Case Report

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Familial Occurrence of Congenital Stricture of Bulbar Urethra

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Key Words

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Abstract

We report on the familial occurrence of congenital bulbar urethral stricture in 2 brothers. Embryological mechanisms are briefly discussed and the literature reviewed.

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Introduction

Strictures of the bulbar urethra in male children are not uncommon and in most cases are correlated with previous surgery, instrumentation, trauma, or infection [1–4]. Congenital strictures of the bulbar urethra have been described in the literature in a few cases [5–10] but, to our knowledge, familial occurrence has been reported previously only in 1 case [7].

We report on 2 brothers with a short stricture of the bulbar urethra without any apparent cause.

Case Report

Case 1. A 21-year-old man was admitted with dysuria, straining and reduced urinary stream of 9 years' duration. There was no history of urinary tract infection, surgery, instrumentation or trauma of the urethra. The cystourethrogram demonstrated a severe narrowing of the bulbar urethra (fig. 1a). At cystourethroscopy a diaphragmatic stricture was seen in the juxtamembranous urethra and internal urethrotomy was performed. The postoperative course was uneventful and the patient voids without complaints at 9 months follow-up.

Case 2. His 19-year-old brother has been complaining of dysuria and diminished urinary stream for 5 years, without a history of inflammation, trauma or instrumentation on the urethra. The cys-

Fig. 1. Preoperative cystourethrograms demonstrating bulbar urethral stricture in 2 brothers, a Case 1. b Case 2.

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tourethrogram showed an identical urethral stricture (fig. 1b). Cys-tourethroscopy demonstrated a diaphragmatic stricture in the juxta-membranous urethra that was treated by internal urethrotomy. The patient was well without recurrence 9 months postoperatively.

Discussion

Congenital stricture of the posterior urethra is quite rare and, in the literature, has been reported in a few cases [5–10]. The embryological explanation generally accepted is that suggested by Cobb et al. [9]: in the male, the urethra forms by joining the cloaca (from which originates the membranous urethra) with the bulbar urethra (which arises from the genital folds). The stricture could result from incomplete rupture of the cloacal membrane at the point of junction between membranous and bulbar urethra and could correspond to submeatal stenosis in girls as described by Lyon and Smith [11]. Duckett and Snow [12] consider that many of the congenital strictures are similar to the congenital urethral membranes or type III Young urethral valves. Harzman et al. [13] reported the association of bulbar congenital stenosis with type I Young urethral valves in 6 children. Congenital stenosis can also result from urethral atresia which usually occurs in conjunction with either Y- or H-type urethral duplication [14–16].

As far as we know there is a single report on the familial occurrence of congenital bulbar urethral stenosis [7]. Familial occurrence in our cases is also stressed by the father of these 2 brothers, who at 25 years of age had undergone surgical repair for urethral bulbar stricture. The man was carefully asked for trauma, infection or instrumentation that could explain the stricture, but he denied any of these events. Internal urethrotomy seems to be the treatment of choice in diaphragmatic congenital urethral strictures [17] and results are generally good.

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