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Case Report

Bilateral duplicated renal collecting systems with lipomeningomyelocoele

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ABSTRACT

Closed neural tube defects are known to be associated with multiple syndromic anomalies. These defects add to the patient's morbidity and have a deleterious effect on their life. After a thorough review of literature, we could not find and reported cases of bilateral duplicated renal collecting systems in association with a closed neural tube defect. We are reporting this case to bring to light its rare nature and promote discussion related to its further management.

Keywords: Duplex system, Neural tube defect, Neurogenic bladder, Spina bifida, Vesico ureteric reflux

INTRODUCTION

Closed neural tube defects have been described with a myriad of syndromes namely cutaneous syndrome, orthopaedic syndrome, urological syndromes (incontinence of urine, abnormal voiding pattern, recurrent urinary tract infections) or neurological syndrome (weakness, numbness of feet or atrophy).[1,2] Associated upper urinary tract anomalies are less commonly described with such defects.[3] We are reporting a previously undocumented case of bilateral duplicated collecting systems with a closed neural tube defect (Lipomeningomyelocoele).

CASE REPORT

A 4-year-old female child presented with the complaints of progressively increasing swelling over the right gluteal region and dribbling of urine since birth. There was no history of discharge from the swelling or associated difficulty in walking or running. The child had sensation of bladder filling, used to start urinating with a dribble followed by some stream mid urination and end with dribbling. There was no history of incontinence while crying, defecation, on standing or running. On examination a soft 8×10 cm swelling was noted over the sacral aspect mainly towards the right side with a 4×2×1 cm appendage attached to it. The swelling had a broad base and fatty consistency with no transillumination. The anal opening was ruggous and pulled towards the right and seemed to share its wall with the swelling. Bilateral lower limbs tone and power were normal.

Ultrasound revealed bilateral hydroureteronephrosis with echogenic debris in urinary bladder and significant post void residual urine. Voiding cystourogram showed overdistended bladder with right sided grade V vesicoureteral reflux into two ureters. This was further evaluated with a Tc99 labelled Di-Mercapto-Succinic Acid scan which showed a differential renal function of 40 % with upper and lower pole scarring on the left side and 60% function with mid and lower pole scarring on the right.

Magnetic resonance urography revealed incomplete duplication of the right collecting system with fusion of the two ureters just before the vesico-ureteric junction along with incomplete duplication of the left collecting system with fusion of upper and lower pole moieties to form a single ureter which is dilated throughout its course. Magnetic resonance imaging of the spine showed posterior defect in the lumbosacral region with a

lipomatous cystic lesion with herniation of dural sac extending towards the right gluteal lesion.

A diagnosis of bilateral incomplete duplicated collecting system with sacral lipomeningomyelocoele was made and she was planned for lipomeningomyelocoele excision and repair along with diagnostic cystoscopy. Cystoscopy revealed a trabeculated neurogenic bladder with a turbid medium and a few diverticuli. The left ureter was grossly normal but the right ureter seemed to be more laterally placed away from the Mercier bar. Its opening was patulous. After excision of said swelling, she recovered well and is on regular follow up.

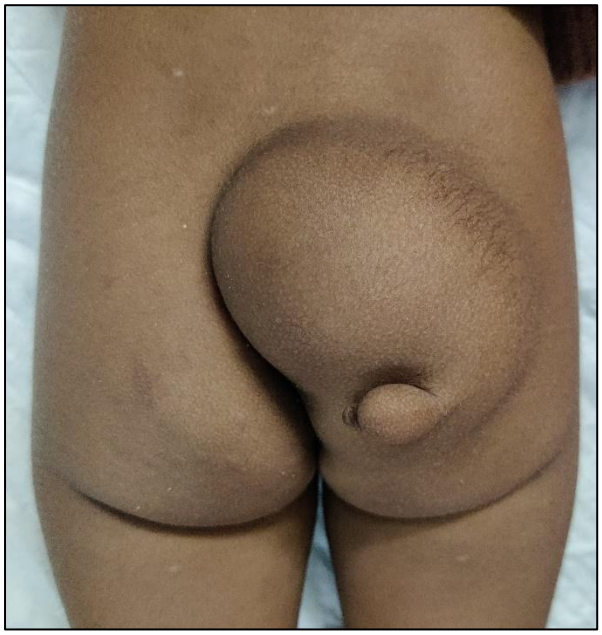

Figure 1: Clinical picture of the sacral-gluteal swelling.

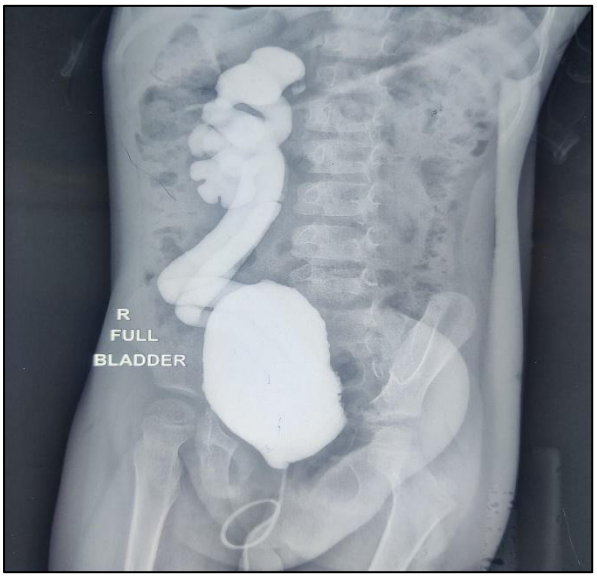

Figure 2: Voiding Cystourogram with vesico-ureteric reflux in both ureters on right side.

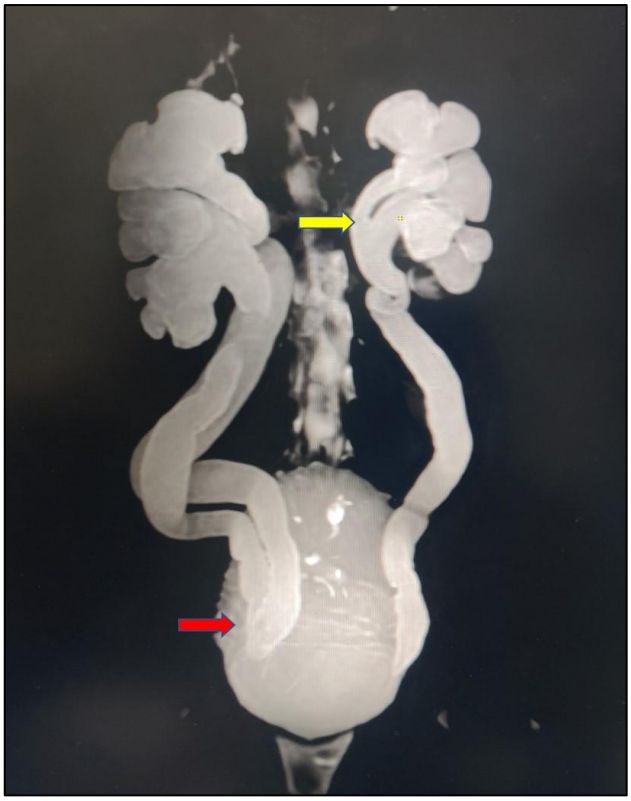

Figure 3: Magnetic resonance urogram showing bilateral duplicated collecting systems with confluence of ureters just proximal to bladder on the right side (red arrow) and of pelvis at the level of mid pole of left kidney (yellow arrow).

DISCUSSION

The occurrence of duplicated collecting renal system along with closed neural tube defects have not been documented in the available English literature. Closed defects have been mostly reported with cutaneous stigmata or neurological weakness symptoms. Congenital or acquired hydrocephalus are also less frequently reported.[4] The presentation with muscular, gait or Orthopaedics deformity is well documented with a defined classification system.[5]

After going through multiple search headings and combinations thereof on Google Scholar and PubMed we were able to find few documents which have tried to correlate the level of sensory deficit in meningomyelocele in specific upper urinary tract anomalies.

This association could be plausible as both systems develop at the same time between 4th to 5th week 6. With the closure of the posterior neuropore on the 28th day and ureteric bud induced metanephric blastema differentiation on 30th to 32nd day, any insult to the embryo during this time may hamper the development of both the neural tube and the metanephros.[7] Open neural tube defects with

sensory deficit at S1 level were seen most frequently involved with ureteric duplication anomalies.[8]

The current treatment recommendations for most variants of closed neural tube defects advocate a waiting period of 6-9 months of age before intervention.[9] As this patient presented to us at 4 years of age, the neurogenic bladder had already set in. The management of the Lipomyelomeningocele and detethering of cord at this age may only prevent further deterioration of bladder dynamics. The ideal plan of care should have included a detailed evaluation of associated anomalies at birth and early detethering of cord.[10] This may have led to the growth and development of a bladder more amenable for ureteric reimplantation.[11]

Due to the paucity of literature on this topic and pre-existing state of bladder at first surgery, the further plan of management will be decided after interdepartmental meeting, further literature review and counselling sessions with parent to discuss the impact of neurogenic bladder, need for surgery and expected benefits/risks of reimplantation.

CONCLUSION

A previously undocumented finding of bilateral duplicated collecting system with an occult spinal dysraphism is reported here to highlight the occurrence of such association and to bring to call to attention the role of early intervention (at birth) in select cases for closed neural tube defects in certain populations.

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