

Transitioning patients with hypospadias and other penile abnormalities to adulthood: What to expect?

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Abstract

Hypospadias patients presenting to adult urologists do so with a wide range of symptoms and problems, including urethral stricture (45–72%), lower urinary tract symptoms (with or without stricture) (50–82%), urethrocutaneous fistula (16–30%), persisting hypospadias (14–43%), micturition spraying (24%), ventral curvature (14–24%), urinary tract infection (15–25%), or lichen sclerosus (13%; range 8–43). Many of these men have concurrent complications as the result of multiple operations and a variety of techniques. Patients with childhood repairs performed by a pediatric urologist are often lost to followup during adolescence and will reemerge in adulthood after what appeared to be a successful pediatric single-stage repair, stressing the need for long-term followup and transitional care. One of the major challenges in successful transitional care is that patients can feel traumatized with feelings of hopelessness surrounding their defects, leaving them hesitant to seek care. As well, these patients often have little knowledge regarding the type of repair or original location of the meatus. Urethral stricture is the most common presenting complication and could be related to various factors, with the clear etiology still under debate. These strictures can fall under four categories based on length, location, and previous surgeries. To lessen the difficulties in transitioning hypospadias patients from pediatric to adult practitioners, followup throughout childhood and adolescence for physical examination, as well as uroflowmetry, is mandatory.

Background: Hypospadias

It has been stated that “it is almost impossible to write a satisfactory journal article on failed hypospadias repair.”¹ This is likely because adults presenting with hypospadias complications do so with a wide spectrum of symptoms, abnormalities, etiologies, and most importantly, expectations.² Hypospadias patients presenting as adults often do so with multiple problems, most commonly urethral stricture (45–72%), lower urinary tract symptoms (LUTS, with or without stricture) (50–82%), urethrocutaneous fistula (16–30%), persisting hypospadias (14–43%), spraying of the

urinary stream (24%), ventral curvature (14–24%) urinary tract infection (UTI) (15–25%), or lichen sclerosus (13%, range 8–43%).²⁻⁷ Less common problems include genitourinary pain (10%), genital dissatisfaction (7%), hair-bearing urethral tissue (6–12%), bladder calculi (2–3%), infertility (4%), urethral diverticulum (1–3%), or a buried penis (1%).²⁻⁸ Many patients have several concurrent complications, multiple previous surgeries (median of five), often performed with a wide variety of techniques (3–12 per individual).²⁻⁸

It has also been a widely held belief that “hypospadias complications are common despite modern techniques,” but this may not necessarily be true.⁹ Current evidence suggests a 1 in 300 incidence of hypospadias.¹⁰ With approximately 175 million men in North America, there should be approximately 600 000 hypospadias cases. Given these statistics, why are all “large” series of adult hypospadias complications so small, with typically less than 100 patients in each series?²⁻⁸ Additionally, studies are limited by a lack of a control group who underwent childhood repair without complication or were managed without surgery and never re-presented to an adult urologist. Given the relative lack of studies examining adult hypospadias and the relatively small sample size in these studies, one might surmise that adult complications from hypospadias are related to a small group of particularly complex patients. Or conversely, it is possible that there is limited long-term data on these patients due to the small number of centres providing transitional care between pediatric and adult urologists. On balance then, it seems that pediatric urologists, in the grand scheme of things, likely do a very fine job of repairing hypospadias, but some problems either persist or develop in adulthood.

Challenges with transitioning hypospadias

Despite advances in hypospadiology, followup of patients with hypospadias should extend into adulthood, as a significant portion of adult presentations ultimately require surgical intervention.¹¹ Bracka was one of the first to examine the difficulties when treating adult hypospadias patients. After contacting 213 men over the age of 15 who had undergone

Case

A 23-year old male with a history of proximal hypospadias and moderate (approximately 60 degree) ventral curvature repaired as a child presents to the urology clinic complaining of urinary hesitancy, decreased force of urinary stream, urinary spraying, and post-micturition dribbling. He often has to strain to void and micturition has never been “normal” to him. He denies any urinary tract infections, but states that his urine has a strong smell. The patient currently reports normal erections, but with some ventral curvature (approximately 20–30 degrees) that occasionally impairs penetrative sexual activity.

You have access to the operative report from his hypospadias repair done when he was one year old. The procedure used was a Duckett repair, which involves transection of the urethral plate to correct the ventral curvature and tubularization of an island dorsal preputial flap to create the neo-urethra.

On physical examination, the penis appears straight, the urethral meatus is located at transition between the lower part of the glans and the corona, and there is a substantial amount of redundant skin on the ventral part of the penis.

Urinalysis and urine culture are normal, without evidence of active infection. Uroflowmetry demonstrates a maximal urinary flow rate (Qmax) of 8 ml/sec, with a prolonged flow curve and a post-void residual urine of 50 mL. During voiding, there is a single urinary stream, with spraying at the start of the micturition, and no obvious urethrocutaneous fistula. You also note some bulging on the proximal ventral shaft of the penis during voiding, and a few drops of urine that come out from the meatus after the patient milks his penis with his fingers post-void.

Cystoscopy and urethrogram demonstrates a long penile urethral stricture from the urethral meatus to the proximal penile urethra with no evidence of urethrocutaneous fistula (Fig. 1).

You explain to the patient that he has a penile urethral stricture involving his prior hypospadias repair. Based on his stricture location, length, and etiology, he is very unlikely to respond to endoscopic treatments (at least in the long-term). In addition, there is a lack of urethral support and elasticity of this preputial flap. You discuss three options with the patient.



Fig. 1. Retrograde urethrogram demonstrating a long penile urethral stricture.

previous surgery, he discovered that 74% of patients felt that they had not received adequate guidance, 60% had not even heard of hypospadias, and 40% felt that their personal relationships had been affected as a result of hypospadias.^{12,13} Additionally, many complications, including infertility, sexual dysfunction, and cosmetic dissatisfaction, only become apparent as patients became sexually active.¹⁴⁻¹⁶

It is a common scenario for patients to undergo childhood repair with a pediatric urologist and then become lost to followup, as teenagers cope with their hypospadias issues or try to hide any genital problem that could be a source of embarrassment. It is challenging to assess these patients for the aforementioned reasons, but it is critically important to do so when almost one-third (29%) of them may present later in life with hypospadias-related problems, even after having undergone what was thought to be an initially

successful single-stage repair.²⁻⁴ When these issues finally arise in adulthood, patients are often thrust into a surgical “no-man’s land” between pediatric and adult specialists.¹⁷

Additionally, when patients present as adults, they tend to have multiple signs/symptoms and frequently require multiple surgical interventions.^{2,3,5,9} A synthesis of the existing literature suggests that patients after hypospadias repair should be followed into adulthood, as they may be at increased risk for urethral stricture, LUTS, urethral diverticulum, recurrent ventral curvature, and UTI as adults, but, as a trade-off, patients undergoing childhood surgery likely have improved satisfaction with regard to genital appearance and may have improved sexual function.^{2,11,18}

Finally, another major challenge when transitioning hypospadias patients who underwent childhood repair into adult urology practice is the fact that some of these young men

Option 1: Multi-staged urethroplasty: In this setting, part or the entire original urethroplasty would be excised and the ventral surface of the penis replaced by oral mucosal grafts harvested from the lower lip or cheek (due to unavailability of preputial mucosa), depending on the extension of the defect to be covered (graft size). This would be done in a first stage, with second stage tubularization of the grafts six months later. With a staged urethroplasty, ventral curvature could be simultaneously addressed. Additionally, the graft is sutured to the corpora cavernosa, once healing is completed, graft fixation to the corpora is achieved, which prevents formation of future urethral diverticulum (Fig. 2). Urinary spraying is also avoided by carrying out the urethroplasty to the tip of the glans, with creation of a slit like meatus (Fig. 3).



Fig. 3. Final cosmetic result of the same patient. Slit-like meatus at the tip of the glans and symmetric mucosal collar. Appearance similar to a circumcised penis.

Option 2: Single-stage urethroplasty: This would involve reconstructing the urethral stricture with oral mucosa in using a single-stage technique. Based on the patient's lack of urethral support and density of the stricture this would be challenging endeavor and would fail to address the meatal location or any residual ventral curvature.

Option 3: Perineal urethrostomy: Based on the stricture length and normal bulbar urethra, this patient would be a potential candidate for a perineal urethrostomy. This would likely establish stricture-free voiding long-term, but given that the patient's young age, could be viewed as a last resort for this patient's specific scenario.



Fig. 2. Oral mucosal graft(s) harvested from both cheeks and lower lip to cover the long distance between the penoscrotal junction and the tip of the glans in a young adult. Once the grafts mature they will be ready for tubularization six months post-first stage.

who have had multiple failed repairs are not only traumatized by their previous bad experiences, but have a sense of hopelessness, as they were told that their defects were too complex to be corrected and that they should simply accept their fate of having to live with either a proximal urethrostomy (peno-scrotal junction meatus) or a "bent" penis with multiple scars and suture tracks. According to the Hypospadias and Epispadias Association (HEA) president (personal communication at the 2013 International Hypospadias workshop in Las Vegas), these multi-operated men, erroneously referred to as "hypospadias cripples" (Fig. 4) in the literature, have been hesitant to look for help from general adult urologists due to the discouragement they received from previous practitioners.

Managing adult hypospadias complications

Assessing adult hypospadias can be challenging, as many patients have no record of how many repairs they had, what type of repair, or the original meatal location, resulting from a delay from surgery until presentation. On examination, adult hypospadias will often have an abnormally located urethral meatus, an attenuated glans cleft, deficient ventral tissue layers (including spongiosum and Dartos), ventral curvature (usually underappreciated), and a dorsally hooded foreskin (if previously untreated). Another important consideration is increased rates of lichen sclerosus even in patients who did not undergo any repair.⁴ The "hypospadias cripple" represents an extreme of these findings, with a more typically deficient glans cleft, multiple scar lines and suture tracks on the penile shaft, a paucity of penile skin, thin ventral



Fig. 4. 13-year-old boy with multi-operated hypospadias (hypospadias cripple).

Dartos typically “soldered” to the urethral repair, and often ventral curvature.¹⁹

In general, adult hypospadias surgery is generally successful, with reported success rates of 75–88%.^{2-5,7-9,20} However, success rates of any single surgery are approximately 50% and thus on average, two or more operations are required to address the adult complications of hypospadias.^{2,4,9} Additionally, these surgeries incur a 26–68% risk of complications.^{2-5,7-9,20} Moreover, revision surgery carries a two-fold risk of complications, likely because the ventral penile skin and Dartos fascia are deficient and poorly vascularized, resulting most commonly in urethral stricture, urethrocutaneous fistula, ventral curvature, and glans dehiscence.^{2-9,20} It is important that all adult urologists be aware of the broad spectrum of symptoms adult hypospadias patients may present with so that prompt recognition of these problems and proper diagnosis can be established, with subsequent referral to a specialist.

In contrast, some pediatric urologists who also have an adult practice have reported encouragingly similar results to those obtained in young children when using newer surgical techniques, generally in the setting of prior untreated hypospadias.^{6,21}

In an attempt to stratify hypospadias complications into different risk groups to allow for transparency and consistency

of reporting, Ching and colleagues presented a classification system for adult hypospadias based on the status of previous surgery. Category I corresponded to patients who has undergone multiple surgeries with significant scarring and tissue loss; Category II involves those with delayed complications after an initially successful childhood repair; and Category III includes patients with no previous repair.⁴

Urethral stricture and hypospadias

Urethral stricture has the highest occurrence in hypospadias patients presenting as adults.^{2,22-24} Urethral stricture has been reported to occur in up to 10% of patients with hypospadias and, in fact, hypospadias is the most common cause of iatrogenic stricture in men under the age of 45.^{22,25} Although the exact incidence remains unclear, development of strictures may be related to the use of preputial flaps, tubularized tissue, or simply related to managing severe, more complex proximal hypospadias cases.¹⁷ The cause of urethral stricture and other hypospadias complications is the source of some lively debate. Some argue that stricture occurs in childhood as a consequence of a poor technical repair or poor technique selection resulting in infection, hematoma, extravasation, and tissue ischemia that is initially undetected and presents later in life.²⁶ Others speculate that hypospadias repairs deteriorate in a *de novo* manner during puberty, with the onset of sexual activity related to the absence of the corpus spongiosum.³ There may also be disproportionate growth of the urethra and surrounding spongiosum, which, over time, may result in urethral stricture. No common factor has been elucidated thus far, and like most puzzles in life, multiple factors likely play a role.

In any event, patients often tend to present insidiously, with a long-standing history of urinary hesitancy, straining to void, and intermittency, with many having concurrent episodes of UTI or acute retention. Endoscopic treatments generally offer little chance of cure, given that urethral dilation and urethrotomy are least successful in penile strictures and strictures >2 cm in length, of which hypospadias strictures are most typically.²⁷ Definitive treatment should often be entertained, especially in young adults of reproductive age.

There are several important aspects to patient evaluation. As mentioned, physical examination is often a key aspect in surgical decision-making, with the size of glans and glans cleft, presence of lichen sclerosus, ventral Dartos thickness, visible scarring, and palpable fibrosis playing a role in treatment. Patients require retrograde urethrogram with or without voiding cystourethrogram to delineate the length of the stricture and concurrent pathology. Cystoscopy using pediatric instruments often plays a crucial role in optimal assessment of these patients with minimal urethral manipulation.

Urethral stricture in adult patients with hypospadias can be difficult to classify in a meaningful way, but generally

speaking fall into one of four categories based on length, location, and previous surgeries. The first and most common is a long “pan-penile” stricture involving the majority of the penile urethra in the setting of previous (and often multiple) hypospadias surgery. The second group comprises a “junctional stricture,” a stricture of variable length at the junction of previous hypospadias repair and native urethra. Another (third) group is that of an isolated and often short bulbar urethral stricture after hypospadias repair. The exact cause of these strictures may be related to the past use of perineal urethrostomy for urinary drainage following hypospadias repair, repeated instrumentation, or associated congenital or possibly ischemic bulbar urethral stricture. The fourth group is urethral stricture developing in the setting of previously untreated hypospadias. These can typically be managed in a single stage using contemporary single-stage hypospadias techniques such as a penile fasciocutaneous flap; decision-making is based on the health of the urethral plate.

Adult hypospadias represents a complex clinical entity requiring further interaction between adult and pediatric urologists. Although it seems intuitive, there has yet to be a study prospectively determining if the urethroplasty success rate for hypospadias-related strictures differs from that of a matched cohort of idiopathic strictures. In the future, multi-institutional collaboration between adult and pediatric urologists will likely be required to answer key clinical questions.

Other penile abnormalities: Epispadias and congenital curvature

Although hypospadias represents the most common congenital penile deformity, other penile abnormalities warrant mention, including congenital curvature of the penis and epispadias complex.

Typically, in children and young adults, ventral penile curvature is associated with hypospadias; however, penile curvature may exist in adolescents and young adults without any obvious hypospadias. This is referred to as congenital curvature of the penis and these curvatures are usually ventral (or ventrolateral) and rarely dorsal. Usually, patients are healthy men that present in adulthood, but describe a lifelong history of penile curvature. Post-puberty and after unsuccessful attempts at sexual activity, most of these men will seek medical attention. Given that most young males do not discuss their sexual function with their parents, most of these cases do not seek medical attention during adolescence. In the case of congenital curvature, the urethral meatus is normally situated, but there can be subtle findings suggestive of hypospadias, including attenuation of the Dartos and other ventral tissue layers often with below-average penile length.²⁸ In contrast, the true (non-hypospadias) congenital curvature is thought to be caused by an asymmetrical

expansion and hyperelasticity of the dorsal tunica albuginea, resulting in a curvature on the contralateral ventral side. In these patients, penile length is not deficient and is, in fact, quite robust. Reconstruction in these patients is performed when the deformity precludes sexual activity and is best achieved by plicating the dorsal tunica albuginea rather than a ventral incision and grafting. Dorsal plication is preferable in these patients to reduce the possibility of graft-induced veno-occlusive erectile dysfunction.²⁸

While patients with congenital curvature of the penis often present as an isolated genital deformity, in contrast, patients with epispadias-related concerns often have a broad spectrum of problems and a long-standing history of prior urological interventions. These patients present with a combination of genital dissatisfaction, erectile dysfunction, ejaculatory dysfunction, reduced penile length, buried penis, urethral stricture, urethrocutaneous fistula, and/or infertility. Similar to hypospadias, these patients treated as children should be followed until adulthood, as they have a very high likelihood of requiring ongoing surgical and psychological care related to both their urinary tract and genital function.²⁹ As adults, these patients require an interdisciplinary and individualized approach with surgery, often best left to those with specialized training in genitourinary reconstruction.

Advice for patients about to transition/have transitioned

Over the last few years, it has become well-established among pediatric urologists that hypospadias patients who underwent childhood repair must be followed until adulthood. Several academic centres, such as the Children’s Hospital of Philadelphia (CHOP), Lurie Children’s Hospital in Chicago, Boston Children’s, and McMaster Children’s Hospital, among others, have started prospective hypospadias data collection. As a result of implementing these long-term databases and adopting rigorous protocols, hypospadias patients are now routinely instructed to return for followup after puberty. At that point, these teenagers will be reassessed with detailed physical examination and uroflowmetry. In addition, completing the Hypospadias Objective Penile Evaluation (HOPE) questionnaire will allow for a more objective and generalizable evaluation of the repair, which can then be sent to the adult practitioner who becomes involved in that patient’s care.³⁰

Teenagers with hypospadias who will be transitioning into an adult urology practice need to have ownership of their condition and be aware of what happened in the past in terms of number of previous surgeries and type of surgical technique used. This type of pediatric patient maturation will allow for a smoother transition to adult specialists, even when their medical records are not available. As proof of

this shift in followup care of hypospadias patients, a recent survey of parents of hypospadias patients showed that more than 90% of the respondents would fully disclose all the details (including type and number of surgeries) to their children so that they would be aware of what happened to them and be able to participate in any future decisions involving their repair.³¹

Case followup

Our case involves a patient with a long penile stricture in the setting of previous hypospadias repair (Group 1). In these cases, the health of the spongiosum and Dartos is critical for a successful outcome of surgery. In the setting of distal hypospadias strictures, there can be a robust glans, healthy corpus spongiosum, and Dartos fascia. These cases may be managed in a single stage using oral mucosa; the biggest challenge is to create a normally situated and shaped urethral meatus. If the problem extends more proximal than the coronal margin or multiple surgeries have been performed, the corpus spongiosum and Dartos are typically deficient. In these cases, a reliable and satisfactory surgical result typically requires a multistaged approach.^{17,32-34} Suprapubic diversion is also commonly employed during the second stage to avoid having any urethral catheter-related glans complications.

In summary, these are complex cases that should be referred to an experienced adult reconstructive urologist, so that timely and proper care can be offered to these patients. Following surgical reconstruction, ongoing followup by an adult urologist is necessary to ensure ongoing success in terms of urinary and sexual function, in addition to surveillance for possible complications.

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