
RESIDENT'S CORNER

Wire in the hole: a case series of eroded intrapubic wire sutures causing genitourinary complications in the bladder exstrophy complex

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Bladder exstrophy and cloacal exstrophy are rare congenital defects of the genitourinary tract that require complex surgical reconstruction. Malrotation of the bony pelvis causes a characteristic diastasis of the pubic symphysis, which is surgically reduced at the time of

initial bladder closure. For a successful primary closure without tension such that the bladder can be placed deep within the pelvis, pelvic osteotomy is often used. However, alternative techniques have been utilized to bring the pubic rami into apposition. The authors present four bladder/cloacal exstrophy patients in which an intrapubic wire was used for pubic apposition, resulting in significant genitourinary complications.

Key Words: bladder exstrophy, cloacal exstrophy, pubic symphysis diastasis, bone wires

Introduction

Formerly thought to be a major bladder anomaly, bladder exstrophy is now known to be a birth defect involving not only the genitourinary system, but also the pelvic floor musculature and bony pelvis. Malrotation of the anterior and posterior pelvic bones and a 30% shortening of the pubic rami causes a characteristic widening of the pubic symphysis,¹ which is typically surgically reduced at the time of initial closure. Various types of osteotomies have been utilized in the repair of the pubic diastasis. Posterior iliac osteotomy was the first procedure developed for the correction of the pubic diastasis seen in the exstrophy-epispadias complex. However, it required placing the patient in the prone position and turning him/her during the repair. Therefore, several methods of anterior osteotomy have been developed in order to shorten operative time and to obviate the need to place the patient in the prone position. These include anterior osteotomy of the superior pubic rami, anterior diagonal iliac osteotomy, and anterior transverse innominate

osteotomy with optional vertical iliac osteotomy. The anterior transverse innominate osteotomy with optional vertical iliac osteotomy is employed most often at the authors' institution. This osteotomy is done entirely with the patient supine. The optional vertical iliac osteotomy is performed in classic bladder exstrophy patients above the age of 2 years due to decreased elasticity of the sacroiliac ligaments and in patients with a particularly wide diastasis. When compared to posterior iliac osteotomy, this technique causes less intraoperative blood loss, better apposition and mobility of the pubic rami at the time of closure, and allows for the ability to place external fixators under direct vision. However, this osteotomy introduces the risk of transient femoral nerve palsy, which occurs in less than 10% of cases and typically resolves within 90 days postoperatively.² Bilateral transverse innominate and vertical anterior iliac osteotomies are recommended when the primary closure is delayed, in cases of extreme diastasis, and/or in the presence of a non-malleable pelvis to allow for adequate placement of the bladder and posterior urethra deep within the pelvis.³ Some surgeons, however, do attempt closure without osteotomy and have used various other techniques for approximation of the pubic rami. After initial closure, surgeons keep patients immobilized via different methods including external fixation with modified Buck's traction, modified Bryant's traction, spica cast, or mummy wrapping. Some opt to not to keep patients immobilized. In the experience of the authors, 6-8 weeks of external fixation with modified

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Buck's traction after osteotomy or 4-6 weeks of modified Bryant's traction after closure without osteotomy lead to the best outcomes.⁴ Cloacal exstrophy is another part of the exstrophy-epispadias spectrum, except the constellation of abnormalities is even more severe.

In this case series, the authors describe three patients born with classic bladder exstrophy and one patient born with cloacal exstrophy who were closed with the use of an intrapubic metal wire suture, all of which eventually eroded into the urinary tract, causing significant genitourinary complications necessitating multiple corrective surgeries. The authors do not use this method for pubic apposition; all cases of intrapubic wire suture placement were performed at various outside institutions.

Case report

Case 1

A 4-year-old boy born with classic bladder exstrophy presented to the authors' institution after a recent adoption from outside the country. His bladder closure, performed abroad as a newborn with osteotomy and a spica cast, was complicated by urinary retention prior to 2 years of age, requiring suprapubic cystostomy and catheter drainage. The child was brought to the authors' institution for examination under anesthesia and urethroscopic examination revealed a metal wire in the urethral lumen. A gravity cystogram showed grade III vesicoureteral reflux bilaterally and a bladder capacity of 46 mL. The radiographic image demonstrated the metal wire that had been seen urethroscopically, sutured into the pubic symphysis for approximation of the pubis at the time of bladder closure, Figure 1. In the operating room, a small incision was made in the distal urethra and the wire was identified, carefully untwisted, and removed. A cystoscope was reintroduced into the urethra, but an impenetrable stricture at the site of suture erosion could not be bypassed by cannulation with a flexible wire. Urethral cannulation through the suprapubic tract was also attempted without success. A suprapubic catheter was left in place with plans for routine exchanges until bladder augmentation, ureteral reimplantation, and creation of a continent catheterizable urinary stoma in 1 year's time.

Case 2

A 3-year-old boy born with classic bladder exstrophy presented to the authors' institution regarding management of recurrent vesicocutaneous and urethrocutaneous fistulae. His bladder was closed overseas, without osteotomy, at 9 days of age with approximation of the pubic symphysis using an



Figure 1. Reason for stent by culture result.

intrapubic metal wire suture and a spica cast. He also underwent an epispadias repair at a later date. A recent kidneys, ureters, and bladder x-ray (KUB) demonstrated that the wire had eroded through the pubic bones and was in an unclear location in the midline. He was having significant pain in his penis and testes, and occasionally would suffer from rectal prolapse. In the operating room, cystoscopy revealed two large bladder stones requiring staged holmium laser lithotripsy to fragment the significant stone burden. Upon lithotripsy, the metal suture seen on KUB was identified in the bladder as the nucleus to the stones, Figure 2. A small suprapubic cystostomy was required to extract the wire because it could not be removed cystoscopically. At that time, the bladder was noted to be located very anteriorly within the pelvis (consistent with a failed closure). Eight months later, the child underwent bilateral transverse innominate and vertical iliac osteotomies, reclosure of bladder exstrophy with repair of two vesicocutaneous fistulae, and repair of three urethrocutaneous fistulae. In

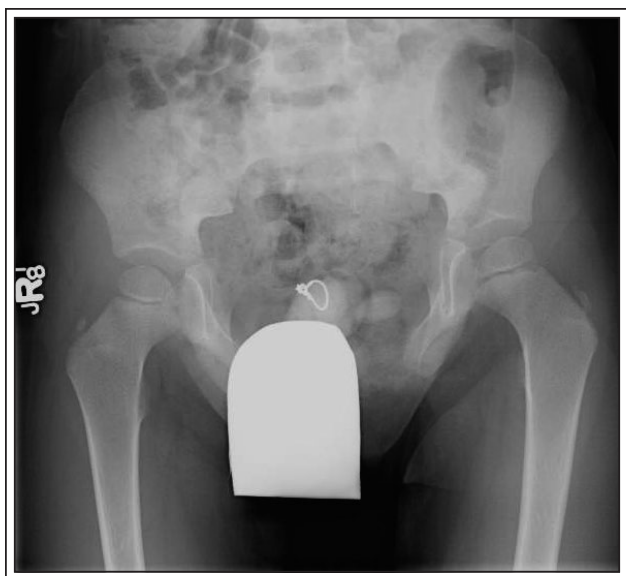


Figure 2. KUB of case 2, demonstrating eroded intrapubic metal wire suture in unclear location in patient's midline.

addition, an intrapubic bar was placed to aid in closure of the bony pelvis and was subsequently removed 6 months postoperatively once the surgical sites had adequately healed.

Case 3

A 12-month-old girl born with classic bladder exstrophy presented to the authors' institution after undergoing primary closure abroad. An osteotomy was not performed at that time. Instead, the pubic symphysis was approximated using a metal wire suture and a spica cast. Unfortunately, the patient suffered a dehiscence soon after closure. The metal wire suture was found in the dehisced bladder, having eroded through the cartilaginous end plates of the pubic symphysis. At 12 months of age, she underwent successful reclosure of her bladder with bilateral transverse innominate and vertical iliac osteotomies at the authors' institution. At the time of reclosure, a #2 nylon suture was used in a horizontal mattress fashion to aid in the approximation of her pubic diastasis. AlloDerm Tissue Matrix (LifeCell Corporation, NJ, USA) was placed between this suture and the bladder in order to provide a protective layer for the bladder tissue.

Case 4

An 8-year-old boy born with cloacal exstrophy presented to the authors' institution after suffering a series of complications following a failed bladder closure. On day 1 of life, the child underwent colostomy with tubularization of the colonic plate, followed by a two-

stage closure of his omphalocele. When his bladder was closed at 22 months old, a pediatric orthopedic surgeon used bilateral osteotomies and a metal wire to close the 10 cm pubic diastasis. After closure, renal ultrasound revealed moderate right and mild left hydronephrosis not present on prior studies. The following year, he developed an enterovesical fistula caused by erosion of the intrapubic wire through both the bladder and bowel discovered at the time of repair. On follow up visits, measured bladder capacity showed a dramatic decrease from 270 mL to 35 mL, presumably due to devascularization of the bladder during fistula repair. Subsequently, his serum creatinine rose from 0.42 mg/dL to 1.82 mg/dL within the course of 2 years in the setting of severe bilateral hydronephrosis. About 4 years after the enterovesical fistula repair, bilateral percutaneous nephrostomy tubes were placed, and his serum creatinine decreased to 1.02 mg/dL and has remained stable. Upon referral to the authors' institution, a cystogram was performed, which revealed a 25 mL capacity bladder with a large diverticulum. An exploratory laparotomy revealed extensive adhesions within the abdomen and pelvis. Due to the extremely small size and poor quality of the bladder, a simple cystectomy was performed with creation of an ileal neobladder and a Yang-Monti type continent catheterizable channel.

Discussion

Successful initial bladder closure in patients with bladder exstrophy is of extreme importance. Multiple studies have shown that a failed initial closure confers significantly worse urinary outcomes in children with bladder exstrophy. Patients with failed closure display lower rates of eventual urinary continence^{5,6} as well as significantly slower bladder growth and smaller bladder capacities.⁷ Failed closures can manifest as bladder prolapse, dehiscence, outlet obstruction, vesicocutaneous fistula,⁸ or as complex failures with loss of the glans, corpora, urethral plate, or penile skin.

Case 1 illustrates an example of outlet obstruction as a result of placement of an intrapubic wire suture. This suture eroded into the posterior urethra, causing an impassable stricture and subsequent obstruction, requiring suprapubic catheterization. In 1999, Baker et al published the outcomes of 41 patients with bladder exstrophy who developed posterior urethral outlet obstruction after closure in the neonatal period. Eleven of these patients were found to have an eroded symphyseal stitch into the urethra. In these children, an average of five therapeutic interventions per patient were required in order to relieve their outlet obstruction. Thirty-one percent suffered from epididymitis, 22% developed upper

tract deterioration, and two patients (4.9%) developed chronic renal insufficiency. Reconstruction of the posterior urethra was also plagued with complications, with a 52% failure rate.⁹

The intrapubic suture in case 2 eroded through the fixation points in the pubic rami and migrated into the bladder, providing a nidus for stone formation. Stone burden was of such magnitude that the patient required two operations to be rendered stone-free. Additionally, he underwent initial bladder closure without osteotomy at nine days of age, and on surgical exploration his bladder was notably anteriorly positioned. This paucity of soft tissue between the bladder and abdominal skin likely resulted in failure of bladder closure with the formation of multiple vesicocutaneous fistulae.⁸

As exemplified in case 3, using a metal intrapubic suture to narrow the pubic diastasis may not adequately counterbalance the tension created by the uncorrected pelvis. Consequently, the intrapubic suture can erode and lead to complete dehiscence of the initial closure.

Case 4 demonstrates how the use of an intrapubic metal wire suture can result in catastrophic complications to both the urinary and enteric tract. Here, not only did the child suffer from a vesicoenteric fistula as a result of wire erosion, but he also lost significant bladder capacity after the surgical repair of the fistula, ultimately leading to the loss of his bladder.

From the authors' experience, osteotomy along with a strong nylon suture is a superior method of correcting the pubic diastasis. In 1997, Sussman et al tested various methods of symphysis pubis repair in 40 pelvises of 1 month-old piglets, as their cartilaginous symphysis and partially ossified pelvis resembled that of human infants. Each repair was tested by applying tensile force until failure, and while no method of repair was equivalent to the strength of an intact pubic symphysis, the #2 nylon horizontal mattress suture was found to provide the best load-to-failure. The authors also found that the better repair methods had large areas of contact between the pubis and repair material. Therefore, maximizing the distribution of load was asserted to be of prime consideration.¹⁰

Due to the tremendous importance of a successful initial closure in patients with bladder exstrophy, surgical techniques must be chosen with great care. The authors believe the use of intrapubic wire suture should be avoided due to potentially severe complications illustrated through this case series. If the patient is older than 72 hours of life, due to relative inelasticity of the pelvis, osteotomy is strongly recommended for adequate closure and long term outcomes. The method of pubic apposition in our practice is a strong nylon suture. In older failed closures with excessive diastasis, a mountaineer internal

fixator bar can be used. In an extreme (> 6 cm) diastasis, we will perform the osteotomy, apply intrafragmentary pins and an external fixator, and gradually crank the pelvis medially over a 2 to 3 week period, then do the soft tissue surgery and place an internal fixator bar at the time of bladder closure.^{11,12} Secure apposition of the pubic rami along with proper immobilization and pain control are the keystones to a successful primary or secondary bladder closure in this interesting group of patients. □

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