Ped Urol Case Rep 2017; 4(2):289-292 DOI: 10.14534/PUCR.2017225091

Ped Urol Case Rep

PEDIATRIC UROLOGY CASE REPORTS

ISSN 2148-2969

Journal homepage: http://www.pediatricurologycasereports.com

Case report: The use of a urethral diverticulum to cover the penis in a hypospadias cripple

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ABSTRACT

A boy presented as a hypospadias cripple, with anatomy that included a very large diverticulum and little residual external penile skin. The degree of excess skin-like tissue of the diverticulum was sufficient to enable eversion of the longitudinally incised diverticulum and formation of a urethra from the diverticular tissue at a second operation with ultimately an excellent result.

Key Words: Hypospadias; urethral diverticulum; hypospadias cripple; diverticulopenoplasty.

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Accepted for publication: 12 December 2016

Introduction

Severe hypospadias cases are challenging at the best of times, let alone when complications occur that require rescue surgery. Experience is required to get good results from even the simplest of cases, and inventive solutions are needed when skin and glans loss have occurred following previous interventions.

Diverticulum formation is a complication of hypospadias surgery that leads to poor urine stream, bulging of the ventral aspect of the penis during micturition and a predisposition to urine infections [1,2]. The formation of the diverticulum appears to relate to the vectors of urine flow producing laterally directed forces that dilate the urethra, partly due to the congenital lack of supporting spongiosus tissue.

The usual management of the complication of urethral diverticulum formation is to resect the ventral aspect of the expanded urethra and tubularize the remaining tissue to produce a more normal calibre urethra.

In this case, the boy had virtually no skin covering the phallus, with an unusually large diverticulum that was able to be incised in the midline, everted, and then used to cover the penis. The first stage of the rescue procedure produced an outcome similar to a first stage repair of a severe hypospadias with chordee; a second stage consisted of tabularization of the urethra, similar to a Duplay tube urethroplasty, which lead to a successful outcome.

Case report

A boy with a known diagnosis of ring chromosome 13 syndrome was noted to have severe hypospadias as birth, consisting of a perineal meatus, hypoplastic, bifid, empty scrotal folds with no palpable gonads; he also had a microphallus embedded in scrotal folds. A micturition cystourethrogram showed no vesicoureteric reflux, and an ultrasound identified the absence of his right kidney.

Prior to moving to Australia, the boy had undergone laparoscopy and gonadal biopsies, at which no female organs were identified, but he had bilateral inguinal hernias and small testes at the pelvic brim. The testes were mobilized and brought into the scrotum at a later operation.

For his penile anomaly, he then underwent a scrotoplasty, release of chordee, tubularized preputial flap urethroplasty, complicated by a proximal urethral fistula.

A further urethroplasty was performed after which the catheter "fell out" two days later, contributing to the development of a fistula at coronal ring. A further operation was described as correction of penoscrotal transposition with multiple Z plasties, followed by additional operations of another closure of urethral fistula and glanuloplasty, then a urethral meatotomy as the last procedure before migrating to Australia.

On review in Australia, he was noted to have a truncated appearance to his penis with marked ballooning at the time of micturition, for which surgery was recommended. After a sub-coronal incision of the outer layer of the skin and degloving of the penis it was evident that there was very little penile shaft skin, but a very large urethral diverticulum, which was then incised longitudinally. The diverticulum was able to be everted, thus providing cover of the penis, albeit with skin of the diverticulum [Fig. 1 A, B].

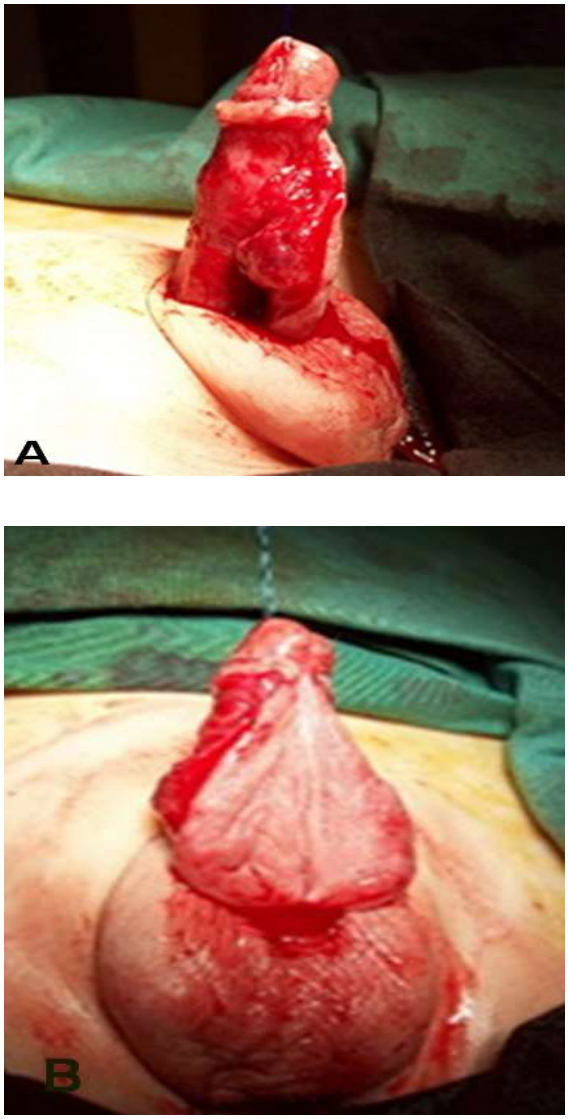

Fig. 1 (A, B). Mobilization of the skin of the penis with degloving. The lack of skin is seen in the lateral view; the size of the splayed diverticulum is evident in the ventral image.

A small amount of skin was also able to be used to place a tube graft through the glans. [Fig. 2]. At a second procedure, there was adequate skin for a Duplay tube urethroplasty [Fig. 3], despite failure of the distal glans graft,

a good functional and cosmetic outcome and no stricture formation, despite a minor infective episode in the early post-operative period [**Fig. 4 A-C**].

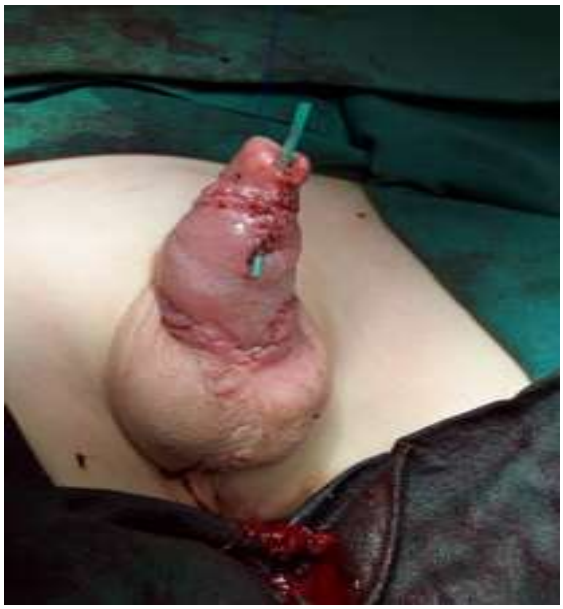

Fig. 2. The penis at the end of the first stage procedure. There is a dorsal longitudinal suture line and the formation of the glans urethra with a free graft.

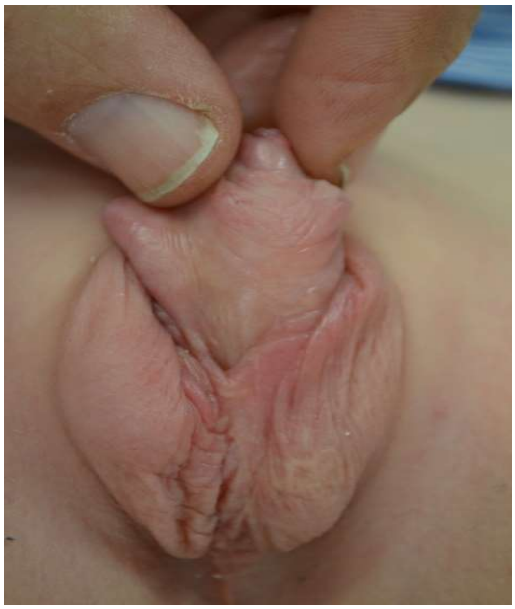

Fig. 3. The appearance of the ventral aspect of the penis after the first of two operations showing the urethral meatus at the penoscrotal junction.

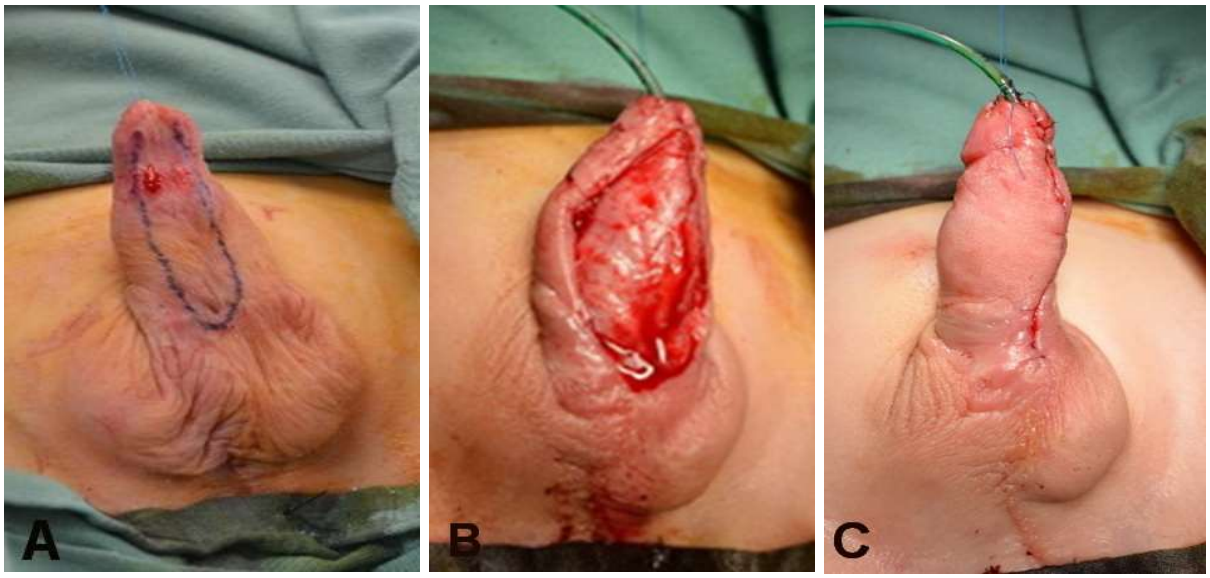

Fig. 4. (A-C). At the second operation, a Duplay tube was marked, mobilized, then closed longitudinally, then the penile skin and glans closed.

Discussion

No two hypospadias cripples are the same, thus the repair should be a combination of concepts and techniques, often with the importation of tissue from another part of the body, including the mouth [3], the bladder [4], or skin from either the penis or other locations [5].

How to include the imported tissue is a further consideration, noting that eversion of a diverticulum has not yet been reported, but is clearly a viable option in a very rare circumstance.

Conclusions

Hypospadias anomalies are many and varied as are the surgical solutions. Likewise, the outcome for major, and minor hypospadias, can have major complications as seen in our case. When a poor outcome does occur, it is important to entertain numerous options for rescue surgery. The option of everting the diverticulum has not previously been reported and, while an unlikely to be applicable option, is obviously an excellent alternative.

Acknowledgements

The author(s) declare that they have no competing interest and financial support.

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