Case Report

180° congenital penile torsion with distal hypospadias mistaken for an epispadias: Optimal outcome with tubularized incised plate urethroplasty and dartos flap rotation

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Introduction: Severe penile torsion of 180° associated with hypospadias is a rare entity. Knowledge of penile anatomy and pathology are necessary as the diagnosis could be missed.

Case presentation: We report a case of severe 180° penile torsion with distal hypospadias that was mistaken for an epispadias which was corrected with surgery.

Conclusion: Tubularized incised plate urethroplasty and dartos flap rotation provided satisfactory result for this association.

Key words: dartos flap, hypospadias, penile torsion.

Keynote message

This report projects the association of a rare association of 180° penile torsion with associated distal hypospadias. Emphasis was placed on physical examination as certain association could be missed leading to mismanagement of the patient. No definitive operative procedure has been randomized for this association. However, we recommend tubularized incised plate urethroplasty and dartos flap rotation for this association as it showed optimal results.

Introduction

Penile torsion is an anomaly of congenital origin where the corpora bodies of the penile shaft is rotated spirally while the proximal attachment remains fixed at the pubis rami.[1] The true incidence of penile torsion with hypospadias is unknown as it is underreported.[1,2] Severe congenital penile torsion is rare[2] and some studies have reported varying degrees of penile torsion 15°–180° with concurrent hypospadias.[1]

The actual etiopathogenesis of penile torsion remains equivocal as few authors have postulated a defect in the dartos fascia[2] and buck's fascia.[3] Studies have elucidated various procedures for penile torsion repair including dorsal dartos wrap rotation,[4,5] diagonal corporal plication sutures,[6] stitch along the pubic periosteum,[7] untwisting plication sutures[8] urethral plate and urethra mobilization[1] and degloving of the penile skin with reattachment.[9]

In 2004, a study by Fisher and Park reported eight pediatric cases of penile torsion corrected by dorsal dartos flap.[4] We report an uncommon case of severe 180° penile torsion with distal hypospadias that was mistaken for an epispadias which was corrected optimally by tubularized incised plate urethroplasty and dartos flap rotation.

Case presentation

A 2-year-old male was referred to our Urology clinic for the management of epispadias. He was reportedly born with an abnormal penis following which he had an abnormal upward urinary stream. He was assessed to have 180° rotation of the penis with a dorsally oriented median penile raphe. The external meatal orifice was aligned dorsally in a sub-coronal plane along with a ventral hood. The testes were palpable in both hemi-scrotal sacs. Ultrasound of

the upper and lower tracts were normal. The patient was counseled for a reconstructive surgery for an 180° penile torsion and distal hypospadias.

He underwent a Snodgrass repair (tubularized incised plate urethroplasty) and dorsal dartos flap rotation to correct the anomaly. The surgery was performed using general anesthesia and endotracheal intubation. A 4.0 polypropylene stay suture was placed along the glans (Fig. 1). A distal sub-coronal hypospadias was seen oriented in a ventral plane due to 180° penile rotation. A circumscribed skin incision was made along the junction of the glans wings to the urethral plate. The penis was degloved down to the penopubic junction (Fig. 2). The urethral plate was found to be flat and healthy, with good vascularization. The urethral plate was incised extending to the underlying corpora. The underlying corpora was normal with no transverse webs along the incision. Based on these findings, tubularization of the incised plate was possible. An 8-Fr stent was placed through the urethra and a tourniquet applied at the base of the penis. The incised urethral plate was tubularized in two sub epithelialized layers using 5-0 polyglactin suture; continuous stitches from distal to proximal, then vice versa for the second layer. A dartos flap was mobilized and rotated anticlockwise onto the penile shaft to correct the torsion and achieve additional coverage of the neourethra. The glans wings were approximated with 6-0 polyglactin and the excess preputial skin excised to complete the circumcision. A Gittes Test showed proper penile alignment. The stent was left *in situ* for 5 days and the postoperative cosmetic outcome was satisfactory.

Discussion

There have been heterogenous reports on the incidence of penile torsion from 1.7% to 27% but severe penile torsion remains a rare entity with incidence 0.7%.[3] The degree of penile torsion with associated hypospadias remarkably varies from 15° to 180° as reported in the literature.[1,5] However, the actual report on 180° penile torsion and associated hypospadias remains unclear. To the best of our knowledge, we present one of the few reports on this association.

The case was mistaken for an epispadias (shown in Fig. 1) and sent for further evaluation. Through thorough clinical examination with anatomical knowledge of the median penile raphe, penile torsion was subsequently ruled in. The distal hypospadias was corrected with a tubularized incised plate urethroplasty (Figs 1,2) as the standard of care from reported data.[1,5] The Snodgrass procedure was preferred to other repairs because of the distal hypospadias having a healthy urethral plate which could be performed in a single procedure with optimal cosmetic outcome. Another advantage of the repair was the vascularized dartos flap was used for both coverage of the neourethra as well as correction of the penile torsion (Fig. 3a). Other repairs for distal hypospadias, like the Mathieus flap technique (urethral plate used as the dorsal wall of the urethra) is less cosmetically appealing compared to the Snodgrass repair because it presents with round meatus rather than slit-like meatus in tubularized incised plate. A few studies[1,4–9] have reported various surgical techniques with success for the management of penile torsion including dorsal dartos wrap rotation, diagonal corporal plication sutures, stitch along the pubic periosteum, untwisting plication suture, urethral plate and urethra mobilization and penile degloving of the penile skin with re-attachment.

Nevertheless, the case in this report was managed successfully with dorsal dartos wrap rotation. It was challenging to determine the direction of rotation as the presentation was an 180° rotation as shown in Figure 1. Therefore, the dartos flap was oriented in a counterclockwise direction which achieved satisfactory correction as displayed in Figure 3. This produces a revolving force that counterpoises that of penile torsion according to Zeid and Soliman.[5]

Conclusion

Severe penile torsion of 180° associated with hypospadias is a rare entity. Knowledge of penile anatomy and pathology are necessary as the diagnosis could be missed. Tubularized incised plate urethroplasty and dartos flap rotation provide satisfactory result for this association.

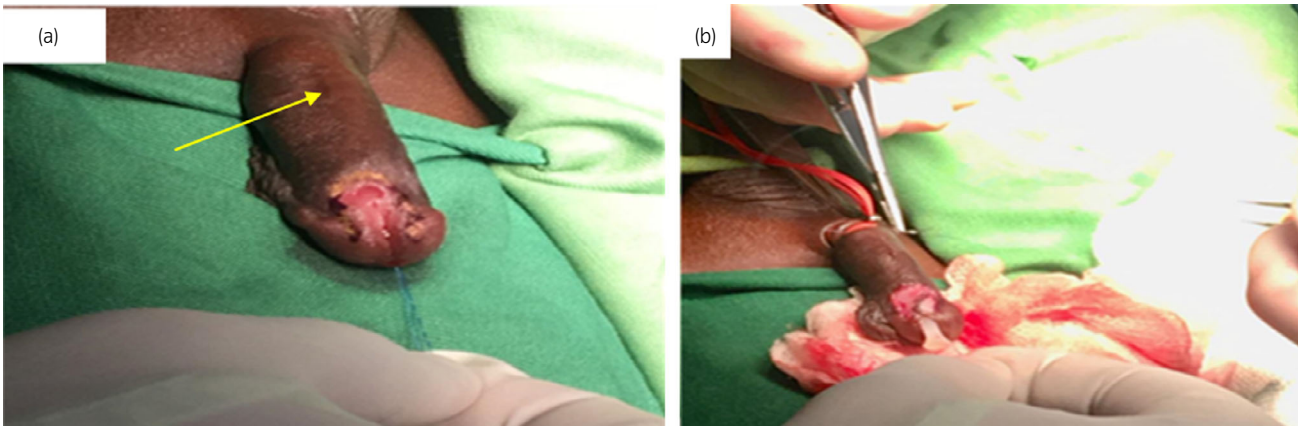

Fig. 1 (a) 180° counterclockwise rotation of the penis with a dorsally oriented median penile raphe (arrow). The external meatal orifice was aligned dorsally in a sub-coronal plane along with a ventral hood. (b) An 8-Fr stent placed through the urethra and tourniquet applied at the base of the penis.

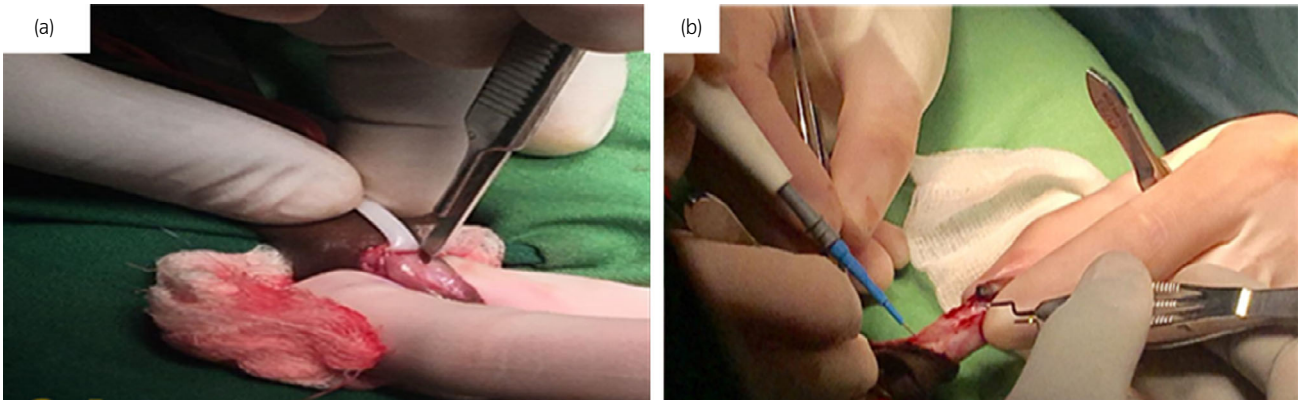

Fig. 2 Tubularized incised plate urethroplasty for distal hypospadias. (a) The glans wing separated, and the urethral plate incised down to the corpora and tubularized in two layers. (b) The penis was degloved down to the penopubic junction.

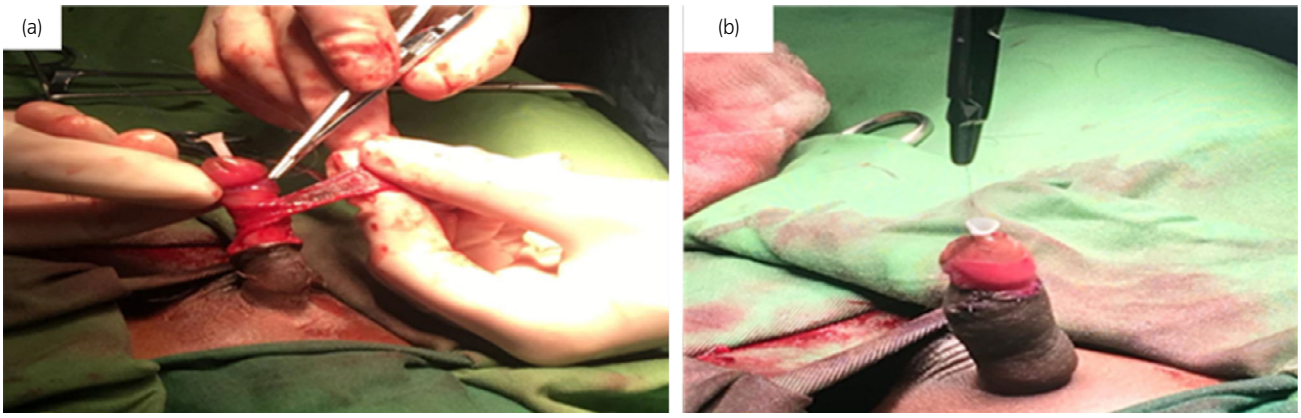

Fig. 3 Dartos flap rotation to correct penile torsion. (a) A dartos flap was mobilized and rotated anticlockwise onto the penile shaft to correct the torsion and achieve additional coverage of the neourethra. (b) Proper penile alignment achieved following correction.

Acknowledgments

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Conflict of interest

The authors declare no conflict of interest.

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