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Widened Pubic Rami in Bladder Exstrophy-Epispadias Complex (BEEC): A Rare Case Report and Surgical Considerations

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ABSTRACT

Bladder exstrophy is a rare congenital anomaly characterized by extrusion of the urinary bladder through a lower abdominal wall defect and is frequently associated with pelvic musculoskeletal deformities. One important abnormality is widening and malalignment of the pubic rami, which complicates reconstruction and requires multidisciplinary management. We report a 6-month-old male infant with two months of hematuria and a congenital suprapubic bulge with a reddish mass. Clinical and radiological evaluation confirmed bladder exstrophy. Pelvic radiographs showed widened, malaligned pubic rami with separation at the symphysis pubis. The patient underwent multidisciplinary surgery involving urologists, orthopedic surgeons, and plastic surgeons. The orthopedic team performed bilateral Salter innominate osteotomies to mobilize the iliac wings, followed by pubic symphysis reduction using the Müller technique and fixation with Vicryl 1 sutures. Bilateral Schanz screws and an external pelvic fixator with backslab were applied to maintain stability. Postoperatively, vascular and neurological status remained normal, with good hip, knee, and ankle range of motion. This case illustrates the importance of early orthopedic intervention in correcting pelvic deformity in bladder exstrophy. Bilateral Salter osteotomies enable medial rotation of the iliac bones, facilitating pubic approximation and contributing to pelvic ring stability when combined with soft tissue and urological reconstruction. Bladder exstrophy with pubic diastasis requires coordinated multidisciplinary care, and timely orthopedic procedures are crucial to support structural reconstruction and optimize functional outcomes in these rare, complex congenital cases.

Keywords: *Widened Pubic Rami, Bladder Exstrophy-Epispadias Complex, bilateral Salter osteotomy*

INTRODUCTION

Bladder exstrophy-epispadias complex (BEEC) is a rare congenital malformation characterized by midline defects involving the lower urinary tract, genitalia, abdominal wall, and pelvic bones.[1] It represents a spectrum of anomalies that range from isolated epispadias to classic bladder exstrophy and the most severe form, cloacal exstrophy. Among these, classic bladder exstrophy occurs in approximately 1 in 30,000 to 50,000 live births, with a male-to-

female ratio of approximately 2.3:1.[2] Epispadias alone is even rarer, occurring in approximately 1 in 117,000 males and 1 in 484,000 females, while cloacal exstrophy occurs in 1 in 200,000 to 400,000 live births.[3]

The exact etiology of BEEC remains unclear, although it is believed to involve a combination of genetic, environmental, and mechanical factors affecting the development of the cloacal membrane during early embryogenesis.[4] Recent studies suggest a multifactorial inheritance pattern, with a recurrence risk of approximately 1 in 100 for first-degree relatives.[5] Some studies have also identified associations with chromosomal abnormalities and candidate genes involved in mesenchymal tissue development and midline closure.[6,7]

A consistent and defining feature of bladder exstrophy is the presence of widened pubic diastasis, resulting from deficient midline fusion of the pelvic bones during fetal development.[8] This skeletal deformity not only contributes to pelvic instability but also poses significant challenges in achieving effective bladder closure and continence. Without orthopedic intervention, the pubic rami remain laterally displaced, impeding soft tissue approximation and increasing the risk of postoperative complications such as wound dehiscence and bladder prolapse.[9]

Management of BEEC is highly complex and requires a multidisciplinary approach, involving pediatric urologists, orthopedic surgeons, and plastic surgeons. Early orthopedic procedures, such as bilateral Salter innominate osteotomy, have been shown to facilitate medial rotation of the iliac bones, enabling tension-free bladder closure and improved pelvic stability.[9] Nevertheless, detailed case reports on the integrated surgical approach to bladder exstrophy with severe pubic diastasis remain limited.

In this report, we present a rare case of a 6-month-old male infant with bladder exstrophy and significant widening of the pubic rami, who underwent a staged, multidisciplinary reconstruction. This case highlights the critical role of orthopedic intervention in the comprehensive management of BEEC and discusses key surgical considerations in optimizing long-term outcome.

CASE PRESENTATION

A 6-month-old male infant was brought to the outpatient clinic with a primary complaint of persistent hematuria over the preceding two months. The hematuria was described as varying in intensity depending on the volume of urine passed and was often accompanied by yellowish mucous discharge. There was no history of trauma, regular medication use, known medical illnesses, or previous surgical procedures. Screening for associated congenital anomalies revealed no evidence of vertebral defects, anal atresia, cardiac abnormalities, tracheoesophageal fistula, renal anomalies, esophageal defects, or limb deformities.

Physical examination revealed a 2 × 2 cm midline defect in the suprapubic region at the level of the pubic symphysis. The base of the defect was consistent with exposed bladder tissue, with urine visibly leaking through the opening. There was associated serous exudate without slough or purulent discharge. The external genitalia showed a buried and bifid penis, with the urethral opening located dorsally on the proximal shaft. Both hemiscrotums were present and palpable. On pelvic examination, the exposed bladder was noted without swelling, bruising, or visible deformity. Palpation elicited tenderness in the bladder area, while vascular examination showed palpable dorsalis pedis arteries and a capillary refill time of less than two seconds. The infant exhibited normal active and passive range of motion in both lower limbs.

The clinical and radiological evaluation confirmed the diagnosis of Bladder Exstrophy-Epispadias Complex (BEEC) with associated pubic diastasis. A pelvic X-ray (anteroposterior view) revealed widening of the pubic rami and midline separation at the symphysis pubis.

Abdominal ultrasound was also performed to assess for visceral and urinary tract anomalies. The case was referred for multidisciplinary management involving pediatric urology, orthopedic surgery, and plastic surgery.

Surgical intervention was performed in a staged, multidisciplinary fashion. The first procedure was conducted by the urology team, who undertook bladder reconstruction under general anesthesia with the patient in the supine position. After sterile preparation and draping, the urethral orifice was identified, and 3.5 Fr catheters were inserted into both ureters and secured. An incision was made circumferentially around the bladder defect and deepened to the fascial layer. The bladder was then mobilized and closed in layers using absorbable sutures, and the surgical site was closed layer by layer.

Following bladder closure, the orthopedic team proceeded with pelvic osteotomy and fixation. Using a lateral pelvic approach, the iliac wings were exposed bilaterally. Bilateral Salter innominate osteotomies were performed on the right and left ilium to allow medial rotation of the iliac bones. The pubic symphysis was approximated using the Müller technique and stabilized with Vicryl 1 absorbable sutures. Schanz screws were inserted into both iliac bones, and external pelvic fixation was applied to maintain pelvic alignment and stability.

Lastly, the plastic surgery team addressed the soft tissue defect in the suprapubic region, which measured approximately 6 × 6 cm and revealed the underlying bladder. The defect had irregular edges and minimal active bleeding. Previous incisions from the pelvic osteotomy in both inguinal regions were well approximated, with good wound healing. A local advancement flap was designed to cover the suprapubic defect. Following infiltration with pehacaine, an incision was made along the marked flap, which was dissected in layers down to the parietal peritoneum. The wound was then closed in layers using absorbable 4-0 multifilament sutures, and the skin was sutured with non-absorbable 5-0 monofilament sutures. The surgery concluded without complications. Postoperatively, the vascular and neurological status of the lower limbs remained intact. The infant exhibited active hip motion from 40° to 150°, knee range of 0° to 135°, and ankle motion from 50° to 70°. This case highlights the importance of a collaborative, multidisciplinary approach in the successful management of BEEC with widened pubic rami.

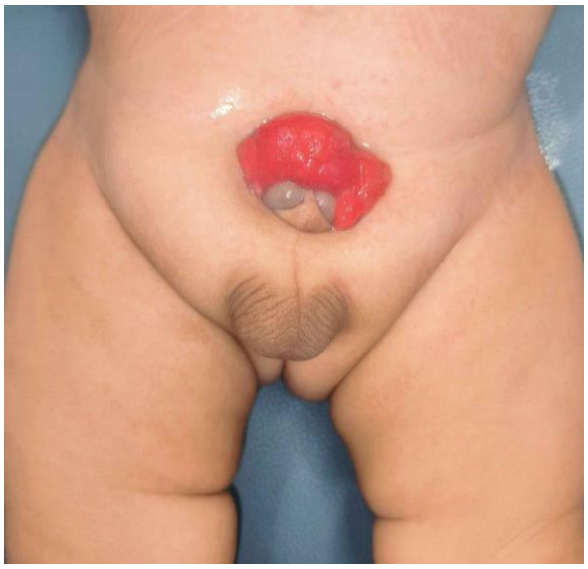

Figure 1. Clinical Presentation

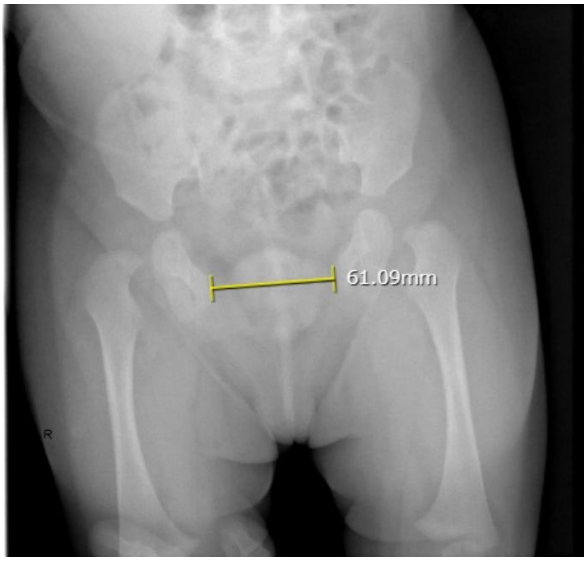

Figure 2. Pelvic X-ray (anteroposterior view)

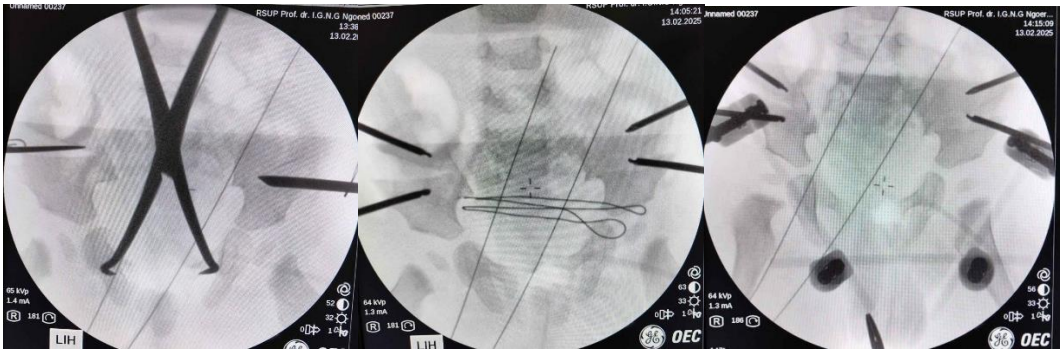

Figure 3. C-arm view inrea-opeation

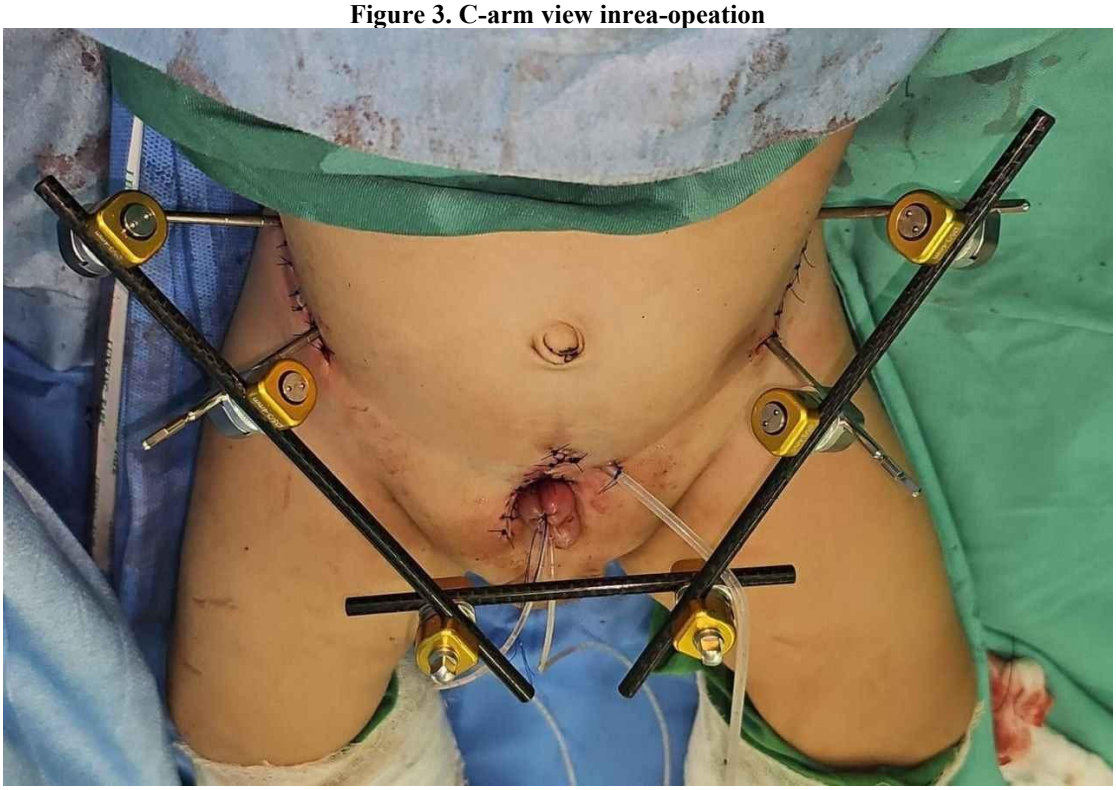

Figure 4. Durante operation

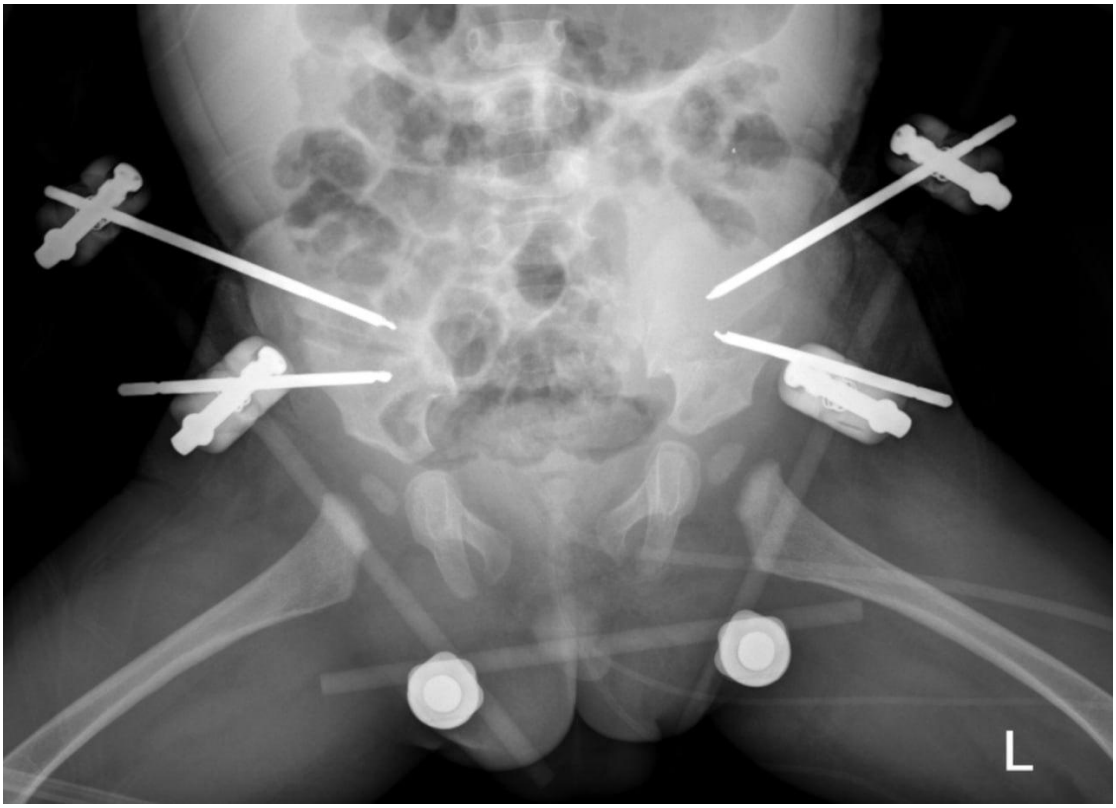

Figure 5. Post-op X-Ray AP Lateral View

DISCUSSION

Bladder exstrophy-epispadias complex (BEEC) is a rare and surgically challenging congenital anomaly involving the urogenital and musculoskeletal systems.[10] The condition presents with a wide range of anatomical deformities that require a carefully orchestrated, multidisciplinary surgical approach.[11] In this case, the patient a 6-month-old male infant presented with classical signs of bladder exstrophy, including exposure of the bladder mucosa through a lower abdominal wall defect and significant pubic diastasis.[12,13] The prolonged presentation of hematuria, alongside mucous discharge and a visible suprapubic bulge, further supported the diagnosis, which was subsequently confirmed through clinical and radiological evaluation.[14]

A defining and frequently overlooked feature of BEEC is the abnormality in pelvic anatomy, particularly the widening of the pubic symphysis.[15] In normal embryogenesis, the pelvic ring undergoes midline fusion to provide structural integrity and support for the genitourinary organs.[16] In bladder exstrophy, failure of this fusion results in lateral displacement of the pubic bones, leading to a widened pubic diastasis that impedes abdominal wall closure, affects urological function, and complicates reconstructive efforts.[17] The degree of pubic separation can be variable, but when significant as in our case it requires orthopedic intervention to allow for successful approximation of the abdominal and bladder components.[18]

Orthopedic management in BEEC typically involves pelvic osteotomy procedures aimed at mobilizing the iliac bones and facilitating medial rotation.[8,19] Bilateral Salter innominate osteotomy, first described by Salter for the treatment of developmental dysplasia of the hip, has been adapted for use in bladder exstrophy repair due to its ability to reorient the pelvic ring.[18,20] In our case, this technique allowed for effective reduction of the pubic symphysis, which was then stabilized using the Müller technique and Vicryl sutures. Additionally, Schanz screws were inserted into both iliac bones, and an external pelvic fixator was applied to maintain post-operative

alignment and stability.[21] This approach not only improved the anatomical configuration of the pelvis but also provided a solid foundation for soft tissue closure and urological reconstruction.[22]

From a urological standpoint, early closure of the bladder is crucial for preserving bladder capacity, protecting the upper urinary tract, and eventually achieving urinary continence.[23] In this case, catheterization of the ureters was performed to identify and protect the ureteral orifices during bladder closure. The bladder wall was then approximated using absorbable sutures in a layer-by-layer fashion, ensuring watertight closure. The presence of buried and bifid penis with dorsal urethral opening (epispadias) is a typical component of the exstrophy-epispadias spectrum and represents an additional challenge that may necessitate future staged surgical correction.[24,25]

Plastic surgery played a vital role in ensuring tension-free closure of the soft tissue defect in the suprapubic region. The use of a local advancement flap allowed for effective coverage of the exposed bladder and surgical site, reducing the risk of wound breakdown and infection.[26] Careful planning of the incision design, followed by multi-layered closure using absorbable and non-absorbable sutures, helped achieve satisfactory aesthetic and functional outcomes.[27]

Postoperatively, the patient demonstrated stable hemodynamic parameters, intact vascular and neurological status, and preserved joint range of motion, indicating a successful multidisciplinary intervention.[28] The use of external fixation proved essential not only for maintaining pelvic alignment but also for minimizing movement at the surgical site, which is particularly important in the healing phase.[29] Importantly, the early involvement of orthopedic surgery ensured that the musculoskeletal deformity was addressed concurrently with the bladder closure, highlighting the significance of a well-coordinated surgical strategy.[30]

This case underscores several key principles in the management of BEEC. First, timely diagnosis and surgical intervention during infancy are crucial to optimizing anatomical and functional outcomes[11]. Second, pubic diastasis should be recognized as a central component of the pathology rather than a secondary deformity, warranting orthopedic correction as part of the initial surgical plan.[31] Third, collaboration among pediatric urology, orthopedic surgery, and plastic surgery is essential in achieving comprehensive and durable reconstruction. Failure to address any one aspect be it bony alignment, bladder integrity, or soft tissue coveragecan compromise the entire surgical effort.[32]

While the literature on BEEC provides a general framework for management, there remains variability in surgical techniques and timing of interventions. Our case contributes to the existing body of knowledge by demonstrating the feasibility and efficacy of integrating bilateral Salter osteotomy, external fixation, bladder reconstruction, and soft tissue repair in a single operative session. Further studies with long-term follow-up are needed to assess continence outcomes, pelvic growth, and psychosocial impact in such patients. Nonetheless, this report adds to the growing recognition of the critical role of orthopedic intervention in the holistic treatment of bladder exstrophy.

CONCLUSION

This case illustrates the critical importance of a multidisciplinary approach in the management of bladder exstrophy-epispadias complex (BEEC), particularly when complicated by significant pubic diastasis. The combination of urological bladder reconstruction, orthopedic realignment through bilateral Salter innominate osteotomy and external fixation, and soft tissue coverage by plastic surgery provides a comprehensive strategy for anatomical restoration and functional rehabilitation. Early orthopedic intervention is paramount in correcting the pelvic

deformity, facilitating tension-free bladder and abdominal wall closure, and promoting long-term pelvic stability.

The successful outcome in this patient underscores the value of collaborative surgical planning and timely intervention. Integration of orthopedic procedures at the initial stage of reconstruction significantly enhances the likelihood of favorable outcomes, both functionally and structurally. This case contributes to the growing body of evidence advocating for the inclusion of orthopedic management in the standard treatment paradigm of BEEC and highlights the need for continued research to optimize surgical timing, techniques, and long-term follow-up.

Clinical Significant

This case report highlights the pivotal role of early orthopedic assessment and surgical intervention in the management of bladder exstrophy-epispadias complex (BEEC). Addressing pubic diastasis through bilateral Salter innominate osteotomy and external fixation enhances the success of bladder and abdominal wall reconstruction. It reinforces the need for multidisciplinary collaboration to achieve optimal outcomes in these rare and complex congenital anomalies

Declaration by Authors

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