

Ruptured Prostatic Utricle Cyst – An Unusual Cause of Isolated Neonatal Urinary Ascites

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1. Abstract

1.1. Background: Urinary ascites is often associated with either rupture of calyceal fornixes, ureter or urinary bladder secondary to distal renal tract obstruction. Prostatic utricle cyst is usually asymptomatic, rarely presents in neonatal period as neonatal ascites.

1.2. Case Description: Term male infant, with antenatal presentation of cystic structure posterior to the urinary bladder, was born with gross neonatal ascites. Postnatal workup confirms diagnosis of perforated prostatic utricle cyst as the cause of isolated urinary ascites. Child was managed with serial abdominal paracentesis and temporary vesicostomy followed by surgical removal of cyst at later date.

1.3. Discussion: Fetal and neonatal ascites is caused by diverse etiologies and requires a broad-based antenatal and postnatal approach in determining its underlying cause. Neonatal urinary ascites occurs commonly due to obstructive uropathies with perforation of prostatic utricle cyst a rare presentation. Peritoneal fluid examination and radiological investigations like ultrasound, MRI and cystoscopy can help to confirm the exact cause of urinary ascites. Basic aim of management of neonatal urinary ascites includes abdominal decompression, urinary drainage with or without vesicostomy or surgical repair of perforation site. Prognosis depends on the age, amount of ascites, degree of compression and extent of changes in urinary tract.

2. Manuscript

2.1. Introduction

Isolated fetal and neonatal ascites is a rare condition of heteroge-

neous origin with urinary ascites been an unusual cause [1]. Dating back to 1952, there are evidence suggesting associations of urinary ascites either due to distal urinary tract obstruction leading to perforation of renal tract or due to severe reflux of urine through urinary tract into the peritoneal cavity [2]. Prostatic utricle cyst is rarely symptomatic in fetal or neonatal period and there are no case reports of its association with isolated fetal ascites. We present a rare, previously unreported case of ruptured prostatic utricle cyst presenting with fetal and neonatal gross ascites requiring peritoneal drainage.

2.2. Case Description

Term male infant with birth weight of 3610 gram was born via planned caesarean delivery to a 33-year old primigravida mother with an uneventful antenatal history. Serial antenatal scans detected a small, left pelvic kidney with a cystic structure posterior to the urinary bladder. The cystic structure which measured 3X3mm at 16 weeks antenatal scan increased in size to 17X21X27mm by 34 weeks of gestation with no evidence of fetal ascites. There was no sonographic evidence of oligohydramnios or polyhydramnios. Non-invasive pregnancy test was positive for Klinefelter syndrome (47, XXY), which was confirmed on an amniocentesis karyotype. At 37 weeks of gestation, just prior to delivery, bedside US scan was unable to locate cystic structure and showed possible fetal ascites.

At birth, baby presented with distended, shiny, tense abdomen with marked ascites. He required non-invasive mechanical support in view of respiratory distress due to splinting effect of gross ascites. On physical examination, child did not appear dysmorphic and

there were no signs of generalized edema, jaundice or pallor. His cardiovascular examination was unremarkable with normal echocardiogram findings. There was no clinical or laboratory evidence of blood group incompatibility and workup for congenital infection including serology for toxoplasmosis, rubella, cytomegalovirus, herpes and parvovirus infection were negative.

A pigtail peritoneal drainage catheter was inserted by pediatric surgeon and he required frequent drainage (up to 120ml/kg/day) of the ascitic fluid, which was replaced with intravenous albumin, fresh frozen plasma or normal saline. Child was also catheterized with size 8fr urinary catheter for strict input/output charting, to prevent urinary bladder distension from pressure effect due to gross ascites and due to initial clinical and biochemical evidence of acute kidney injury. There was gradual improvement in ascites with reduction of abdominal girth and urinary and peritoneal catheters were removed on day 21 of life.

Peritoneal fluid analysis was suggestive of transudate, with low

levels of protein, lactate dehydrogenase, triglycerides and white blood cells. Fluid cytology was negative for any malignant cells and triglycerides levels were persistently low despite child on enteral breast milk feeds. Peritoneal fluid was negative for acid-fast bacilli smear, fungal smear, gram stain, and bacterial culture. Raised levels of ascitic fluid creatinine (176umol/L) compared to plasma creatinine (108umol/L) confirmed urinary ascites.

Abdominal CT scan on day 1 of life showed gross ascites without any evidence of cystic structure or urinary tract obstruction. Later, postnatal US scan done after resolution of ascites showed anechoic (3X2.4X2cm) structure posterior to bladder whereas Micturating Cysto Urethrogram (MCU) confirmed abnormal configuration of urinary bladder with a distended posterior urethra and no vesico-ureteric reflux. MRI scan revealed large (3X3X2.2cm) midline cystic structure located postero-inferior to urinary bladder communicating with prostatic urethra, representing an utricle cyst (Figure.1).

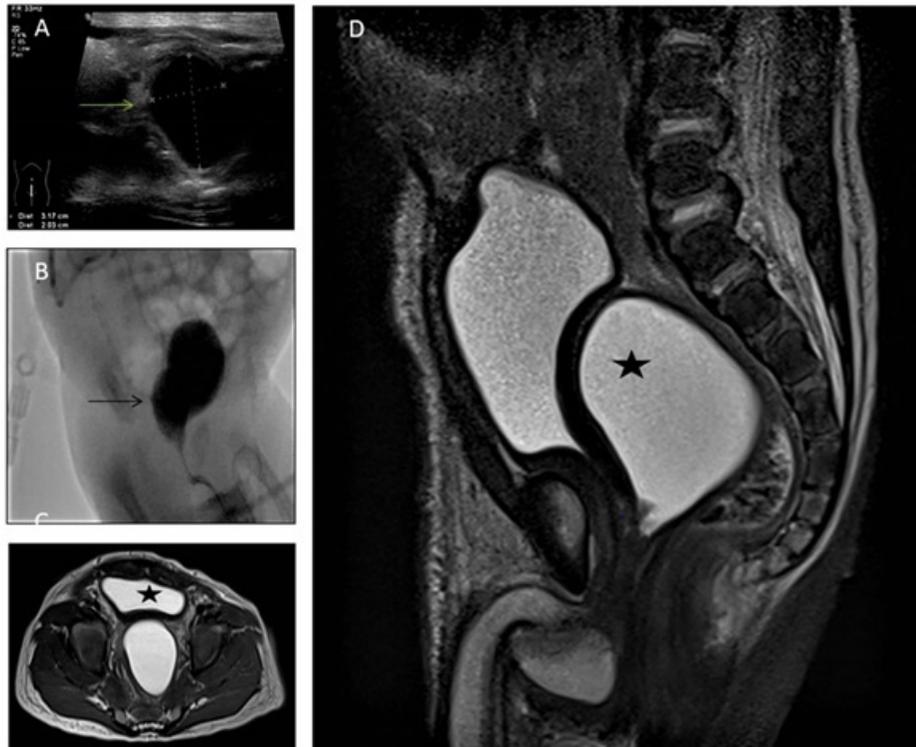


Fig:1 A) US scan showing 3X2.4X2cm anechoic structure posterior to bladder (white arrow),

B) MCU showing abnormal configuration of urinary bladder with a distended posterior urethra (black arrow),

C & D) MRI showing large midline cystic structure located postero-inferior to urinary bladder communicating with prostatic urethra, representing a utricle cyst (star)

US – Ultrasound; MRI – Magnetic Resonant Imaging; MCU – Micturating Cystourethrogram

Operative cystoscopy performed in 4th week of life showed a floppy utricle cyst likely causing partial bladder outlet obstruction at posterior urethra with a possible leak site at the posterior cyst wall. Intra-operative cystourethrogram showed distension of cyst with contrast with no identifiable leak. Child underwent vesicostomy with stent left in situ and he was discharged on day 36 of life with

oral urophylaxis antibiotics.

On follow up, child was growing age appropriately and had an episode of urinary tract infection at 7 months of age which was treated with intravenous antibiotics. At 1 year of age, he underwent elective cystoscopy, open removal of utricle cyst and closure of vesicostomy uneventfully.

2.3. Discussion

Fetal ascites commonly occurs associated with or as a precursor of fetal hydrops fetalis. However, it can also occur in isolation when there is fluid collection in abdominal cavity without accumulation in serosal cavities or subcutaneous tissue [1]. Isolated fetal ascites is rare entity with unknown incidence and thought to be occurring as a result of imbalance between capillary and interstitial hydrostatic and oncotic pressure [4]. It is a condition with heterogeneous etiology which can be caused by maternal, fetal or placental abnormalities. Fetal cardiac, genitourinary, hepatobiliary, gastrointestinal malformations, metabolic storage disorders, blood isoimmunisation, congenital intrauterine infections and chromosomal disorders are common causes of fetal ascites [2].

Neonatal urinary ascites is a rare presentation occurring commonly due to either obstructive uropathies or neurogenic bladder [3]. There is a male preponderance with male to female incidence ratio of 7:1. Urinary ascites is unusual and can occur due to disruption of integrity of urinary tract from buildup of intrarenal pressure leading to rupture of calyceal fornixes, ureter or urinary bladder often associated with distal obstruction [5]. Posterior Urethral Valve (PUV) is the most common cause of urinary ascites occurring in about 70% of cases. Severe vesico-ureteric reflux (VUR) can also cause ascites due to reflux of urine through urinary tract into the peritoneal cavity. Traumatic or non-traumatic rupture of urinary bladder is another example causing urinary ascites. Neonatal urinary ascites is actually a protective urinary tract decompressing measure preventing renal function deterioration and secondary changes to bladder from raised intrarenal pressure [6].

Prostatic utricle cyst is a dilated cystic cavity seen posterior to the prostatic urethra. Prostatic utricle cyst, an embryonic remnant of Mullerian duct is usually asymptomatic in majority of cases but may present as urinary tract infection, urinary incontinence, stone formation, dysuria or constipation [7]. Its incidence is reported at about 10-15% in male infants with hypospadias, cryptorchidism or intersex abnormalities. It occurs due to delay in the male hormonal production leading to incomplete regression of the Mullerian ducts [8]. To date there have been no reports in literature suggestive of its association with fetal or neonatal ascites following rupture.

On antenatal evidence of fetal ascites, mother should be systematically investigated for prenatal workup including blood group incompatibility with indirect Coombs test, intrauterine infections, serial ultrasound scans, fetal echocardiography and amniocentesis. Once the neonate is born, extensive investigative approach to establish an etiology of ascites should be carried out. Simultaneous analysis of ascitic fluid, serum and urine urea and creatinine leads to diagnosis of ascites to be of urinary origin. Like in our case, in urinary ascites, the serum values of urea and creatinine will be lower than peritoneal fluid values suggesting presence of urine in peritoneal cavity. Besides routine diagnostic workup with perito-

neal fluid examination, radiological investigations like ultrasound, MCU, MRI and cystoscopy can help to demonstrate exact anatomy of lower urinary tract leading to the cause of urinary ascites [9].

Once confirmed the cause of ascites been of urinary tract origin, basic aim of management should include decompression by abdominal paracentesis, catheter drainage with or without vesicostomy or surgical exploration and repair of perforation site in symptomatic cases [5,6].

In our case, the prostatic utricle cyst ruptured prior to birth of the baby leading to gross fetal and neonatal ascites. On insertion of indwelling urinary and peritoneal catheter for urinary drainage and serial abdominal paracentesis, the cyst shrunk in size with self-sealing of leakage site. With the cystoscopic evidence of utricle cyst causing partial bladder outlet obstruction, the child underwent vesicostomy as temporary measure with the aim for definitive surgical correction at later date.

Our case was confirmed to have Klinefelter syndrome on antenatal and postnatal karyotype. There have not been any case reports or any known association between Klinefelter syndrome and prostatic utricle cyst. As mentioned earlier, due to available incidence of association of utricle cysts with intersex anomalies, it is probable that the underlying genetic make-up of the baby could have predisposed him to develop utricle cyst.

Prognosis of urinary ascites depends on the age of occurrence, degree of compression and extent of changes in urinary tract. Early diagnosis and appropriate urinary drainage forms the cornerstone of favorable outcome. Overall, prognosis of isolated fetal ascites is good if it occurs at late gestational age, if ascites is of mild to moderate degree and of treatable cause [10]. An extensive investigative work up is required to determine etiology of isolated ascites, which will provide for appropriate treatment and successful outcome. Lastly, we recommend considering rupture of prostatic utricle cyst as a rare cause of isolated urinary neonatal ascites.

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