13 The Surgical Approach to Two-Stage Hypospadias Repair

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13.1 Background, Epidemiology, and Introduction

Hypospadias is amongst the most common congenital male anomalies, with an incidence ranging from 1:150 to 250 of male births [1, 2]. The vast majority of boys present with a mild, distal variant, and surgical reconstruction, when indicated, is highly successful [3, 4]. Proximal hypospadias is reported to occur in 10–25% of patients with hypospadias, and its increased severity presents several unique management challenges to the surgeon [5, 6]. In addition to worsened curvature and shorter penile length, anatomic studies have found less elastic tissue and relative androgen resistance in boys with proximal hypospadias, suggesting more dysplastic tissue which further complicates surgical reconstruction [7–9]. Although traditionally thought to have a complication rate ranging up to 25%, recent publications indicate a significantly higher complication rate for proximal hypospadias repair, with secondary surgery rates as high as 70%, and many of these complications presenting after puberty, and into adulthood [10–13]. This high complication rate demands that we focus our efforts on identifying risk factors to improve our outcomes for these patients. For these reasons, we consider proximal hypospadias a different disease process when compared to distal hypospadias, one that warrants unique considerations for assessment, management, and follow-up.

13.2 Indications and Considerations for Surgical Repair

Proximal hypospadias can present in a variety of ways. One is the classic appearance with obvious severe curvature and a proximal meatus (Fig. 13.1). The surgeon must also be aware of other variants. A boy can present with a distal meatus, but a more severe proximal variant is identified in the operating room (Fig. 13.2). These pictures suggest hypoplastic ventral penile shaft skin and severe ventral penile curvature after degloving. This boy would benefit from a two-stage repair.

Because of these variants, the surgeon should never enter the operating room with a rigid plan for a hypospadias repair, instead of keeping their plan fluid as the anatomy unfolds in the operating room. Indications for utilizing a two-stage repair at our institution include severe penile curvature (defined as 30 degrees or greater after penile

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Fig. 13.1 A boy with proximal hypospadias. The meatus is located at the penoscrotal junction, there is severe curvature and severe penoscrotal transposition

degloving) and/or a lack of penile tissue that would prohibit a tension-free repair. A step-wise approach should be used to assess penile curvature, beginning with penile degloving and excision of chordee tissue. Artificial erection is then performed. This can be challenging in this patient population, especially if penile concealment is present. One must be aware of proximal penile curvature that may be obscured with a tourniquet. If a ventral corporal lengthening procedure is required, we feel that a staged repair should be performed, regardless of the initial location of the urethral meatus. We believe that the priority is to correct the penile curvature at the first stage, while at the same time establishing a healthy bed of tissue on the ventral penile shaft for the second stage repair and urethroplasty. As the management of a simple distal variant contrasts greatly with the approach and complication rate for a two-stage repair, the surgeon must educate the parents properly at the time of the office consultation, particularly given the longer recovery process and the possibility of requiring a secondary procedure.

The traditional approach of grading the complexity of hypospadias based solely on the location of the meatus is an inadequate exercise. Staging the phenotype is a vital exercise in understanding the full spectrum or the degree of severity of hypospadias and allows us to convert the art form of hypospadias into a common language for comparison sake. Staging systems such Glans-Urethral Meatus-Shaft (GMS) score objectively assess the penile anatomy with measurement of the glans width, assessment of the quality of the urethral plate, the degree of chordee, and the location of the urethral meatus, and correlate a higher score with an increased complication risk [14]. We believe that the universal acceptance of the GMS or a similar standardized evaluation system such as the Hypospadias International Society scoring system will advance our understanding of hypospadias in hopes of improving our outcomes. Regardless of the method used, one must incorporate the location of the meatus, the quality of the ventral penile shaft tissue, the degree of penile curvature, and any associated anomalies in their assessment.

13.3 Preoperative Assessment

As mentioned above, the office assessment begins the process of evaluating the degree of penile curvature, the quality of the ventral penile shaft skin, the width and quality of the glans, and the urethral plate to determine the potential need for a staged procedure. Patients must also be screened for the presence of undescended testes as this may indicate the presence of a disorder of sexual development, particularly in patients with proximal hypospadias. This will guide the preoperative discussion with the family, including the possible need for a karyotype or additional testing to identify a complicating underlying disorder. Given the high risk for postoperative complications after a proximal hypospadias repair, it is imperative that the family is counseled on the potential for a secondary procedure as in our experience this discussion in the preoperative period guides family expectations.

A glans width <15 mm has been associated with an increased risk of urethroplasty complication development [15]. Supplemental testosterone administration prior to the procedure can increase

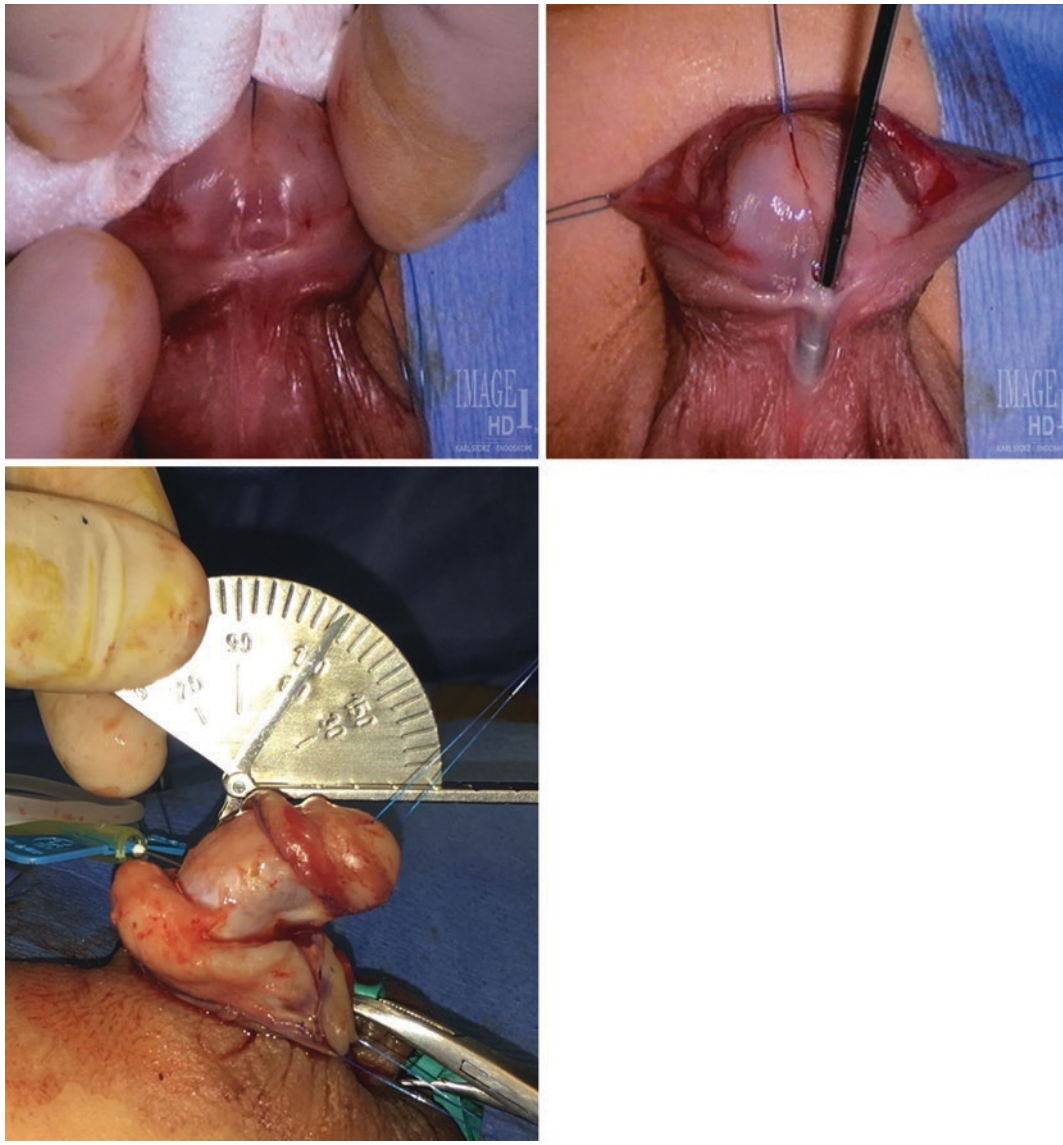

Fig. 13.2 A boy that presented with a distal urethral meatus but after further assessment in the operating room was identified as true proximal hypospadias with poor ventral penile tissue

glans width in pre-pubertal boys [16, 17]. Its use is controversial as some feel that there is little benefit or even a risk of worse surgical outcomes when administered [18]. Others have raised concerns about a blunted response to testosterone in those with a severe form of proximal hypospadias due to androgen insensitivity [9]. Our personal experience does not mirror the aforementioned negative concerns regarding testosterone supplementation, and we feel that testosterone enhances our technique with no increase in complications compared to the literature.

Our use of testosterone in this patient population has changed over time and is patient dependent. Our typical practice is to administer preoperative testosterone 5 and 2 weeks prior to the planned surgery date to boys with a glans with <15 mm. In patients with a small penis with limited penile shaft skin, we administer preopera-

tive testosterone prior to the first stage repair. If glans width remains an issue at the second stage, this can be repeated.

Finally, the preoperative consultation is an important time to center patient and parental expectations. The ultimate goal in any penile reconstruction is to have a functional penis that will act as a sufficient conduit for normal voiding and to achieve full sexual potential in adulthood. Achievement of the so-called perfect penis in distal hypospadias, with a slit-like meatus within the distal glans is much more attainable than it is in proximal hypospadias [3]. Given the degree of dysplasia of the penile tissue in proximal hypospadias, it may not be possible to achieve the same cosmetic results attainable in distal hypospadias. Placing the urethral meatus within the coronal margin, although not the normal anatomic location, may result in a normal functional penis and forestall complications such as meatal stenosis that may complicate attempts to place the meatus within the glans in cases where the glans is underdeveloped. We pursue the approach of placing a urethral meatus in a coronal or proximal glans location in the patients of a narrow urethral plate and glans that otherwise would prohibit glans closure. Consideration must be given to balancing what we may think is a "perfect penis" as surgeons and what ultimately will result in the optimal outcome for patients. Alternatively, we may plan for a dorsal inlay graft at the time of the second stage of the repair to widen the glans in order to advance the meatus into a more distal location [19]. These possible outcomes are conveyed to families in order to set expectations.

13.4 Elements of Surgical Repair

The sequence of surgical decision-making begins with an assessment in the operating room prior to making an incision. The location of the true urethral opening and the quality of the associated ventral shaft tissues is assessed to determine if a more severe variant is present. Although often obvious on the exam, proximal hypospadias can masquerade as distal hypospadias with severe penile curvature and hypoplastic ventral shaft skin. The surgeon must recognize this in the operating room to ensure that the appropriate repair is performed.

The circumcising incision is made, and the penis is degloved to the penoscrotal junction. Care should be taken to develop a mucosal collar, rotating redundant dorsal hooded foreskin to the ventrum as this will aid in ventral shaft skin coverage and yield an improved cosmetic result [20]. One can also leave a shorter mucosal collar in order to preserve inner preputial skin for ventral shaft coverage. It is important to resect all dysplastic tissue on the penile shaft during the degloving process. Once degloved, an objective assessment must be performed as the degree of penile curvature is a major deciding factor between proceeding with a single versus two-stage repair (Fig. 13.3). After degloving and resection of chordee tissue, an artificial erection is performed, and the degree of penile curvature is determined. We use a goniometer to objectively measure the degree of curvature, although we encourage any objective measure of curvature to obtain the most objective assessment [21]. Most surgeons would perform a dorsal plication for curvature <30° [22]. If curvature persists after plication or is ≥30°, the next step would involve either mobilization or division of the urethra. Persistent curvature ≥30° at this point would warrant a corporal lengthening the procedure by incising the ventral corporal tunica albuginea and placing a graft to cover the defect [23]. Options include autologous tissue (dermal graft [24], tunica vaginal flap [25] or graft) or non-autologous tissue (Alloderm, small intestinal submucosa (SIS) [26, 27]). Alternatively one could perform ventral fairy cuts or several ventral incisions through the tunica albuginea to release the ventral curvature without an additional graft or flap insertion to close the gap [28]. Completion of the first stage of the repair is outlined in the next section and is dependent upon the approach taken.

Alternatively, a corporal lengthening procedure utilizing a tunica vaginalis flap, followed by urethroplasty and glansplasty can be performed to complete a single-stage repair utilizing inner

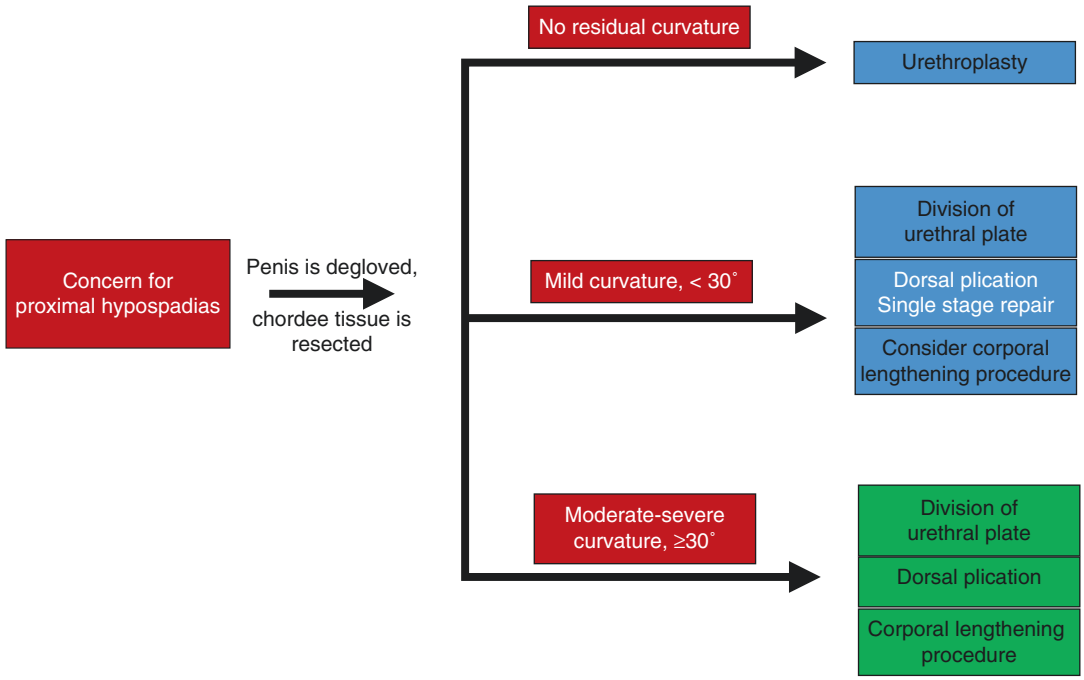

Fig. 13.3 Algorithm for assessment and repair of penile curvature

preputial tissue as a flap, or tubularizing a mobilized urethral plate [25]. We have concerns about the healing potential of a simultaneous graft overlying a flap and feel that this will place both tissues at risk for scarring and poor wound healing [29]. Our preferred approach in the setting of persistent curvature ≥30° is the two-stage repair in order to maximize the healing potential of both the corporoplasty and the subsequent urethroplasty.

Regardless of the approach to corporal lengthening, it is important to ensure that it is corrected fully at the time of the first stage, which can be verified with repeat artificial erection after correction. If necessary, a plication can be performed at the time of the second procedure to correct minor recurrent curvature. The timing between surgical reconstructions should range from a minimum of 6 months to allow sufficient graft take. We often allow 8–12 months between procedures, with interval exams to ensure satisfactory graft uptake, to monitor for recurrent curvature, and to determine if the ventral skin is supple enough for second-stage urethroplasty closure.

13.4.1 Surgical Techniques for Two-Stage Hypospadias Repair

Modern technical approaches to the two-stage repair of proximal hypospadias can be divided into three categories, each separated by technical variations on the approach to urethroplasty. The first procedure addresses penile curvature with the differences outlined below.

The Bracka two-stage repair utilizes a free graft, harvested from either the inner prepuce or the buccal mucosa, which is placed into the ventral penile shaft for eventual closure at the second stage (Fig. 13.4) [30, 31]. The Staged Tubularized Autograft Repair (STAG) is a variation of this technique and has been popularized more recently [28]. The urethral plate is divided, and a midline incision is made within the glans to act as a receiving bed for the graft. The graft can then be quilted into place on the ventral penile shaft to prevent hematoma formation and to facilitate graft take. The second stage is performed at least 6 months later, during which a U-shaped incision, similar to the Thiersch–Duplay approach, is made and the

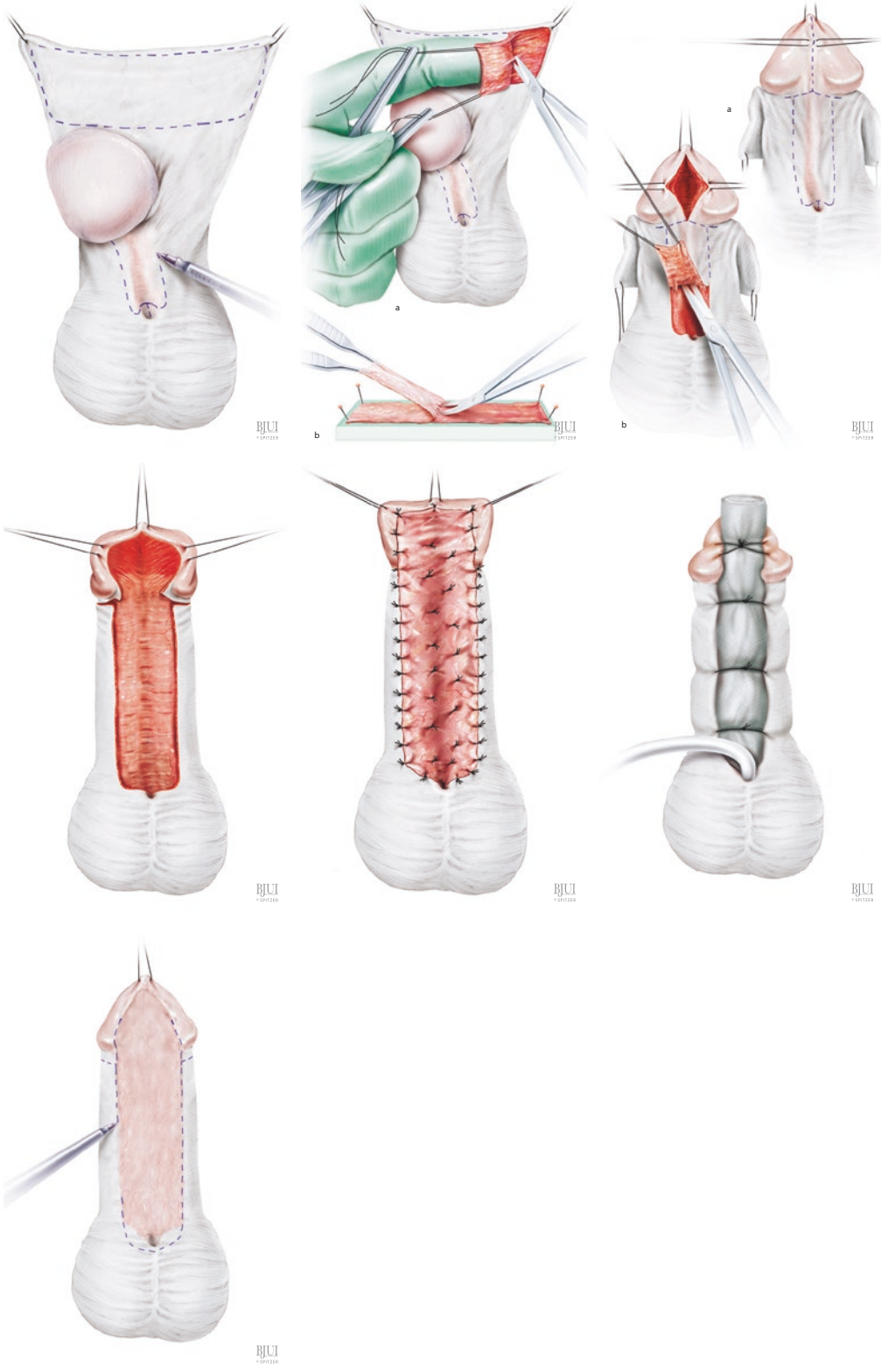

Fig. 13.4 Bracka two-stage repair

urethra is tubularized, and the glans is closed [31]. Multilayer closure is performed to maintain vascular flow to aid the healing process. This technique is ideal for patients in whom a previous circumcision was performed, or a lack of foreskin is present to perform an alternate repair. With the STAG repair, an inner preputial free graft is harvested in lieu of a buccal mucosa graft. In the setting of severe penile curvature requiring a corporal graft procedure, we do not recommend this approach as overlap will impede graft uptake [29]. Graft contracture, particularly over open corporotomies, has been reported to increase graft loss and would require additional procedures.

The Byars flap procedure utilizes redundant dorsal preputial skin, which is incised in the midline dorsally and rotated ventrally at the first procedure, as the scaffold to form the urethra (Fig. 13.5) [32, 33]. Similar to the Bracka repair, at the second stage, the neourethra is closed by making a long-U-shaped incision with glansplasty using a standard Thiersch–Duplay technique. Some key technical components include a water-tight closure and establishing a lumen of equal caliber throughout the length of the urethroplasty. A two-layer urethroplasty, followed by multiple layers of adjacent dartos tissue as an overlay is required to ensure that the neourethra

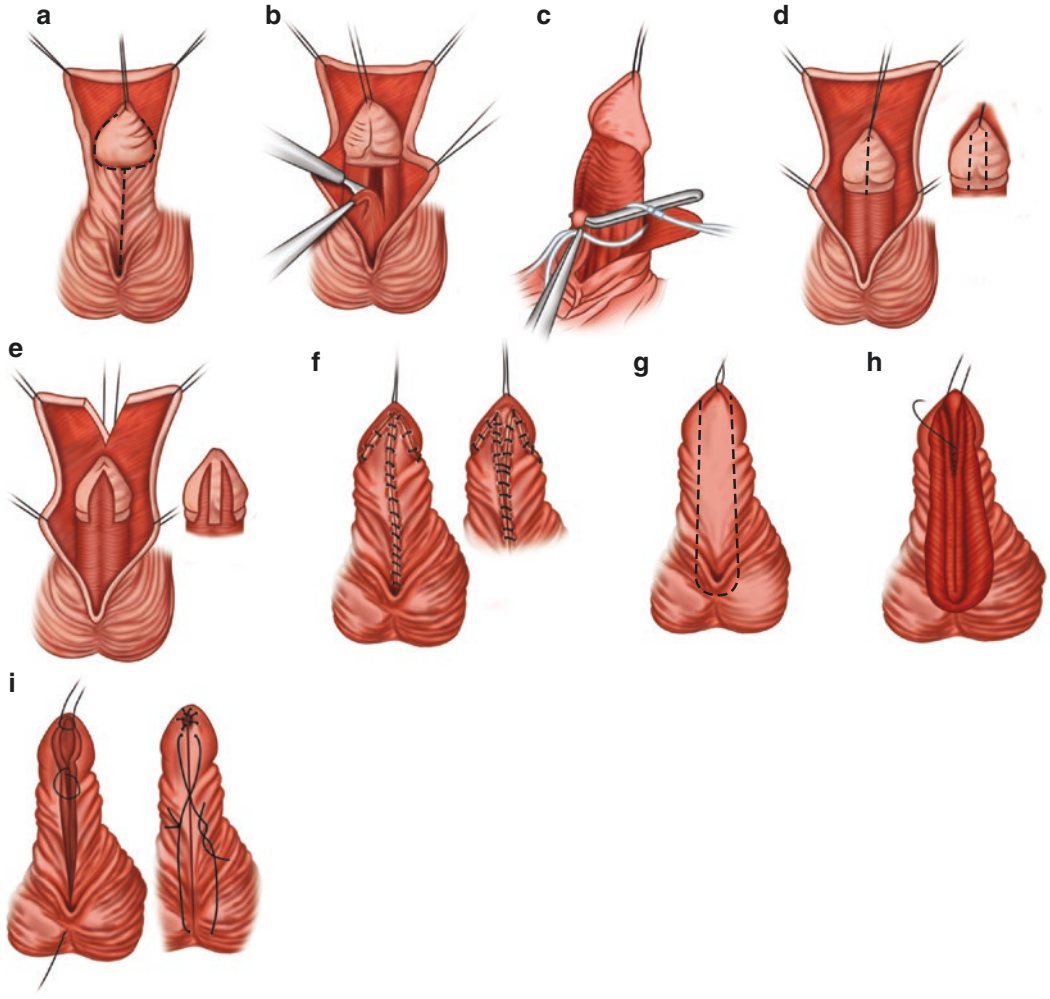

Fig. 13.5 2-stage Byars flap repair. The penis is marked and circumscribing incision is made (**a**). The penis is degloved and the urethral plate is divided (**b**). Artificial erection is performed to confirm that curvature is corrected (**c**). The glans can be incised to allow for flap placement as part of the first stage repair (**d**, **e**). The flap is secured in the mid-line of the penile shaft (**f**). AT teh time of the 2nd stage repair, a ventral U shaped incision is marked and incised (**g**). The urethroplasty is performed (**h**), followed by skin closure (**i**)

maintains adequate blood supply. In particular, establishing a supple dartos bed overlying the corporoplasty at the first stage will provide sufficient vascular flow to allow safe urethroplasty at the second stage.

The third approach involves correction of the penile curvature, closure of the distal urethra and glans, and rotating an interposing flap or graft to replace the urethra at the first stage repair (Fig. 13.6). For the distal urethra, one approach

Fig. 13.6 Patient with a severe, proximal hypospadias (**a**). Penis is degloved (**b**). An inner preputial island flap is developed (**c**) and tubularized (**d**). The glans is incised and the tubularized flap is tunneled into the glans (**e**, **f**). The dorsal penile shaft skin is incised in the midline, creating Byars flaps (**g**). The Byars flaps were rotated ventrally to close the penile shaft skin, leaving a gap between the native urethral meatus (proximal) and the distal, tubularized urethra (**h**)

includes a tubularized incised plate type of closure urethra and glans [34]. Alternatively, in the Ulaanbaatar repair, a transverse island interposition tube can be developed from the preputial skin, which is tunneled into the distal portion of the glans [35]. For both of these variants, the urethra is matured into a proximal position, and Byars flaps are rotated ventrally to allow for completion urethroplasty of the interposing portion of the urethra at the second stage of the closure.

13.4.2 Postoperative Care and Follow-Up

The immediate postoperative period consists of urethral diversion with a stent and compressive dressing. In our practice, a compression dressing is left for 72 h, after which time the stent is removed with the compression dressing, and petroleum jelly is applied to facilitate grafts and/or flaps healing. If a Buccal mucosa graft is performed, we suture a compression dressing 5–7 days directly over the graft to minimize the risk of hematoma formation and subsequent graft loss. A suprapubic tube can be placed, if desired, to maximize urinary diversion, particularly in the setting of a buccal mucosa graft. We utilize this approach more commonly in toilet trained children.

Urinary diversion at the time of urethroplasty is typically done for 7–14 days. Prolonged stent duration has been shown to decrease complication rates although one must determine if this is applicable for all patients and must be balanced against the risk for infection, patient discomfort, and the potential for stent encrustation [36]. Antibiotic prophylaxis is typically given while the stent is in place, although this is controversial [37].

Flap or graft care is a key aspect of the postoperative period. Topical petroleum jelly application with a digit and manual pressure to the ventral penile shaft can help prevent contracture and uneven healing. We initiate this process within 2 weeks of the procedure and continue for 6–8 weeks after the first stage repair. Topical steroid application can be performed if there is skin contraction or other healing concerns.

A minimum of 6 months should separate the first and second stage surgeries to allow sufficient graft and flap uptake and neovascularization and the surgeon should not hesitate to wait an additional time to ensure proper healing has occurred. A critical evaluation is a thorough postoperative assessment to examine for appropriate skin fit, monitoring for persistent curvature, and to determine the viability of the ventral penile shaft skin for potential urethroplasty. The viability of the graft and/or flaps should determine if additional procedures are necessary and the appropriate timing for the next procedure.

13.4.3 Outcomes with Brief Pertinent Literature

Successful outcomes in distal hypospadias are high, ranging from 87 to 95% [3, 4, 38]. Unfortunately, proximal hypospadias reveals a much higher complication rate—and complications may become manifest several years after the initial repair [10–13, 39]. The degree of associated tissue hypoplasia and severity of the phenotype increases with more proximal hypospadias likely explains the higher predilection for complications with proximal hypospadias, since the smaller width of the glans and longer length of the urethroplasty are contributors to this increased complication rate.

Comparison of technical approaches to curvature correction is difficult. Curvature assessment is often subjectively assessed by the surgeon and is categorized into mild, moderate, or severe variants which limits our ability to compare techniques across institutions. Only recently has the focus shifted toward a more objective approach to defining the degree of curvature, which if incompletely corrected, has the potential to create long-term concerns or complex surgical reconstructions [40]. One study directly compared dorsal plication versus penile lengthening procedures to correct curvature in patients with proximal hypospadias [27]. The rate of recurrent curvature at 17 months of follow-up was higher for dorsal plication versus penile lengthening (27.9% vs 9.4%, $p = 0.03$), with insufficient shaft

skin the etiology for the latter group suggesting adequate correction of corporal disproportion with a lengthening procedure.

Regardless of technique, the rate of recurrent penile curvature for the two-stage repair of proximal hypospadias in the current literature is thought to be relatively low, ranging from 0 to 5% of patients [10–13, 28]. Two-stage repair with corporoplasty was associated with increased penile length and improved cosmetic results [41]. Penile length is an important aspect in the repair, particularly with proximal hypospadias, as a survey of adult patients after infant repair reveals concerns primarily for shortened penile length [42].

The Bracka repair, or technical variations thereof, has an overall reported complication rate of <25% [28, 41, 43]. The most common complication encountered was glans dehiscence and/or urethrocutaneous fistula. The rate of recurrent penile curvature is low. Additional layers of closure, increasing the local blood supply, have been shown to decrease the complication rate [44]. A comparison of four approaches to proximal hypospadias repair, including tubularized incised play, island preputial onlay, and a two-stage Bracka repair, found that the repair approach did not correlate with complication development [41]. Concerns for a skin graft incorporation over a corporal graft can be circumvented with dorsal plication techniques combined with proximal urethral mobilization to correct the majority of curvature [45]. Otherwise, if severe curvature persists, a corporal graft should be performed, and this would increase the risk for skin graft loss.

Several reports have recently indicated a high complication rate associated with the Byars flap two-stage repair [10–13]. Complication rates ranged from 30 to 70%, with urethrocutaneous fistula representing the most common complication noted, followed by glans dehiscence. Urethral diverticulum is also a potential complication, thought to occur as a result of a combination of a lack of spongiosum and lack of fixation of the flap onto the ventral penile shaft at the time of the urethroplasty. Compared to the graft utilized for the ventral penis in the Bracka repair, the theory would suggest that the improved vascularity of the preputial flaps should improve wound healing. However, the high complication rates suggest a yet unknown cause for complications. One technical modification, the advancement of the urethra to the coronal margin, may prove to decrease the complication rate after the second stage repair.

Although mostly represented by case series, the two-stage approach incorporating distal urethra closure and correction of curvature at the first stage has reportedly favorable outcomes [34, 35]. The authors have not seen a comparative increase in concerns for distal urethral stricture or stenosis, a concern given the increased likelihood of a small glans at the time of the initial procedure and the lack of urine flow through this area after the initial procedure.

To the best of our knowledge, no direct comparison of the three approaches above has been performed that would determine the ideal approach to proximal hypospadias with severe ventral penile curvature. Regardless of the approach taken, long-term outcomes in the literature are lacking and only raise the possibility that more complications will be captured as these patients are followed over time [46, 47]. We strongly agree with this principle and have modified our follow-up protocols accordingly to extend follow-up for these boys well into the pubertal period.

13.4.4 Future Advances and Directions

Much work remains to be completed in the advancement of surgical reconstruction of severe hypospadias [48]. These efforts need to focus on improving the relatively high complication rate, extending our routine follow-up into adulthood, and using long-term analysis to guide patient and parental expectations of the repair.

A survey of the literature reveals a limited number of studies with patients undergoing hypospadias repair with follow-up that into puberty. This severely limits our ability to determine the real outcomes and complications of our

repairs. Extending our follow-up will allow us to answer several persistent questions concerning hypospadias repair. The first is to determine the ideal approach to correction of penile curvature, specifically which approach maintains effectiveness through the accelerated penile growth during the pubescent period and into adulthood. If a boy has a straight penis as a child but has painful, curved erections as an adult, then we would argue that we have failed that patient. If it is determined that dorsal plication in a patient with 20° of ventral curvature is associated with a higher rate of recurrence as an adult, then this approach must be abandoned. In a similar fashion if corporal grafting results in a higher rate of erectile dysfunction due to increased venous leak as an adult, then we must consider this when a repair is performed.

In addition to surgeon assessment of outcomes into adulthood, the patient-reported outcomes must also become increasingly important. Unification of patient-reported outcomes with surgical outcomes will advance our understanding of our surgical penile reconstruction. Questions to be answered include the correlation of cosmetic features with functional outcomes. Are patients happy with a urethral meatus located at the coronal margin in the setting of proximal hypospadias? If the patient is able to stand to void with a strong urinary stream and has straight erections sufficient for intercourse, will it matter if the meatus is slightly lower than normal? This question is an important one in the sense that the creation of the so-called perfect penis, with a slit-like meatus within the distal glans, in the setting of proximal hypospadias may be detrimental to the patient. Extension of the meatus into the distal glans may be responsible for, at least in some part, the high complication rates we are seeing in proximal hypospadias. We theorize that the relative rigidity of the spongiosum of the glans acts as a bottleneck for urine passage, increasing proximal voiding pressure, and driving complications such as a urethrocutaneous fistula and diverticulum, amongst others. Finally, as proximal hypospadias is associated with severe tissue dysplasia, we need to determine what normal sexual and voiding function is for these patients as they enter adulthood [49]. Doing so will allow us to fully determine the course of the repair of this complex disease process in infancy in order to translate into achieving its maximal potential as an adult.

13.5 Conclusion

Proximal hypospadias is a complex variant with a high risk for complications. Proper anatomic assessment and determination of complexity is a crucial aspect of the repair. The two-stage repair has emerged as a more reliable approach to ensure adequate correction for chordee at the first stage of the repair, optimizing graft or flap healing in preparation for urethroplasty at the second procedure. Many unanswered questions about hypospadias, and it is clear that long-term follow-ups of these patients into adulthood is not only suggested but should be mandatory.

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