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Urethral Duplication associated with Epispadias in a Child

O B Shittu*, T B Kamara**

Summary

Shittu OB, Kamara TB. Urethral Duplication associated with Epispadias in a Child. *Nigerian Journal of Paediatrics* **1999; 26: 53.** A case of duplication of the urethra associated with epispadias in a seven-year old boy is reported. This to our knowledge, is the first case to be reported in a Nigerian child. The child presented with urinary incontinence and an abnormally shaped penis due to severe chordee. The epispadiac urethra was excised and the chordee corrected to give a normal looking penis. The boy also became continent of urine.

Introduction

URETHRAL duplication is a rare and intriguing congenital abnormality. Symptoms could be mild and manifest merely as double urinary streams, or may be severe and result in uraemia due to urinary outflow tract obstruction by one of the urethrae. We present a case of duplication of the urethra associated with an epispadias that was recently managed in our unit and which to our knowledge, is the first of its kind to be described in a Nigerian child.

Case Report

A seven-year old primary four pupil was referred on account of urinary incontinence and an abnormal penis. He weighed 25.5 kg and was 1.32m tall. There was peno-pubic epispadias and severe chordee, with the penile shaft lying parallel to the anterior abdominal wall. He leaked urine intermittently from the epispadias opening, but there was also a meatal opening in the normal location on the glans penis.

Ultrasound examination revealed that the kidneys were normal in position, outline and size and the urinary bladder was within normal limits. Radiograph of the pelvis showed symphysial diasthesis measuring 25mm (normal <10mm). The intravenous urogram (IVU) was essentially normal. Surgical exploration revealed a ventral urethra as well as an epispadic urethra both of which communicated with a single bladder, but the epispadic urethra did not go through the external sphincter muscle. The epispadic urethra was excised through a subpubic and retropubic approach and the proximal stump was ligated at the bladder neck. The chordee was also corrected and a preputial flap was used to cover the skin defect on the dorsal surface of the penis. The groove on the dorsum of the glans penis, distal to the epispadic meatus was excised and the splayed glans was approximated to achieve as normal an appearance as possible. The post-operative course was normal, and the boy now has a straight penis, and has also become continent of urine.

Histological examination of the excised specimen showed an abnormal urethra lined by transitional epithelium with focal areas of squamous metaplasia.

Discussion

Urethral duplication is a very rare congenital abnormality. The few references to the condition in the English literature have been case reports,[1-4] while a popular textbook of paediatric surgery in the tropics did not even mention the condition.[5] Several classifications for urethral duplication have been

University College Hospital, Ibadan

Department of Surgery
* Senior Lecturer
** Senior Registrar

Correspondence. O B Shittu

suggested, but the most useful classification for the management of the condition is a modification of that of Effmann *et al*,[4] who recognized five types namely: Types I, IIA, IIB, IIC and III. In Type I, there is a blind incomplete urethral duplication in addition to a patent urethra; such cases are usually asymptomatic. In Type IIA, there are two non-communicating urethras arising independently from the bladder with two patent meatal openings (Fig 1A). In Type IIB (Fig 1B), there is a second channel arising from the first and coursing independently to a second meatus. In Type IIC, there are two urethrae arising from the bladder or posterior urethra and uniting to form a common distal channel (Fig 1C). In Type III, there is urethral duplication as a component of partial or complete caudal duplication in which the bladder, the penis and structures derived from the embryologic hindgut are also duplicated.

Clinical symptoms will vary from one patient to the other depending on the type of duplication.

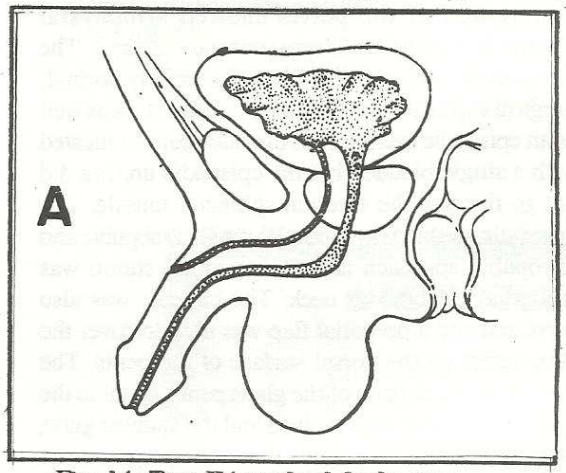

Fig. 1A: Type IIA urethral duplication

Patients with complete patent urethral duplication can be asymptomatic, or can present with a double urinary stream,[6] urinary incontinence as in our case, urinary tract infection or outflow obstructive symptoms.[7] Physical examination may reveal two meatal openings along with some other abnormalities such as epispadias, hypospadias, ambiguous genitalia, and chordee. However, not all these abnormalities may be found in an individual case. Investigation of a patient with suspected double urethra should include a retrograde or voiding cystourethrogram to outline the urethra. In addition, an ultrasound of the kidneys and an IVU are required to exclude upper tract abnormalities,[8-10] and where available, a urethroscopy should be done to fully assess the calibre of the urethra. An abnormally wide symphysis pubis (more than one cm) as seen in our patient is sometimes found as in patients with isolated epispadias.[4]

Complete excision of the duplicate urethra, as done in our case, is the most definitive procedure

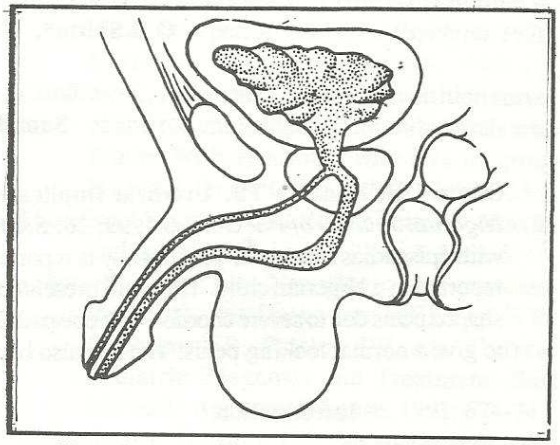

Fig. 1B: Type IIB urethral duplication

and should be attempted when feasible. This could be done by a penile or a combined retropubic and penile approach,[11] as was performed in the present case. Intraurethral fenestration and transurethral excision of the intraurethral septum are other surgical options that have been described.[12] However, in operating on patients with significant symptoms or abnormalities, it is important to know that the ventral channel, regardless of its absolute position usually prove to be more functionally normal. It is where the vedrumontanum resides and the genital ducts empty, and is also subject to the standard congenital anomalies that can affect a single urethra, such as posterior urethral valves.[13] Failure to remember this

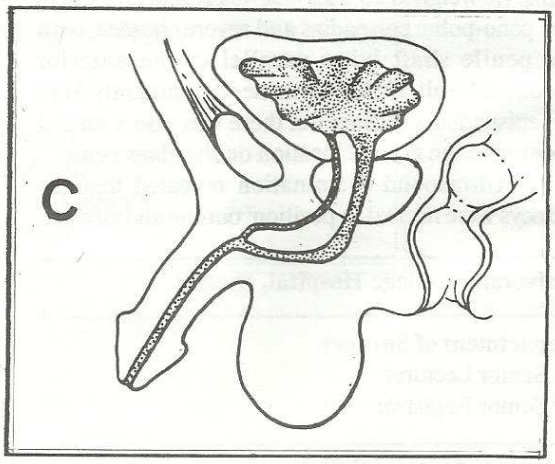

Fig. 1C: Type IIC urethral duplication

fact, with excision of the ventral channel may result in urinary retention or persistent incontinence.

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