Case Report

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Congenital Bulbar Urethral Stricture Occurring in Two Brothers

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Key Words

Congenital urethral stricture

Abstract

Two cases of apparently congenital bulbar urethral stricture occurring in brothers are presented. Such a finding has been rarely reported. Congenital strictures may be more common than previously thought and a full history is essential in such cases when the aetiology remains uncertain. Biopsy of the stricture may be of value.

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Most urethral strictures arise from iatrogenic, inflammatory or traumatic causes. It has been suggested that in those patients where no aetiological factors have been identified the stricture may be congenital in origin.

Case Report

A 30-year-old fast-jet navigator presented with a long history of worsening urinary stream, frequency and terminal dribbling. There was no previous medical history and in particular no history of trauma, previous surgery or sexually transmitted disease. A midstream specimen of urine was sterile, a peak flow rate was measured at 9 ml/s and ultrasound urography was normal apart from demonstrating a significant residual urine after micturition.

Urethrography showed a short tight bulbar urethral stricture (fig. 1). Urethroscopy confirmed this stricture and a biopsy was taken. The bladder was trabeculated but otherwise normal. An optical urethrotomy was performed and a silastic urethral catheter left indwelling for 4 days. On removal a flow rate was measured at 31 ml/s. Follow-up at 3 months showed no deterioration in urinary stream and at 9 months a further peak flow rate was 28 ml/s. He declared himself asymptomatic. Biopsy of the urethral stricture showed a normal urethral mucosa with no evidence of fibrosis or inflammatory reaction in the submucosal tissue.

Fig. 1. Ascending urethrogram showing tight bulbar stricture.

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His 35-year-old brother had complained of testicular pain, backache and dysuria with occasional urinary tract infections since the age of 18. Intravenous urography was normal. He presented to another hospital with a further urinary tract infection in 1984 and at that time gave no history of trauma, previous surgery or venereal disease. Urethroscopy demonstrated a tight bulbar urethral stricture. This was treated by optical urethrotomy. Follow up at 2 and 6 months showed no recurrence of the stricture and, as the patient declared himself asymptomatic, he was discharged.

Discussion

Congenital urethral stricture is not a commonly diagnosed condition. It is normally assumed to be of an inflammatory or long forgotten traumatic aetiology.

The presence of a similar stricture in 2 brothers who gave no previous history despite specific questioning makes a congenital origin highly likely. In addition, the absence of fibrous or inflammatory changes at the site of the stricture supports this view.

Such strictures have been described previously in a father and son [1] and in cases of urethral duplication when the normally placed urethra may be narrowed from the site of the duplication distally [2]. A review of urethral strictures in male children suggested that congenital strictures may be located at single or multiple positions and are probably due to embryologic narrowing of the channel or atresia [3]. A series of 26 children with urethral stricture showed the most common presenting symptoms to be enuresis, recurrent urinary tract infections, abdominal pain, behavioural problems and hae-maturia [4]. Of these 26 cases, 10 had severe associated congenital abnormalities, 3 had hypospadias and 4 had urethral valves in addition to the stricture. It is suggested that as the proximal urethra, formed of endodermal tissues, and the distal urethra, formed by ectodermal infolding of the ventral surface of the penis, join up at the bulbomembranous junction, a congenital stricture represents incomplete rupture of the cloacal membrane. This may be analogous to distal urethral stenosis in the female. In support of this suggestion, a urethral membrane causing obstruction has been demonstrated at the junction of the bulbar and membranous urethra in a series of 6 cases [5].

Bulbar urethral stricture in 2 brothers has been previously described [6], and it was suggested that congenital strictures were either juxtamembranous or associated with atresia and urethrocutaneous fistula. In addition, a family of a father and 2 sons has been described with bulbar urethral strictures of unknown aetiology [7].

It is clear from these reports and that of the brothers described in this case report, that strictures often present when the aetiology is unknown. In the absence of any possible factors, a careful family history must be taken. If biopsy of the stricture and histological examination show a normal urethral mucosa with no evidence of fibrosis or inflammation, then the likely diagnosis is that of congenital stricture.

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