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ORIGINAL ARTICLE

A less invasive technique for delayed bladder exstrophy closure without fascia closure and immobilisation: can the need for prolonged anaesthesia be avoided?

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Abstract

Introduction It is believed that the main factors enhancing security of the bladder exstrophy closure are use of osteotomy, pubic bones approximation or transferred flaps for rectus fascia closure. However, these methods increase operating time, surgical trauma and carry risks for the patient.

Objectives To demonstrate that the goal of secure bladder exstrophy closure can be achieved easier technically and safer for the child than previously thought. The paper examines the hypothesis that less invasive bladder exstrophy closure achieved without fascia closure can reduce pain and avoid the need for immobilization and prolonged analgesia.

Study design Patients aged 34 days to 15 years ($n = 36$) from 37 who consecutively referred to the institution with classical bladder exstrophy between 2004 and 2016 underwent modified delayed primary (25) or redo (11) closure. One boy with low weight was excluded. Patient and treatment features were analysed to determine needs for immobilisation and anaesthesia in the postoperative period, and outcomes.

Procedure Bladder exstrophy closure with proximal urethroplasty was performed with the detachment of crura from the ishiopubic rami and levators—from obturator internus muscle. Abdominal wall closure was accomplished with skin and subcutaneous fat mobilisation without rectus fascia closure. No method of immobilization was applied.

Results and limitations Bladder closures have been successful in all 36 children in this report after 37 months (22–138) follow up. The surgeries took time between 126 and 215 min (mean − 148). After 1 day in the ICU the majority of the patients (34/36) were returned to the ward. No bladder spasms or signs of acute pain were noted in the ward; therefore, no local anesthesia or opioids were needed. Intravenous analgesia with non-narcotic analgesics was used for all patients in the ward for an average period 2.2 days (95% CI 2–4 days).

Complications Minor complications: two fistulas, which closed spontaneously; three bladder outlet obstructions, each required one endoscopic incision. No major complications of exstrophy closure such as dehiscence or bladder prolapse were occurred.

Conclusions The proposed less invasive technique with relieved postoperative program is the way to obtain successful bladder exstrophy closure as well as to reduce some risks for the patients. Absence of major complications, and avoiding the need for immobilisation and prolonged analgesia, contribute to the benefits of this approach.

Keywords Bladder exstrophy · Immobilization · Anaesthesia · Bladder spasms · Bladder exstrophy and epispadias complex

Introduction

Bladder exstrophy is one of the most challenging conditions in paediatric urology practice. As with many other rare diseases, it has not yet been possible to apply the principles of evidence-based medicine and develop clinical guidelines with recommendations shared by most experts [1]. Secure initial bladder closure is the key factor for achieving long-term success in incontinence treatment [2]. Wound

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dehiscence or bladder prolapse after bladder exstrophy closure varies from 0 to 37.5% [3–6]. The classical approach to bladder exstrophy treatment assumes primary closure during the first 48–72 h of life. However, some experts have recently advocated for delayed repair, performed several months after birth, to facilitate family bonding, simplify the procedure technically, and decrease risks related to anaesthesia [7]. A return to the concept of delayed exstrophy closure, which predominated in the 1960s, appears to be feasible, as recent evidence indicates that delayed closures performed within 1–3 months of life do not compromise bladder growth [8, 9].

Closure of the fascial defect between recti muscles without tension is considered by most experts to be one of the main goals in providing successful bladder closure. It can be achieved by pubic bone approximation, with or without osteotomy, or by the use of transferred flaps [1–3, 6]. This is followed by pelvic immobilisation for several weeks, with intensive analgesia required throughout the entire immobilisation period [10]. Immobilisation in newborns is sometimes achieved by elective paralysis and ventilation for 7–8 days [11]. In any case, the use of immobilisation and analgesia for long periods of time presents various side effects and complications [3, 6]. Opioid infusion and benzodiazepine sedation are associated with dose-dependent respiratory and gastrointestinal complications. In addition, neonates and young infants can quickly become dependent on narcotics. Regional anaesthesia techniques reduce opioid requirements, but they have their own limitations and risks. The drawbacks include failure during the placement of tunnelling catheters for epidural and caudal anaesthesia and bacterial colonisation [12, 13].

Here, we suggest a less invasive technique of delayed midline bladder exstrophy closure without the need for pubic bone approximation or flaps. We tested the hypothesis that the modified technique is safe and that the need for immobilisation and prolonged analgesia during the postoperative period can be avoided.

Materials and methods

Delayed midline exstrophy closure procedures without osteotomy, pubic bone approximation or flaps were performed in 36 of 37 consecutive patients with classical bladder exstrophy (CBE) who were referred to our institution between 2004 and 2016. The patients in the redo group were after failed exstrophy closures, which resulted in complete bladder dehiscences. All patients underwent a similar surgical procedure performed by the same surgeon, regardless of the size of the bladder plate or the degree of pubic diastasis. Osteotomy was recommended for a 15-year-old girl with a large pubic diastasis following a failed exstrophy closure at 2 years of age, but she refused. An underweight 5-year-old boy with no subcutaneous fat was closed with osteotomy, and thus was excluded from this study. Patients with cloacal exstrophy or rare anomalies were also excluded.

I adhered to a staged exstrophy reconstruction, with bladder exstrophy closure representing the first stage of surgical treatment. Antireflux procedures, epispadias repair (in males) and bladder neck reconstruction (in incontinent children) were performed afterwards as required.

The medical records for this cohort were reviewed. The data regarding both operative and postoperative features were collected. It included the surgical findings and time of surgeries, transfusion risk, postoperative care, lung ventilation, duration of stay in the intensive care unit (ICU), analgesia, oral feeding, nutritional support, management of urinary drainage tubes, antibiotic prophylaxis and surgical complications. The focus was on the secure bladder closure, the need for postoperative analgesia and the surgical complications over a 1-year postoperative follow-up. Surgical success was defined as a repair that did not require re-operations, with no prolapse or dehiscence.

Surgical technique

Mobilisation

The bladder plate and the urethral plate were dissected from the skin, subcutaneous fat and rectus fascia caudally down to the vaginal orifice (females) or the penis (males). The peritoneum was detached from the dome of the bladder and pushed posteriorly.

To avoid tension at the bladder neck region, subperiosteal incomplete detachment of the crura of the penis and levators detachment from the medial walls of pelvis, together with tendinous arch (ATLA) and with the obturator fascia was carried out (Fig. 1). Because levators are mobilised with the obturator fascia, but inferior part of the obturator fascia and the posterior parts of crura remain attached to the ischium, and the Alcock canal is not opened, such mobilisation does not lead to injury of pudendal neurovascular bands, that can happen with the Kelly technique [14].

Reconstruction

An identical technique was used in primary and redo cases, in both young and old patients. No suprapubic tube was used. For most of the patients the ureteral stents were not required. Antireflux bilateral Innes Williams procedures [15] were performed, when catheter 8Fr freely passed through ureteral orifices. If the bladder plate was not pliable enough, the plate was inverted by carefully compressing with fingers to extrude the oedematous fluid, and indwelling ureteral stents were left for 12 days.

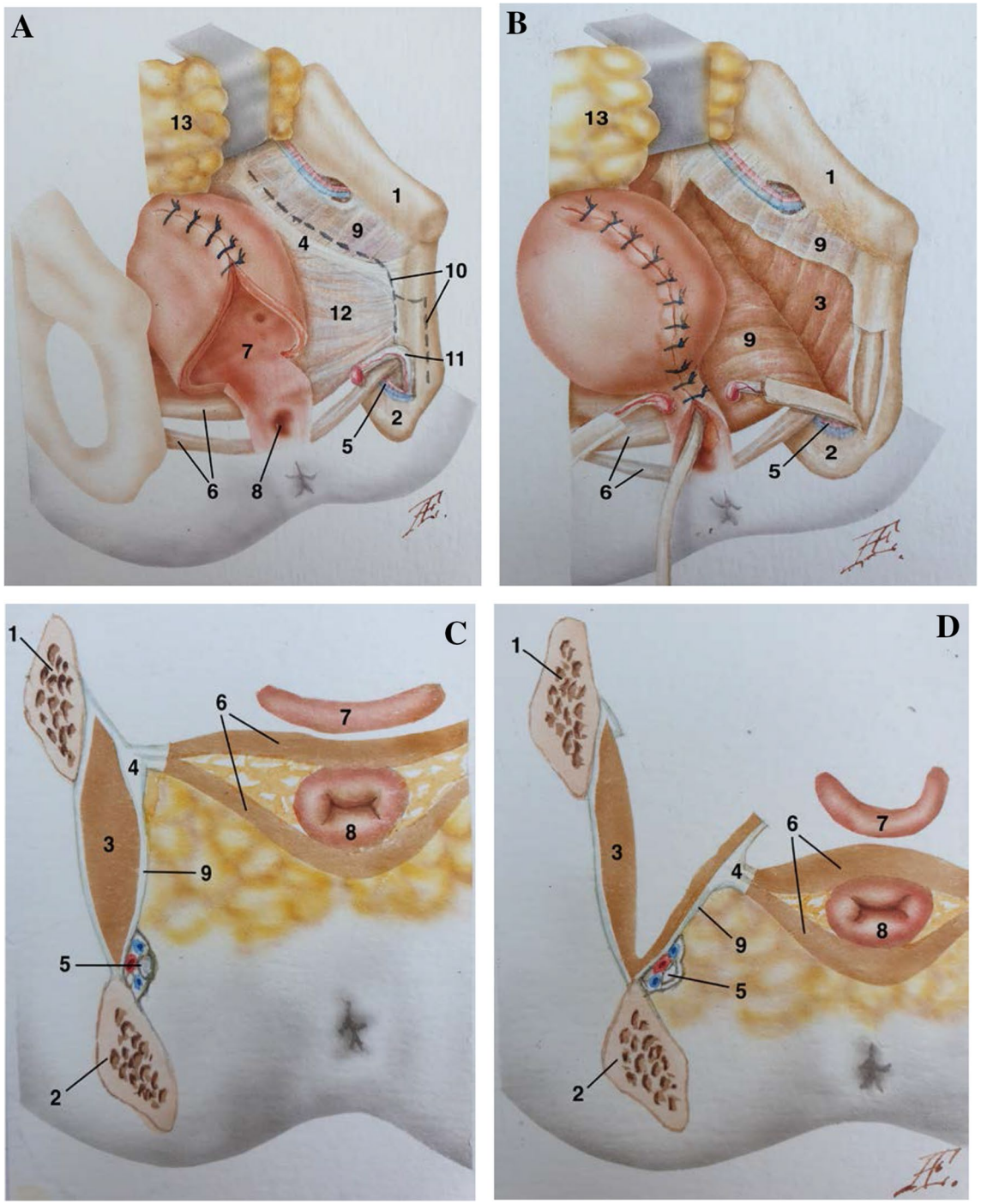

Fig. 1 **a**–**d** Principle of partial pelvic tissues mobilisation without Alkock canal openning. 1—os pubis; 2—os ischium; 3—m. obturator internus; 4—arcus tendineus levator ani; 5—Alcock's canal; 6—urogenital diafragm fibers and levators; 7—bladder plate; 8—vagina; 9—obturator fascia, 10—incision line; 11—crus clitoris; 12—endopelvic fascia: 13—paravesical fat. **a**, **b** Mobilization of the urogenital diaphragm begins with subtotal crura (11) mobilisation from ishiopubic rami. Incision (10) along the lateral edge of the arcus tendineus levator ani (4) allows the levators (6) with obturator fascia (9) to be peeled apart from obturator internus muscle (3). With this manoeuvre, the lateral edge of the levator ani muscle (12) is released, freeing the urogenital complex without the need to transect the urogenital diaphragm fibres in most patients. **c**, **d** Coronal section. Levators are mobilised with the obturator fascia (9), but inferior part of the obturator fascia and the posterior parts of crura remain attached to the ishium. The Alcock canal (5) is not opened

Urethrocervicoplasty was performed by excision of full thickness triangles at the level of the bladder neck and tubularization of the formed strip by two rows of interrupted figure-eight sutures (Polidioxanone 5/0) around an 8–10Fr pig-tail catheter.

The bladder plate was closed by one row of sutures. Then, the bladder was placed deep into the pelvic cavity. On the anterior aspect of the formed urethra, parts of the urogenital diaphragm that were already detached from the pubic bones were stitched together when it was possible. Grafts of paravesical fat were bilaterally bluntly separated from the pelvic walls and levators, and closed in front of the bladder neck and the lower part of the anterior bladder wall with interrupted sutures, to form additional tissues layer between the skin and the bladder neck.

Abdominal wall closure

Skin with subcutaneous fat and Camper's fascia was mobilised from muscular sheathes up to the level of anterior superior iliac spine, to allow appropriate midline closure without tension. Transferred flaps or pubic approximation was not used (Fig. 2). The inguinal canals were explored on each side, and division of the patent vaginal processus was performed. The dead paravesical spaces and spaces between Camper's fascia and muscular sheathes were drained within 8–12 days until healed.

The defect between the recti fascia was closed on the midline only cranially with 2–3 sutures without tension. The anterior bladder wall was then sutured with 3–4 stitches to the contiguous divergent edges of the recti fascia. The wound was closed in layers with interrupted sutures in the superficial fascia and dermis, with the formation of the urethral meatus in a typical place for the staged repair. A slightly compressing bandage was applied on the operated site. Epispadias repair was not performed at this stage.

Postoperative care

Patients were allowed to feed orally immediately after transfer from the ICU. Breastfeeding in their mother's arms was encouraged for infants. No method of pelvic immobilisation was applied. Special attention was given to monitoring blood protein levels and wound infection. All patients received broad-spectrum intravenous antibiotics for 8–10 days. Routine urine culture was taken on

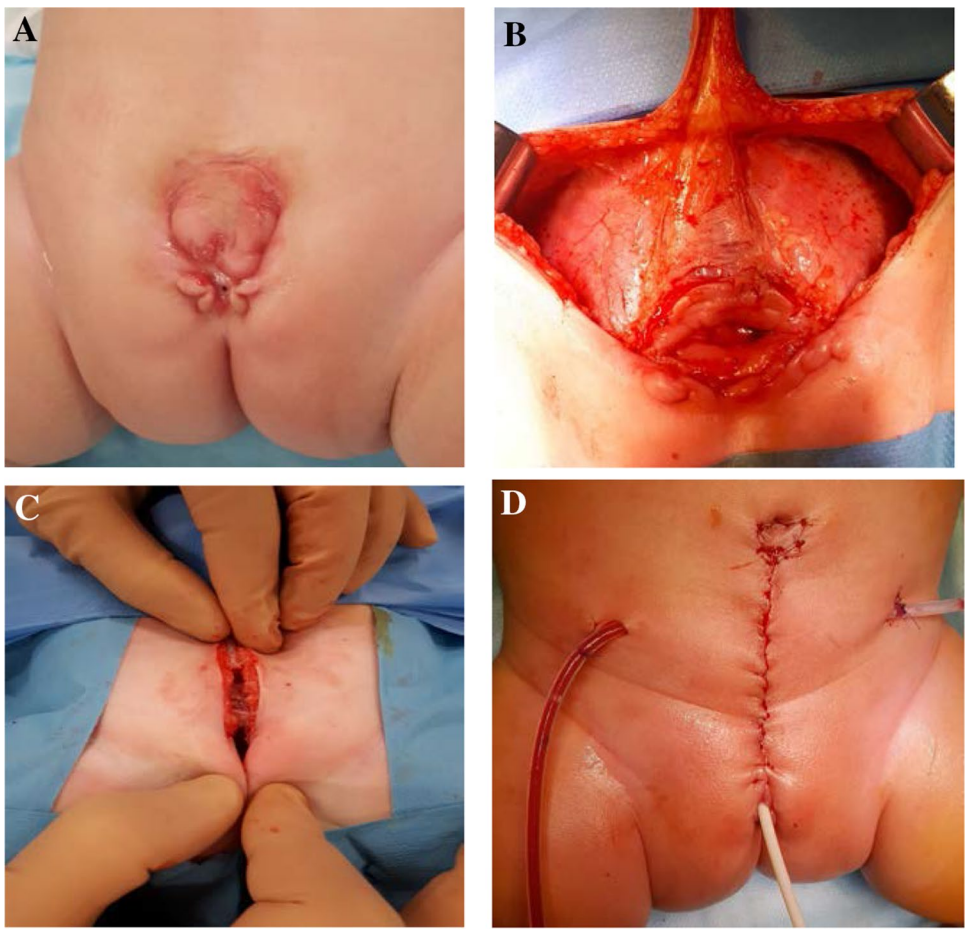

Fig. 2 Bladder and abdominal wall closure without pubic approximation and flaps. **a** External appearance before the closure. **b** Skin with subcutaneous fat widely mobilised from muscular sheathes to the anterior superior iliac spines to avoid tension. **c** There is no tension before abdominal closure. **d** Final appearance after bladder and abdominal wall closure. No suprapubic tube or ureteral stents. No immobilization. Umbilicoplasty is done

the 4–5th postoperative day. Additional antibiotics were administered if there were any signs of wound infection or symptomatic urinary tract infection.

When removing the urethral catheter, several children had signs of obstruction, which required endoscopic incision. In the remaining patients, the urethral catheter was replaced with a 5-Fr pig-tail catheter inserted with fluoroscopy. I previously used endoscopic incisions in cases of obstruction but later leaned in favor of prolonged drainage inserted with fluoroscopy intentionally.

All children continued to be monitored after discharge, and were followed up from 2 to 12 years after bladder closure. During the first year after the operation, ultrasound examinations were performed at 3, 6 and 12 months. Static renal scintigraphy with 99mTc-DMSA for the assessment of renal damage was performed within 2 years after bladder closure. Afterwards all patients were examined at least once a year.

Patients received staged surgical treatment including epispadias repair, antireflux procedures and bladder neck reconstruction. In children with progressively expanding pubic diastases, osteotomy was performed either simultaneously with bladder neck repair or in a separate procedure.

Results

Primary ($n = 25$) and redo ($n = 11$) bladder exstrophy closures were performed in 17 (47%) girls and 19 (53%) boys aged 34 days to 15 years (95% CI), median age of 112 days. The degree of pubic diastasis was 38–118 mm (95% CI median 45 mm). The large 118-mm diastasis belonged to a teenage girl in the re-closure group. During the examination, less compliant bladder plates that did not distend with an increase in abdominal pressure were noted in 15 patients (42%). Bladder plate oedematous and polypoid occurred in nine children (25%). All five children with small bladder plates (17%) were in the primary closure group (Table 1).

The surgeries lasted from 126 to 215 min (median 148 min), and the mean blood loss was 16 ml (95% CI 10–20 ml). Antireflux procedures were performed in eight children. In 34/36 patients, ureteral stents were not required. Indwelling ureteral stents were left in two patients, whose bladder plates were not pliable enough. Elective paralysis and ventilation were not applied in the ICU for more than 6 h. Transfusion was performed in only five children.

The majority of patients 34/36 (94%) were returned to the surgical ward after 1 day in the ICU, and began oral feeding immediately following the procedure.

Table 1 Characteristics for patients with bladder exstrophy

Characteristic	Value
Gender (*n*)	
Male	19/36 (53)
Female	17/36 (47)
Primary	25/36 (69)
Bladder plate oedematous and polypoid	4
Small and polypoid	6
Secondary (after failed closure in other institutions)	11/36 (31)
Bladder plate oedematous and polypoid	5
Small and polypoid	0
Reason for delay (*n*/all)	
Late referral	15/36 (42)
Failed primary closure	11/36 (31)
Small bladder template	6/36 (17)
Prematurity	4/36 (10)
Vertebral and skeletal malformations	2
Age at surgery (weeks)	
Range	34 days–15 years
95% CI	37–318 days (mean = 112)
Pubic diastasis (mm)	
Range	38–118
95% CI	39–54 (mean = 45)

Data presented as number of patients per total, with percentages in parentheses

CI confidence interval

No bladder spasms or signs of acute pain were noted in the ward, therefore, no child required opioids, benzodiazepines or epidural anaesthesia. Intravenous analgesia with non-narcotic analgesics was used for all patients in the ward for an average period of 2.2 days (95% CI 2–4 days).

Patients' bowel function was restored soon after transfer to the ward. A nasogastric tube was left in one patient with peritonitis, and parenteral nutrition was provided for 4 days. Five patients with small bladder plates and two patients who underwent re-do procedures received prophylactic anticholinergics. The surgical drains were removed between 8 and 12 days (median of 9.1 days) after the procedure.

The modified closures were successful in all 36 children within 2 year after closure. No major complications such as dehiscence or bladder prolapse were observed, and there was no need for re-closure (Table 2). Peritonitis occurred in one patient, probably as a result of jatrogenic intestinal perforation on the 3rd day after surgery. All bladder and urethral sutures were competent, despite exudate getting into the paravesical space via a peritoneal defect. Loop ileostomy was performed and ileostomy closure was carried out at 8 weeks.

Table 2 Postoperative complications

Complications	Value
Major complications	0/36 (0)
Dehiscence	0
Bladder prolapse	0
Urethral stricture	0
Minor complications	5/36 (14)
Bladder outlet obstruction	3/36 (8)
Fistula (subsequent healed)	2/36 (6)
Occasional complications (peritonitis)	1/36 (3)

Data presented as number of patients per total, with percentages in parentheses

The urethral catheter was removed between day 14 and 21 in 20 children (Fig. 3). Minor complications occurred in five patients (14%). In three patients in this group, a bladder outlet obstruction occurred due to inadequate opening of the bladder neck, despite showing free permeability for an endoscope (8 Fr). Minor incisions were performed in the area of the bladder neck at the 12 o'clock position for each of the three patients, with subsequent restoration of free micturition. Only one patient, a 15-year-old girl, required intermittent catheterisation for a year after the surgery. Within this period, voluntary voiding was resumed. In the remaining 16 patients, the 8–10-Fr urethral catheter was replaced with a 5-Fr pig-tail catheter inserted with fluoroscopy on day 14–15. These were removed at home 2–7 weeks after discharge when the urine began to go not only via the catheter but also beside him. Under such management, no signs of urethral obstruction were noted.

Eight patients (22%) required additional antibiotics for the treatment of minor wound infection (6/36, 18%) or symptomatic UTI (2/36, 6%). All patients received vesicoureteral reflux antimicrobial prophylaxis after discharge. In two patients (6%), a tiny fistula was noticed, but in both cases it closed spontaneously without urethral catheter assistance.

In the follow-up period, 32 of the 36 patients (89%) had normal ultrasound scans and normal renal outcomes based on eGFR in 2 years after closure. There was no herniation in the gap between the pubic bones in 34 children. In three cases, repairs were performed due to inguinal hernia recurrence.

The skin and subcutaneous fat and Camper's fascia midline closure without rectus fascia closure provided good cosmesis and security of the abdominal wall. No bulging of the anterior bladder wall through the fascial defect was noted in 34 of the 36 children. The midline scar with a flat scar between the anterior bladder wall and Camper's fascia safely filled the defect between the recti fascia, which

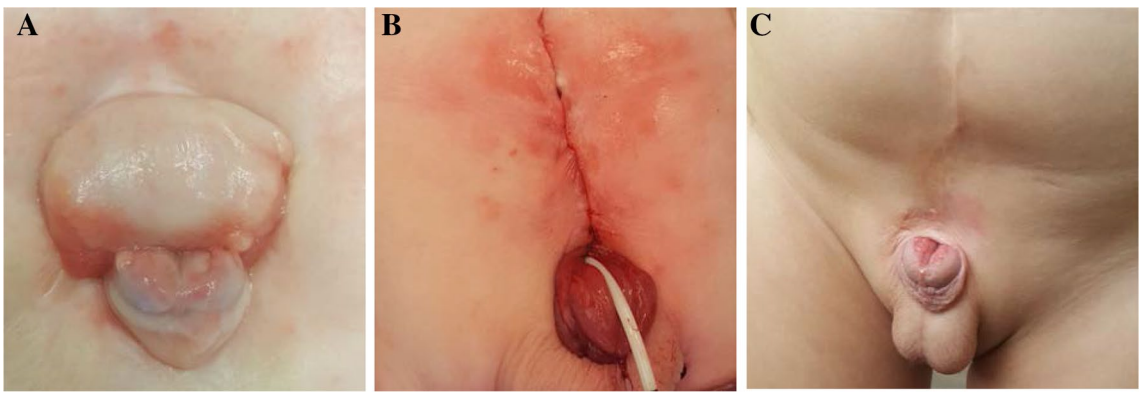

Fig. 3 Bladder exstrophy closure in boy without fascia closure and outcome. **a** At 1 month of life. **b** Normal blood supply and no oedema of the penis (3 days after the closure). **c** Firm low abdominal wall before the epispadias repair (2 years after the closure)

became stronger (Fig. 3) after the subsequent antireflux surgery and bladder neck reconstruction in most patients.

Laxity of the abdominal wall was noted in two patients due to atrophic scars, which appeared within 2 years of bladder closure in the teenage girl with the large pubic diastasis and in a boy 13 years old. In both cases, the space between the recti fascia and between the pubic bones was reinforced with propylene mesh (Fig. 4). Antireflux procedures were performed in 25 (69%) patients, usually 1–2 years after closure (16/25) or at the same time as bladder neck reconstruction (9/25). Osteotomy was performed in three children aged 5–6 years old who had progressively expanding pubic diastasis up to 7–9 cm.

Discussion

Delayed bladder exstrophy closure, performed after 1–2 months of life, has increased in popularity [6] following recent evidence that closure during the first months of life does not negatively influence bladder development [8, 9]. Furthermore, this delayed procedure has been detailed in several publications [6, 9, 10, 16]. Secure initial bladder exstrophy closure is an important predictor of long-term bladder growth and urinary continence [4]. Most authors agree that the success of primary bladder closure is dependent on secure recti fascia closure [2, 3]. The cost of performing fascial closure is additional injury with significant risks, as well as the need for postoperative immobilisation for 3–8 weeks. This requires prolonged intensive pain management, which comes with its own side-effects and complications [12, 13].

In this report, we evaluated the results of a modified bladder closure technique without fascia closure. The focus of this study was on the suggested repair technique, secure abdominal wall closure and complications. We tested the hypothesis that this less invasive delayed closure technique, without postoperative immobilisation, would eliminate the need for prolonged intensive anaesthesia.

Bladder exstrophy closure is considered the first stage of treatment in this technique. Epispadias repair occurs when the child is older and there is a smaller risk of penile ischemia, and hormonal preparation for surgery could be prescribed if needed [17].

The distinctive technical features of the modified procedure and postoperative care compared to other well known techniques are detailed below. The procedure is performed in children after 1 month of life when there is a sufficient layer of subcutaneous fat, which is necessary for safe abdominal wall closure. Urethral closure is achieved after subtotal mobilisation of the crura of the penis or clitoris without penetration into the Alcock's canal, thereby avoiding the pudendal vessels and the perineal and rectal branches of the pudendal nerve injury, which can occur after the radical soft tissue mobilisation (Kelly technique) [14, 18].

Performing an incision along the lateral edge of the arcus tendineus levator ani allows the levators to be peeled apart from obturator internus muscle. With this manoeuvre, the lateral edge of the levator ani muscle is released, freeing the urogenital complex without the need to transect the urogenital diaphragm fibres and neurovascular branches in most patients. The paravesical fat was mobilised bilaterally and closed anterior to the formed urethra, which provides additional closure security, prevents fistula formation and lessens scarring between the urethra and the midline scar. The rectus fascia was closed only on the upper part of the fascial defect without tension, but the lower part of the defect was partially filled with the caudal part of the anterior bladder wall and with the perivesical fat covering the bladder neck and lower part of the bladder. I believe that the coating of the urethra and the bladder

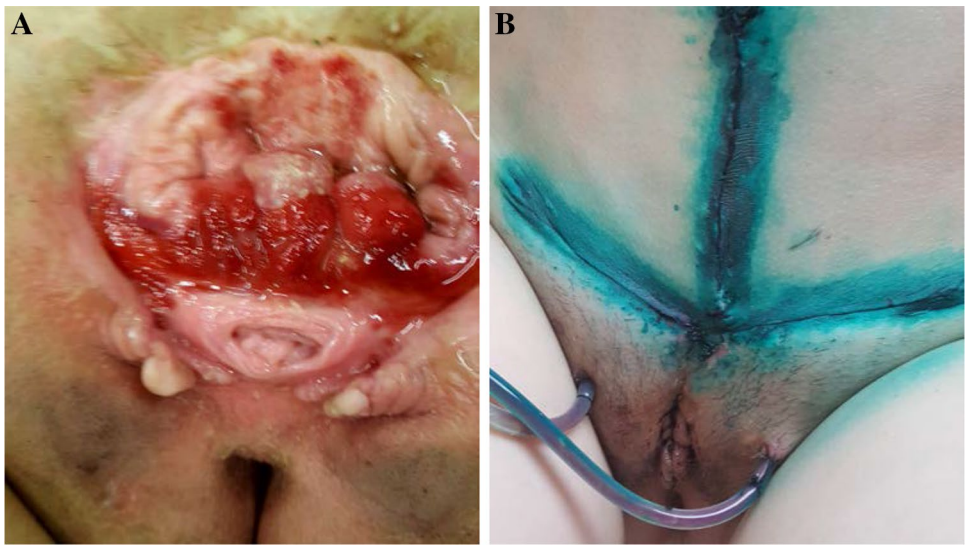

Fig. 4 Teenage girl before the bladder reclosure (**a**) and after strengthening the abdominal wall with mesh on the third day (**b**)

neck with the paravesical fat increases security of closure. The anterior bladder wall was stitched to the divergent edges of the recti fascia to prevent peritoneal protrusion and hernia appearance. Midline closure, without flaps or pubic approximation, reduces tissue damage and improves cosmesis. Surprisingly, midline closure was possible even with a very large diastasis, which was achieved without the need for osteotomy or flaps.

The suggested technique for delayed bladder exstrophy midline closure was 100% successful. The avoidance of fascia closure, pubic bone approximation, osteotomy, ureteral catheters, use of transferred flaps and immobilisation led to a reduction in perioperative treatment, especially with regard to analgesia, immobilisation, transfusions and parenteral nutrition [10]. Moreover, the list of complications experienced by patients was also decreased. Only minor complications occurred, including tiny fistulas that closed spontaneously (6%) and bladder outlet obstruction (8%), which was easily resolved with small endoscopic incisions. There were no signs of acute pain during the postoperative period, and no analgesics were needed after 2–3 days. Therefore, complications were minimised and our proposed hypothesis was confirmed.

Considering that our findings contradict the generally accepted ideas about the need for immobilisation and anaesthesia, it is important to discuss the possible causes of discrepancies. The absence of severe pain could be explained by the less invasive surgery technique, which avoided closure of the fascia, and the lack of immobilisation allowed earlier onset of enteral nutrition, especially breastfeeding, skin-to-skin contact and the complete absence of bladder spasms.

Bladder spasms after bladder exstrophy closure are considered a serious problem, demanding the use of a whole armamentarium of measures including epidural analgesia, lung ventilation with elective palsy, anticholinergics, opioids and benzodiazepines, to reduce pain and subsequent transient rises in intra-abdominal pressure that can lead to wound dehiscence [19, 20]. It is known that bladder spasms usually occur as a result of the detrusor muscle reaction on a bladder mucosa irritation or as a reaction to external irritation [21]. The reduced mucosal irritation in our patients can be explained by avoiding the use of a suprapubic tube and ureteral catheters, which, given the small volume of the closed bladder, may induce mucosal irritation and bladder spasms. Moreover, the proposed technique involves a lack of external compression of the bladder, which usually occurs after pubic approximation with rectus fascia closure, causing pressing of the bladder wall on the catheters. The delay in closure probably also plays a role in reducing mucosal irritability. During the delay, the mucosa of the bladder plate is in contact with a protective film or diapers, therefore, becoming less sensitive and more resistant to irritation.

Osteotomy is reserved for patients with a large pubic diastasis, mainly for those with cloacal exstrophy, and for patients with classical exstrophy who experience a marked increase in diastasis over time. Our three patients with expanded diastasis of 7–9 cm underwent osteotomy 5–6 years after closure, while in most patients diastasis did not increase to the same extent by this age. Thus, due to variation in the severity of diastasis among patients, many can avoid osteotomy. Considering osteotomy as a method for improving appearance and continence, it seems reasonable to perform osteotomy as a stage of staged exstrophy repair in select children at an age when their bone density is higher than that of newborns [1, 22].

This research has certain limitations. Firstly, this is a retrospective analysis, and the surgeries were performed over a long period of time. Another limitation is the absence of a control group. Despite these limitations, this report describes a reliable approach to obtain successful surgical closure of the bladder in children with classical bladder exstrophy.

Conclusions

In this study, we show that successful classic bladder exstrophy closure can be technically easier and safer for the patient when performed without full fascia closure. The results confirm our proposed hypothesis that the less invasive technique for bladder exstrophy closure eliminates the need for immobilisation and prolonged anaesthesia in the postoperative period.

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Compliance with ethical standards

Ethical appproval The study complied with principles of the declaration of Helsinki (1964), and received approval from the institutional Ethical and Clinical Research Committee (2010).

Informed consent Parents of patients gave informed consent to the work.

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