Incomplete bladder duplication with multiple congenital anomalies: A rare presentation

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Abstract

We report a rare case of incomplete bladder duplication associated with multiple congenital malformation. The patient presented with a symptomatic left pelvi-ureteric junction obstruction which was surgically managed. To the best of our knowledge, this type of bladder duplication with such syndromic association is not yet reported in literature.

Key Words: Bladder duplication, scoliosis, septate uterus

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INTRODUCTION

Bladder duplication is an extremely rare condition. Incomplete duplication of the bladder is an unusual congenital anomaly and is usually associated with other congenital anomalies. [1] It is found more commonly in males than in females. [2] We present a case of incomplete bladder duplication in a female with associated septate uterus, uterine fibroid, anorectal malformation, with unilateral (left) pelvi-ureteric junction obstruction, and poorly functioning kidney. To the best of our knowledge, this type of bladder duplication with such syndromic association is not yet reported in literature.

CASE REPORT

A 20-year-old female presented with the complaint of left-sided flank pain for last 6 months with occasional stress urinary incontinence and primary infertility. On physical

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examination, a patulous urethral opening was seen and anal opening was separated from the vagina by a thin septum. A single urethral meatus was observed. Ultrasound showed grossly hydronephrotic left kidney [Figure I] with uterine fibroid [Figure 2]. Intravenous urography suggested septate bladder with scoliosis [Figure 3]. Magnetic resonance imaging showed septate bladder [Figure 4] and uterine septation [Figure 5]. Diehylene Triamine Penta Acetic acid (DTPA) renogram showed poorly functioning left kidney with type 2 curve. Left transperitoneal laparoscopic pyeloplasty was done with double-J stenting. Postoperative recovery was uneventful and the stent was removed after 4 weeks. At 6 months follow-up, patient is doing well without any flank pain and with improved left renal function on follow-up DTPA diuretic renogram. Patient was advised for treatment of septate bladder and septate uterus, but she was unwilling to undergo any further investigations or interventions.

DISCUSSION

Bladder duplication may be complete or incomplete. Depending on the axis of the septum, it can occur at the sagittal or coronal plane^[3] Abrahamson classified the various bladder duplication anomalies and found complete duplication in the sagittal plane to be more common^[3] compared to the coronal one, in a ratio of 2.5:1.^[4] Females are more likely to present the sagittal type

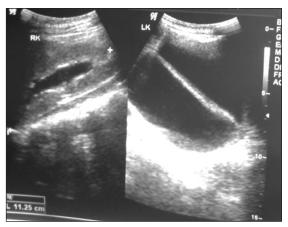


Figure 1: Ultrasound Kidney Ureter Bladderl (KUB) showing left hydronephrosis



Figure 3: IVU showing incomplete bladder duplication with scoliosis

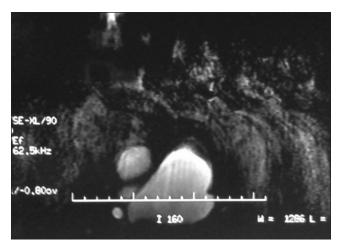


Figure 5: MRI showing septate uterus

of duplication.^[3] In incomplete bladder duplication, two bladders communicate with each other and drain into a common urethra. To determine the anatomic situation, a complete preoperative diagnostic evaluation with karyotype, ultrasound, Intra Venous



Figure 2: Ultrasound showing fibroid uterus

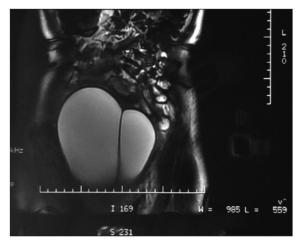


Figure 4: MRI showing incomplete bladder duplication

Urography (IVU), video-urodynamic studies, genitogram, and gastrointestinal tract imaging is helpful.^[3]

Kossow and Morales reviewed 40 cases of complete bladder duplication and 90% of cases showed associated duplication anomalies of the external genitalia. Associated duplication anomalies of the lower gastrointestinal tract have been reported in up to 42% of cases and spina bifida, meningocele, or myelomengocele has been reported in 15% of cases. Berrocal *et al.* also showed associated duplication anomalies of the external genitalia, gastrointestinal malformations, duplications of the spine, spina bifida conditions, and various fistula formations between the urogenital and gastrointestinal tract. [6]

Due to the variety of presentations with other associated anomalies and rarity of the disease, the treatment cannot be standardized and should be individualized. Renal preservation and prevention of infections by relieving obstructed genitourinary tracts should be the aim of initial treatment. Further treatment should be reconstruction of the internal and external genitalia with an aim to achieve continence. Incomplete duplications may not require surgical procedures if both bladder halves are sufficiently drained by a common urethra.^[3]

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