ORIGINAL ARTICLE

Closure of Exstrophies: Avoiding Bladder Dehiscence

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ABSTRACT

Introduction: Bladder exstrophy (BE) is a complex malformation involving urinary, genital, and musculoskeletal system. BE repairs are challenging and failure can manifest as complete bladder dehiscence (BD). The rate of BD is still high in our institution. Therefore, since 2017 we have implemented a protocol for BE. The objective of this study is to describe the procedures for BE and how the new protocol reduced the incidence of BD. **Methods:** This is a retrospective descriptive study of patients diagnosed with BE in Dr. Hasan Sadikin Hospital, during 2009 - 2022. Since 2017, protocol for bladder closure was implemented: timing of surgery >6 months old, complete incision of the urogenital diaphragm, osteotomy, and modification of stent placement and dressing. Patients' characteristics, incidence of BD, and factors affecting its occurrence were analyzed using unpaired-t, Mann Whitney, and Fisher exact test, p<0,05 = significant. **Results:** There were 30 BE patients (13 before, 17 after protocol). After implementing the protocol, patients were significantly younger (p=0,018), incision of urogenital diaphragm, use of large ureteral stent, and simple transparent dressing were more frequently performed (p<0,001 each). BD was significantly reduced after implementation of protocol (84,6% vs 41,2%; p=0,016) and more frequently occurred in <6 months patients (p=0,030; PR(95%CI)=2.00(1.02-3.91). Incision of urogenital diaphragm (p=0.026; PR=0.49(0.26 – 0.90)) and the use of simple transparent dressing (p=0.026; PR(95%CI)=0.49(0.26 – 0.90)) were associated with reduced BD. **Conclusion:** Age >6 months old, incision of urogenital diaphragm, and application of simple transparent dressing significantly reduce the incidence of BD.
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INTRODUCTION

Bladder exstrophy (BE) is a part complex of malformation involving the urinary tract, genital tract, the musculoskeletal system and sometimes the intestine. The most common malformations are related to defects of the abdominal wall, bladder, genitalia, pelvic bones, rectum, and anal tract (1). The incidence of BE is estimated to be 2.15 cases per 100,000 livebirths in the United States and 0.5 cases per 100,000 livebirths in China. However, incidence of BE in Indonesia is not available (1,2). Failure of mesodermal reinforcement of the abdominal wall and/or the cloacal membrane has been postulated to play a major role in the pathogenesis of BE, often BE is associated with other mesodermal anomalies such as omphalocele, hernias, renal agenesis, and imperforate anus, requiring complex reconstruction (3). The primary objective of surgical management of BE is to achieve a successful primary closure due to decreased overall costs, decreased bladder inflammation and fibrosis, improved bladder growth, and decreased need for diversion (4). The multiplicity of BE repairs that exist to close an exstrophy bladder reflects the fact that it is one of the most challenging tasks for the pediatric surgeon (5).

Surgical reconstruction of BE requires numerous reconstructive surgeries. The initial surgeries involve the closure of the bladder, bony pelvis, and anterior abdominal wall, with subsequent surgeries for epispadias and bladder neck repair. It is a complex process that demands precision and accuracy (4). The primary objective in functional primary closure, the most important part of this procedure is the bladder, posterior urethral and abdominal wall closure to convert BE into a complete epispadias with the urethra well up to the proximal shaft or mid-shaft of the penis. Failures can manifest as bladder prolapse, neourethral stricture, obstruction soft-tissue loss, vesicocutaneous fistula, and the main failures complete bladder dehiscence (BD) (1). The incidence of BD in previous studies were high with 4 out of 25 cases (16%) and 10 out of 21 cases (42%) (6,7). The rate of BD dehiscence is high in our institution,

so since 2017 we have implemented protocols in our institution to prevent BD such as timing of surgery > 6 months of age, complete incision of the urogenital diaphragm, routine osteotomy, modification of stent placement and dressing. The objective of this study is to describe our surgical procedures for BE and how the new protocol may reduce the incidence of BD.

MATERIALS AND METHODS

This is a retrospective descriptive study of patients diagnosed with BE in Dr. Hasan Sadikin Hospital, a referral provincial hospital in West Java, Indonesia during 2009 - 2022. On 2017, in response to high incidence of postoperative BD, we modify the protocol for management of patients with bladder exstrophies, including: 1) timing of surgery at >6 months of age; 2) complete incision of urogenital diaphragm; 3) routine osteotomy; 4) large caliber of ureteral stents which placed separated from the wound site; and 5) transparent wound dressing. Data such as diagnosis, sex, age of surgery, osteotomy procedure, and the incidence of BD was recorded. The protocol was discussed and agreed among 6 pediatric surgeons in our institution. However, in the first two years after its implementation, some pediatric surgeons had not fully adhered to the new protocol, especially in terms of timing of surgery and osteotomy procedure. Repeated discussion regarding the protocol had undertaken to reassure its implementation by the staffs.

Patient's characteristic such as age at surgery, sex, distance of pubic diastasis, intraoperative measures, and the incidence of BD, as well as the difference between both groups were studied. Factors affecting the occurrence of BD was also analyzed using unpaired-t, Mann Whitney, and Fisher exact test, p<0,05 = significant.

This study was approved by Research Ethics Committee Hasan Sadikin General Hospital, Bandung, West Jawa, Indonesia No. LB.02.01/X.6.5/KEP/368/20.

RESULTS

There were 30 patients of BE treated during 2009 – 2022, 13 patients were treated before the implementation of protocol for BE, and 17 patients were treated after it. Demographic data is shown in Table I.

The median age of BE patients was significantly higher after implementation of new protocol than before it (4.4 m.o. vs 1 m.o.; p=0.018). There was also an increase percentage of patients aged >6 months old (p=0.139) and who underwent osteotomy (p=0.123) after the implementation of the protocol. Incision of urogenital diaphragm, the use of large caliber ureteral stent, and simple transparent dressing were significantly higher after the protocol (p<0.001 each).

Table I: Characteristic of BE patients before and after the implementation of new protocol.

Characteristic	Group		p value
	Before new protocol n=13	After new protocol n=17	
Age (months)			
Median (IQR)	1 (0.2 – 16.5)	4.4 (4.0 – 5.1)	0.018[b]*
Min – Max	0.1 – 60.0	3.0 – 5.5	
Age criteria, n (%)			
<6 months	9 (69.2)	5 (29.4)	0.139[c]
≥6 months	4 (30.8)	12 (70.6)	
Sex, n (%)			
Male	4 (30.8)	3 (17.6)	0.666[c]
Female	9 (69.2)	14 (82.4)	
Pubic diastasis (cm)			
Mean ± SD	4.3 ± 0.8	4.2 ± 1.2	0.565[a]
Min – Max	3.0 – 5.5	2.5 – 8.0	
Osteotomy, n (%)			
Yes	7 (53.8)	14 (82.4)	0.123[c]
No	6 (46.2)	3 (17.6)	
Complete incision of urogenital diaphragm (%)			
Yes	0 (0)	17 (100)	<0.001[c]*
No	13 (100)	0 (0)	
Large caliber ureteral stent, n (%)			
Yes	4 (30.8)	17 (100)	<0.001[c]*
No	9 (69.2)	0 (0)	
Simple transparent dressing, n (%)			
Yes	0 (0)	17 (100)	<0.001[c]*
No	13 (100)	0 (0)	
Incidence of BD, n (%)			
Yes	11 (84.6)	7 (41.2)	0.016[c]*
No	2 (15.4)	10 (58.8)	

Note: Statistical analysis using [a]Unpaired t-test, [b]Mann Whitney test, [c]Fisher-Exact test

The median age of patients with BD was significantly younger than those without BD (1.8 m.o. vs 10.5 m.o.; p=0.044). Children <6 months old had a higher risk to develop BD compared to those older than 6 months old (80.0% vs 40%; p=0.030). The prevalence ratio of 2,00 shows that children <6 months old had twice the risk of developing BD. The mean pubic diastasis distance was narrower in the group with BD compared to without BD (4.1 cm vs 5.1 cm; p=0.007) (Table II).

Incision of urogenital diaphragm was significantly more common in patients without BD compared to with BD (10% vs 7%; p=0.025). The prevalence ratio of 0.49 indicates that the incision may reduce the risk of BD by 51%. The use of simple transparent dressing was significantly more common in patients with BD (10% vs 7%; p=0.026). Prevalence ratio of 0.49 indicates the procedure can reduce the risk of Bladder Dehiscence by 51%.

Table II: Association of demographic factors, intraoperative procedures, and the incidence of BD.

Characteristic	Total n=30	Incidence of BD		P value	PR (95% CI)
		Yes n=18	No n=12		
Age (months)					
Median (IQR)		1.8 (0.3 – 13.5)	10.5 (5.5 – 78.0)	0.044[b]*	
Min – Max		0.1 – 60.0	0.1 – 156.0		
Age criteria, n (%)					
<6 months	15	12 (80.0)	3 (20.0)	0.030[c]*	2.00 (1.02 – 3.91)
≥6 months	15	6 (40.0)	9 (60.0)		
Sex, n (%)					
Male	7	5 (71.4)	2 (28.6)	0.669[c]	1.26 (0.70 – 2.28)
Female	23	13 (56.5)	10 (43.5)		
Pubic diastasis (cm)					
Mean ± SD		4.1 ± 0.8	5.1 ± 1.1	0.007[a]*	
Min – Max		2.5 – 5.5	4.0 – 8.0		
Osteotomy, n (%)					
Yes	21	12 (57.1)	9 (42.9)	0.704[c]	0.86 (0.47 – 1.55)
No	9	6 (66.7)	3 (33.3)		
Complete incision of urogenital diaphragm (%)					
Yes	17	7 (41.2)	10 (58.8)	0.026[c]*	0.49 (0.26 – 0.90)
No	13	11 (84.6)	2 (15.4)		
Large caliber ureteral stent, n (%)					
Yes	21	11 (52.4)	10 (47.6)	0.249[c]	0.67 (0.39 – 1.15)
No	9	7 (77.8)	2 (22.2)		
Simple transparent dressing, n (%)					
Yes	17	7 (41.2)	10 (58.8)	0.026[c]*	0.49 (0.26 – 0.90)
No	13	11 (84.6)	2 (15.4)		

Note: PR=Prevalence Ratio, CI=Confidence Interval, statistical analysis using [a]Unpaired t-test, [b]Mann Whitney test, [c]Fisher-Exact test

DISCUSSION

BE is a severe congenital malformation caused by a developmental disorder of the ventral part of the body that results in the opening of the bladder from the abdominal cavity (8). Based on previous studies, there is a higher incidence in male pediatric patients compared to female pediatric patients (9). The sex ratio will vary according to the study location, the sex ratio in pediatric patients in Sweden had a sex ratio of 53.7% male and 46.7% female (10). In this study, BE is more common in female than male patients. The epidemiologic difference may be related to difference in the number of patients and the geographical location of the study site.

This study showed that after implementation of the new protocol, median age at surgery was significantly older (p=0,018). However, after the new protocol, 5 patients still underwent operation before the age of 6 months (1 months old: 3 patients, 5 months old: 2 patients). This was happened because in the first 2 years some surgeons did not fully implement the protocol, especially in term of age at surgery. Incision of urogenital diaphragm and the use of large caliber ureteral stent, as well as simple transparent dressing were more frequently performed after the new protocol (p<0,001 each). The mean pubic diastasis width was not statistically different between two groups, but osteotomy procedure was more frequently performed after the new protocol. There were still 3 patients aged 1 months old who did not undergo osteotomy after the protocol. In such cases, the surgeons assessed non-tension bladder closure intraoperatively.

In this study, the incidence of BD was significantly reduced after implementation of the protocol (84,6% vs 41,2%; p=0,016). We analyzed factors that may influence BD in patients with BE. We found that children with BD was significantly younger than without BD (p=0.044) and <6 months babies had twice the risk of developing BD. This may be due to smaller bladder template, which cause bladder dissection and closure without tension difficult. Bueno at al. found higher BD incidence after neonatal repair (15% neonatal repair vs 0% delayed repair) (11). Delayed closure of the smaller bladder template in age over 6 months may allow sufficient time for native bladder tissue to grow to a size suitable for successful closure (12,13).

In our study, the mean width of diastasis was 4,3 cm. If pubic bones are more than 4 cm apart, or if the pelvis is not malleable, osteotomy should be performed (1). Interestingly, in this study pubic diastasis was significantly narrower in patients with BD. This may be due to the greater proportion of patients <6 months

who have BD, who may have a narrower pubic diastasis compared with older children. Before implementation of protocol, the width of diastasis is not the parameter for performing osteotomy for patients with BE, but rather the age of the patients. We did not routinely perform osteotomy on newborns because the pelvic considered malleable.

Osteotomy provides a positive outcome in patients with BE, as it reduces the risk of wound dehiscence, posterior iliac osteotomy is safe and can be repeated if necessary (2). Pelvic osteotomy performed at the time of initial closure confers several advantages, including (1) easy approximation of the symphysis with diminished tension on the abdominal wall closure and elimination of the need for fascial flaps; (2) placement of the posterior vesicourethral unit deep within the pelvic ring, enhancing bladder outlet resistance; and (3) bringing the large pelvic floor muscles near the midline, where they can support the bladder neck and aid in eventual urinary control (1). We found that performing osteotomy was not associated with the incidence of BD. This may be because osteotomy is not the main determinant of successful bladder closure and must be supported by other factors, such as incision of urogenital diaphragm, effective drainage, and good wound care.

Complete urogenital diaphragm incision was performed in all cases after implementation of the protocol. We did not have sufficient data in medical record regarding this procedure was performed or not before. However, from the discussion among surgeons, they did not explore deeply into the urogenital diaphragm before, and only dissected around the bladder until it can be closed without tension. In this study, we found that urogenital diaphragm incision reduced the risk of BD by 51%. Intact urogenital diaphragm fibers can lead to unsuccessful bladder closure. In the absence of a complete division of these fibers, it is difficult for the surgeon to lower the posterior vesicourethral unit deep into the pelvis. Therefore, when the anterior pelvis is brought together, the intact fibers move the posterior vesicourethral unit to a superficial and anterior position, making the unit more susceptible to prolapse or dehiscence (1). Study by Davis et.al. found that the urogenital fibers were completely intact bilaterally in repeat closures in 74 patients (79.6%) (14).

Although the use of large caliber ureteral stent was more frequently used after the new protocol (p<0,001), we found that its application was not associated with lower incidence of BD. We used the largest size that fits in the ureter (feeding tube no. 3,5 Fr to 8 Fr.). The stent was fixated at the skin, away separated from the surgical wound. Large stent lets the urine drain completely from the ureter with minimal spillage to the bladder, thereby preventing ineffective drainage or urinary obstruction, bladder distension, stretched stitches, and urine spillage to the wound (14).

Wound dressing after bladder closure is still rarely discussed. After implementation of the protocol, we used simple transparent dressing using Tegaderm® in all cases (p<0,001), which replaced every 3 days or when the wound becoming wet. Patients who cared with simple transparent dressing showed a lower risk of BD and this procedure can reduce the risk of BD by 51%. This type of dressing facilitates direct wound observation without frequent removal and prevents urine spillage from urethra to the wound, thereby supporting wound healing. It also prevents accidental removal of the stent in case with bulky gauze dressing.

In this study, although the incidence of BD was significantly reduced after implementation of the new protocol, but its incidence was still high. The cause of BD was taught due to surgical site infection, in which occurred in all 7 patients with BD. Two of them also had positive urine cultures. A case report by Lowe et al observed 20 patients who developed BD after initial closure. Bladders prolapse (46%) and wound infection (42%) were the main causes of dehiscence. Prolapse is usually due to poor pelvic fixation and failure to maintain bladder outlet reconstruction. Wound infection is usually caused by inadequate urinary drainage and inadequate catheter or urethral passage (15).

This study provides important data in identifying risk factors and future preventive measures to reduce the rate of BD. However, it has some limitations. The sample size is relatively small considering that the incidence of BE is quite rare. Some surgeons did not fully follow the new protocol in the first two years, especially in terms of operating age and osteotomy. In addition, several accompanying conditions were not analyzed, such as nutritional status, infectious and chronic diseases, anatomic detail genitourinary and musculoskeletal system, and other postoperative complications. Further studies need to be conducted to analyze the outcomes of this new protocol in the treatment of BE, in a longer term, with larger sample sizes, and with assessment of a wider range of risk factors that may influence the development of BD.

CONCLUSION

Age >6 months old, incision of urogenital diaphragm, and application of simple transparent dressing significantly reduce the incidence of BD.

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