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Single-staged male bladder exstrophy–epispadias complex reconstruction with pubic bone adaptation without osteotomy: 15-year single-center experience

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Abstract

Purpose To represent the 15 years' experience of an academic referral center for the reconstruction of bladder exstrophy–epispadias complex with a modified single-stage approach. Single-staged reconstruction techniques are commonly used for classic bladder exstrophy. However, combined bladder closure and epispadias repair have been taken into great consideration in patients with initially failed reconstruction or delayed primary closure.

Methods A total of 49 boys underwent 1-stage bladder and epispadias repair with pubic bone adaptation and without the application of pelvic osteotomy. The mean $\pm$ SD age at surgery was 5.23 ± 2.04 months. Continence and social dryness were assessed in the follow-ups with 3 months intervals for the first year and biannually thereafter.

Results The mean $\pm$ SD of follow-up was 127.25 ± 71.32 months. Urethrocutaneous fistula, stricture, wound infection, and hemiglans were developed in six distinct patients. However, no other major complications were noted. Three patients (6.1%) remained incontinent; while 32 (65.3%) children were socially continent and 14 (28.6%) children were waiting for toilet training. All the patients without previous failed closure were socially continent, while all incontinent patients had two failed closures in their history. No patient was rendered hypospadiac.

Conclusion Based on the experience of this institution, the application of single-stage reconstructive techniques can lead to continence, cosmetically pleasing appearance with promising outcomes, and reduction of overall operations, hospital stay and costs in the majority of cases as compared to multiple surgical procedures.

Keywords Bladder exstrophy · Epispadias · Single-staged surgical procedures · Male

Abbreviations
BEEC Bladder exstrophy–epispadias complex
BNR Bladder neck reconstruction

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Introduction

The bladder exstrophy–epispadias complex (BEEC), as a rare midline defect, is commonly accompanied by several anomalies including omphalocele, spinal defects and imperforate anus [1]. This malformation usually develops at around 4–6 weeks of gestation, when prematurely invasion of the cloacal membrane and mesoderm is accomplished to build the abdominal wall for the genitourinary system development [2]. The primary defect in this surgically correctable complex congenital anomaly is a derangement in midline developmental with a range of complications. In epispadias, dorsal urethral unit failed to shape into a tube and it is not fused which is considered as the mildest form. In classic bladder exstrophy, the bladder and urethra are open and uninterrupted with the abdominal wall. In this situation, the abdominal muscles, pelvic floor musculature, and pelvic ring, failed to fuse in the midline. BEEC is associated with rigorous degree of simultaneous congenital derangements

of musculoskeletal, genitourinary, gastrointestinal, and neurological systems [3]. Imaging such as magnetic resonance imaging (MRI) has been also considered as a preferred, useful and cross-sectional modality of choice in the primary evaluation of BEEC defects [4]. Urinary incontinence and unattractive genitalia remain the greatest concerns to male patients with classic BEEC [5]. Secure closure of the abdominal wall, satisfactory reconstruction of the external genitalia, adequate bladder and prostate placement sufficiently deep within the pelvis, and obtaining urinary continence with the conservation of renal function are the main goals of surgical interventions [6]. Fruitful initial closure of bladder, posterior urethra, and abdominal wall accompanied with early epispadias and bladder neck reconstruction (BNR), are required to achieve an appropriate bladder capacity and continence in the bladder exstrophy population.

Initial bladder closure failure and delayed primary repair due to an inadequate template at birth will decrease the continence rate and the ability of bladder growth [7]. These obstacles may result in several additional surgeries for obtaining satisfactory cosmetic and functional outcomes as well as a considerable economic cost [8]. Abdominal wall and bladder closure in the neonatal period will increase surgical and functional outcomes, compared to delayed surgical interventions [8]. However, both initial bladder closure and epispadias repair are performed concomitantly in some cases with satisfactory outcomes [9].

In this study, we focused on the single-staged repair of BEEC without osteotomy in a group of male population.

Patients and methods

After the Committee of Human Research and institutional review board approval and obtaining informed consent from all caregivers, charts of all surgical records from September 2002 to November 2017 were reviewed retrospectively. Forty-nine male patients who underwent single-staged reconstruction of classic BEEC were included in this study. The data were gathered, including age, presence of bladder dehiscence or prolapse, urethral fistulization, number of previous failed attempts in closure and epispadias repair, use of osteotomy at former closure attempts, previous surgical technique, outcomes and complications of reclosure techniques. Renal ultrasonography and pelvic bone 3D-CT were initially performed in all children. Two patients were noted with a severe waddling.

No pelvic osteotomy, external fixators or traction devices were used at the time of primary closure or after reconstruction. Our approach to the single-staged reconstruction of this complex malformation is described in our previous articles [10, 11]. Briefly, the bladder plate was circumscribed by a bisturi knife and urethral boundaries

were also defined precisely to prevent the verumontanum existence inside the bladder. Any imprecision in the exact localization of verumontanum may leave splayed fibers of urinary sphincter inside the bladder and compromise eventual continence. A 0.5–1 cm skin margin was left inside the boundaries as an anchoring place for subsequent harnessing stay sutures. Silk 3–0 stay sutures applied to steer the bladder plate. The plate was meticulously dissected off the underlying rectus fascia and peritoneum down to the central vascular pedicle of the bladder plate. Simultaneously, urogenital diaphragm fibers were sharply dissected off the pubic arches down to the corpora cavernosa. The bladder plate was freed liberally prior to pubic approximation; so, it could be closed in two layers without any tension. The urethral plate was dissected off the corpora. The corpora were also detached up to the glans sparing distal 1 cm in a Cantwell-Ransley manner. Doing this before closure will allow deep placement of the bladder, portending a better continence state. After resecting the anchoring skin margin, the bladder plate was sewn lip to lip in two layers with Vicryl sutures. In case of adequate space, an appropriate size cystostomy tube was inserted inside the bladder. Ureteral catheters were inserted inside ureteral orifices and anchored to bladder mucosa with fine absorbable sutures. Pubic rami were gradually approximated with a Bailey rib retractor and an absorbable C-plate and fixing heavy Vicryl sutures. Urogenital fibers were wrapped around the bladder neck on both sides to recapitulate a circular sphincter with stronger fibers at 12 o'clock. Urethral tubularization and ventralization were undertaken, applying IPGAM incision and cavernosal medial rotation and suturing. The rotating Vicryl sutures on corpora cavernosa was not too extreme to cause excessive twisting of central cavernosal fine arteries. The wrapping urogenital diaphragm flaps around the bladder neck also were not tight excessively. Postoperative leg plaster or leg traction was not required after this procedure, but the patients remained in a frog leg bandage for 1–3 weeks post-operatively, as described by the previous article of the senior author (AMK) [10] (Fig. 1) (Video 1).

The patients were discharged 2–3 weeks post-operatively when they were catheter free. The home location, distances from the hospital, and the health care facilities nearby patient's residential area were especially taken into consideration before the patient's discharge. During the first year, patients were followed-up every 3 months. The follow-ups were then continued biannually. Clinical examinations were performed post-operatively to evaluate the bladder closure success and continence, the cosmetic appearance of the external genitalia, gait outcomes, renal/bladder ultrasonography, and 3D-CT scan. Continence was also assessed during follow-up, and was defined as daytime and nighttime

Fig. 1 Surgical steps of the single-stage reconstructive technique

dryness, with no stress incontinence for at least 3 h. Social continence was defined as having the ability to stay dry for more than 3 h during the day.

Results

The mean ± SD ages for the operation was 5.23 ± 2.04 years (range 14.5 months to 13 years). Some patients were referred from different centers; following failed closure attempts. Of these 49 children, 31 patients (63.2%) had a history of failed initial bladder closure with 1–2 osteotomies, 7 patients (14.3%) underwent failed initial bladder closure without an osteotomy, and 11 (22.4%) patients had no history of surgery. The mean operation time was 321 min and the mean ± SD hospital stay was 17.2 ± 1.07 days. The median and the mean ± SD follow-up from date of surgery were 105 and 127.25 ± 71.32 months, respectively. There were no patients lost to follow-up.

As summarized in Table 1, all children had uneventful postoperative periods without the development of wound infection, except for two children with a superficial infection around the suprapubic tube. However, no dehiscence or bladder prolapse occurred. None of our patients were presented with recurrence of the bladder prolapse or dehiscence after undergoing this method. The upper urinary tract was normal in all the patients confirmed by ultrasound. The bladder remained closed throughout the follow-up in all children. Furthermore, the cosmetic appearance of the external genitalia was satisfactory in all patients according to observation and the parents' opinion (Fig. 2). None of the patients developed orthopedic and neurological complications. Furthermore, those children with waddling gait ($n = 6$) (difficulty climbing stairs, running, jumping, and standing up from a squatting position) showed improving results after 5 months postoperatively without outward rotation of the lower limbs. In three boys, either urethrocutaneous fistula or stricture was developed in whom fistula repair and urethroplasty were accomplished, respectively. Besides, hemiglans ischemia (discoloration that was resolved within 2 weeks of operation) was notified in one patient with a history of two failed bladder closure. The pubic separation was not noted in any of the patients. The 3D-CT scan of a patient before and after the surgery is shown in Fig. 3.

Of these 49 patients, 32 (65.3%) were socially continent during day and night and did not require augmentation or clean intermittent catheterization. All the patients without previous failed closure were socially continent ($n = 11$); while other patients who were socially continent at the last follow-up, had the history of previous failed closures (one or two failures, with or without osteotomy). However, three (6.1%) patients remained incontinent. All incontinent patients had two failed closures in their history. The other 14 patients (28.6%) were waiting for toilet training. All the patients in this group had the history of one or two previous failed closures with or without osteotomy. Cystometric bladder capacity, compliance, Pdet at cystometric capacity, and detrusor muscle function were recorded. The VCUG

Table 1 History of male patients before and after the single-staged bladder exstrophy-epispadias complex reconstruction

Variable	No. of patients (%)	
Previous surgeries		
No surgeries (GI)	11 (22.4)	
One FBC with osteotomy (GII)	14 (28.5)	
Two FBC with osteotomy (GIII)	17 (34.7)	
FBC with no osteotomy (GIV)	7 (14.3)	
Mean ± SD age (years)	5.23 ± 2.04	
Mean Hospital stay ± SD (days)	17.2 ± 1.07	
Complications		
Dehiscence	0	
Major bladder prolapsed	0	
Minor bladder prolapsed	0	
Wound infection	2 (4.1)	
Urethral stricture	1 (2.1)	
Pubic separation	0	
Hemiglans ischemia	1 (2.1)	
Fistula	2 (4.1)	
Mean ± SD Follow-up (months)	127.25 ± 71.32	
Continence outcomes		
Socially continent	32 (65.3)	11 (GI), 10 (GII), 6 (GIII), 5 (GIV)
Waiting for toilet training	14 (28.6)	4 (GII), 8 (GIII), 2 (GIV)
Incontinence	3 (6.1)	3 (GIII)

FBC failed bladder closure

Fig. 2 Cosmetic results after undergoing this surgical technique

and voiding cystogram of a patient is depicted in Fig. 4; showing satisfactory postoperative outcomes (Video 2) and increased bladder capacity with increased compliance,

which demonstrated improvement in the same patient over time after closure.

Discussion

While many staged approaches are used for reconstruction of BEEC, a continued interest in combined techniques of reconstruction exists. Whether using a modern or single-staged approach, successful initial bladder closure is the most critical point for developing a satisfactory bladder capacity and continence in patients with bladder exstrophy. Radical dissection of the posterior urethra and bladder from environing pelvic structures, immobile closure of the abdominal fascia and pubis, prudent suturing of the bladder, posterior urethra, and last centimeters of the penis; steady immobilization; and protection from distraction have been mentioned as certain long-standing principles.

Although encouraging outcomes were obtained from Grady and Mitchell for a single-staged procedure, subsequent ureteral reimplantation was required in 50% of patients [12]. Moreover, hypospadias occurred in several boys as a consequence of complete disassembly of the urethra from the corporeal bodies [12]. According to Gearhart and Jeffs, urethroplasty in boys with bladder exstrophy enhances urethral resistance and bladder growth [13]. In one study in 1998, wound separation and bladder prolapse were prevented and posterior urethra was reconstructed following concomitant bladder closure and epispadias reconstruction with minimal lateral distracting forces [9]. In another study, prolapse was prevented and outlet resistance was improved to stimulate bladder growth after epispadias repair, in which the bladder outlet distal segment was narrowed [14].

The end results of achieving continence rates after bladder augmentation and CIC in resource-poor setting situations are intricate. Illiteracy and scarcity are powerful issues for selecting a single-staged BEEC operative procedure and obtaining low-pressure reservoir for preservation of upper tract and satisfactory primary continence. In Yogesh's cystorectostomy which is an alteration of Heitz–Boyer–Hovelacque method, the bladder plate is directly anastomosed to the recto-sigmoid pouch, without mobilizing the ureters from their native region. It has been indicated that the achievement of total urinary continence and preservation of upper tract function were encouraging in short-term follow-up [15].

The modified Cantwell–Ransley repair was applied as a substitute for the modified Young repair that was accompanied by several complications including high fistula rate, persistent and recurrent dorsal chordee, and resultant tortuosity of the urethra [16]. Moreover, in another study, initial penile straightening was performed in this technique for male neonates with exstrophy at primary bladder closure,

Fig. 3 The pelvic bone 3D-CT before and after the single-staged repair of BEEC without osteotomy
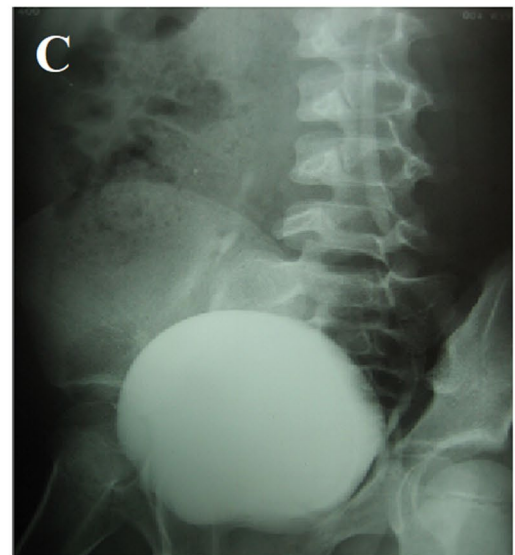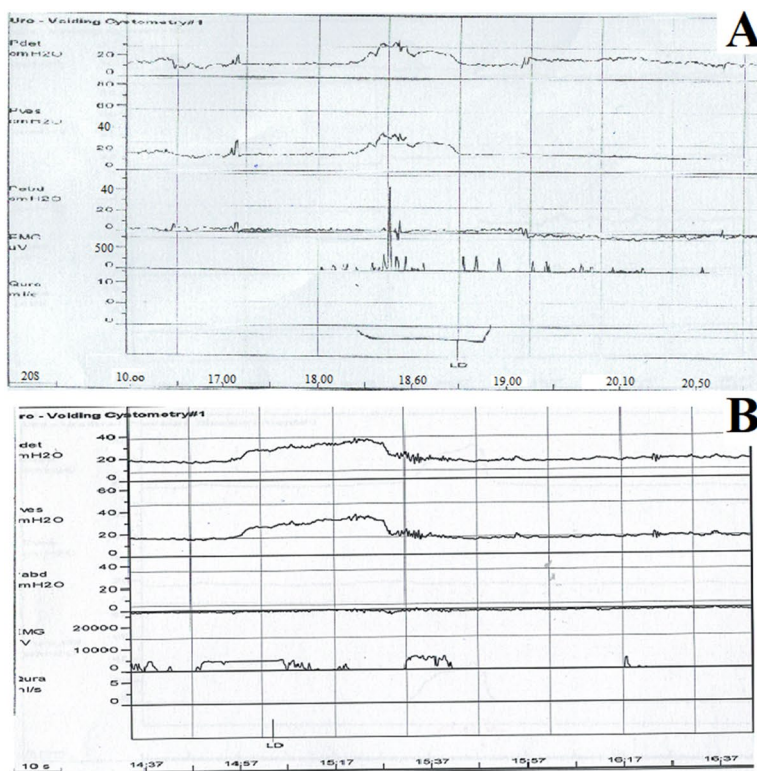

Fig. 4 Voiding cystography of a patient **a** before and **b** after bladder reconstruction. **c** VCUG of the same patient after the surgery

following some additional penile lengthening at the time of penile repair by the application of para-exstrophy skin flaps for lengthening the urethra [17]. However, the application of para-exstrophy skin flaps did not show any significant difference in fistula formation; 21% versus 25% in the para-exstrophy flap group [17]. Additionally, the dissection was gotten directly onto the corpora with preservation of the neurovascular supply and approximation of the pubic rami; resulting in adjusted intrinsic corporeal chordee and increased penile length [17].

Dehiscence, multiple closures, bladder prolapse, and calculi are among the several factors which can affect long-term outcomes, regardless of the type of technique for bladder repair [18]. Accordingly, posterior urethral outlet obstruction, bladder prolapse, and dehiscence are recognized as major complications of primary bladder closure that can decrease the satisfactory outcomes of staged bladder reconstruction, as well as influencing ultimate fertility. Shearing forces within the pelvis and abdomen are among the most crucial factors that can result in the subcutaneous

displacement of the bladder and posterior urethra or their complete dehiscence [14]. Devascularization of the penis has been reported as a serious consequence of failure in bladder exstrophy and epispadias closure following technical error, prolonged dissection or venous congestion that will eventually result in loss of function and a poor cosmetic appearance [19]. Loss of penile and corporal tissue are other concerns about using these methods [19, 20]. Lack of suitable tissues is the most frequent obstacle in satisfactory reconstruction of corporeal and glandular tissues [20]. However, the quality of the bladder template, depth, and width of the urethral groove and pubic diastasis, size of the phallus, and experience of the surgeon are among the associated factors determining the postoperative outcomes in different methods of bladder exstrophy closure. The Kelly radical soft-tissue mobilization has been recently considered as a safely method, which can be combined with delayed bladder closure without osteotomy in classic bladder exstrophy cases with satisfactory outcomes. However, short-term follow-up and not addressing the continence and voiding were considered as the limitations [21]. According to the Seattle experience, single-stage repair of BEC cases was performed with and without osteotomy in 39 patients with 84% success rate. Major complications were two dehiscence and five fistulas (16%) [22]. The results of modern-staged repair with osteotomy in male patients was also stated. Although the success rate of primary closure was 95.4%, much longer hospital stay, ICU stay and immobilization period was reported compared to the present study [23]. Dickson et al. reported the results of modern-staged repair with osteotomy in 67 patients, and compared early (infancy) versus delayed (at 3 months of age) reconstruction. The success rate was 87% in infancy compared with delayed repair (100%) [24].

A failed attempt for closure can have an undesirable impact on final continence. Documented reports have proved that the likelihood of sufficient capacity achievement for BNR in a previously failed bladder closure is nearly 60%; while only 50% of these children will be eventually continent [25]. These caches are considerably decreased in third and subsequent closures. Furthermore, it has been demonstrated that a large amount of fibrous scarring was present around the bladder after a failed closure and the amount of detrusor muscle was decreased in the second closure that can lead to incontinence [25].

Osteotomy has been strongly recommended in such clinical situations for achieving bladder and epispadias closure [26]. Wound infection, the absence of osteotomy in patients with a large pelvic diastasis, and abdominal distension have been identified as critical factors contributing to the failure of the initial bladder closure [14]. Single-staged closure with osteotomy may be technically challenging in some patients due to unfavorable anatomy in spite of the fact that pelvic osteotomy has shown promising results in decreasing the complications after primary pelvic closure [27]. However, favorable results have been revealed by the application of a specific approach in which pelvic osteotomy and closure were also separated into staged procedures [28]. Reports noted in the literature demonstrated that combining bladder closure with epispadias repair facilitates medial rotation of the corporeal bodies over the urethra by adding pelvic osteotomy to subsequent bringing together of the pubic symphysis [9, 29]. Although it has been mentioned that osteotomy is mandatory in preoperative exstrophy closure and in delayed primary closure [30], it was not used in any of the patients in this series because of the fact that it can cause orthopedic and neurological injuries and increase the morbidity of bladder exstrophy management. Additionally, our previous study authenticated the feasibility of biodegradable miniplate and screws for secure anterior pubic bone adaptation in the animal model of pubic diastasis [31]. No need for long-term traction, simple surgical handling, and no skeletal growth disturbance were among the benefits of this technique.

Our study highlights some possible advantages of single-staged management of BEEC in which the fistula rate was similar to that of staged reconstruction. Although Gearhart et al. have suggested that the occurrence of stricture in the posterior urethra may arise after combined neonatal bladder closure and epispadias repair with great impaired access to this area [9], none of our patients showed such complications. Initial data of complete one-staged closure, penile disassembly, and epispadias repair, suggest that early continence may be achieved in some newborns [32]. According to the results obtained from our previous experiment, the modified Cantwell technique can result in satisfactory cosmetic outcomes with a low complication rate in almost all cases [33]. In spite of the fact that epispadias repair and bladder exstrophy closure may be safely performed simultaneously, this combination is not recommended in newborns and should be limited to older infants and those with initially failed closure. Furthermore, potential soft tissue complications of these methods should not be underestimated.

In conclusion, if initial closure is performed efficiently in the neonatal period with adequate postoperative care and a well-executed bladder closure, a satisfactory rate of continence will be gained. However, the probability of a required further surgery to the urethra or phallus is not far from the mind as only 50% of patients may eventually achieve continence while a significant number ultimately requires continent urinary diversion [26]. Limited numbers of patients and not comparing the results with previous standard surgical techniques are amongst the limitations of the current investigation. Nevertheless, multiple surgical procedures are required for achieving continence and a cosmetically pleasing phallus.

Conclusion

Although bladder exstrophy reconstruction is usually performed in staged procedure accompanied by subsequent epispadias repair and BNR, the combination of epispadias repair and bladder closure in selected patients does not show adverse effects on the results of surgical reconstruction. While this approach has decreased possible common complications and provided adequate outlet resistance with promising postoperative outcomes, meticulous follow-up of the whole genitourinary tract remains the foundation of success.

Compliance with ethical standards

Conflict of interest The authors declare that they have no conflict of interest.

Informed consent Informed consent was obtained from all individual participants included in the study.

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