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Correspondence

Case report: congenital urethrocutaneous fistula

A 2-year-old boy presented with a suspected urethrocutaneous fistula. He had been born at 26 wk gestation and spent 14 wk in the special care baby unit with severe respiratory problems and an episode of meningitis. He had not had a urinary catheter in situ or urinary tract infections. Bilateral inguinal hernias had been repaired. At presentation he was below the 3rd centile for height and weight but had reached all other developmental milestones.

On examination he had a 2 mm skin defect on the ventral aspect of the penis in the midline at the midshaft. Distal to this the skin was normal and he had a normal prepuce. He voided urine from a normal meatus. Both testes were present in the scrotum. Probing under anaesthetic at the age of 3 y confirmed the presence of a midpenile fistula (Fig. 1). A normal meatus was found at the tip of the glans. The urethral tube from and including the fistula to the glans appeared to consist of mucosa only, without a corpus spongiosum. Proximal to the fistula the corpus spongiosum was apparently deficient as far as the penoscrotal junction. An artificial erection test (Fig. 2) showed marked ventral curvature of the penile shaft, and ventral angulation of the glans on the corpora cavernosa.

The distal penile urethra was excised to correct the chordee and remove the mucosa-only urethra. This disection confirmed the absence of corpus spongiosum throughout the length of the penile urethra. The distal urethra was reconstructed with a neo-urethra fashioned from a split preputial flap (Harris, 1984). Four months later the patient was passing urine comfortably from the meatus and there was no recurrence of the fistula.

Up to 1979 only 13 cases of congenital urethrocutaneous fistula had been reported; 12 of these were reviewed by Welch (1979) along with a case of his own. He indicated that many of these had been subcoronal in location and that none had associated chordee. Our case more closely resembles that reported by Goldstein (1975) in that both were midshaft and had associated chordee. Ours is the first reported case in a boy born prematurely.

It is generally accepted that this lesion is a variant of hypospadias but the exact mechanism is not completely clear. The penile part of the male urethra is made up of 2 parts. The proximal (major) part is formed from a forward growth of a cord of cells from the cloaca on the ventral surface of the elongating genital tubercle. This cord is termed the urethral plate. Urethral plate cells mingle with genital tubercle cells. A gutter forms on its under surface, the edges of which tubulate to produce the urethra. Associated genital tubercle cells form the remaining penile structures. (It is postulated that if the growth of the plate is arrested, the genital tubercle tissue continues its forward growth causing ventral penile curvature.) This process occurs proximally to distally, being complete by about the 12th wk. During the 3rd mo an ingrowth of epithelial tissue from the glans forms the most distal portion of the penile urethra. Fusion of these 2 segments is the final part of the process (Johnson, 1920; Glenister, 1958).

In explaining the group of subcoronal fistulae without associated chordee, Welch surmised that the growth of the proximal urethra had been arrested (as in normal hypospadias) and compensatory overgrowth of the glandular urethra had occurred with failure of fusion at the site of the fistula. The cases reported by ourselves and Goldstein demonstrated chordee and deficient subcutaneous ventral coverings distal to the fistula. The fistula was too far proximal for the above to provide a satisfactory explanation.

In cases of congenitally short urethra with associated chordee but no hypospadias, it was suggested by Goldstein that a deficient distal urethral plate may form the urethra but be incapable of inducing the corpus spongiosum or penile fascia to close ventrally over this urethra. The case we report may be a variant of this: either that the urethral plate

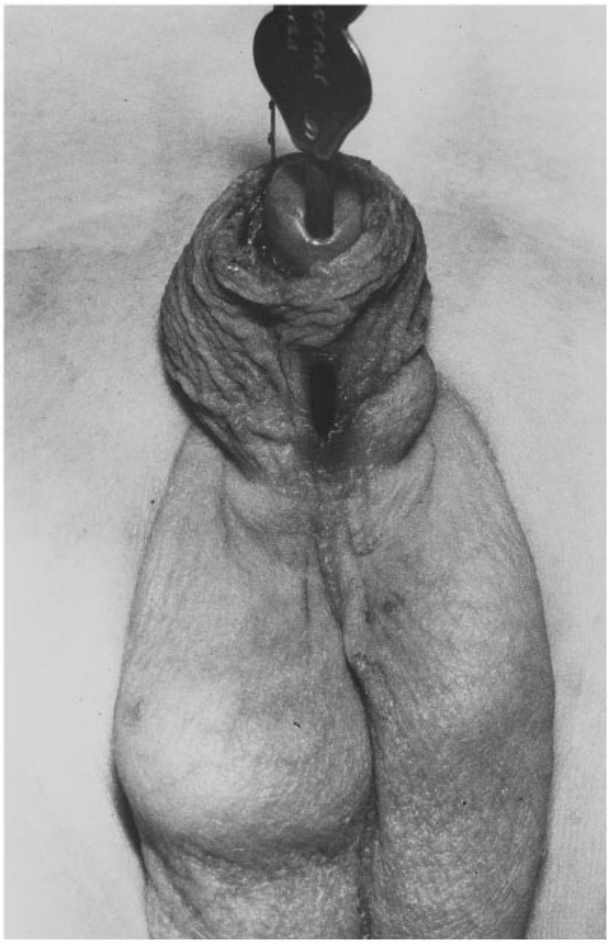

Fig. 1. A probe demonstrates normal glandular meatus and preputial skin distal to the midpenile fistula.

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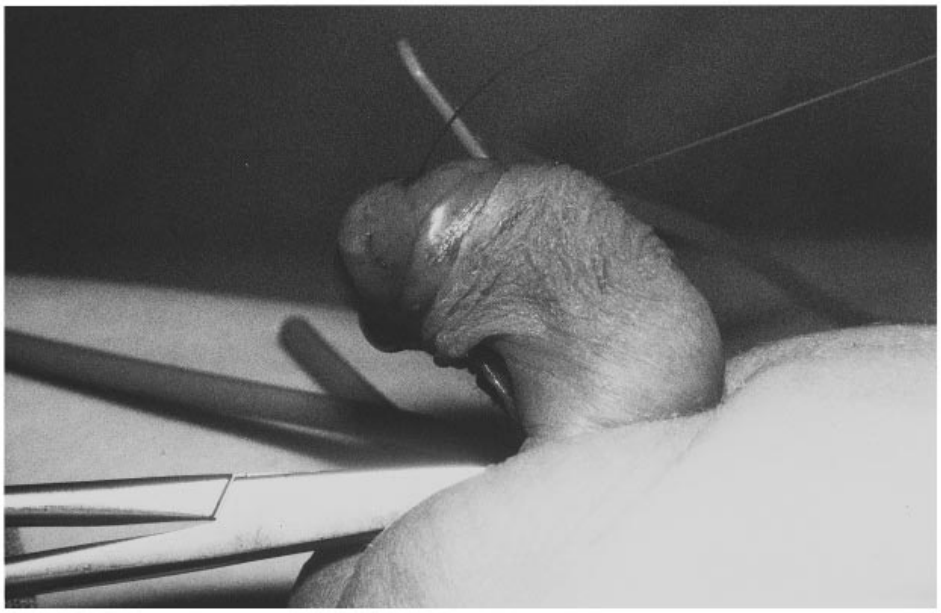

Fig. 2. An artificial erection test demonstrates ventral chordee.

was even more deficient at one segment, leading to an isolated failure to complete the closure of the urethra (and therefore fistula formation), or that minor local trauma or infection caused fistula formation between the skin and a poorly covered urethral tube beneath.

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