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Double trouble: A rare case of posterior urethral valve and Cobb's collar

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ABSTRACT

Posterior urethral valves (PUV) are the most common cause of bladder outlet obstruction (BOO) in boys. Cobb's collar, a rare narrowing of the bulbar urethra, is one of the lesser-known causes of congenital urethral obstruction. We present a case of both congenital anomalies occurring concomitantly, in a preterm baby presenting with urinary ascites.

Introduction

Cobb's collar (CC) is a membranous stricture of the bulbar urethra. In contrast to posterior urethral valves (PUV), this stricture lies distal to the external sphincter and is distinct from the verumontanum. Although the precise prevalence is unknown, CC is a type of anterior urethral valve (AUV), which are suspected to be 7–8 times less common than PUV. The association of posterior and anterior urethral valves is uncommon, but recognised. We report on a case of concomitant PUV and CC. To the best of our knowledge, this has only been documented once before, in a 10 year old boy where delay in diagnosis resulted in chronic renal failure. This is the first case report of a preterm baby with coexistent PUV and CC.

Case report

We present a baby boy, born at 35 weeks gestation in good condition. His antenatal scans exclusively reported a large abdominal girth. He had normal passage of meconium, but on examination was found to have abdominal distension with a poor urinary flow and a creatinine that was moderately raised at 45 μmol/l. Abdominal X-ray and ultrasound displayed extensive abdominopelvic ascites. Among the battery of tests that were undertaken to determine the cause of fetal ascites, echocardiography showed a small patent foramen ovale and patent ductus arteriosus; upper gastrointestinal contrast ruled out malrotation; and an ascitic tap, of golden yellow aspirate eliminated chylous ascites. With a possible diagnosis of urinary ascites, a catheter was inserted. Micturating cystourethrogram (MCUG) showed a posterior bladder diverticulum and a dilated posterior urethra extending down to the bulbar urethra (Fig. 1). With a working diagnosis of PUV we performed a cystourethroscopy, during which we were surprised to find a circumferential membranous narrowing at the bulbar level that was subsequently successfully fulgurated (Fig. 2). In addition to this, a PUV was noted and successfully fulgurated at 5, 7 and 12 o'clock (Fig. 3). The following day, the baby was discharged home. Repeat cystourethroscopy at two-month follow-up did not show any residual PUV or obstructing membranous narrowing.

Discussion

CC is the eponymous name given to a bulbomembranous obstruction of the anterior urethra. Cobb et al. (1968) were the first to describe a bulbar urethral narrowing; using retrograde urethrography, calibration bougienage and cystourethroscopy they observed a bulbar abnormality in twenty-six boys.[1]

The anterior urethra starts at the membranous region, continues to encompass the bulbar and penile urethra, and terminates at the fossa navicularis. The most common anterior urethral valves (AUV) are those in the bulbar urethra (40%), followed by penoscrotal (30%) and penile (30%). Although rare, strictures of the fossa navicularis have been documented.

PUV is the most common cause of bladder outlet obstruction (BOO) in boys, and the most common obstructive cause of end stage renal disease in children. The association of PUV and CC is rare: we found only one case report, in which the CC was not recognised during fulguration of a PUV resulting in chronic renal failure.

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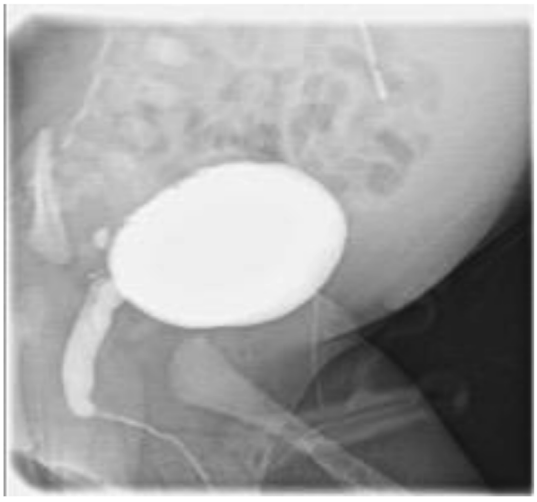

Fig. 1. MCUG showing a posterior bladder diverticulum and dilated posterior urethra extending down to bulbar urethra.

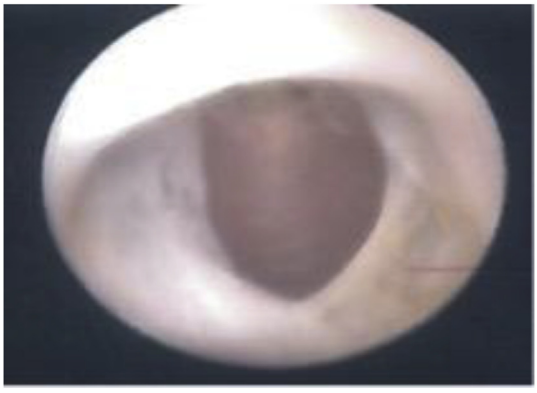

Fig. 2. Cystoscopic photograph of circumferential Cobb's collar prior to fulguration.

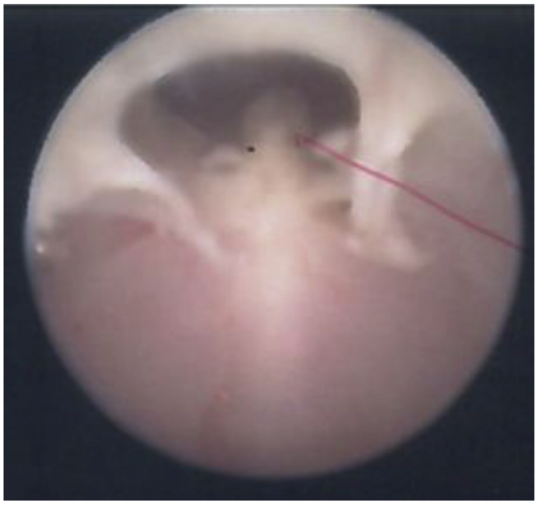

Fig. 3. Cystoscopic photograph of PUV post fulguration.

Both PUV and CC are congenital anomalies, however, they are conjectured to occur through different embryological aberrations. Posterior valves result from abnormal anterior placement of the mesonephric ducts at the urogenital sinus, which prevents duct migration leaving behind obstructing urethral ridges.[2] CC occurs distal to this, and is thought by some to result from a narrowing at the urogenital ostium - the embryonic junction site of the pelvic to phallic urethra. Others have suggested an incomplete dissolution of the urogenital membrane as the cause of narrowing.[3]

With modern surgical diagnostic techniques, Young's three-part PUV classification has been simplified and updated under a single unifying term: congenital obstructive posterior urethral membrane (COPUM). COPUM encompasses any obstructing posterior membrane with paramedian folds emanating from the crista urethralis. This should assuage the conflation of type-3 PUV and CC; the later being defined as a proximal bulbar stricture, distinct from the crista urethralis.

In contrast to PUV, where most boys present in the neonatal period, the presentation of CC is bimodal with peaks in infancy and the second decade of life. CC is classified as mild, moderate and severe based on degree of urethral encroachment, which provides some explanation for presentation in adulthood.

One would expect the prominent clinical features of a double urethral narrowing, such as documented in this report, to originate from the most proximal obstruction. PUV classically presents either antenatally with oligohydramnios and respiratory distress secondary to pulmonary hypoplasia, or postnatally with BOO. The presenting features of CC include poor urinary stream, urinary ascites, urinary retention, urinary tract infection and enuresis. Urinary ascites, as found in our case, can be a feature of both PUV and CC – where a “pop-off” mechanism relieves upper tract pressure.

Over the past fifty years, since its description, CC has been investigated in a variety of ways. Sugimoto et al. found uroflowmetry to be ineffectual in detecting CC and recommended a MCUG.[4] Similarly, on suspecting infravesical obstruction at our institution, we place a catheter and undertake an MCUG. In our case, this showed a small posterior bladder diverticulum, trabeculations, and a uniform upstream urethral dilatation starting at the anterior urethra. A stricture was assumed at the point of dilation. Our later discovery of a second stricture appears to indicate that the upstream dilation caused by the CC disguised the posterior obstruction on MCUG.

With suspicion of an infravesical obstruction, diagnosis is confirmed by cystourethroscopy, where transurethral ablation of the obstructing membranes can be undertaken. Improved outcomes have been reported with a cold knife technique; in comparison to an electrode knife, where an increased incidence of submucosal burning can lead to urethral stricture.[5]

Conclusions

CC is an uncommon congenital narrowing of the bulbar urethra. Although rare, it can occur concomitantly with PUV. Missing this double pathology can result in serious long-term consequences. Therefore, when making a diagnosis of infravesical obstruction, an anterior urethral stricture should be considered. We recommend a MCUG as the best initial diagnostic tool, followed by cystourethroscopy and transurethral ablation, to confirm and treat the condition.

Disclosures

The authors have no conflicts of interest to declare.

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Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.eucr.2019.100848.

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