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The staged approach to bladder exstrophy closure and the role of osteotomies

Abstract Since the 1970's, the staged reconstruction of bladder exstrophy has yielded consistent surgical success. The Johns Hopkins Hospital approach begins with early pelvic ring approximation with abdominal wall, bladder, and posterior urethral closure. Within the first 72 hours of life, the malleable pelvis can sometimes be approximated without osteotomies. Beyond this age, the author's prefer a combined vertical iliac and horizontal innominate osteotomy. Second, we typically perform the epispadias closure at 1 year of age. A modified Cantwell-Ransley technique is performed, usually yielding an increase in bladder capacity and very satisfactory results. In the last phase, the modified Young-Dees-Leadbetter continence procedure along with transtrigonal/cephalotrigonal ureteroneocystostomies are performed when the urethra is catheterizable, the bladder capacity is 60cc or greater, and the child will participate in a postoperative voiding program (typically 4–5 years of age). This applied approach usually results in a continent, voiding patient with pleasing external genitalia and preserved renal function.

Whereas it was once deemed “impossible to restore the bladder” (H.J. Jewett, patient communication, 1957) in patients with exstrophy, now, with contemporary management, this profound anomaly has successfully been reconstructed. While some controversy continues concerning not only the timing but also the techniques of reconstruction, the staged reconstruction of the exstro-

phy patient, as performed at the Johns Hopkins Hospital, has proven to withstand the test of time, yielding acceptable renal preservation rates and continence [11, 18, 34, 40, 45]. In this review, the current strategy of staged reconstruction and the application of osteotomies in this reconstruction will be delineated. Management options in the exstrophy reconstruction failures will also be addressed.

Staged exstrophy reconstruction

The staged approach to exstrophy reconstruction consists of early bladder and posterior urethral closure, later epispadias repair, and finally bladder neck reconstruction with an antireflux procedure. This approach can yield a secure bladder and abdominal wall closure, urinary continence without renal compromise, and reconstruction of a cosmetic penis with good sexual function. Others have advocated a single- or two-stage approach to reconstruction with good preliminary results [7, 15, 19] but the numbers are limited. However, the risk of complications is significant for any one stage and, clearly, one complication could jeopardize other aspects of the repair. This staged approach has been recommended and performed at our institution since the mid-1970s and long-term results seem to justify this approach.

Stage 1: Initial early exstrophy closure

With rapid referrals and, in some cases, prenatal diagnoses [26], neonates with bladder exstrophy should be evaluated for surgical reconstruction promptly. At stage 1 of exstrophy reconstruction, the surgical objectives include: (1) approximation of the pelvic ring, (2) closure of the bladder and posterior urethra inside the pelvic ring, and (3) closure of the abdominal wall.

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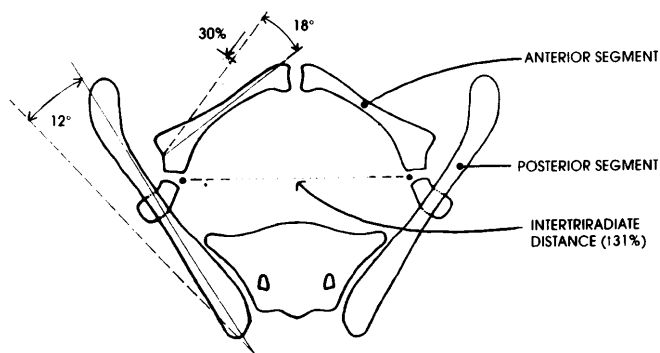


Fig. 1 The anatomy of the normal bony pelvis is seen in transverse view. Superimposed are the deviations observed in the pelvis of classic bladder exstrophy patients. In the posterior segment, each half is externally rotated 12°. The acetabulae are retroverted, yielding an increased intertriradiate distance. In the anterior segment, each half is externally rotated 18° and is on average 30% shorter

Approximation of the pelvic ring

The anatomy of the bony pelvic ring in classic exstrophy has been defined by Sponseller et al. using CT scan reconstructions [30, 49]. There is a 12° external rotation of the innominate bones on the sagittal axis, retroversion of the acetabula, and the pubic bones are on average 30% shorter with an 18° external rotation at the junction of the ischial and iliac bones [49] (Fig. 1). This yields the wide public symphyseal diastasis.

It has become clear that approximation of the open pelvic ring eases abdominal wall closure and diminishes the rate of bladder dehiscence and abdominal wall dehiscence [19, 34, 45]. This therefore allows the construction of an intrapelvic urethra and reapproximates the pelvic floor musculature, likely contributing to long term continence [1, 18, 45]. Additionally, in females, osteotomy may reduce the likelihood of uterine prolapse [3, 45]. Therefore, pelvic ring reapproximation is useful at initial bladder closure as well as at later stages of

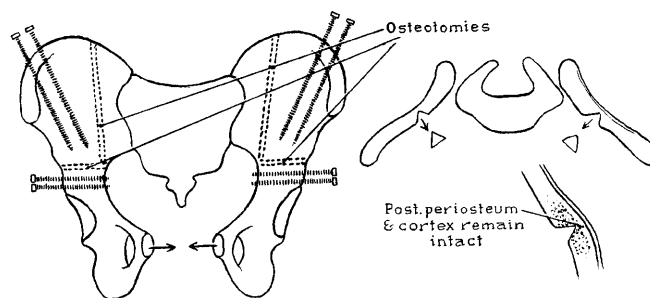


Fig. 3 Osteotomy sites for combined vertical iliac and transverse innominate osteotomies showing the placement of intrafragmentary pins

reconstruction if satisfactory reapproximation was not achieved initially.

Pelvic closure in the neonate younger than 48–72 h old can be achieved without osteotomies due to the effects of maternal relaxin; immobilization is achieved by modified Bryant's traction for 3–4 weeks (Fig. 2). However, beyond this age or at this time if the pelvis is not sufficiently malleable, osteotomies are performed at the time of initial closure. An interesting predicament for the surgeon is the situation of the newborn with a large bladder template. In this case, with a bladder having great potential for later continence, the public diastasis will be wider and require osteotomy regardless of when closure is attempted. While the authors have performed posterior iliac osteotomies, anterior innominate osteotomies or public ramotomies at initial closure with success, a combined approach of vertical iliac osteotomy with horizontal innominate osteotomy (Fig. 3) has recently been used in over 68 patients at our institution with good results [30]. Femoral nerve palsy is a possible complication which must be monitored. However, with gradual tightening of the fixator, this complication has not been seen in the last 36 procedures. These osteotomies are secured by intrafragmentary pins, external fixation (4 weeks for newborns; 6–8 weeks for older patients), and light Buck's traction for 2–4 weeks.

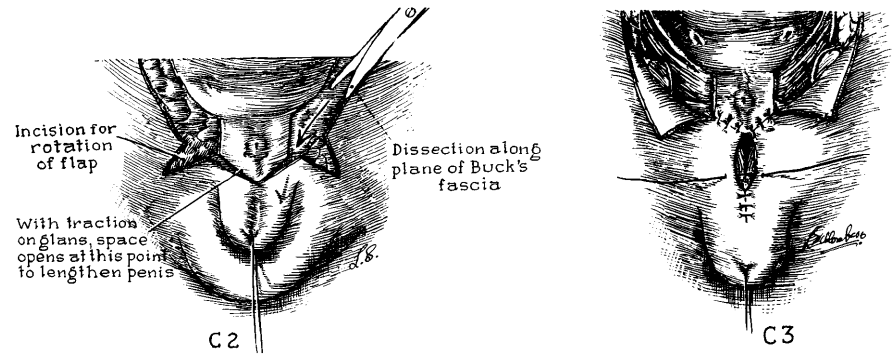


Fig. 2 Patient seen with an external fixator and in Bryant's traction

Exstrophy closure

The initial decision as to whether a bladder template is amenable to closure is currently based on subjective data. Even a small bladder template, if distensible and contractile, can enlarge to a useful size once closed. However, if an examination under anesthesia reveals a small (<5 cm³) acontractile, nondistensible bladder patch, an attempt should not be made to close this bladder. While ureterosigmoidostomy has proven useful in this group of patients [39], our preference is to wait and reexamine the child at 4–6 months of age under anesthesia. If at later evaluation the bladder remains inadequate, a nonrefluxing colon conduit is used to divert the urine along with the abdominal wall closure and epispadias repair. This can be converted at a later age to

Fig. 4 Steps in primary bladder closure following osteotomy or without osteotomy in the newborn younger than 72 h age. C2 Following bladder and posterior urethral mobilization, lateral skin incisions are made into the thick, healthy paraexstrophy skin; C3 flaps of skin are rotated and anastomosed to the posterior urethral plate



a continent diversion or anastomosed to the rectosigmoid.

If the bladder template is amenable to closure, early exstrophy closure is advantageous for multiple reasons. Socially, for the family, a prompt closure begins the road toward normalcy. In addition, early closure saves the bladder from prolonged exposure which can lead to such histological changes as acute and chronic inflammation, squamous metaplasia, cystitis glandularis and cystitis cystica, and muscular fibrosis [2, 46]. Functionally, it is likely that this results in a bladder with low capacity and poor compliance [29]. Ongoing research at our institution has shown that the newborn exstrophy bladder is “immature” when compared to control newborns, having fewer small nerve fibers [43], less smooth muscle, and a three-fold increase in type III collagen content [41]. While it is unclear whether these changes are a part of the primary pathology or secondary to the lack of bladder cycling in utero, it is conceivable that prompt closure could also “mature” the bladder by restoring bladder cycling. Similarly, recent clinical data from the Mayo Clinic has verified the importance of immediate postnatal exstrophy closure; infants closed before 7 days of age required fewer bladder augmentations to achieve eventual continence [44].

The surgical technique of the midline closure of the bladder, posterior urethra, symphysis, and fascia has been previously described [21]. However, a few caveats that can add to surgical success follow. Inguinal hernias, seen in up to 82% of males, should be surgically repaired at the time of initial exstrophy closure [10]. If the child has severe chordee and a short urethra that inhibits attempts at penile lengthening, an incision of the urethral plate may be necessary. Management of this is best assessed after extensive mobilization of the bladder and prostatic plate from the rectus fascia, the pubic bones, and the inferior ramus of the pubis. Once radical division of the urogenital diaphragm fibers from the posterior urethra/bladder neck area is accomplished, the prostatic urethra can be displaced posteriorly into the pelvis. Division of the anterior attachments of the crura and the suspensory tissue of the penis then displays the gap to be bridged between the prostatic plate and the penile plate. While this has been reconstructed after the manner of Johnston or Duckett, paraexstrophy skin flaps should be used with great caution. The au-

thors have noted a 40% complication rate associated with their use and significant comorbidity [24]. This lack of urethral plate length can be managed by dividing the prostatic plate distal to the verumontanum in a V-shaped fashion. After corporal mobilization and midline reapproximation, the penile urethral plate is fashioned from a lateral rotational flap generated after a single lateral incision into the paraexstrophy skin (Fig. 4). Less tissue redundancy and dissection is therefore necessary. Paraexstrophy skin flaps are unnecessary in female exstrophy closures and are associated with a very high complication rate [6].

In all cases of initial closure, suprapubic tubes and ureteral stents are placed and urethral tubes/stents should not be used. Additional factors that are important to the success of the initial closure include pelvic immobilization and fixation, adequate pain control, prevention of abdominal distention, and postoperative antibiotics [34, 42]. Adequate urethral caliber is assured prior to removal of the suprapubic tube at 4 weeks. The status of the upper tracts is documented by ultrasound prior to discharge and is followed by ultrasound at 3 months and every 6 months to 1 year.

The successfully closed child is continued on prophylactic antibiotics, for essentially all have vesicoureteral reflux. Between the ages of 1 to 3 years, yearly cystograms under anesthesia are performed to assess the degree of reflux and, more importantly, to assess the growth of the bladder. By this age, it is hoped that the capacity will achieve 50 to 60 cm³.

Stage 2: Epispadias repair

Epispadias repair should result in a cosmetic, straight functional penis with a urethra capable of catheterization if necessary. The modified Cantwell-Ransley epispadias repair has been the preferred technique at our institution since 1988, being performed typically between the age of 1 and 1.5 years [28, 37]. This repair offers a straight urethra with a 15% fistula rate at 3 months while simultaneously incorporating a chordee repair [28]. Performing the epispadias repair prior to bladder neck plasty provides a gradual increase in outlet resistance with the added benefit of enhancing bladder capacity [17].

In preparation for this repair, the following questions must be addressed: (1) Would preoperative stimulation of penile growth with testosterone benefit the repair? (2) Is penile lengthening useful at this time? and (3) Is the current posterior urethral meatus ideal, that is, without and/or not prone to stricture?

Most of our male patients undergo testosterone stimulation before epispadias closure. Testosterone enanthate in oil (2 mg/kg) is administered at 5 and 2 weeks prior to epispadias closure. The decision as to the need for penile lengthening can then be made after the response to testosterone has been observed. Penile lengthening can almost always be achieved at this time by division of all remnants of the suspensory ligament and scar tissue as well as further release of the crura from the inferior pubic rami. It is surprising how much of this tissue is intact at the onset of the epispadias repair. In some situations, further urethral lengthening may require free skin grafts, buccal mucosal grafts, ureteral grafts or pedicle skin flaps [33, 50]. These same maneuvers can be used to repair the scarred or stenosed posterior urethra.

Postoperative care includes bladder antispasmodics, broad-spectrum antibiotics and removal of the urethral catheter at 2 weeks. Long term experience with over 125 patients with the modified Cantwell-Ransley repair finds a conical glans, a straight penis and urethra below the corporal bodies with an acceptable complication rate.

Stage 3: Bladder neck plasty with or without Osteotomies

A continence procedure is indicated when (1) the urethra is stricture-free and capable of catheterization if necessary, (2) under anesthesia, the bladder capacity has achieved a minimum of 60 cm³, and (3) the child is of the maturity to desire continence and to participate in the postoperative voiding program. This is typically around age 4–5 years. The modified Young-Dees-Leadbetter bladder neck plasty [36] and the transtrigonal/cephalotrigonal bilateral ureteral reimplantation [8, 9] are the surgical techniques employed at the Johns Hopkins Hospital. Again, it is necessary to reassess whether osteotomy might stabilize the intersymphyseal bar and improve continence at the time of bladder neck reconstruction. It is important to completely free the bladder neck area at this time so that urethropexy can be achieved. This may require excision of a portion of the intersymphyseal bar for visual access.

Urine is drained via bilateral ureteral stents and a suprapubic tube for 3 weeks. The urethra is calibrated with a soft 8-Fr. catheter and voiding trials are begun with the suprapubic tube in place after the urine has been sterilized. If no urine is passed, cystoscopy and urethral stenting may be required for a short period. A bladder readjustment period may span several months before day and subsequently night continence is achieved.

Combined exstrophy and epispadias closures

In the event of a significantly delayed primary closure or an initial bladder closure failure, a simultaneous closure of the exstrophy bladder and the epispadias can be performed. The advantages to this approach include one less anesthetic exposure and increasing outlet resistance to stimulate bladder growth. Again, in the scenario, the combined horizontal innominate and the vertical iliac osteotomies are necessary for a secure bladder, abdominal wall, and posterior urethral closure and can allow transposition of the urethra to beneath the corpora with some dissection. This combined approach of simultaneous exstrophy and epispadias closures has been performed in 17 patients at this institution with comparable results (i.e. fistula rate, bladder neck reconstruction rate, augmentation rate, and continence rate) to age-matched patients who underwent the classic staged approach after failed initial closure [31]. However, this is only necessary in select patients.

Outcomes of staged exstrophy reconstruction

Accumulation of the data from several series of staged reconstructions reveals the following conclusions:

1. Early exstrophy closure, only if in conjunction with pelvic ring approximation, results in a higher rate of continence achieved without augmentation; delayed closure, especially if without osteotomies, resulted in more bladder augmentations [34, 44].
2. Paraexstrophy skin flaps are at high risk for complications; the use of such flaps should be minimized in the male and is unnecessary in the female [24].
3. Bladder capacity under anesthesia, prior to bladder neck plasty, is the most important single predictor of eventual urinary continence [25]. A recent review of our 21 years of experience with 60 consecutive exstrophy closures found that 72% are dry and voiding urethrally without augmentation or clean intermittent catheterization [40].
4. In the Hopkins experience, the Cantwell-Ransley epispadias repair results in a straighter penis and urethra but is associated with a 15% fistula rate at three months [28].
5. In adult exstrophy men, all experience normal erections and 75% have satisfactory orgasms. Fertility is still an issue, however, with only 63% having antegrade ejaculation and few pregnancies achieved [5].

Exstrophy reconstructive failures

Failed initial closure

At the primary closure, failures can manifest as bladder dehiscence, bladder prolapse with obstruction,

neourethral stricture and obstruction. It is prudent to consider referral of these complex management situations to an exstrophy center. Dehiscence, which may be precipitated by incomplete approximation of the pelvic ring, inadequate pelvic immobilization, wound infection, abdominal distention, or urinary tube malfunction [34, 42], necessitates a 6-month recovery period prior to a second attempt at closure. A tension-free closure with osteotomy and immobilization are important factors in both an initial and any subsequent closures [29]. Unfortunately, the chance of obtaining an adequate bladder capacity for bladder neck plasty and eventual continence, following multiple reclosures, is markedly diminished [20, 23, 29]. Similarly, bladder prolapse is considered a failure and will require bladder reclosure/revision. Neourethral stricture is most often associated with the paraexstrophy skin flap use [24]; pubic suture reaction and erosion or urethral stents may contribute in this problem. This diagnosis may be somewhat subtle; however, warning flags consist of urinary tract infections, detectable bladder volumes on ultrasound, bladder stones, and prolonged dry periods. While all closed exstrophy bladders have vesicoureteral reflux, upper tract deterioration is the ultimate fate of significant outlet obstruction. At this point, the management options include upper tract diversion (bilateral cutaneous ureterostomies or diversion into bowel conduit, which might be incorporated into augmentation or continent diversion at a later date), urethral dilation/incision, or open urethroplasty with or without osteotomies as indicated [3, 21]. Clearly, if renal function is compromised, the choice must achieve unquestionable free drainage for a period of time to allow the upper tracts and the kidneys to fully recover. No further bladder neck or urethral reconstruction should be performed until the

posterior urethral stricture is clearly repaired and free drainage has been achieved.

Failure to gain bladder capacity

Despite a satisfactory initial bladder and posterior urethral closure, some bladders never attain sufficient capacity to act as functional bladders. It has become clear that multiple bladder closures, bladder prolapse, dehiscence, bladder calculi, recurrent infections, and vesicostomy have a negative impact upon the potential of the exstrophy bladder [16]. Bladder augmentation is recommended in this situation. If the bladder neck and/or urethra are problematic, a catheterizable abdominal stoma with or without bladder neck plasty or transection is performed with the augmentation [3, 21].

Failed bladder neck reconstruction

If urinary continence, defined as a 3-hour dry interval, is not achieved within 2 years of bladder neck reconstruction, this is considered a failure. Some success has been seen in repeating a Young-Dees-Leadbetter repair if the bladder neck is patulous and the bladder capacity is adequate [22]. The majority of bladder neck failures require eventual augmentation or a continent diversion [27]. Either the bladder neck is transected proximal to the prostate with a Mitrofanoff substitution or a continence procedure is performed, such as artificial sphincter [13, 32] or collagen injection [4].

Delayed urinary continence has been reported at the onset of puberty in some males and has been attributed to prostatic growth [41]. By MRI, the prostate of

Fig. 5 Axial T2-weighted images through the mid-prostate gland. A 22 year-old normal individual demonstrates the usual relations between the components of the puborectalis sling (54°); B 30-year-old classical exstrophy patient demonstrates the altered relations between the components of the splayed puborectalis sling (109°) (F, fibrous band; OI, obturator internus; P, bony pubis; Pr, prostate gland; Sf, symphysis; SV, seminal vesicle; R, rectum, U, urethral)

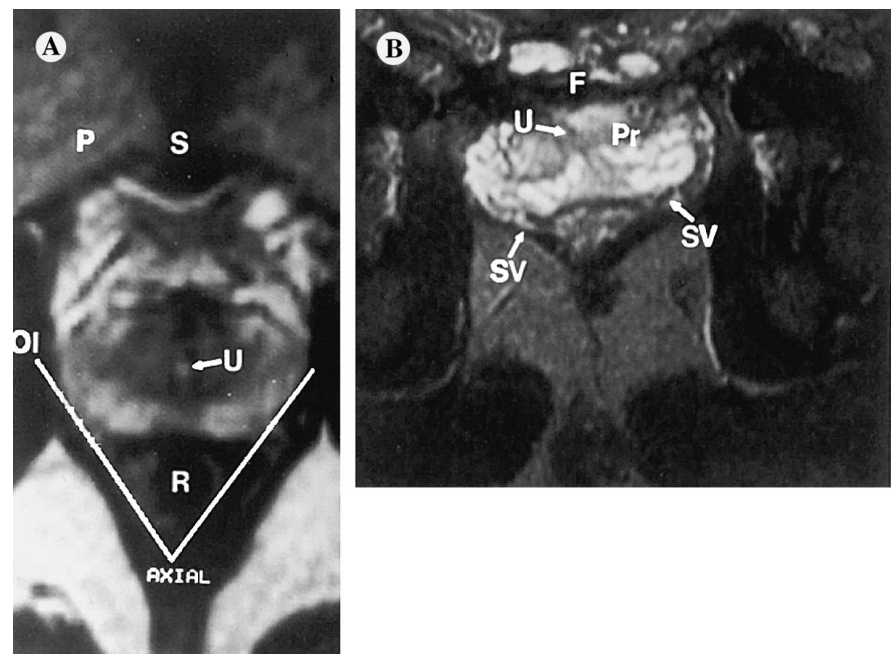
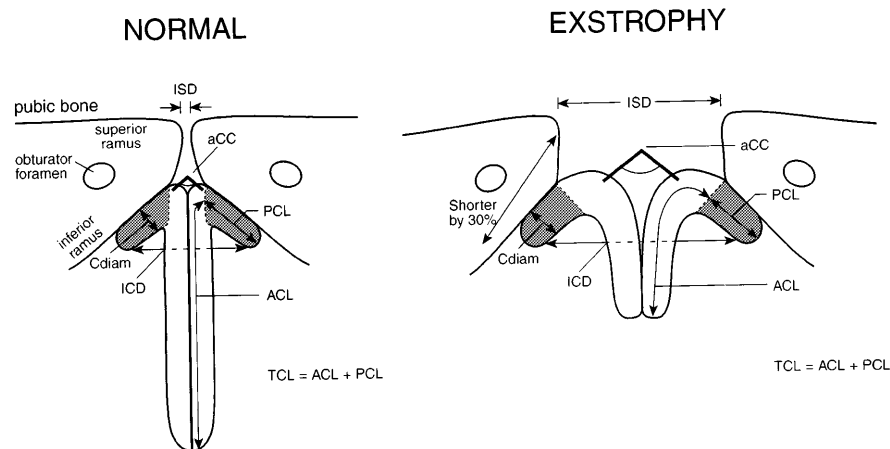


Fig. 6 Penile and pelvic measurements in normal men and patients with exstrophy (*ISD*, intersymphyseal distance; *ACC*, corpora cavernosa subtended angle; *Cdiam*, corpus cavernosum diameter; *PCL*, posterior corporeal length; *ICD*, intercorporeal distance; *ACL*, anterior corporeal length; *TCL*, total corporeal length)



exstrophy patients (Fig. 5) is clover-shaped and absent anteriorly while its mean weight, maximal cross sectional area, and volume are normal [25]. Therefore, it is doubtful that growth of this abnormally configured prostate can impact upon urinary continence.

Failed genital-urethral reconstruction

The most common complication of Cantwell-Ransley epispadias closure, urethrocutaneous fistula formation, was seen in 15% of patients at 3 months postoperatively [28]. Urethral tortuosity with difficult catheterization and strictures are uncommon with this repair.

At an older age, unsightly penile scars and a short phallus may prompt further surgical intervention. Scar excision can be closed in a plastic fashion if penile skin is available; otherwise, flaps or full-thickness skin grafts can be used [21]. Freeing all scar tissue and suspensory ligament tissue can maximize penile length. A dorsal dermal corporal graft [51] or ventral corporal plication and rotation of the Cantwell-Ransley type [28, 38] may additionally help lengthen as well as correct any chordee. However, it must be recognized that the exstrophy penis, when compared to age and race-matched controls, is congenitally deficient in anterior corporeal tissue as assessed by MRI (Fig. 6). The posterior corporeal segments, attached to the inferior pubic ramus, are the same length as the controls. Therefore, overly aggressive attempts at lengthening the penis may only result in corporeal denervation and devascularization without additional lengthening [48].

Conclusion

Forty years ago, bladder exstrophy was considered a hopeless state, which could not be reconstructed. As delineated above, the staged surgical management of exstrophy has evolved, through much work and frank experimentation, into a precise scheme. Clearly, urinary continence can be achieved with renal preservation and

the external genitalia can be fashioned to be functional as well as cosmetic by implementing this multi-stage approach. Other approaches such as single-stage bladder closure and epispadias repair in the very young child await the test of time as far as status of the upper tracts, ability to gain sufficient capacity for bladder neck reconstruction, and long-term continence results are concerned.

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