Anterior Pelvic Osteotomy of Superior Pubic Rami as Adjunctive Measure to Repair the Primary and Recurrent Cases with Bladder Extrophy

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Abstract
Bladder extrophy (ectopia vesicae) it’s a congenital anomalies that exist along the spectrum of the extrophy – epispadius complex and involve protrusion of the urinary bladder through abdominal wall defect with variable presentations including abnormalities of the bony pelvis, pelvic floor and genitalia. It is a rare congenital anomaly and observed in one of every 10,000-50,000 live births, and about four times more often in boys than in girls. The classical approach for the repair has been disbelieving the need for pelvic osteotomy adjunct to bladder extrophy repair performed within 72 hours of life due to the relative malleability. The current surgical approach consists of early closure with pelvic osteotomy and later surgery for continence to achieve satisfactory urinary storage, continence and bladder emptying by the time the child enters school. Several types of pelvic osteotomies used adjunctively to primary repair. Anterior pelvic osteotomy of the superior pubic ramus, seems to be a safe and quick method combined with the repair to obtain tension-free approximation of the symphysis pubis and better results. This study conducted to evaluate the feasibility of the bilateral anterior pubic osteotomy as adjunctive measure to simple closure of the bladder extrophy and achieving a tension-free approximation of the symphysis pubis and abdominal wall. Prospective study between January 2006 and January 2017, involve a total of 14 patients (12 males and 2 females) with bladder extrophy, 2 of them were recurrent. All cases underwent elective surgery for repair, which include dissection of the extrophic bladder from the abdominal wall, closure of the bladder and reconstruction of the urethra, with identification of pubic bone from attached tissues, which makes osteotomy of the superior pubic ramus easy, thus tension free approximation and less dehiscence. Fourteen patients with Bladder Extrophy were operated. Twelve 85.7% of the patients were males and only two (14.2%) was female. Their ages at the time of the procedure ranged from 2 to 5 days for the primary cases and (7, 8) months for the recurrent two cases. Successful bladder closure was achieved in 13(92.8%) of the 14 patients operated. The bilateral anterior pelvic osteotomy was safe, quick with minimal blood loss and can be performed by the pediatric surgeon without the need to an orthopedic surgeon. Bilateral anterior pelvic osteotomy of superior pubic rami is a sufficient adjunctive measure to bladder extrophy repair, to obtain tension-free approximation of the symphysis pubis, and hence diminishes the risk of wound dehiscence whether primary or re-do cases. It provides further advantages namely, ease and rapidity, minimal blood loss, and no need for an extra skin incision or patient’s repositioning.

Keywords: Bladder extrophy, pelvic osteotomy, Primary, Recurrent.
INTRODUCTION
Bladder exstrophy (ectopia vesicae) it’s a congenital anomalies that exist along the spectrum of the exstrophy - episadius complex and involve protrusion of the urinary bladder through abdominal wall defect with variable presentations including abnormalities of the bony pelvis, pelvic floor and genitalia\textsuperscript{[1]}. It is a rare congenital anomaly and observed in one of every 10,000-50,000 live birth, and about four times more often in boys than in girls. It is a complex defect that affects commonly the urinary and musculoskeletal systems in addition to the gastrointestinal and genital systems \textsuperscript{[2]}. Cloacal membrane hypertrophy arise between the 6th and 10th week of gestation prevents migration of the mesoderm between the ectoderm and endoderm. Development of the lower part of the abdominal wall arrested that lead to diastasis with extension of the muscles and fascias of the anterior abdominal wall and pelvic diaphragm. The anus and vagina are anteriorly displaced. Reduction and bending of the penis in boys with total cleft of the clitoris in girls result of extension and rotation of the pubic bones. Additionally, the urogenital septum, in the form of two lateral bundles that attach the bladder neck region to the pubic bones, is also cleft. Total dissection of these bundles is necessary to position the bladder deep within the pelvis\textsuperscript{[2]}. Classic bladder extrophy patients are born with a symphyseal diastasis of a mean value of 4 cm at birth that increases steadily with age to a mean of 8 cm at age 10, compared with a mean normal width of 0.6 cm at all ages. The sacral width and length of the posterior (iliac) segment of the pelvis in extrophy patients are normal; however, the anterior (ischio-pubic) segment of the pelvis is a mean 30% shorter and both the anterior and posterior segments are externally rotated compared to normal\textsuperscript{[3]} as shown in fig. 1.

The current surgical approach consists of early closure and later surgery for continence aiming; if possible, to achieve satisfactory urinary storage, continence and bladder emptying by the time the child enters school\textsuperscript{[4]}. Primary closure with approximation of the pubic bones is generally regarded as the initial procedure of choice for patients with this malformation. Even if this is accomplished during the first few days of life, primary closure usually requires some form of osteotomy if the pubic bones are to be brought together without undue tension \textsuperscript{[5]}. Fusion of the symphysis with reconstruction of all layers of the anterior abdominal wall is the basic element of the surgical procedure. It is thought that osteotomy is not necessary in newborns up to 72 hours of life because the pubic bones are elastic enough to be brought together\textsuperscript{[6]}. Pelvic osteotomy performed at the time of initial closure is recommended for a variety of reasons: It aids in tension-free approximation of the bladder, posterior urethra, and abdominal wall. It allows placement of the vesico-urethral complex deep within the pelvic ring to enhance bladder-outlet resistance. Finally it provides alignment of the large pelvic floor muscles to support the bladder neck \textsuperscript{[7],[8]}. Bilateral posterior iliac osteotomy has been the most commonly used technique in patients.

Figure 1: 2 days female with classic bladder extrophy.
undergoing primary closure of bladder extrophy. Another osteotomy technique, first described by Frey and Cohen in 1988 and consisting of anterior pelvic osteotomy of the superior pubic ramus, seems to be a safe and quick alternative method to obtain tension-free approximation of the symphysis pubis \(^{[5], [9]}\). In this study we evaluate the feasibility of the bilateral anterior pubic osteotomy as adjunctive measure to simple closure of the bladder extrophy and achieving a tension-free approximation of the symphysis pubis and abdominal wall.

**PATIENTS AND METHODS**

In this prospective study between January 2006 and January 2017, a total of 14 patients (12 males and 2 female) with bladder extrophy, 2 of them were recurrent. All underwent elective surgery with bilateral anterior pelvic osteotomy for repair. Age range were 2-5 days for the primary cases and (7, 8) months for the recurrent two cases. Abdominal ultrasound obtained prior to surgery for all patients and no further associated urinary tract anomalies were recorded.

With the initial bladder closure, attention was directed to the pelvic osteotomy, in which both crura were dissected from their attachment to inferior pubic wing. The technique of anterior pelvic osteotomy was the same as described by Frey and Cohen \(^{[9]}\).

The superior pubic rami were identified at the lateral border of the rectus abdominis sheath, and then divided with a bone-cutting forceps with care taken to avoid injury to the inferior periosteum and obturator nerve and vessels. Sometimes diathermy was used. After completing the pubic osteotomy bilaterally, both pubic bones are tilted medially and approximated using one or two polyglactin-1 or 2 sutures. Before securing the polyglactin sutures, it is important to make sure that the proposed bladder neck and urethra are positioned deep to the approximated pubic bones. Wound closure done with suturing the easily approximated both rectus muscles using interrupted absorbable sutures. Finally, the skin is closed as shown in fig. 2.

**Figure 2:** Same 2 days female patient after repair with bilateral anterior pelvic osteotomy.

Postoperatively, recurrent cases were immobilized by means of hip spica cast as a fixation for 2-3 weeks, and then discharged home with legs joined together with an elastic bandage for another three weeks and then movement was allowed as shown in fig. 3,4, while the primary cases were immobilized by a plaster and pressure bandage for 3 weeks. Broad spectrum antibiotics were given. The vesical catheter was removed at day 10 and the two ureteric stents were removed at day 11 and 12 respectively.

**Figure 3:** 7 month male with recurrent extrophy
RESULTS

Twelve 85.7% of the patients operated on were males and only two (14.2%) were female. Their ages at the time of the procedure ranged from 2 days to 5 days for the primary cases and (7, 8) months for the recurrent two cases. Reconstruction of the recurrent cases was more difficult than for the remaining primary cases. Successful bladder closure was achieved in 13(92.8%) of the 14 patients operated. Three patients suffered from soft tissue infection which was treated conservatively. Two of them by frequent wound dressing and appropriate antibiotic therapy and the repair remained intact. Only one patient had complete dehiscence of the repair. Failure of closure was noted in 1 patient (5 day’s age) compared to 0% among patients aged 2-4 days and even 0% for the recurrent two cases [Table 1] and [Graph 1].

Table 1: Age at time of repair with sex & fate

| Age        | No. of patients | male | female | Failure |
|------------|----------------|------|--------|---------|
| 1st day    | 0              | 0    | 0      | 0       |
| 2nd day    | 4              | 3    | 1      | 0       |
| 3rd day    | 4              | 4    | 0      | 0       |
| 4th day    | 3              | 2    | 1      | 0       |
| 5th day    | 1              | 1    | 0      | 1       |
| 1-12 month*| 2              | 2    | 0      | 0       |
| Total      | 14             | 12   | 2      | 1       |

1-12 month age group represents the recurrent cases enrolled in this study.

They were primarily operated during the first 3 days without osteotomy.

Graph 1: Age at repair with sex and fate
The operative time for both osteotomies ranged from 30 to 45 minutes. Patients intraoperative repositioning was not needed. Blood loss was minimal and no blood transfusion was required in all patients, and tension-free approximation of the symphysis pubis was achieved. Patients with successful repair were followed for about one year postoperatively and perfection of the repair was confirmed in all of them.

**DISCUSSION**

Aim of the initial extrophy repair is to close the pubic bones and soft tissue of the anterior abdominal wall without tension. One of the most important surgical principles is approximation of the sutured tissues without tension for better healing and results. With classic approach for bladder extrophy repair; there was an acceptance to close the symphysis without osteotomy, especially in newborn up to 72 hours of life due to the relative malleability of the pelvic bone [6]. Classic bladder extrophy patients are born with a pubic diastasis that increases steadily with age from a mean value of 4 cm at birth to a mean of 8 cm at age 10, compared with a mean normal width of the pubic symphysis of 0.6 cm at all ages [3], so it is apparent that any type of osteotomy seems recommended to prevent dehiscence of the reconstructed bladder and abdominal wall.

Different types and techniques of pelvic osteotomies have been described by many authors, which can be chosen according to the surgeon’s preference and experience [7]. [9]-[14]. Bilateral posterior iliac osteotomy described by Schultz in 1958[7]. The anterior innominate osteotomy described by Sponseller et al in late 1980s and was used to approximate the pubic bones using an external fixator [10]. They became well-known techniques for closure and repair of cases with bladder extrophy. However; in addition to repositioning the patient with posterior iliac osteotomy, both remains major time-consuming procedures and usually done by orthopaedic surgeons, increased blood loss and postoperative pain.

Other approaches like, combined vertical with horizontal pelvic osteotomy [12], or oblique iliac wing osteotomy [13], were also described with some promising results. Anterior pelvic osteotomy was first described by Frey and Cohen in 1985 [9], for repair of patients with bladder extrophy. This type of osteotomy has many advantages; the operative duration is reasonable, with no need to change the patient’s position, minimal blood loss, can be used for both primary and re-do cases, regardless of patient age and no need for an orthopedic surgeon [14],[15]. Frey in 1996 again documented further experience with anterior pelvic osteotomy on 16 cases after extrophy repair. He reported excellent results in all patients except two, who developed soft-tissue infection and bladder dehiscence in one, and transient obturator nerve palsy in the other [15].

Chiari et al reported in his study using anterior pelvic osteotomy [5], that all five procedures were successful, tension-free complete approximation of the symphysis and uncomplicated healing without palsy of the obturator nerve or postoperative hemorrhage. Elsayed et al reported simple closure with anterior pubic osteotomy is a feasible and effective adjunctive means to facilitate both bladder and abdominal closure for patients with bladder extrophy and with success rate; 93% [16].

As comparison to Chiari and Elsayed studies who reported a male incidence of 80% and 86.6% respectively [5],[16], in present study 85.7% of the patients were males. Successful repair using bilateral anterior pelvic osteotomy of superior pubic rami in this study was achieved in 92.8% of the patients operated on. This result is comparable to what is reported by Elsayed et al [16]. It is slightly lower than what is reported 100% by Chiari et al [5], which may be due to the small sample size in the later study[table 2and graph 2].
Table 2: Comparison of Present Study (no., sex, success rate) to Other Studies

| Study            | No. of patients | Male % | Female % | Success rate % |
|------------------|-----------------|--------|----------|----------------|
| Present study    | 14              | 85.7   | 14.3     | 92.8           |
| Chiari et al.    | 5               | 80     | 20       | 100            |
| Eseyed et al.    | 15              | 86.6   | 13.4     | 93             |

Graph 2: Comparison of present study (no., sex, success rate) to other studies

The operative time for both osteotomies ranged from 30 to 45 minutes. The recurrent cases get longer time than primary cases. These results were comparable to what is reported in Chiari et al and Elsayed et al studies [5][16]. Failure rate of closure among male patients was (8.3%) compared to 0% among female patients. Failure of closure was noted in 1 patient (5 day’s age) compared to 0% among patients aged 2-4 days and even 0% for the recurrent two cases.

The procedure was safe, relatively quick, repositioning of the patients not required and was performed by the pediatric surgeon alone. Blood loss was minimal and blood transfusion was not required in all cases.

CONCLUSION

Bilateral anterior pelvic osteotomy of superior pubic rami is a sufficient adjunctive measure to bladder extrophy repair, to obtain tension-free approximation of the symphysis pubis, and hence diminishes the risk of wound dehiscence whether primary or re-do cases. It provides further advantages namely, ease and rapidity, minimal blood loss, and no need for an extra skin incision or patient’s repositioning.

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