Oblique pelvic osteotomy in the exstrophy/epispadias complex

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We reviewed retrospectively 45 patients (46 procedures) with bladder exstrophy treated by bilateral oblique pelvic osteotomy in conjunction with genitourinary repair.

The operative technique and post-operative management with or without external fixation are described. A total of 21 patients attended a special follow-up clinic and 24 were interviewed by telephone. The mean follow-up time was 57 months (24 to 108).

Of the 45 patients, 42 reported no pain or functional disability, although six had a waddling gait and two had marked external rotation of the hip. Complications included three cases of infection and loosening of the external fixator requiring early removal with no deleterious effect. Mid-line closure failed in one neonate managed in plaster. This patient underwent a successful revision procedure several months later using repeat osteotomies and external fixation.

The percentage pubic approximation was measured on anteroposterior radiographs pre-operatively, post-operatively and at final follow-up. The mean approximation was 37% (12% to 76%). It varied markedly with age and was better when external fixation was used. The wide range reflects the inability of the anterior segment to develop naturally in spite of close approximation at operation.

We conclude that bilateral oblique pelvic osteotomy with or without external fixation is useful in the management of difficult primary closure in bladder exstrophy, failed primary closure and secondary reconstruction.

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The exstrophy/epispadias complex has a spectrum from epispadias, through classic bladder exstrophy to devastating cloacal exstrophy, which includes intestinal prolapse and spinal anomalies (Fig. 1). The prevalence of classic bladder exstrophy is approximately 1 per 40 000 children, and cloacal exstrophy, 1 per 200 000 children.¹ Those born with bladder exstrophy have an exposed bladder plate and wide diastasis of the pubic rami.^{2,3} The epidemiology, anatomy and need for multidisciplinary treatment in the exstrophy/epispadias complex have been well described.¹⁻⁵

Exstrophy itself is not a lethal condition, but in the past complications and treatment often resulted in death. Recent advances in anaesthesia and surgery have contributed to a dramatic fall in mortality. Nowadays, death from exstrophy alone is uncommon. The children have normal intelligence and cardiorespiratory function. They can expect a normal lifespan and respond well educationally and socially.^{1,2,4-7} They are often extraordinarily motivated in spite of their physical problems. Different types of pelvic osteotomy have been described

to facilitate relaxation of the abdominal wall during closure of exstrophy and to improve the outcome of genitourinary reconstruction.^{1,8-21} The traditional osteotomies described are posterior⁸ and transverse,¹ or a combination of both^{1,12} (Fig. 2). McKenna et al²² compared the different osteotomies using models and described an oblique/diagonal osteotomy, which they thought would be more effective. However, they do not appear to have used this in a major clinical study.

At our hospital osteotomies were initially posterior¹³ and later transverse.¹⁶ Since 1996, we have developed an oblique osteotomy and a system of external fixation. We used computer models to predict the displacement (Fig. 3) and improve the technique as the series progressed. We now report the results.

Patients and Methods

We reviewed the notes and radiographs of 45 children who had undergone bilateral oblique osteotomies by the senior author (DJ) as part of reconstruction of exstrophy between 1996 and 2003. Of these, 21 were able to attend a

Fig. 1

Photograph of a patient with severe exstrophy showing the exposed bladder and uterus.

Fig. 2a

Fig. 2b

CT scan images of the infant pelvis showing a) frontal and b) superior views with the lines of posterior, transverse and oblique osteotomies.

special follow-up clinic and the remainder were interviewed by telephone by one of the authors (SP). This reflected the widespread referral pattern of our hospital. There were 19 girls and 26 boys with a mean age at operation of 15.5 months (3 weeks to 10 years) (Fig. 4).

The patients were grouped according to age at the primary or revision surgery, and by whether fixation was used (Fig. 4). The operating time, blood loss and urological procedure were noted. Complications which included pin-site infection, early loosening of pins requiring removal of the frame, malunion, nonunion, nerve palsy and scarring were reviewed.

Clinical review included symptoms, limitation of activity, leg-length discrepancy, abnormality of gait, power of hip flexion, wound healing and palpation for symphyseal separation. Anteroposterior radiographs of the pelvis taken

before operation, at four to six weeks after surgery and at the latest follow-up were analysed.

The diastasis was measured as the distance between the two most medial points of the pubic rami. The hips were examined for dysplasia and the sites of osteotomy for healing. The percentage of pubic approximation was calculated according to the following formula:

$$\frac{\text{Pre-operative diastasis (mm)} - \text{post-operative diastasis (mm)}}{\text{Pre-operative diastasis (mm)} - 10 \text{ mm}} \times 100$$

The denominator corrects for the 1 cm symphyseal diastasis which is present throughout life in normal individuals. Post-operative assessment of diastasis was based on the measurements at the latest follow-up. In patients who required revision osteotomy, only the results of the last procedure were analysed.

Fig. 3a

Fig. 3b

Fig. 3c

Computer models (3-D reconstruction) showing an oblique osteotomy a) before and b, c) after displacement of the osteotomy. The models predict prominence of the anterior superior iliac spine and flattening of the perineum secondary to the osteotomy and derotation.

Fig. 4

Graph showing the groups of patients according to age and whether fixation was used.

Operative technique (Fig. 5). The osteotomies are usually carried out after mobilisation of the bladder and surrounding soft tissues, and reconstruction of the bladder and bladder neck. The patient is prepared for osteotomy with placement of a rolled towel beneath the buttock. The incision is just below and parallel to the iliac crest, which in the exstrophy patient is laterally placed. The interval between tensor fascia lata and sartorius is developed to the anterior superior iliac spine and the lateral cutaneous nerve of the thigh retracted medially. The iliac apophysis is split and the ilium exposed on both sides down to the sciatic notch, in which the periosteum is mobilised by rotatory movements of a bone spike in order to give maximum exposure. On the medial side, psoas is divided at the pelvic brim. On the lateral side, the flat part of the ilium above the hip is exposed.

The osteotomy is between points as high as possible in the notch to the iliac crest, approximately 1 to 2 cm behind

Fig. 5a

Fig. 5b

Fig. 5c

Fig. 5d

Fig. 5e

Fig. 5f

Intra-operative images. Figure 5a – The anterior superior iliac spine is removed, the pin has been passed from the back of the ilium, exiting the front and lower part of the notch. This track allows maximum bone behind the tip of the pin and in front of its shaft. Figure 5b – Model of the pelvis showing the osteotomy and the track of the pin. Figure 5c – Elevation of the pin internally rotates the distal fragment, approximates the symphysis and brings the pin into the line of the wound. Figures 5d and 5e – Photographs showing the frame applied. The limbs are bandaged together and a nylon tape is tied between the bandage and frame. This prevents any tendency for the pins to lean back against the abdominal wall. Figure 5f – Post-operative antero-posterior radiograph showing the osteotomies, placement of the pin and fixator assembly. A spanner is taped to the frame.

Fig. 6

Photograph showing broomstick plasters applied to a neonate when external fixation is not possible.

the anterior superior iliac spine. In the neonate, the osteotomy is made from crest to notch, using a bone cutter and osteotome, rotatory movements of which mobilise the inner cortex to allow maximum displacement of the distal fragment. In the infant and young child, the osteotomy is from the notch to crest, using a Gigli saw. Its passage can be facilitated by passing a tape through the notch and pulling the saw through the notch with the tape. It is necessary to narrow and bend the eye of the saw to help it through.

In the older child the osteotomy is from crest to notch, using a power saw. An osteotome completes the cut and mobilises the inner cortex as in the neonate. In all cases the anterior superior iliac spine is removed with a bone cutter down to an area just above the anterior inferior iliac spine.

The route for the fixator pin is predrilled (2 mm) from the back of the ilium, exiting the front and lower part of the notch (Figs 5a and 5b). This track allows maximum bone behind the tip of the pin and in front of its shaft. The pin is inserted on a Jacob's chuck. The diameter of the pin depends on the size of the patient and is usually 3.5 mm in the neonate and infant and 4.5 mm in the older child.

At this stage the pin is horizontal to the table. Elevation of the pin internally rotates the distal fragment, approximates the symphysis and brings the pin into the line of the wound, which is closed after repair of the iliac apophysis (Fig. 5c).

The procedure is repeated on the other side. It is better to use short pins to avoid their impingement against the table when the patient is turned. The pins are propped on rolled towels to maintain relaxation in the mid-line. The urological reconstruction is completed along with approximation of the symphysis using 0 polydioxanone sutures. An A-frame is applied, along with anterior slabs to the lower limbs, taking care that plaster does not impinge on the groin (Fig. 5d).

The limbs are bandaged together and a nylon tape is tied between the bandage and the frame (Fig. 5e). This prevents any tendency for the pins to lean back against the abdominal wall. A spanner is taped to the frame in case adjustments are necessary on the ward after a post-operative anteroposterior radiograph is taken (Fig. 5f) and routine pin care given.

The plaster slabs reduce limb movement in order to facilitate mid-line healing. They are removed at three weeks and the pins at six weeks. In the neonate, if fixation is not possible, the baby is immobilised in broomstick plasters (Fig. 6).

Results

The mean age of the group treated by fixation was 24 months (3 months to 10 years) and in those managed in broomstick plasters was 5.5 weeks (3 weeks to 15 months). Stabilisation by an A-frame pelvic fixator occurred in 29

procedures and in 17 by plaster casts. One of these was a neonate who had mid-line breakdown after osteotomies without fixation and several months later underwent successful reconstruction, which included revision osteotomies with fixation.

The mean follow-up was 57 months (24 to 108) and the mean blood loss was 467.5 ml (120 to 760). There were 41 patients with bladder exstrophy and four with cloacal exstrophy. The simultaneous urological procedures were varied and included one or more of the following: bladder closure, bladder neck reconstruction, ureteric re-implantation, Mitrofanoff formation, genitoplasty, closure of cloacal exstrophy, repair of epispadias, omphalooplasty and augmentation cystoplasty.

All six patients who underwent revision osteotomy had external fixation. The frames were removed at a mean of 5.7 weeks (3 to 7). Three patients required removal at three weeks because of early loosening. However, there was no mid-line breakdown in these patients.

A total of nine patients had oozing from the pin-site, but only in two were microbial pathogens grown. These were *Staphylococcus aureus* in one patient and β -haemolytic streptococcus in the other. All pin sites healed with no sequelae. Radiographs at follow-up showed that all the osteotomies healed. There was no case of neurological or vascular injury.

Symptoms and function. There were three children with mild pelvic pain in relation to extended activity; the remaining 42 had no pain. In 40 children there was no limitation of activity when compared with their peers, as described by their parents. The parents of six of the children described their walking as waddling, which became more apparent when they were tired. All patients and parents were satisfied with the appearance of the surgical scars. In the 21 patients who attended the special follow-up clinic there was no discernable leg-length discrepancy and no pelvic instability was detected on stressing the pelvis.

Of the six children with a waddling gait, two had increased external rotation of the lower limbs with foot progression angles of 30° and 25°, respectively, and two had associated spina bifida. All had full power of hip flexion indicating a satisfactory return of the function of psoas.

Diastasis and pubic approximation. The mean symphyseal diastasis for all patients was 42.3 mm (32 to 75) before and 27.8 mm (0.8 to 40) after operation. The mean percentage of pubic approximation for the series was 37% (12% to 76%). For all age groups in which an external fixator had been used the mean approximation was 51.2% (42% to 76%) whereas with broomstick plasters it was 12.2% (12% to 34%). The percentage of pubic approximation varied markedly with age. There were 21 patients undergoing surgery at between 0 and 6 months of age who had a mean approximation of 21.3% (18% to 29%). Osteotomies at between 6 and 18 months of age (15 patients) had a mean approximation of 56.5% (40% to 64%), at between 18 to 60 months (four patients) of 74.25% (72%

to 76%) and at over five years of age (five patients) of 49.6% (38% to 56%).

The patients undergoing pelvic osteotomy as a revision procedure had a mean pubic approximation of 73% (70% to 75%).

The wide range of percentage approximation reflects the inability of the anterior segment to develop naturally, even though close approximation was achieved at operation. One patient had a pin-site infection but did not grow a pathogen or demonstrate early loosening. This group of patients had no musculoskeletal problem clinically.

Discussion

The role and timing of pelvic osteotomy in the reconstruction of the exstrophy complex depends on the requirements of the urologist. The traditional sequence of reconstruction of exstrophy involved primary closure of the bladder and abdominal wall in the newborn period, repair of epispadias around the age of one or two years in boys, and reconstruction of the bladder neck around the age of three or four years, when the child was mature enough to achieve urinary continence.¹

During our study, urological techniques and preferences were evolving at our hospital. In the earlier years, pelvic osteotomy was routine in the primary closure of bladder exstrophy. At present, osteotomies are reserved for patients with a difficult primary closure (usually cloacal exstrophy), a failed primary bladder closure and secondary reconstruction. These major operations are becoming more comprehensive, because of excellent anaesthesia and intensive care.

We believe that in the first three years of life oblique pelvic osteotomy can be a major help in securing mid-line closure. In later years the operation can be used to improve the appearance of the abdominal wall, mons pubis and penis. It may also have a role in preventing uterine prolapse in adults. However, this group of patients has not yet been analysed.

We have not been able to confirm a relationship between symphyseal closure and continence. This is because soft-tissue surgery for continence continues to evolve and there are too many variables to propose a definite relationship between osteotomy and continence.

The early osteotomies extended from the iliac crest to the sciatic notch, with stabilisation using broomstick plasters. However, there were difficulties with access to the perineum. The solitary mid-line breakdown occurred in this group and prompted development of a system of external fixation, which at our hospital had previously only been used in older children.

The AO minifixator (Synthes AO; Stratec Medical Ltd, Welwyn Garden City, United Kingdom), usually applied to the adult wrist, was suitable for the infant pelvis and the adult version for children. The A-frame assembly proved to be effective at all ages and the system was accepted and easily managed by theatre personnel. The A-frame is a stronger

Fig. 7a

Fig. 7b

Fig. 7c

Post-operative CT of united osteotomies. They show (a and b) the displacement and c) flattening of the front of the pelvis.

construct than a simple spanning rod between the pins. It also gives better rotational control. The diameter of the pin varied according to the size of the pelvis but, at all ages, a single pin proved to be adequate. The children were well sedated with good relief from pain and were comfortable within a few days.

The pelvis is congenitally abnormal and therefore oblique pelvic osteotomy, although helping with recon-

struction, does not create a normal pelvis. Rotation of the anterior segment approximates the symphysis, but leaves the front of the pelvis flattened (Fig. 7). This is of particular significance in boys in whom the corpora cavernosa are attached to the inferior pubic rami.

The preliminary computer model of the oblique osteotomy predicted that the anterior superior iliac spine would be prominent and likely to provoke counter pressure from

iliopsoas. Excision of the anterior superior iliac spine, and division of the psoas tendon allowed the distal segment to rotate neatly into the defect in psoas. However, no symptoms of weakness of iliopsoas were reported and no clinical evidence of weakness was observed in the children who attended for follow-up.

We routinely retained the pelvic fixator for six weeks. This may be overcautious since in those patients in whom it had to be removed early, no problems were seen. We are therefore considering fixation for four weeks only.

Advances in urological practice require the orthopaedic surgeon to be flexible in the role of assistant to the urologist in order to achieve a successful reconstruction. The technique of oblique osteotomy with or without fixation has proved to be useful and applicable to all ages and compares well with more complex osteotomies.¹

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