Quadruple Orchidopexy for Torsion Testis in an Adolescent With Polyorchidism: A Case Report

Hany Ibrahim, Matthew J. Roberts, and David Hussey

We report a case of testicular torsion in an adolescent with polyorchidism, specifically quadorchidism. Sonographic and intraoperative images are provided to describe the presentation and management of this case, while the challenges and pitfalls with this clinical scenario are discussed. UROLOGY 87: 196–199, 2016. © 2015 Elsevier Inc.

Polyorchidism represents a congenital disorder resulting in the presence of supernumerary testicle/s, with the most commonly reported variant being triorchidism in asymptomatic patients. Whereas the prevalence of torsion with anatomically normal testes is 0.025%,[1] testicular torsion in the presence of polyorchidism is exceedingly rare. Polyorchidism with 4 testes has been reported for 8 patients to date.[2-4] Here, we report a unique case of testicular torsion in an adolescent with 4 testes that was successfully preserved intraoperatively. This case outlines the challenges faced with diagnosis and management of this condition.

CASE REPORT

A healthy 14-year-old boy presented to the emergency department with a 2-hour history of atraumatic, first-episode left scrotal pain following a football game. He denied any recent or concurrent fever, urinary symptoms, nausea, and family or travel history. His birth history was uncomplicated following conception using in vitro fertilization.

On physical examination, his vital signs were within normal limits and he appeared well, alert, and comfortable with a soft, nontender abdomen without organomegaly or abdominal herniation. External genitalia examination revealed no hypospadias, rash, or other abnormality, as well as no scrotal swelling, erythema, hydrocele, or inguinal lymphadenopathy. On examination of the left hemiscrotum, the patient was uncomfortable because of moderate generalized tenderness; 2 separate, mobile masses were palpable in each hemiscrotum, of which the left superior mass was firm. On repeated examination, the initially superior firm mass was palpated in the inferior hemiscrotum, indicating unrestricted movement. Leukocytosis (13.9×10^9/L) and neutrophilia (11.24×10^9/L) were demonstrated in otherwise normal serum investigations. A urinalysis was not performed.

Scrotal ultrasonography demonstrated polyorchidism, specifically 2 testes located within each hemiscrotum (Fig. 1A). Vascularity was absent and reduced in the upper and the lower left testes, respectively (Fig. 1B,C).

Following informed consent and general anesthesia, scrotal exploration including complete opening of the tunica vaginalis was performed. Intraoperatively, the sonographic findings were confirmed to be a result of clockwise 720° torsion of the upper left testis (Fig. 2A), which appeared ischemic. The testis was connected to a smaller inferior testis and ductus deferens by a long epididymis, with duplicated vascular supply observed (Fig. 2B). This anatomical arrangement was present bilaterally (Fig. 2B,C). Following detorsion and warming, the left upper testis was deemed viable. Three-point orchidopexy was performed with a vertically oriented (superior-, mid-, and inferior pole) approach using an absorbable braided suture (3.0 Vicryl) on all 4 testes, which were fixed to the tunica vaginalis in close approximation nearest to the most natural configuration.

The patient experienced an uncomplicated recovery, and outpatient abdominal and renal tract ultrasound did not demonstrate further congenital or other abnormality.

COMMENT

The etiology of polyorchidism may be from aberrant division of the genital ridge in early gonadogenesis before 8 weeks' gestation (Fig. 3).[2,5] Anatomical and functional classification models have been proposed based on the presence of testicular drainage by ductus deferens and arrangement of the epididymis.[2,6] Anatomically, this case illustrates a bilateral type A3 (Bergholz) or type II (Leung) polyorchidism (Fig. 2D). Based on the Leung

Financial Disclosure: *The authors declare that they have no relevant financial interests.*

From the Department of General Surgery, The Prince Charles Hospital, Brisbane, Australia; the Department of Urology, The Royal Brisbane and Women's Hospital, Brisbane, Australia; the School of Medicine, The University of Queensland, Herston, Australia; and the Centre for Clinical Research, The University of Queensland, Herston, Australia

Address correspondence to: Matthew J. Roberts, M.B.B.S., B.Sc., Department of Urology, The Royal Brisbane and Women's Hospital, Brisbane 4006, Australia. E-mail: m.roberts2@uq.edu.au

Submitted: August 2, 2015, accepted (with revisions): September 10, 2015

http://dx.doi.org/10.1016/j.urology.2015.09.011
0090-4295

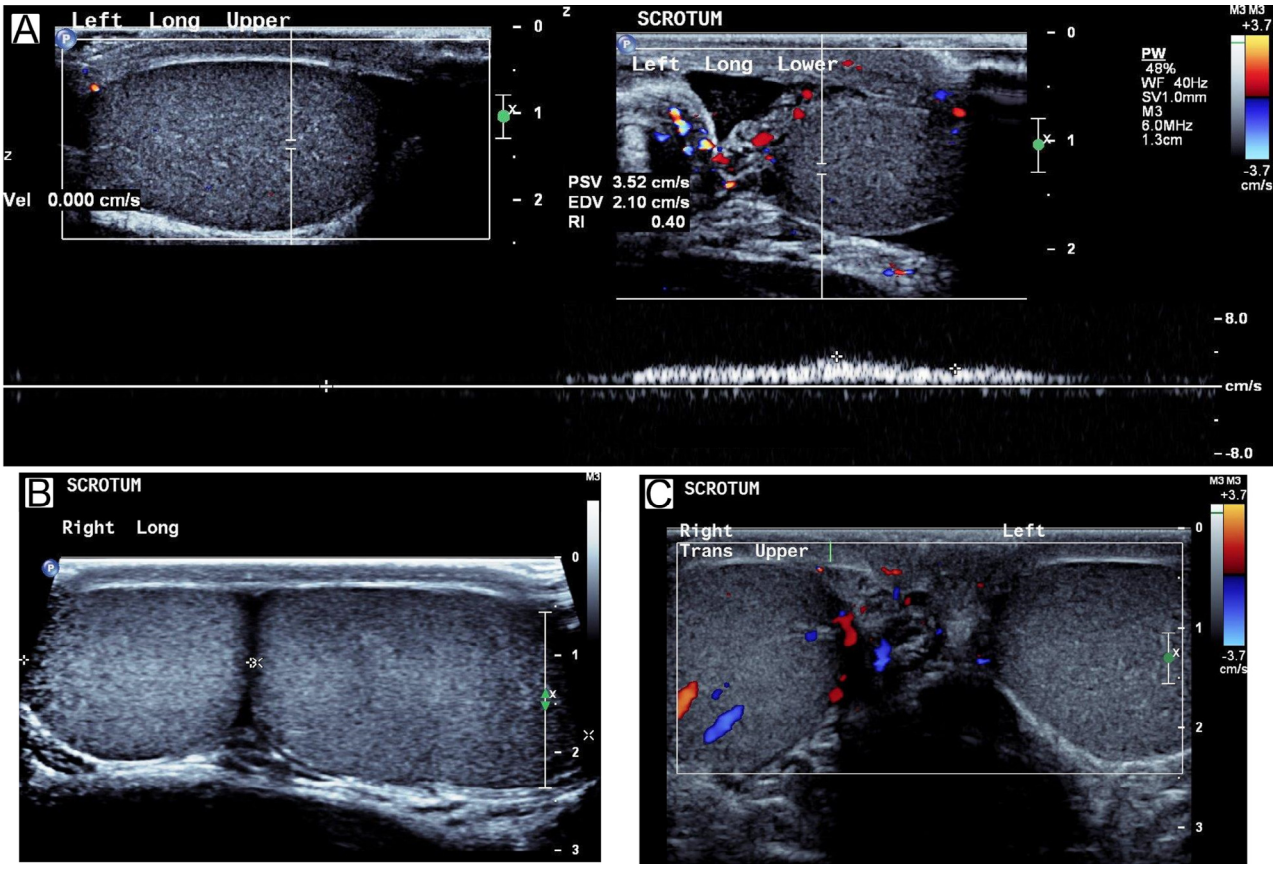

Figure 1. Scrotal ultrasonography; **(A)** left hemiscrotum (longitudinal section) with corresponding Doppler measurement, demonstrating absent blood flow in upper left testis, and diminished vascularity in left lower testis; **(B)** right hemiscrotum (longitudinal section) demonstrating 2 testes; **(C)** transverse section across upper scrotum showing right and left upper testes, demonstrating normal and reduced/absent blood flow, respectively. (Color version available online.)

classification, it is hypothesized that this case is the result of bilateral genital ridge division at the primordial gonads without affecting the mesonephros and metanephric ducts.[6,7] Functionally, this case represents a Singer type 1A, being that the intrascrotal polyorchid testes are drained by a vasoepididymis and have reproductive potential.[6]

Although polyorchidism is rare, more than 200 cases have been reported to date. According to a meta-analysis of 191 cases in 2009,[2] triorchidism (95%) and left-sided anomalies (65%) occurred most commonly, with malignancy detected in 5% of histologically confirmed cases, predominantly located in a maldescended position. Other associated anomalies include inguinal hernia (30%) and maldescended testis (22%), whereas other congenital anomalies (hypospadias, varicocele, and anomalous urogenital union) were uncommon. Testicular torsion occurred in 13% of cases, which considering the small population size, is considerably higher than general incidence. The "bell clapper deformity," where the tunica vaginalis attaches higher than usual, is central to the etiology of testicular torsion. This developmental defect may be more pronounced in polyorchidism with a greater testicular volume transmitted with the peritoneum through the inguinal canal. This developmental aberration intuitively predisposes these patients to a higher incidence of testicular torsion.[7] This was a feature of our case, with regular positional change of the testes between serial examinations. Furthermore, conception via in vitro fertilization and later development of polyorchidism in this case may be associated, although evidence for this is scarce.

Management of asymptomatic uncomplicated polyorchidism usually comprises a conservative approach with regular ultrasonography and follow-up.[3] As the reproductive potential of a supernumerary testis is estimated to be 50%-65%,[8] testicular preservation is encouraged.[7] However, with concurrent undescended testes (inguinal or intra-abdominal; Singer type B), excision of the affected testis should be considered because of high risk of malignant transformation. When scrotal exploration is performed, orchidopexy is encouraged to minimize risk of future testicular torsion. In our case, orchidopexy of 4 testicles was challenging owing to uncertainty of important anatomical landmarks, such as the sinus of the epididymis and available surface area of the tunica vaginalis. We experienced that allowing the testes to return to their resting position provided the most appropriate location and were accordingly fixated to the tunica vaginalis. Reconstructive vasoepididymal approximation using a nonabsorbable suture has