

## Bilateral Anterior Osteotomy for Bladder Exstrophy at Older Age: A Case Report Respati Suryanto Dradjat<sup>1</sup>, Panji Sananta<sup>2\*</sup>, Andryan Hanafi Bakri<sup>3</sup>

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### ABSTRACT

**Introduction:** Pubic diastasis in bladder exstrophy is a complex condition requiring intervention as soon as possible to achieve a tension-free abdominal wall. While most literature highlights the successful procedures conducted early, more studies need to report the management in older age. We herein report the management of pubic diastasis for bladder exstrophy at older age.

**Case Presentation:** A 12-year-old girl presented with bladder exstrophy requiring surgical reconstructive surgery with a team comprising of Orthopaedic, Urology and Plastic Surgery Departement. Intraoperatively, bilateral anterior osteotomy was performed to correct the 10 cm pubic diastasis. The correction then facilitated bladder reconstruction and subsequent skin closure overlying it. A modular external fixator was installed to maintain the correction. Then, a post-operative radiograph was obtained, and 1.5 cm of pubic diastasis was revealed.

**Clinical Discussion:** Bladder exstrophy is a congenital anomaly where the bladder is everted outside the body, affecting 2.07 of 100,000 births, with male predominance. Risk factors include alcohol, misoprostol, heparin, valproic acid, and diazepam exposure during pregnancy. The condition has a high potential for malignancy, with squamous cell carcinoma (SCC) as high as 3-40% due to intestinal epithelial metaplasia. Surgical procedures like pelvic osteotomy are advised due to advances in prenatal diagnosis.

**Conclusions:** Bilateral anterior osteotomy combined with an external fixation device could be a solution to correct pubic diastasis in bladder exstrophy among older age patients.

**Keywords:** bladder exstrophy, osteotomy, pediatric, pubic diastasis, reconstruction

### INTRODUCTION

Bladder exstrophy is a congenital abnormality characterized by a completely opened bladder at the lower part of the abdomen, and the inner surface of the bladder is fused to the abdominal skin.<sup>1</sup> This troublesome malformation may involve the genitourinary tract, musculoskeletal elements and, occasionally, the gastrointestinal tract. Considering the complexity of the anatomic structure, special consideration must be given by the treating physicians, who may involve pediatric urologists, orthopaedists, or plastic surgeons.<sup>2,3</sup> The goal of the treatment is to achieve a closed bladder and the anterior abdominal wall, as well as urinary continence.<sup>3,4</sup> The role of orthopaedic intervention is to reduce pelvic diastasis to create a tension-free abdominal wall so that it is possible to close the bladder and abdominal wall.<sup>2,4</sup> Regardless of the method used, in most reports, it is usually performed at early ages, with a few days following birth as the most recommended period.<sup>4</sup> Of note, a lack of literature presented the management of this potentially malignant lesion in older ages.<sup>5</sup> Thus, in this report, we present the management of bladder exstrophy in teenage patients.

### CASE PRESENTATION

A 12-year-old girl presented with a complaint of a reddish mass in her lower abdomen since birth (Figure 1). She hadn't had her complaint consulted with any healthcare professional. The patient underwent an examination by the Urology Department and was diagnosed with bladder exstrophy. Then, a bladder biopsy from the dome, right and left lateral wall, posterior wall, around the urethral orifice, trigonum, and the bladder neck was performed with the result of metaplasia. A multi-disciplinary approach, consisting of the Orthopaedic and Plastic Surgery Department, was conducted to close the defect. Our department consulted the patient to close the diastasis, which was 10 cm, as evaluated radiographically (Figure 2). Thus, bilateral anterior osteotomy was planned to achieve this.



**Figure 1.** Clinical picture of bladder exstrophy



**Figure 2.** Radiograph workups of (A) pelvic x-ray and (B) pelvic CT scan

During the surgery, a 5 cm incision was made bilaterally at the anterosuperior iliac spine (ASIS). Then, the incision was deepened to expose the iliac wing and secure the lateral cutaneous nerve. Afterwards, a window is created at both iliac using an osteotome under an image intensifier (Figure 3). The pins (Schanz screws of 3.5 x 180 mm) were inserted two superiorly and two inferiorly at both sides. Following the screw insertion, the surgery was proceeded by bladder reconstruction by the Urology Department



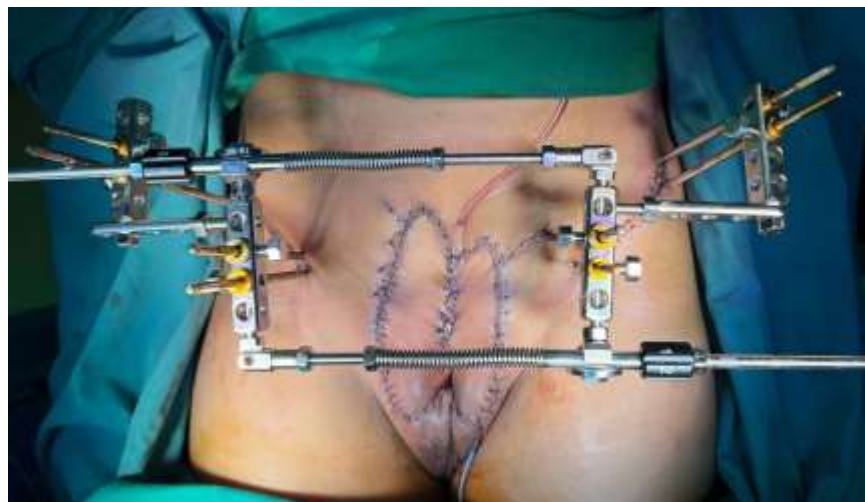
**Figure 3.** Pelvic osteotomy under image intensifier

The bladder was released from the rectus abdominis muscle during the reconstruction procedure. The pediatric urologist could refresh and reconstruct the bladder neck, followed by cystostomy and catheter insertion. Then, the plastic surgeon resumed the surgery by defect closure using mesh and a double keystone flap (Figure 4).



**Figure 4.** Bladder reconstruction and closure procedure

After the reconstruction and defect closure, the pelvis was brought together and held using the modular external fixation device (Figure 5). Afterwards, the post-operative pelvic radiograph was ordered, and the diastatic measured, which became 1.5 cm (Figure 6). The surgical wound care was performed periodically, and the patient was discharged in stable condition 7 days following the surgery.



**Figure 5.** External fixation device holding the pelvis



**Figure 6.** Post-operative radiograph of the pelvis

## DISCUSSION

Bladder exstrophy is a congenital anomaly in which the bladder is everted outside the body at the lower abdominal part, the area between the umbilicus and symphysis pubis. This colloquial terminology comes from the Greek word “exstrophy”, which means turned inside out. The prevalence of this anomaly, first described in 1595, is estimated to be 2.07 of 100,000 births, with male preponderance. Despite the extensive efforts to describe it, scientists still fail to prove the exact cause, albeit alcohol, misoprostol, heparin, valproic acid and diazepam exposure during pregnancy are considered risk factors for developing this potentially malignant lesion.<sup>1</sup> The importance of this troublesome malformation is its malignant potential, with squamous cell carcinoma (SCC) as high as 3-40% because of the metaplasia of intestinal epithelial elements and often has a poor prognosis. Therefore, we performed a bladder biopsy, resulting in a metaplasia of the lesion. This result is relevant in most cases where the biopsies show well-differentiated lesions.<sup>6</sup>



As a consequence, surgical procedure consisting of closure of the urological defect in conjunction with pelvic osteotomy is strongly advised. Due to the advance in prenatal diagnosis, this surgical procedure, which involves a multidisciplinary team, is usually done within the first days to weeks of life.<sup>1,2,4</sup> This is to avoid the environmental injury of the bladder mucosa and call for a collaboration of pediatric urologist, orthopaedist and plastic surgeon.<sup>1</sup>

Nevertheless, the condition is only sometimes ideal, particularly in developing countries, as shown in our case. Even though no current literature describes the actual epidemiology of adulthood with bladder exstrophy, Nerli et al. have shown their experience in managing this congenital disorder in adults ages, with an average age of 19 years. During their 15-year retrospective study, they found that the mean age was 19 years old. Researchers also noted that most of their patients had a hindrance to reaching the healthcare system, either a lack of knowledge or access to it. In addition, they also mentioned that the majority of the patients came from rural areas. This fact is somewhat similar to our patient, who lives in a remote area of East Java and doesn't have much understanding of her condition, so she presented to us at a late age. Fortunately, because of the nation's healthcare insurance system, she managed to get her ailment treated.

However, delivering surgery in this age group has no obstacles, especially for orthopaedic surgeons. Bladder exstrophy presenting at older ages is associated with more difficulty in pubic diastasis correction because there is an increase in pubic diastasis distance, from approximately 4 cm at birth to just about 8 cm by age 10. Symphyseal ligament disruption might be responsible for this burdensome phenomenon. In our case, the symphyseal diastasis is 15 cm, which calls for pelvic osteotomy to decrease the pelvic distance, which is reduced by one-tenth postoperatively. Our consideration in performing this manoeuvre is to minimise the tension of the overlying abdominal skin so that the risk of wound breakdown can be lessened, considering the wide pubic diastasis in our patient.

Whilst bladder reconstruction is the core of the correction surgery, pelvic osteotomy is still a topic of debate in this complicated surgery. The proponents advocate the need for osteotomy since it is associated with reduced pelvic organ prolapse, lessened wound dehiscence rate and improved continence.<sup>4,7,8</sup> On the other hand, some opponents didn't recommend its role because of cosmetic issues and increased morbidity as a consequence of increased surgical time, which is linked to a longer anaesthesia period and more blood loss.<sup>1,3,9</sup> They avoid pelvic osteotomy because their patients are young, so pelvic approximation can be achieved without osteotomy by utilizing the ligamentous laxity.<sup>1,10</sup> This method isn't feasible for our case because the diastasis distance is too wide to be approximated directly.

Pelvic anomaly in bladder exstrophy comprises the undergrowth of the pubic rami, resulting in ischiopubic segment shortening along with the outward orientation of the iliac, with the result being the pelvic diastasis, external rotation of the posterior part of the pelvis of 12° as well as retroversion of acetabulum.<sup>2,10</sup> As mentioned, the role of Orthopaedic is to assist bladder closure by approximating the pelvic diastasis, even though the diastasis doesn't seem to affect the pelvic and hip function, particularly during the walking process.<sup>4</sup> To achieve this, a pelvic osteotomy can be implemented, especially for severe cases, as shown in our patient whose pelvic diastasis was wide enough. Some also stated that osteotomy is amenable for those who present after the first month of age. Several osteotomy methods, such as iliac osteotomy, the proximal end of the greater sciatic notch osteotomy followed by a rotation of distal fragment, osteotomy of the lateral side of pubic bone or the staged osteotomy, have been proposed.<sup>8</sup> The anterior osteotomy is one of the most commonly used methods because it is considered safer and easier. The idea of the procedure which Sponseller introduced is to perform horizontal osteotomy anteriorly at the iliac, then the lower segment of the pelvis could be moved medially.<sup>2,9</sup> For young children, some surgeons prefer to conduct vertical- hinged osteotomy at the posterior side near the sacroiliac. The idea is to create a hinged greenstick fracture, which the children would benefit from. In some successful cases, the post-operative diastasis ranges from 1-3 cm.<sup>2,8,10</sup>

## CONCLUSION

Bladder exstrophy is a congenital anomaly which requires a multidisciplinary approach. The role of an orthopaedist is to assist in bladder reconstruction and closure by decreasing pubic diastasis, usually performed during the early days or months of life. Surgery conducted at older age, as shown by very little literature, might need to be revised. In our case, we performed pelvic diastasis correction in older patients with bilateral anterior pelvic osteotomy, as shown by decreased diastasis to 1.5 cm postoperatively. However, thorough follow-up is entailed to perceive the long-term outcome, particularly the function and healing of the structure involved.

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