

Cobb's collar occurring in two brothers in a family: A rare entity revisited

Partha Pal, Sayantan Ray, Arunansu Talukdar¹, Nikhil Sonthalia¹, Sumit Chakraborty²

Departments of Medicine, Calcutta National Medical College and Hospital, ¹Medical College and Hospital, ²Department of Radiology, Institute of Post Graduate Medical Education and Research and Seth Sukhlal Karnani Memorial Hospital, Kolkata, West Bengal, India

Correspondence: Dr. Sayantan Ray, Department of Medicine, Calcutta National Medical College, 24, Gorachand Road, Kolkata - 700 014, West Bengal, India. E-mail: sayantan.ray30@gmail.com

Abstract

Most urethral strictures arise from iatrogenic, traumatic, or inflammatory causes. We report the familial occurrence of a congenital bulbar urethral stricture in two brothers. Retrograde and voiding cystourethrography was performed. A Cobb's collar was diagnosed after radiological and endoscopic evaluation in both cases and was successfully managed with urethroplasty. Cobb's collar is a rarely recognized cause of a membranous stricture of bulbar urethra that can lead to several urinary problems. In cases of adolescent and young adults presenting with symptoms of progressive urinary obstruction and enuresis with or without urinary tract infection, Cobb's collar can be seen as a minor constriction in the bulbar urethra, but is not frequently symptomatic, and the familial occurrence of such a stricture is even rarer.

Key words: Bulbar urethra; Cobb's collar; congenital strictures; cystourethrography; urethroplasty

Introduction

Cobb's collar^[1] or Moormann's ring^[2] is a largely underestimated cause of male bulbar urethral strictures. It can potentially lead to acute urinary retention, upper urinary tract dilatations, enuresis, infection, poor streaming, and hematuria. Embryologically, this focal bulbar urethral stricture develops due to failure of canalization of the cloacal membrane during fetal development.^[3] In Cobb's collar, the site of obstruction is more distal than that seen with posterior urethral valves (PUV).^[4] A prompt diagnosis with minimally invasive treatment may avoid upper and lower tract deterioration in these patients. We present two cases of congenital bulbar urethral obstruction (Cobb's collar) occurring in brothers. The anatomy, embryology, and clinical features are discussed in order to turn the spotlight on to this rare disease.

Case Reports

Case 1

A 30-year-old man presented with a long history of a worsening urinary stream and a sense of incomplete voiding and straining at micturition. He had two episodes of near retention before admission. There was no history of traumatic injury, instrumentation of the urethra, or sexually transmitted disease. Abdominal and genitourinary examination revealed an enlarged and distended urinary bladder with normal-looking external genitalia.

Renal function tests and urine routine examination were within normal limits, and urine culture showed no growth. USG of the abdomen revealed bilateral mild hydronephrosis [Figure 1a], presence of hydroureters, a dilated prostatic urethra, and a thick-walled over-distended bladder. Pre-void (1972 ml), first (957 ml) and second post-void (790 ml) residual volumes were significantly raised [Figure 1b]. Urodynamic study could not be performed as catheterization failed due to the tightness of the urethral stricture. Subsequent retrograde urethrogram (RGU) showed a 3-cm stricture in the bulbar urethra [Figure 2a]. Micturating cystourethrogram (MCU) using the suprapubic route showed a narrowing in the proximal bulbar urethra, without reflux into the collecting

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system [Figure 2b]. Endoscopic evaluation revealed a bulbar-obstructing membrane. The urinary bladder wall was mildly trabeculated. A Cobb's collar was diagnosed. Suprapubic catheterization (SPC) with visual internal urethrotomy (VIU) was performed and the patient was discharged. Later, he was readmitted for a voiding trial and Teflon dilatation. In view of the persistent urinary problems, the patient was planned for buccal mucosal onlay urethroplasty. The postoperative course was uneventful, and the patient voids without complaints at 6 months follow-up.

Case 2

The first case's 25-year-old younger brother also complained of poor urinary stream and straining at micturition. He had no history of urethral trauma, instrumentation, or infection. Physical examination of the abdomen and genitalia was unremarkable. Renal function tests and urine routine examination were within normal limits. USG showed mild hydronephrotic changes, hydroureters, and a thickened bladder wall, with significant post-voiding residue. RGU was performed which showed a short segment stricture in the bulbar urethra [Figure 3]. A Cobb's collar was diagnosed and successfully managed with VIU. The patient is asymptomatic at 6 months follow-up, with normal serial USG evaluations.

Discussion

The proximal prostatic and membranous part of the male urethra is formed by endodermal tissues and the penile part is formed from the phallic portion of the urogenital sinus. Congenital idiopathic urethral stricture may result from incomplete rupture of the cloacal membrane at the junction of membranous and bulbar urethra which may correspond to submeatal stenosis in girls.^[3]

Cobb *et al.* (1968) were the first to draw attention to the entity of congenital stricture of the bulbar urethra.^[1] They analyzed 52 cases of proximal bulbar urethral obstruction in male patients for a 3-year period. Twenty-six out of 52 patients were boys under 16 years of age; of these 26 boys, 16 had a trabeculated bladder, 15 showed a dilated prostatic fossa, whereas 6 had a secondary hydroureter, and the most frequently referred symptoms were enuresis, urinary tract infection, hematuria, and failure to thrive. Three different types of Cobb's collar have been identified. All these forms of obstruction are located just below the external sphincter^[5] [Figure 4].

Such strictures of the bulbar urethra had been underestimated for many years until the first half of the 1990s when Dewan *et al.*^[6] and later Nonomura *et al.*^[7] redefined and classified the bulbar urethral narrowing by reviewing video-recorded cystoscopies as well as radiological pictures in the patients. Both studies showed that the majority of patients were below 1 year of age with a large number

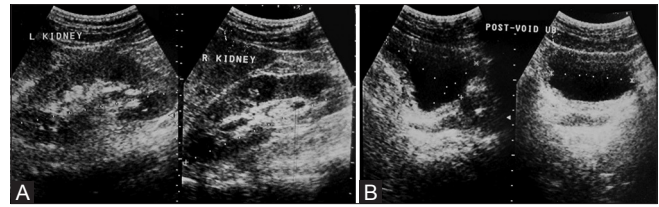


Figure 1 (A, B): (A) Bilateral mild hydronephrosis (B) significant post-void urine residue (Case 1)

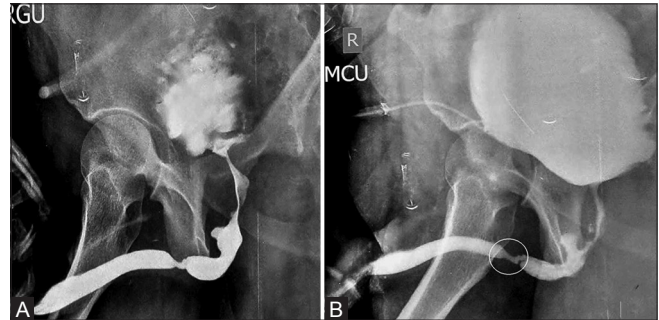


Figure 2 (A, B): (A) Retrograde urethrogram demonstrating a narrowing in the proximal bulbar urethra, (B) micturating cystourethrogram (MCU) showed a narrowing in the proximal bulbar urethra (white ring) (Case 1)



Figure 3: Retrograde urethrogram showing bulbar urethral stricture (Case 2)

showing vesicoureteral reflux (VUR), with no history of urethral trauma, catheterization, or instrumentation. VUR in association with clinical findings and endoscopic findings of vestigial remnants at the level of bulbar urethra indicates that this narrowing has a congenital onset. Presentation at a later age, as occurred in our cases, may represent delayed presentation of a congenital stricture. Their response to treatment is clearly different from that of the under 1 year olds, since majority require buccal mucosal onlay urethroplasty or scrotal or penile island flap urethroplasty. The more common causes of congenital

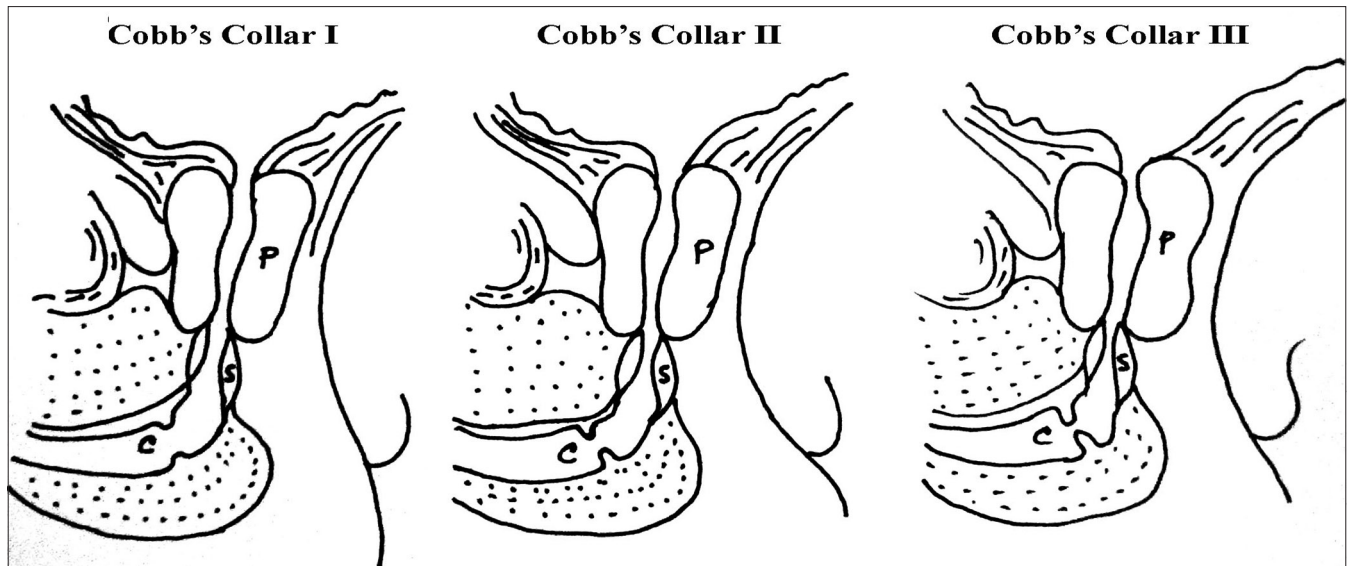


Figure 4: Sagittal section through prostate and urethra showing the area of Cobb's collar. It can appear either as a ridge of tissue (Cobb's collar I) or as a definite stricture in the area described (Cobb's collar II) or even as a very tight pinhole opening (Cobb's collar III). P, prostate; S, external sphincter; C, Cobb's collar

urethral stricture are different types of posterior urethral obstruction, which was originally described as PUV (three types) by Young *et al.*, but these obstructions were found to be diaphragmatic obstruction as suggested by Parkkulainen on endoscopic studies.^[8] Congenital attachment of the verumontanum to the anterior wall of the posterior urethra is the embryologic forerunner to the congenital obstruction of the posterior urethra. Congenital obstructing posterior urethral membrane (COPUM), first described by Dewan *et al.*, is a new terminology of the same. More distal bulbar urethral abnormality without folds running up to the verumontanum is likely to be a remnant of urogenital diaphragm, commonly known as Cobb's collar.^[9]

Banks *et al.*,^[10] in their series of 12 pediatric patients with bulbar or posterior urethral stenosis, identified a sort of bimodal distribution of age at presentation, with 50% of patients presenting during the first year of life, whereas 5 of the remaining 6 patients presented after the age of 11. This late presentation may represent a delayed presentation of a congenital stricture, or may be due to a forgotten urethral trauma or asymptomatic inflammation. Clinical presentation may be variable and, often, may be related to the use of blind urethral catheterization in these patients. Chronic urinary retention and bilateral hydronephrosis may be a presenting feature in some patients, as in our case.^[10,11] These adult patients may also complain of poor ejaculation.^[5] Late presentation of Cobb's collar (caused by a persistence of urogenital membrane) can be differentiated from acquired stricture by the absence of the characteristic folds between COPUM anomalies and verumontanum.^[12]

Pediatric urologists continue to debate whether the Cobb's collar or Moormann's ring represents a true urethral

stricture or not. Some consider Cobb's collar to be a benign physiological narrowing of the bulbar urethra, easily passable with a cystoscope, although Cobb describes a series of bulbar strictures in a pediatric population in his original paper. So, although rare, Cobb's collar has to be considered in the differential diagnosis of progressive urinary flow obstruction and enuresis, especially in pediatric and adolescent patients where acquired traumatic, iatrogenic, or inflammatory strictures are more common. Radiologically indistinguishable dysfunctional voiding due to involuntary contraction of urethral sphincter or pelvic floor muscles and Hinman syndrome (functional non-neurogenic bladder) are also important differential diagnoses to be considered. Contrast-enhanced conventional radiological studies can identify this anomaly and also be helpful in postoperative follow-up of this entity which can be easily treatable by minimally invasive endoscopic corrective surgery.^[12] Late-onset cases may need urethroplasty. Familial occurrence of Cobb's collar is exceedingly rare and only few cases are reported in the literature, indicating that this entity may have a familial basis.^[13] Screening for family members is an important part of managing these patients. In the absence of traditional risk factors, careful family history should be taken. If the biopsy of the stricture and histological examination shows a normal urethral mucosa and the absence of any inflammation or fibrosis, then Cobb's collar becomes the likely diagnosis.

Conclusion

From the limited data available and from the cases reported here, it is clear that urethral strictures often present with an unknown etiology. Although extremely uncommon, in cases of adolescent or pediatric patients presenting with

symptoms of progressive urinary obstruction and enuresis with or without urinary tract infection and/or hematuria, a bulbar urethral obstruction such as Cobb's collar should be considered.

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