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Persistent vesicocutaneous fistula after repair of classic bladder exstrophy: A sign of failure?

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Abstract *Objective:* To describe the potential predisposing clinical characteristics at initial closure, the treatments of and outcomes associated with vesicocutaneous fistula (VCF) after primary bladder closure.

Materials and methods: Eighteen patients were referred for VCF after primary bladder exstrophy closure. Aspects from the primary closure such as gender, timing of diagnosis and repair, osteotomies, pelvic immobilization, layers of closure, use of tissue adjuncts, and complications in addition to details from their required treatment of VCF were retrospectively extracted from the medical record.

Results: A diagnosis of failed closure was made at the time of evaluation in 13 of 18 patients who presented with VCF. All 13 patients underwent delayed single-layer closures and had a widened pubic diastasis. Five patients with secure closures who underwent successful simple fistula repairs were all closed early in life, had multi-layered closures with pelvic osteotomies, and had minimal change in pubic diastasis.

Conclusion: VCF may represent a failed bladder closure. Factors which may suggest failure are a fistula tract in the lower abdominal midline between the pubic rami, a pubic diastasis

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increased from pre-closure measurement, and cystoscopic evidence of an anteriorly positioned bladder.

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Introduction

Multiple surgical options exist worldwide for the modern treatment of bladder exstrophy. Complications such as failure of the primary closure either with complete dehiscence, bladder prolapse, or penile and corporal soft tissue loss can severely impact the potential for voided continence. Recent data by Schaeffer et al. shows that a successful primary bladder closure represents the best chance to create a native bladder with reasonable functional potential and eventual voided continence [1].

While prolapse, dehiscence, and soft tissue loss remain major detriments to voided continence, other complications have been reported which also may impact overall bladder growth and development. One complication reported by Shnorhavorian et al. and then emphasized by Alpert et al. in a multi-institutional study is the development of vesicocutaneous fistula (VCF) after primary closure [2,3]. VCF has been reported in all major types of exstrophy repair [2–7]. Here we describe our experience with a cohort of patients who had undergone complete or staged primary closure and were referred to our institution for recurrent fistulae or continence evaluations and subsequently found to have VCF. The aim of this study is to describe the presentation, treatments and eventual outcomes of this unique complication.

Materials and methods

The records of all patients treated between 1999 and 2011 from an IRB-approved database were reviewed. Eighteen patients were identified with VCF. All patients had classic bladder exstrophy and were referred for a primary ($n = 15$) or recurrent ($n = 3$) VCF. Two patients had a modern staged repair of exstrophy (MSRE), and 16 underwent complete primary repair of exstrophy (CPRE). Gender, age at initial closure and diagnosis, use and type of osteotomies, type and length of immobilization, pre-closure diastasis, number of layers in closure, and any complications were noted at the time of primary closure from the referring institutions' medical records. Details regarding age and presentation at diagnosis of VCF along with technical aspects of treatment were also reviewed.

All patients were male and were initially diagnosed with classical bladder exstrophy. Median age at the time of referral was 19 months (range: 4–38). Upon referral, critical attention during examination under anesthesia was placed on measuring the pubic diastasis with a pelvic x-ray, identifying the location of the bladder relative to the pelvis and skin with bimanual examination and cystoscopy, and assessing the quality of the lower abdominal fascial/subcutaneous closure. Patients with a VCF, a pubic diastasis wider than pre-closure diastasis, a subcutaneous position of the bladder, and attenuated lower abdominal fascia were considered failed primary closures. Otherwise they were considered to have a simple VCF and underwent excision of the fistula.

Results

Of the 18 referred patients with VCF, 13 (72%) were diagnosed with a failed primary closure and 5 (28%) had secure closures with simple VCF. Among the 13 with failed primary closures, 10 underwent re-closure, 2 underwent augmentation cystoplasty and continent urinary diversion (CUD), and 1 patient awaits diversion. Median age was 26 months (range: 8–38). Three of these patients were referred for recurrent VCF (underwent prior fistula repair) and all underwent re-closure. The remaining 5 patients with secure abdominal closures underwent simple fistula repair alone as described below. Median age was 13 months (range: 4–18). At a median follow-up of 33 months (range: 3–94), all patients have been successfully closed, and none of the VCF have recurred.

Initial closure was completed in the first 72 h of life in 13 patients and after 72 h in 5 patients. Of the 13 closed in the first 72 h of life, 6 had osteotomies. Three patients had anterior innominate osteotomies with Spica Cast and external fixation for 4 weeks, and 3 patients had combined anterior innominate and vertical iliac osteotomies with external fixation and Buck's traction for 4 weeks. Of the 7 patients who did not undergo osteotomy but were closed in the first 72 h of life, 2 had modified Bryant's traction for 3 weeks, 2 had a mummy wrap for 3 weeks, and 1 for only 2 weeks. Two boys had Spica Casts for 3 weeks. Of the five patients who underwent delayed closure after 72 h of age at median of 31.8 days (range: 7–81), all had osteotomies. One had posterior iliac osteotomies with Spica Cast for 3 weeks, one a pubic ramotomy and modified Bryant's traction for 4 weeks, 2 had anterior innominate osteotomies and Spica Cast for 4 weeks, and 1 had oblique iliac osteotomies with Spica Cast and external fixation for 4 weeks. All 5 patients who were found to have simple VCF were closed in the first 72 h of life with osteotomies and external fixation.

In all cases, the pubic rami were brought into apposition at the time of surgery according to the operative notes. In all patients who did not undergo osteotomy with primary closure, pubic diastasis was noted to be < 4 cm. Those with a failed closure were found to have a pubic diastasis that increased from a median of 4.0 cm (range: 3.0–5.0) pre-operatively to a median of 5.8 cm (range: 4.5–6.8) at the time of referral. Among patients with a secure abdominal closure, median pre-operative pubic diastasis was 4.2 cm (range: 3.2–4.8) and 2.7 cm (range: 2.0–3.0) at the time of referral.

A VCF occurred postoperatively in the area just below the junction between the pubic rami and abdominal wall at a median of 7 weeks (range: 2–28) after surgery. VCF developed at a median of 8 weeks (range: 3–28) in 4 patients who had a two-layer urethral closure and a median of 6 weeks (range: 2–12) 14 patients with a single-layer urethral closure. Among the patients identified with a two-layer closure, none had a failed primary closure and were

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