

Research Article

PERIOPERATIVE OUTCOMES IN PEDIATRIC BLADDER EXSTROPHY-EPISPADIAS COMPLEX: CASE SERIES OF MODERN STAGED REPAIR AT A TERTIARY CARE CENTER IN BANGALORE

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Received 20th November 2024; Accepted 27th December 2024; Published online 31st January 2025

Abstract

Introduction: Bladder Exstrophy-Epispadias Complex (BEEC) constitutes a rare congenital anomaly that profoundly affects the genitourinary system, posing unique challenges in surgical management. The Modern Staged Repair of Exstrophy (MSRE) has emerged as an attempt to achieve functional and anatomical correction. This study reviews the perioperative and long-term outcomes of patients with BEEC treated with MSRE at a tertiary care centre in Bangalore. **Methods**: his retrospective case series comprises eight patients diagnosed with BEEC and managed with MSRE. Data were extracted from electronic medical records, focusing on patient demographics, clinical characteristics, surgical complications, and continence outcomes. Patient and surgical factors were systematically categorised. Descriptive statistics were employed to summarise the data and identify trends. **Results:** The study cohort comprised six males and two females, all diagnosed postnatally. Only one patient had a favourable bladder plate, with 87.5% having poor bladder quality, complicating primary repair. Wound leakage occurred in all patients, with fistula in 25 % of cases. Infections were seen in 25% of cases, which were managed conservatively. Continence was achieved in only one patient (12.5%), while the remaining patients remained incontinent. One patient was lost to follow-up. Key challenges included anatomical variability, timing of interventions, and comprehensive follow-up. This study accentuates the necessity for early diagnosis and recommends multidisciplinary care, long-term follow-up, and tailored management plans to enhance surgical outcomes and address psychosocial factors affecting patients.

Keywords: Bladder Exstrophy, bladder plate, continence, surgical outcomes, ureteric reimplantation.

INTRODUCTION

Bladder exstrophy is a rare and complex congenital anomaly within the exstrophy-epispadias complex (EEC) spectrum. This spectrum includes various genitourinary malformations, such as epispadias and cloacal exstrophy [1]. The incidence of bladder exstrophy is estimated at approximately 3.3per 1,00,000 live births, with a notable higher prevalence in males[2]. The condition is characterised by the failure of the lower abdominal wall to close during embryogenesis, leaving the bladder exposed on the abdominal surface. This defect presents significant challenges in urinary and reproductive functions as well as social integration for affected individuals, often necessitating multiple surgical interventions throughout their lives to address both functional and aesthetic concerns [3,4].

Aetiology

The aetiology of bladder exstrophy is multifactorial, involving both genetic and environmental factors. Notable genetic mutations in the p63 genes have been associated with impaired development of the lower abdominal wall and the urogenital system[1]. Disruptions in mesodermal growth surrounding the cloacal membrane during gestation may impede the proper fusion of body wall folds, ultimately resulting in bladder exstrophy [5,6]. Additional theories propose that factors such as premature rupture of the cloacal membrane, abnormal fluid retention, and mechanical influencesmay further contribute to the condition [7,8]. While the precise cause of bladder exstrophy remains unclear, genetic predisposition and mechanical disruptions occurring during foetal development are considered significant contributing factors.

Clinical Presentation and Management of Complexity

Clinically, bladder exstrophy is characterised by a bright red bladder mucosa visible on the lower abdomen, a shortened urethra, and often a widened pelvic ring caused by pubic symphysis diastasis. Males often display a short, dorsally curved penis (chordee), while females may present with a bifid clitoris and shortened urethra [9].

The standard approach to bladder exstrophy management is multidisciplinary and staged, aimed at restoring bladder function, achieving urinary continence, protecting renal function, and reconstructing genitalia. The Modern Staged Repair of Exstrophy (MSRE) involves a series of procedures:

1. **Bladder and abdominal wall closure:** This initial surgery is performed shortly after birth to close the bladder and abdominal wall, thereby protecting the bladder and promoting its growth. Osteotomies, such as bilateral innominate or vertical iliac osteotomies, may be necessary to enhance pelvic alignment [10].

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- 2. **Epispadias repair**: Conducted typically between six months and one year of age, this procedure focuses on reconstructing the urethra and genitalia for improved voiding function and aesthetic outcomes [3,11].
- 3. Bladder neck reconstruction and antireflux surgery: Usually performed between ages four and five, these procedures aim to enhance urinary continence and safeguard renal function by preventing vesicoureteral reflux [12,13].

The success of these procedures is contingent upon various factors, including the quality of the bladder plate, intervention timing, and the surgical team's expertise. Despite advancements in techniques such as MSRE, complications including wound leakage, fistula formation, and persistent incontinence remain prevalent challenges [14]. Consequently, there is a need for further research to optimise management strategies for bladder exstrophy and enhance the quality of life for affected individuals. The present study was conducted to retrospectively review the perioperative and long-term outcomes of patients with classic bladder exstrophy-epispadias complex (BEEC) treated at a local tertiary care hospital in Bangalore, India, from 2018 to 2024. This research aimed to contribute to the existing literature on MSRE and provide valuable insights to inform future clinical practices and patient management strategies for BEEC. The specific objectives included identifying challenges, evaluating complications associated with BEEC management, andcategorising and assessing surgical outcomes, with a particular emphasis on complications such as repair breakdown, wound leaks, and incontinence.

METHODOLOGY

Study Design

This clinical study represents a retrospective case series conducted over six years, spanning from 2018 to 2024, aimed at evaluating the perioperative and long-term outcomes of patients diagnosed with classic BEEC who underwent the MSRE technique. The study comprises cases treated with a uniform surgical approach at a single centre, thereby ensuring data comparability. The retrospective design facilitates an indepth examination of treatment outcomes within a rare patient population, acknowledging the limited sample size, where each case contributes valuable insights into the patient and surgical factors relevant to this uncommon condition. The patient selection was based on the following criteria:

- *Inclusion Criteria:* Patients were considered eligible if they possessed a confirmed diagnosis of classic BEEC and received treatment utilising the MSRE technique. Only those patients with comprehensive perioperative and follow-up records were included in the study to ensure a thorough analysis of all relevant data concerning surgical and postoperative outcomes.
- *Exclusion Criteria:* Patients were excluded from the study if they had incomplete medical records. Moreover, cases involving complex or atypical forms of exstrophy, such as cloacal exstrophy, were excluded to maintain a focused analysis of classic BEEC cases.

Ethical approval was obtained from the institutional review board (IRB) of the study hospital before the commencement of the study (Ref No: SIMS & RC/EC-19/SS- 12/2024-25).

Informed consent was not applicable for this retrospective analysis as the data were collected from electronic medical records while maintaining confidentiality and anonymity of patient information by deidentifying patient details during data processing.

Data Collection and Categorisation

Data were obtained from the hospital's electronic medical record (EMR) system, which provided access to detailed perioperative and follow-up information for each patient. The data collection focused on extracting information regarding preoperative assessments, the MSRE surgical procedures performed, immediate and long-term postoperative complications, and follow-up outcomes. All patients were advised to return for regular follow-up visits to monitor urinary function, wound healing, and any potential complications requiring intervention. The follow-up period ranged from 6 months to 5 years postoperatively, depending on the timing of the procedure. Continence and quality of life were assessed in ongoing follow-ups, with social and psychological support provided as part of the standard protocol.

To enhance clarity and facilitate analysis, the data were categorised into two major groups:

- **Patient factors:** This category included demographic information, clinical presentation, and specific characteristics that may influence outcomesto ascertain whether patient-related characteristics could impact the likelihood of complications or successful outcomes.
- *Surgical factors*: This category emphasised outcomes explicitly associated with the MSRE procedure, including intraoperative and postoperative complications.

The detailed categorisation of patient and surgical factors is presented in **Table 1.**Psychological evaluation and mental health support to patients and families support were incorporated as part of the standard treatment protocol to address the social and emotional challenges associated with BEEC.

 Table 1. Factors affecting surgical outcomes in the management of patients with bladder exstrophy

Factors	Variable	Categorisation
	Timing of	Prenatal or postnatal, with postnatal diagnosis
Patient factors	diagnosis	being the case for all patients in this series.
	Age at presentation	Grouped as <1 year, 1–5 years, 6–10 years, 11–15 years, >15 years, as later presentation can impact repair complexity.
	Bladder plate quality	Assessed intraoperatively as favourable* or unfavourable.
Surgical	Wound leaks and infection	Recorded incidence of wound leakage, which can increase infection risk and affect healing outcomes.
factors	Incontinence following repair	Recorded to assess urinary control post-repair, with some patients requiring additional management.

*A favourable bladder plate was typically a healthy, well-vascularised tissue of adequate size, allowing for effective surgical closure and reconstruction and minimal scarring or fibrosis to enhance surgical outcomes.

Statistical Analysis

Descriptive statistics were used to summarise the data, using frequencies and proportions to illustrate the distribution of key variables and identify trends in surgical outcomes and complications. Tables and charts were generated to compare patient and surgical factors across cases.

RESULTS

This case series analysed eight patients diagnosed with BEEC and treated using the MSRE. A summary of the patients, the complications and the procedures followed are captured in Table 2 and Figure 1.Demographic trends and clinical characteristics are delineated in Table 3, detailing the distribution of bladder exstrophy by sex, age, timing of diagnosis, and bladder plate quality within the current cohort (patient factors). The cohort comprised six males (75%) and two females (25%). Moreover, a larger proportion of patients (75%) presented after the age of two years, and no cases were identified prenatally. The timing of diagnosis indicated that none of the patients in this study received a prenatal diagnosis. When assessed for the bladder plate quality, only one patient (12.5%) had a favourable bladder plate during intraoperative assessments, while the remaining seven patients (87.5%) had unfavourable bladder plates.

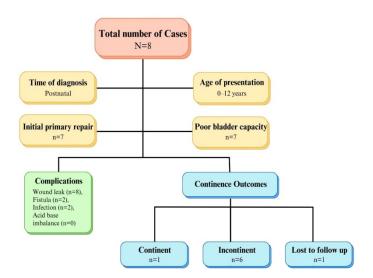


Figure 1. Flow chart summarising the demographic trends, clinical characteristics, and surgical outcomes of patients diagnosed with bladder exstrophy-epispadias complex (BEEC) within the current cohort

Table 2. Diagnosis and surgical management in patients with bladder exstrophy and exstrophy-as	associated com	artions

Case	Date of surgery	Age/Sex	Diagnosis	Procedure
1	26/10/2018	5 years/F	BEEC s/p primary repair	Exstrophy bladder stage 2 surgery (ileocystoplasty with bladder neck repair) under GA+CA
2	14/4/2020	2 days/M	BEEC	Primary closure of bladder exstrophy under GA+CA
3	12/2/2020	12 years/M	BEEC s/p primary repair	Laparotomy and augmentation ileocystoplasty with bilateral ureteric reimplantation bladder neck closure with appendicular Mitrofanoff under GA+CA
4	4/5/2019	1 year/F	BEEC	Primary closure of bladder exstrophy/exstrophy repair with bilateral ureteric reimplantation under GA+TAP block
5	2/10/2021	2 years/M	BEEC	Primary closure of bladder exstrophy repair under GA+CA block
6	9/3/2022	2 years/M	BEEC	Bladder exstrophy closure with augmentation ileocystoplasty done under GA+CA
7	15/03/2024	7 months/M	BEEC	Primary closure of bladder exstrophy/exstrophy repair with bilateral ureteric reimplantation under GA
8	7/6/2023	12 years/M	BEEC s/p primary repair	Stage 2 repair with ileocystoplasty bilateral ureteric reimplantation

BEEC = Bladder exstrophy and epispadias complex; CA = combined general plus neuraxial anaesthesia; GA = general anaesthesia; s/p = status post; TAP = transabdominal plane

Table 3. Distribution of bladder	• exstrophy in t	the patient cohort
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Patient Demographics and Clinical Characteristics	No. of cases, n (%)
Sex	
Male	6 (75%)
Female	2 (25%)
Age of Presentation	
<1 year	2 (25%)
1–5 years	3 (37.5%)
6–10 years	0 (0%)
11–15 years	1 (12.5%)
>15 years	0 (0%)
Time of Diagnosis	
Prenatal	0 (0%)
Postnatal	8 (100%)
Quality of Bladder Plate	
Favourable	1 (12.5%)
Unfavourable	7 (87.5%)

Surgical factors

The MSRE technique was applied to all patients within this series, with primary repair focused on closing the bladder and abdominal wall to restore structural integrity. Based on the complexity of the surgical procedures performed, the cases were divided into two groups:

- **Primary closure of bladder exstrophy:** Cases 2, 4, 5, 6, and 7 represent instances of primary closure in an age group ranging from newborns to toddlers. These procedures typically involve circumferential incisions around the bladder plate and aim to achieve bladder closure and reimplantation of ureters, which is pivotal in establishing urinary continuity.
- Augmentation procedures: Cases 1, 3, and 8 included more complex interventions characterised by bladder augmentation and reimplantation. Case 3 demonstrated a laparotomy approach combined with ileocystoplasty, highlighting the necessity of advanced surgical techniques in older children with previously repaired exstrophy experiencing complications such as small bladder capacity and penopubic fistula. The re-operation in Case 8 further emphasises the importance of managing long-term complications from earlier surgeries.

The complications observed are captured in **Table 4**, and the surgical factors and outcomes are described below.

- *Wound leakage and infection:* All patients (100%) encountered wound leakage, which was further complicated by fistula formation (25 %) in some of them. However, postoperative infections were observed in only two patients (25%), which were managed conservatively with antibiotics and wound care.
- *Continence outcomes:* Of all the patients, only one female patient (12.5%) attained continence, and one (12.5%) lost to follow-up. In contrast, the remaining six patients (75%) continued to experience incontinence despite surgical intervention (Figure 2). For these patients, additional strategies, including clean intermittent catheterisation or the consideration of future bladder augmentation, were being contemplated to improve continence outcomes. Representative figures from these cases are presented in Figures3,4 and 5, respectively.



	Primary Complications	No. of cases, n (%)
	Wound Leakage	8 (100%)
	Fistula Formation	2 (75%)
	Presence of Infection	2 (25%)
	Incontinence	8 (100%)
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Figure 2. Continence outcomes in patients with bladder exstrophy-epispadias complex (BEEC) post-surgical intervention

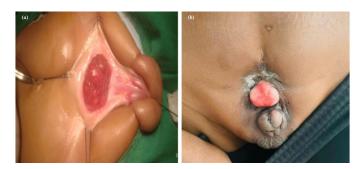


Figure 3. Preoperative image showing the exposed bladder and associated anatomical features characteristic of bladder exstrophy-epispadias complex (BEEC) before surgical intervention (a) in a 5-year-old female; (b) in a 12-year-old male



Figure 4. Intraoperative image depicting the surgical steps involved in the modern staged repair of exstrophy (MSRE), demonstrating tissue mobilisation and alignment techniques used to achieve closure in a 2-day-old male



Figure 5. Post-repair image illustrating the completed repair, showing the closed bladder and abdominal wall, along with the reconstructed genitourinary structures in a 12-year-old male

DISCUSSION

This series examines eight cases of bladder exstrophy managed using diverse surgical techniques, including primary closures, ileocystoplasty, and bladder augmentation. Our cohort comprised a higher proportion of male patients, which aligns with existing literature indicating a greater incidence of BEEC among males[15,16]. The ages of patients varied significantly, ranging from 2 days to 12 years, which underscores the diverse presentation and severity of this condition. It is noteworthy that a substantial number of patients received their diagnosis after the age of two years. Such delayed presentation is a significant factor that results in a delayed intervention that potentially exacerbates the complexity of management, as older patients generally possess more developed musculoskeletal structures, potentially affecting the outcomes of primary closure and necessitating additional procedures such as osteotomies [6,17]. Additionally, no cases were detected through prenatal screening, which indicates that early assessments during pregnancy may be inadequate for identifying this condition, possibly due to limited screening options or challenges in recognising BEEC through standard prenatal imaging. This shortfall in early detection raises concerns regarding the effectiveness of current prenatal screening methods and emphasises the need for enhanced diagnostic approaches.

The analysis of surgical factors demonstrated a significant prevalence of wound leakage, which is often attributable to high tissue tension and insufficient vascularity in the reconstructed bladder plate, necessitating additional management and potentially prolonging recovery time. A smaller subset of patients exhibited signs of infection. This association can be attributed to the inherent risks associated with exposing the urinary tract and the complexities of surgical manipulation involved in these procedures. Understanding the microbiological profiles of postoperative infections in this context may enhance the development of targeted prophylactic strategies to reduce infection rates. The tailored surgical approaches employed for each patient underscored the importance of personalized treatment in such intricate procedures. Variability in anatomical features and the presence of potential complications necessitated a flexible surgical strategy, which included bladder neck repair, ureteric reimplantation, and augmentation techniques. Managing the penopubic fistula case required precise dissection techniques, emphasising the necessity for surgical dexterity and experience in achieving favourable outcomes in complex repair situations. multiple instances involving bilateral In ureteric reimplantation, the associated risks of urinary tract infections, reflux, and obstruction highlighted the need for diligent postoperative monitoring. Augmentation procedures were crucial for patients with small-capacity bladders, though they carry risks such as bowel complications. Consequently, an indepth understanding of these complications is crucial for successful surgical outcomes.

Achieving urinary continence remains critical to managing BEEC, yet it presents considerable challenges. Our findings revealed that within this cohort, only one patient achieved satisfactory continence, while another was lost to follow-up. For those patients grappling with incontinence, alternative strategies as management such clean intermittent catheterisation or consideration of future bladder augmentation were being deliberated. Future studies should aim to refine these strategies and investigate their efficacy in enhancing continence outcomes, thereby contributing to knowledge surrounding BEEC surgeries. Furthermore, it is worth noting that managing BEEC is challenging, as achieving optimal patient outcomes relies profoundly on surgical expertise. Advanced surgical approaches, such as MSRE, require high surgical proficiency and are frequently limited in low- and middle-income countries (LMICs). To mitigate this disparity, surgical training programs must incorporate extensive training focused on the complexities associated with BEEC. Moreover, older children with BEEC frequently encounter social and psychological challenges related to their condition, underscoring the necessity for a comprehensive approach that integrates psychological support alongside surgical intervention. This underscores the need for a holistic care approach that integrates psychological support alongside surgical management. By addressing both the surgical and psychosocial aspects of BEEC, we can foster better overall outcomes for these patients.

Clinical Recommendations

Considering the challenges identified and the surgical outcomes observed in this case series, the following clinical recommendations are proposed to facilitate better patient care and effectively address the complexities of treatment.

- 1. *Comprehensive preoperative assessment*: A thorough preoperative assessment should encompass a detailed physical examination, appropriate imaging studies, a psychological evaluation, and open discussions regarding the family dynamics influencing patient care.
- 2. *Multidisciplinary approach*: A collaborative engagement of a multidisciplinary team, including surgeons, urologists, psychologists, and paediatricians, is essential for the holistic management of patients to address both the physical and emotional challenges.
- **3.** *Tailored management plans:* Individualised management strategies should be developed to account for patient-specific anatomical and psychosocial factors. Tailoring these plans should consider age, prior surgical history, and overall health status to optimise care and outcomes.
- 4. *Long-term follow-up:* Consistent postoperative surveillance should be instituted for the early identification of complications. This should include periodic imaging studies and functional assessments. Establishing standardised follow-up protocols may enhance long-term bladder function and continence outcomes.
- 5. *Specialised Surgical Training Programs:* To improve outcomes in managing BEEC, surgical training programs should integrate extensive training and practice focused on managing the complexities of BEEC, including advanced surgical techniques like MSRE.

Strengths and Limitations of the study

Strength: The study provides valuable case-based insights into BEEC management in a specific clinical setting, contributing to the limited literature on this rare condition.

Limitations: The small sample size of eight cases limits the generalisability of the findings. Broader research with larger cohorts is needed to validate these observations. Additionally, the focus on a single institution may reduce the applicability of the results in diverse settings with varying resources and surgical expertise.

Conclusion

This case series elucidates the considerable challenges ofmanaging BEEC, particularly in instances characterised by delayed presentation and suboptimal bladder plate morphology. The findings emphasise the critical necessity for early diagnosis, given that timely intervention can significantly enhance surgical outcomes and overall patient prognoses. The ongoing complications of wound leakage, fistula formation, and limited success in achieving continence highlight the

technical intricacies inherent in the MSRE technique, reinforcing the need for ongoing advancements in surgical methodologies. While this investigation provides essential insights into the management of BEEC, the necessity for larger, multicentre studies and continuous innovation in this field remains paramount to enhance the quality of life and long-term outcomes for affected individuals.

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