ACH Med J
(2025) 1: 010-014
ISSN:2822-5872
Doi : 10.5505/achmedj.2025.85057

RESEARCH ARTICLE

Bladder Exstrophy Reconstruction Without Osteotomy; Evaluation of 50 Cases

Suleyman Tagci[1], Gokhan Demirtas[2], Bilge Karabulut[1], Tugrul Tiryaki[1]

[1]Department Of Pediatric Urology, Ankara Bilkent City Hospital, Ankara, Turkiye
[2] Department Of Pediatric Urology ,Sincan Training And Research Hospital, Ankara,Turkiye

Article Info

Received Date: 25.01.2025
Accepted Date: 09.02.2025

Keywords:
Bladder Exstrophy,
Osteotomy,
Urinary incontinence

ORCIDs of the authors:
ST :0000-0002-5763-8916
GD :0000-0003-0787-2330
BK :0000-0002-3638-2253
TT :0000-0002-9544-1137

Abstract

Introductıon: To evaluate the outcome of our patients who underwent BE (bladder exstrophy)repair without osteotomy and to determine whether they are continent, upper urinary system functions, and complication rates.

Methods: The data of 50 patients who applied to our clinic due to bladder exstrophy between 2010 and 2022 were analyzed retrospectively. The gender of the patients, the age of the operation, the surgical interventions, the complications detected, whether they were continent or not, and their upper urinary system functions were recorded.

Results: The data of the 50 patients diagnosed with classical bladder exstrophy were evaluated and included in the study. Nine of these patients were female, and 41 were male. The patients' mean age was 71.2 months(6-264 months). The mean follow-up period was determined to be 70.8 months. Bladder closure was performed in 13 of 50 patients, bladder closure + bladder neck repair + epispadias repair was performed in 29 patients, and augmentation was performed for eight patients. Augmentation was recommended for five patients in the follow-up, but patients did not accept it. Skin dehiscence occurred in 2 patients, and a primary suture was performed for them. Three patients were reoperated for bladder dehiscence. During the follow-ups, cystolithotripsy was performed in 3 patients,and csytolitotomy was performed in 2 patients. Bladder dehiscence developed in 1 patient after csytolitotomy, and he was operated on.25 of the 50 patients included in the study were under five years of age. The continence status of the remaining 25 patients was evaluated. Fifteen (60%) of 25 patients were found to be continent.

Conclusion: Although strong advocates of bladder exstrophy repair with osteotomy exist, we recommend repair without osteotomy as an easy-to-apply and low-complication approach.

Correspondence Address: Üniversiteler Mahallesi 1604. Cadde No: 9 Çankaya Ankara - Türkiye
Phone: +90 530 938 35 22 / **e-mail:** suleyman_tagci@hotmail.com

Introduction

Bladder exstrophy (BE) is a congenital developmental disorder of the bladder. The anterior bladder wall is undeveloped, and the posterior bladder wall protrudes from the anterior abdominal wall.[1] The prevalence is about 2 in 100,000 births.[2] The condition is seen two times more frequently in boys than girls. However, some studies have also shown a very high male preponderance, with male to female = 6 to 1.[3]

Surgical treatment of patients with bladder exstrophy is still controversial. Some surgeons, particularly in males, recommend a single-stage repair in the neonatal period. Conversely, other surgeons advocate limiting neonatal surgery to bladder closure and postponing epispadias repair until later in life, usually between 6 months and 2 years.[4,5] It is also controversial whether osteotomy should be performed during bladder closure. The use of pelvic osteotomy to close bladder exstrophy has a long medical history. It is widely accepted, except in the first period of neonatal life (less than 72 hours), when osteotomies may not be required due to the malleability of the bones in early life.[6]

The primary aims of bladder exstrophy repair are to achieve urinary continence and to ensure the long-term preservation of the upper urinary tracts. There is considerable variation in the surgical techniques employed in BE reconstruction. It is regrettable that, despite the considerable technical advances made over the years, the effective management of this condition continues to present a significant challenge. The role of osteotomy in initial bladder closure remains a topic of significant debate and investigation. Some authors especially emphasize that osteotomy should be added to repair bladder exstrophy after the first 72 hours of life.[7] Some authors suggest that the success rate concerning closure in the repair of bladder exstrophy without osteotomy is similar to the series with osteotomy.[8] In our clinic, the repair of our bladder exstrophy cases is performed by approximating the pubic bones without osteotomy in the neonatal period. After the neonatal period, the repair is provided without osteotomy and approximating the pelvic ring.

This study seeks to evaluate patients who have had bladder exstrophy repair to establish whether they are continent, to determine the functionality of the upper urinary system, and to identify any complication rates who did not undergo osteotomy (pelvic ring approximated and nonapproached).

Material and Methods

After obtaining approval from the institutional ethics committee, we retrospectively analyzed the data of all patients who presented to our clinic with bladder exstrophy between 2010 and 2022. Patients with a follow-up period of fewer than six months who were operated on for duplicated bladder exstrophy and underwent ureterosigmoidostomy were excluded from the study. Patients were followed up every three months. It was recorded whether the patients were continent or not, how many hours they stayed dry if they had continent, whether they had fever urinary tract infection, used prophylactic antibiotics, used anticholinergics, used a clean intermittent catheter, or not. After physical examination, biochemistry, urinalysis, and ultrason are performed, bladder capacity measurement is performed once a year, and control urodynamics is performed when necessary. The patient's gender, age at the time of surgery, surgical procedures, complication rates, continence success rate, and upper urinary system functions were documented. Although continence is defined differently in many studies, daytime urinary continence was evaluated by the age of five years in our study. Patients were considered continent if completely dry for ≥3 hours, with or without a clean intermittent catheterization. [9] The patients were divided into three groups: those who had the first surgery in our clinic in the first 3 postnatal days, those who had the first surgery in our clinic after the third postnatal day, and those whose first surgery was performed in another center and was unsuccessful and underwent secondary closure by us. We performed the surgeries of all our patients using modern staged repair of bladder exstrophy techniques (MSRE).[10] Primary closure was performed in the newborn, epispadias correction at 12 months of age, and bladder neck reconstruction (BNR) at 4-5 years of age. Bladder augmentation (BA) was recommended and performed in patients with low bladder compliance and incontinence.

Statistical analysis

All statistical analyses were performed using SPSS version 25 (IBM Corporation, NY, USA). The chi-square test was used for categorical comparisons. Mann-Whitney U and Kruskal-Wallis tests were used to analyze independent variables. A p-value <0.05 was considered to be statistically significant.

Results

Three patients who were operated on for duplicated bladder exstrophy and three patients who underwent ureterosigmoidostomy operation were excluded from the study. Fifty patients operated on for classic bladder exstrophy were included in the study without gender discrimination. Nine of these patients were female and 41 were male. The mean age of the patients was calculated as 71.2 months (6-264 months) SD: 65.516. The mean follow-up period was 70.8 months (6-264 months) SD: 65.057. Primary operations were performed in 13 patients in the first three days of newborns, and 5 patients after the first 3 days of newborns due to bladder exstrophy in our clinic. Secondary bladder closure and complementary surgeries were performed in 32 patients whose first operations were performed in other centers.

Bladder closure was performed in 13 of 50 patients, bladder closure + bladder neck repair + epispadias repair in 29 patients (augmentation was recommended for 5 patients in the follow-up, but patients did not accept), and augmentation was performed for 8 patients (2 girls, 6 boys). Skin dehiscence occurred in 2 (4%) patients, and a primary suture was performed for them. Three patients (6%) were reoperated for bladder dehiscence. During the follow-ups, cystolithotripsy was performed in 3 patients (6%), cystolitotomy was performed in 2 (4%) patients (bladder dehiscence developed in 1 patient after cystolitotomy, and he was operated on.), and stone excision from the urethral diverticulum was performed in a patient. Bladder closure with the staged approach is expected to reach the appropriate age for the planned surgical interventions in thirteen patients. 25 of the 50 patients included in the study were under five years of age. The continence status of the remaining 25 patients was evaluated. 15 (60%) of the patients were found to be continent, and 10 (40%) were found to be incontinent. Of the 15 continent patients, 8 were augmented bladder patients. All patients who underwent augmentation were observed to be dry. When augmented patients were excluded, 7 (%41) of the remaining 17 patients were found to be continent. There was no significant difference in terms of continence between those who had only bladder closure and those who had epispadias repair in addition to bladder closure (p = 0.79).

Expected bladder capacity for age was normal or above normal in only two patients. The mean bladder capacity for age was -90 cc. The distribution of the ratio of the current bladder capacity of the patients to the expected bladder capacity by age is as in Table 1, and its median value was calculated as 55 percent.

Table 1. Bladder capacity of the patients to the expected bladder capacity by age

Bladder capacity	Frequency	Percent
<%10	2	% 4
%10-%25	7	% 14
%25-%50	14	% 28
%50-%75	15	% 30
>%75	12	% 24
Total	50	% 100

Discussion

Trendelenburg said that "All the patients of exstrophy are born with the potential of continence." at 1906.[11] Woodhouse and Kellett said that "All the patients of exstrophy bladder are born with the potential for fertility and continence." at 2006.[12]

The primary objectives of bladder exstrophy repair are to achieve urinary continence and to ensure the long-term preservation of the upper urinary tracts. There is considerable variability in the surgical techniques employed in the reconstruction of bladder exstrophy.

It is regrettable that, despite the considerable technical advances that have been made over the years, the management of this condition remains a significant challenge. Patients frequently report a lack of satisfaction and a sense of profound frustration. The outcomes of bladder exstrophy repair are often suboptimal, necessitating multiple specialized surgeries and frequent hospital admissions. In traditional reconstructive surgery for exstrophy bladder, the defect in the anterior abdominal wall can be repaired either with or without osteotomy, following bladder closure and epispadias repair.

A variety of osteotomy techniques have been described that facilitate the approximation of the pubic bones to the midline, thereby providing efficient protection of the closed bladder.[13] Additionally, osteotomy assists in the restoration of the transformation of the urogenital diaphragm from a rectangular to a triangular shape, which in turn facilitates the increase in corporal length following corporoplasty.

The role of osteotomy continues to be an im-

portant subject in bladder closure. The preference for osteotomy over non-osteotomy methods is based on several factors. Firstly, the closed bladder is secured within the reconstructed pelvic ring, ensuring stability. Secondly, the levator ani base is positioned to support the bladder base. Thirdly, the length of the corpora is not lost in corporoplasty, which is an important consideration. The prevailing view is that osteotomy is not required as an adjunct to bladder exstrophy closure performed within 72 hours of life due to the relatively malleable nature of the pelvic bone. After 72 hours of life, osteotomy is strongly recommended by most of the authors. There are also studies indicating that there is no difference in the success rate of bladder closures.[3,8,14-16] Nevertheless, symphysis diastasis is commonly seen to recur after the pelvic closure methods used.[17] In addition, due to complication rates, technical difficulties, and the need for postoperative immobilization, methods without osteotomy have begun to be preferred.

Özcan et al. demonstrated that a notable proportion of patients who underwent anterior diagonal iliac osteotomy exhibited a recurrence of diastasis of the pubic bones at a mean follow-up period of 34 months.[18]

Only a few studies investigate pubic diastasis following different pelvic osteotomy procedures with adequate follow-up.[19-22] Castagnetti et al. compared prospectively patients with and without osteotomy after initial closure. The mean recurrent pubic diastasis distance does not show a significant difference in the long term patients with and without osteotomy.[23]

During long-term follow-up, there were no significant differences observed in the width of pubic diastasis, the number of surgeries related to exstrophy, incontinence rates, or the need for clean intermittent catheterization for bladder emptying.[23]

According to Kertai et al., despite the hip morphology specific to bladder exstrophy, long-term hip function in adolescent adults was not impacted after symphysis approximation without osteotomy in infancy. The recurrence of symphysis diastasis after this procedure was in line with the long-term results seen after osteotomy.[17]

According to a case series by Mushtaq et al., 70 out of 74 patients (95%) achieved successful bladder closure through primary closure without osteotomy and postoperative immobilization.[14]

The data of 29 patients were evaluated in the study of J. S. Ellison et al. Continence was achieved in 3 out of 10 patients with osteotomy and 8 out of 17 patients without osteotomy (osteotomy status unknown in 2 patients); no significant correlation was found between osteotomy and continence status.[8] Ahmed et al.'s study, the continence rates were determined as 78.7%, spontaneous voided continence 17.0%. In our clinic, the continence rates were determined to be 41% in patients who did not undergo osteotomy, which is consistent with the literature.[24]

In the study of Marco Castagnetti et al., the data of 14 exstrophy patients were examined. Osteotomy was performed on eight patients, and 4 of these eight patients were found to be continent. Six patients did not undergo osteotomy, and 3 of these six patients were found to be continent. No difference was found in continence status in patients with and without osteotomy ($p=0.07$).[23]

In the study by Preeya et al. 286 bladder exstrophy patients, 186 of whom were in the neonatal period, were operated on by osteotomy. In 23 of the patients who were operated on during the neonatal period and in 3 of the patients who were operated out of the neonatal period; bladder dehiscence was observed in 26 (9%) of 286 patients who were operated on by osteotomy.[25] In the study by Hofmann et al., bladder dehiscence was not observed in 66 patients who underwent extrophy repair without osteotomy.[26] In our series, our bladder dehiscence rate was 6%, which is correlated with the literature.

Pelvic ring closure without osteotomy was accomplished in all infants with classical BE under one week of age in our department. We do pubic approximation in children older than one week.

Conclusion

In conclusion, despite the presence of Exstrophy Epispadias Complex-specific hip morphology, long-term hip function is preserved in adult and adolescent patients after symphyseal approximation without osteotomy in infancy. The pubic approximation is significantly less invasive and more straightforward than pelvic osteotomy.

Studies have shown that osteotomy does not affect continence and bladder dehiscence. In our study, we found that our results were consistent with the literature regarding continence, bladder dehiscence rate, and the preservation of upper urinary system functions, all of which are crucial for patients with bladder exstrophy. We argue that successful bladder exstrophy repair can be performed without osteotomy.

References

1. Buyukunal CS, Gearhart JP (2011). A short history of bladder exstrophy. Semin Pediatr Surg 20(2):62-5
2. Siffel C, Correa A, Amar E, et al. (2011). Bladder exstrophy: an epidemiologic study from the International Clearinghouse for Birth Defects Surveillance and Research, and an overview of the literature. Am J Med Genet C Semin Med Genet 15;157C(4):321-32
3. Ebert AK, Reutter H, Ludwig M, et al (2009) The exstrophy-epispadias complex. Orphanet J Rare Dis 30;4:23
4. Mitchell ME (2005). Bladder exstrophy repair: complete primary repair of exstrophy Urology 65:5–8
5. Baird AD, Nelson CP, Gearhart JP (2007). Modern staged repair of bladder exstrophy: a contemporary series. J Pediatr Urol 3(4):311-5
6. Mohan S Gundeti (2019). Surgical techniques in pediatric and adolescent urology.Jaypee Brothers Medical Publishers s.269-270
7. Sirisreetreerux P, Lue KM, Ingviya T, et al. Failed Primary Bladder Exstrophy Closure with Osteotomy: Multivariable Analysis of a 25-Year Experience. J Urol. 2017 Apr;197(4):1138-1143.
8. JS Ellison, M Shnorhavorian, K Willihnganz-Lawson, et al. (2016). A critical appraisal of continence in bladder exstrophy: long-term outcomes of the complete primary repair. J Pediatr Urol12(4):205. e1-7
9. Lloyd JC, Spano SM, Ross SS, et al. How dry is dry? A review of definitions of continence in the contemporary exstrophy/epispadias literature. J Urol. 2012;188:1900–4.
10. Diamond DA, Jeffs RD. Cloacal exstrophy: a 22-year experience. J Urol. 1985;133(5):779–782.
11. Trendelenberg F (1906). The treatment of ectopia vesicae. Ann Surg 44:981–9
12. Woodhouse CR, Kellett MJ (1984). Anatomy of the penis and its deformities in exstrophy and epispadias. J Urol 1984; 132:1122–4.
13. Wild AT, Sponseller PD, Stec AA, Gearhart JP. The role of osteotomy in surgical repair of bladder exstrophy. Semin Pediatr Surg. 2011 May;20(2):71-8.
14. Mushtaq I, Gariboli M, Smeulders N, et al. (2014). Primary bladder exstrophy closure in neonates: challenging the traditions. J Urol 191:193–8.
15. Rösch WH, Promm M (2016) Blasenekstrophie: qualität der primärversorgungundlangzeitprognose. Urologe 55:53–7.
16. Husman DA, McLorie GA, Churchill BM (1989). Closure of the: an evaluation of the factors leading to its success and its importance on urinary incontinence. J Urol 142:522–4.
17. Kertai MA, Rösch WH, Brandl R, et al. (2016). Morphological and Functional Hip long-term results after exstrophy repair. Eur J Pediatr Surg 26:508–13.
18. Özcan C, Ulman I, Kara S, et al (2000). Clinical results with anterior diagonal iliac osteotomy in bladder exstrophy. J Urol 163: 1932, 2000
19. Woodhouse CR, North AC, Gearhart JP (2006). Standing the test of time: long-term outcome of reconstruction of the exstrophy bladder. World J Urol 24:244–9.
20. Silver RI, Yang A, Ben-Chaim J, et al. (1997). Penile length in adulthood after exstrophy reconstruction. J Urol 157:999–1003.
21. Gearhart JP, Leonard MP, Burgers JK, et al. (1992). The Cantwell Ransley technique for repair of epispadias. J Urol 148:851–4.
22. Benz KS, Dunn E, Solaiyappan M, et al (2018). Novel observations of female genital anatomy in classic bladder exstrophy using 3-dimensional magnetic resonance imaging reconstruction. J Urol 200:882–9.
23. Castagnetti M, Gigante C, Perrone G, et al. (2008). Comparison of musculoskeletal and urological functional outcomes in patients with bladder exstrophy undergoing repair with and without osteotomy. Pediatr SurgInt 24:689–93.
24. Haffar A, Hirsch AM, Morrill CC, Crigger CC, Sponseller PD, Gearhart JP. Classic Bladder Exstrophy Closure Without Osteotomy or Immobilization: An Exercise in Futility? Urology. 2023 Nov;181:128-132.
25. Preeya Khandge, Wayland J Wu, Saran A Hall (2021).Osteotomy in the newborn classic bladder exstrophy patient: A comparative study. J Pediatr Urol 17(4):482.e1-482.e6.
26. Hofmann A, Haider M, Promm M, et al (2021). Delayed primary closure of bladder exstrophy without osteotomy: 12 year experience in a safe and gentle alternative to neonatal surgery. J Pediatr Surg 57(10):303-308.