypospadias might be teased about the appearance of his penis and/or for sitting to urinate, then it should be remembered that it is not his penis that is the ‘problem’ in need of ‘fixing,’ but rather those individuals who are doing the teasing. As Dreger (2006) succinctly commented, “What to do about bullies? Fix *them*” (p. 79, original emphasis). In this respect, the recommendation to operate on

hypospadias in order to avoid social stigma misidentifies the ultimate source of the problem. Rather than surgically altering children's bodies to fit morally questionable social norms, we could instead endeavor to change the harmful social attitudes and practices that lead to such psychosocial suffering in the first place. For a start, we could teach children to be more empathetic and accepting of those who look different and increase accountability for those who tease and bully.

Of course, social change can take many years. We would not propose that the well-being of children with physical differences, including hypospadias, should be “sacrifice[d]...on the altar of [such] noble aspirations” (Parens, 2006, p. xxix). However, it is important to clarify that, although we are skeptical about the moral permissibility of early hypospadias surgery, we are not arguing in favor of doing *nothing*. Should the affected male go on to experience psychosocial issues related to his hypospadias, and want help in dealing with them, these could be addressed using less invasive and less risky psychosocial interventions. In addition to psychological counseling, peer support groups can be a valuable source of emotional support and information for individuals with genital differences (see, e.g., Cull & Simmonds, 2010).

To summarize, parents' and surgeons' concerns regarding the future, possible effects of hypospadias on the affected male may prove unfounded. It is far from certain that a boy with hypospadias *will* go on to experience harm as result of his condition, and, if he does, that this will bother him enough for him to want to undertake the risks and potential complications of surgery. This is particularly evident when one considers the empirical evidence we cited earlier demonstrating that many men with hypospadias do not experience the various ‘problems’ that are commonly attributed to the condition (Dodds et al., 2008; Schlomer et al., 2014). Consequently, an unknown number of boys undergoing hypospadias surgery will never go on to experience problems related to their hypospadias and are consequently being exposed to the risks and pain associated with the surgery for no overall benefit.

### **Best interests vs. other ethical standards**

So far, our discussion has assumed a “best interests” standard for evaluating the moral permissibility of authorizing and/or performing surgery on an individual who is not yet competent to provide valid consent (see Maslen et al., 2014). However, the best interests standard has been criticized on a number of grounds in the pediatric bioethics literature. One criticism is that, due to its narrow focus on the *child’s* well-being, the best interests standard does not allow parents to take into account the interests of other family members, including their own. Another is that the “best interests” standard seems to imply (by its very name) that parents must choose the objectively “best” option for their child—which may be far more stringent than the standards typically used to assess other parental decisions concerning their children’s lives (for example, decisions about where to raise the child, or to send him or her to school) (Diekema, 2004; Gillam, 2010). In light of these criticisms, some ethicists have advanced alternatives to the best interests standard, such as Diekema’s (2004) “harm principle” (for an overview, refer to McDougall & Notini, 2014). Although the harm principle was originally put forth in the context of state intervention regarding parental *refusals* of medically recommended procedures, it has also been discussed in the context of evaluating the ethical permissibility of parental *requests* for procedures which may not be recommended, but could be carried out in a medical context (see, e.g., Van Howe, 2013). According to the harm principle, parents’ decisions regarding their children’s medical care (even suboptimal ones) should be respected, providing they do not place the child at “significant risk of serious harm” (Diekema, 2004, p. 252).

Of course, the harm principle, too, is not without its critics (see, e.g., Ouellette, 2010; Pope, 2011; Van Howe, 2013). One problem with the harm principle is that it fails to specify the appropriate threshold for harm, which opens the door for interested parties to argue, for any intervention, X, that it is “not harmful enough” to warrant the placement of limitations on parental decision making (e.g., Van Howe, 2013; Earp, in press). However, in the case of surgery for hypospadias, it seems to us, such a concern does not really arise. This is because it is more than likely that any parents actually requesting

such an intervention would be doing so precisely because they did, as a matter of fact, think that it would be in the overall best interests of their child (given the range of available options that they perceive as being manageable for them). Given this, it is problematic that the option of *refraining* from surgery—and exploring alternative, psychosocial interventions if necessary—is not typically presented to parents as being a possibility, much less a desirable one. Thus, it is our contention that even parents who do wish to do what is (manageably) “the best” for their child—and who authorize surgery for hypospadias on just those grounds—may inadvertently be subverting their own intentions.

In fact, these parents may even be violating the seemingly less stringent harm principle. That is, they may be placing their child at “significant risk of serious harm.” This is because a male who undergoes medically unnecessary hypospadias surgery during infancy or early childhood has a significant chance of ending up worse off (i.e., harmed overall) than one whose genitals were left intact. In part, this is due to the risk of complications for hypospadias surgery (whenever it is performed) being as potentially great as we have documented, but also because irreversible, early surgery poses a threat to the child’s future autonomy as well (see Maslen et al., 2014), including his (future) ability to make an informed decision about how his “private parts” should look and function (see, e.g., Darby, 2013; Darby 2015). Yet even if one were concerned only about the ‘physical’ issue of surgical risk, it must be acknowledged that “when a surgery is medically unnecessary [and performed before an age of consent], the tolerance for risk/complication [should be] incredibly low and should weigh strongly against intervention” (Ungar-Sargon, 2013, p. 3).

Of course, a decision to refrain from surgery is still a decision, and is one that the child may later disagree with or even resent. Thus, whether surgery is pursued or not, the parent(s) and healthcare professional(s) involved are closing off *some* future option for the child. Specifically, parent(s) who decide in favor of early surgery close off the child’s future ability to make his own decision regarding surgery (and run the risk of the child experiencing surgical complications, resentment, and so on), while parent(s) who refrain from early surgery close off the option for the affected male to undergo the

surgery *during infancy or early childhood* (and run the risk of the child experiencing physical or psychosocial issues related to his hypospadias). It is possible that the affected male whose parents opt against early surgery in order to allow him to make his own decision in the future may later decide in favor of surgery. Others will experience adverse outcomes, and may wonder if the “cure” was worse than the “disease.” The morally relevant difference between these cases, however, is that anyone who does indeed experience problems as a result of hypospadias can always request a surgical intervention to address them, and will be safe in the knowledge that he is undertaking a certain amount of risk to achieve his goals, under his own volition. The adult whose early surgery may not have been desired, and indeed led to poor outcomes including functional difficulties or even more severe medical problems, however, does not have an equivalent recourse (i.e., to “undo” the surgery once it has been done). This asymmetry seems to us to be of central ethical importance.

### **Age-appropriate consent?**

One final consideration concerning consent is that an individual’s ability to provide meaningful, informed permission for a self-affecting operation does not necessarily “switch on” at the age of 18. Indeed, it is possible that a child may express a strong desire to undergo medically unnecessary hypospadias surgery well before he reaches an age of legal majority. For example, a seven-year-old with hypospadias who is unable to urinate in a standing position may experience significant psychosocial distress at not ‘fitting in’ with his male peers. This raises the question: is it ethically justifiable to perform hypospadias surgery with the age-appropriate agreement of a child who is not yet competent to provide legally or ethically valid informed consent, in order to alleviate actual (rather than merely anticipated) psychosocial harm that the child does in fact face? (See Levy et al., 2003; Maslen et al., 2014 for a discussion of assent/consent in minors.) As we highlighted earlier, the first line of defense for addressing issues that are psychosocial in nature should be interventions that are similarly in the psychosocial domain, such as counseling, since these are much less invasive and less risky than

surgery (for further discussion, see Earp, Wudarczyk, Sandberg, & Savulescu, 2013). It must be acknowledged, however, that some children with hypospadias may find such measures to be ineffective. Therefore, surgery may be permissible as a “last resort” in extreme cases before an age of legal majority. However, given the significant risks associated with surgery for hypospadias, including the risk of loss of sexual sensation due to damage to sensitive nerve tissue, we believe it would be prudent not to perform the surgery at least until the individual has reached an age of sexual debut. He would therefore be (relatively) competent to understand the potential long-term risks involved, and what would be at stake in such a surgery.

## **Conclusion**

Given the potential harms associated with hypospadias surgery, empirical evidence suggesting that hypospadias surgery can be performed later in life without significantly increasing the absolute risk of surgical complications to an ethically meaningful degree (see Earp, 2015a for further discussion), and the existence of less invasive and less risky options to address hypospadias-related psychosocial issues (should they arise), we conclude that there are no compelling reasons to justify performing medically unnecessary hypospadias surgery on individuals prior to an age of meaningful consent. After being fully informed regarding the potential benefits and risks associated with hypospadias surgery and alternative options, it should be up to the affected male (upon reaching an age of understanding) to conduct his own risk-benefit analysis and decide for himself whether his hypospadias presents problems for him and whether these problems are severe enough to outweigh the risks and pain associated with surgery.

Parents who are contemplating hypospadias surgery for their children should be informed that not all individuals with hypospadias go on to experience problems (physical or psychosocial) related to their condition, and should be explicitly presented with the option of *no surgery*. They should also be provided with information regarding psychosocial interventions (e.g., counseling and peer support

groups) that the child may take advantage of if necessary. It is time to reconsider the routine referral of children with hypospadias for surgery, and instead allow the affected male to make his own informed decision regarding surgery when he is in a position to express his desires and preferences and understand the important issues that are at stake.

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