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Ectopic Insertion of a Duplicated Ureter into a Seminal Vesicle: A Case Report of a Giant Retroperitoneal Cyst and Restored Fertility After Removal

Ektopična insercija podvostručenog uretera u sjemeni mjehurić – prikaz slučaja velike retroperitonealne ciste i obnove fertiliteta nakon njezina uklanjanja

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Abstract. ***Aim:*** We report a rare case of a symptomatic, inflamed giant retroperitoneal cyst. The lesion was found to represent an afunctional, sac-like upper pole of the right kidney with a dilated, duplicated collecting system and a megaureter ectopically inserted into the ipsilateral seminal vesicle, resulting in male infertility. ***Case report:*** A 26-year-old male patient presented to the emergency department with the right flank pain, nausea, and high body temperature. A giant retroperitoneal cyst was diagnosed which proved to be afunctional, sack-like upper pole of the right kidney with its dilated, separated collecting system and megaureter ectopically inserted into the ipsilateral seminal vesicle. Although the patient's symptoms were relieved after percutaneous drainage of the cyst, the symptoms recurred one month later, and an open surgical excision was performed. The patient's symptoms disappeared and his impaired fertility was fully recovered after surgery**.** ***Conclusion:*** There is a close relation between congenital anomalies of the urinary tract and the seminal vesicle cysts due to their close embryological origin which often cause concurrent malformations of the ipsilateral upper urinary tract. Cystic formations of the retroperitoneum require thorough evaluation and extensive imaging to rule out congenital urogenital malformations. Additionally, male fertility testing is required if there is any indication of genitourinary system development anomalies because early detection and treatment of anomalies can lead to a partial or full recovery of fertility.

Keywords: bifid or double ureter; diagnostic imaging; male infertility; seminal vesicles; urogenital abnormalities

Sažetak. ***Cilj:*** Prikazati rijedak slučaj velike, simptomatske i inflamirane retroperitonealne ciste za koju se, nakon opširne dijagnostičke obrade, utvrdilo da je nefunkcionalan, vrećasto proširen gornji pol desnog bubrega s proširenim, podvostručenim kanalnim sustavom i megaureterom koji se ektopično ulijevao u ipsilateralni sjemeni mjehurić, uzrokujući neplodnost kod muškarca. ***Prikaz slučaja:*** Muškarac u dobi od 26 godina javio se u hitnu službu zbog boli u desnoj polovini trbuha, mučnine i povišene tjelesne temperature. Dijagnosticirana je velika retroperitonealna cista za koju se ustanovilo da je nefunkcionalan, vrećasto proširen gornji pol desnog bubrega s proširenim, podvostručenim kanalnim sustavom i megaureterom ektopičnog ušća u ipsilateralnom sjemenom mjehuriću. Iako su simptomi nakon perkutane drenaže ciste nestali, zbog njihova je relapsa učinjena otvorena kirurška ekscizija. Nakon operacije simptomi su potpuno nestali, a patološke vrijednosti spermiograma u potpunosti su normalizirane. ***Zaključak:*** Postoji uska embrionalna i anatomska povezanost u razvoju mokraćnog i genitalnog sustava. Zato se malformacije ovih

dvaju sustava najčešće prezentiraju konkomitantno i ipsilateralno. Cistične tvorbe retroperitoneuma zahtijevaju temeljitu obradu i detaljnu radiološku dijagnostiku kako bi se isključile kongenitalne urogenitalne malformacije. Također, ispitivanje muške plodnosti nužno je kada postoji sumnja na anomalije razvoja urogenitalnog sustava jer rana dijagnoza i liječenje mogu dovesti do djelomičnog ili potpunog oporavka plodnosti.

Ključne riječi: muška neplodnost; seminalne vezikule, slikovne dijagnostičke metode; bifidni ili dupli ureter; urogenitalne anomalije

INTRODUCTION

There is a close relation between congenital anomalies of the urinary tract and the seminal vesicle cysts due to their close embryological origin which often cause concurrent malformations of the ipsilateral upper urinary tract. Ectopic insertion of the ureter is frequently conjoined with ureteral duplication. Herein, we report a rare case of male infertility due to ectopic insertion of right duplicated ureter into ipsilateral seminal vesicle, primarily shown as inflamed giant retroperitoneal cyst.

CASE REPORT

A 26-year-old male patient presented to the emergency department with right flank pain, nausea, and high body temperature. He also reported symptoms of dysuria, frequency, and urgency. He did not report any sexual dysfunction, such as erectile or ejaculatory disorders. The right hemiabdomen was slightly tender on palpation while the physical examination of genitals and prostate was unremarkable. Blood and urine tests showed slightly elevated leukocytes ($10.3x10^9$/L, *norm. 3.4 – 9.7*), high CRP (284 mg/L, *norm. <5*) and leukocyturia. Abdominal ultrasonography revealed a large hypoechoic, cyst-like mass closely associated with the right kidney, extending into the pelvis. The kidney was displaced laterally and the liver cranially. The left kidney appeared normal. CT scan confirmed a large, thin-wall cyst in the right hemiabdomen arising from the enlarged ipsilateral seminal vesicle with no contrast enhancement. The cyst measured 45x13 cm (Figure 1).

There is a close embryological relationship between urinary tract and seminal vesicle anomalies. Cystic retroperitoneal masses therefore require thorough imaging studies and an evaluation of male fertility, as early diagnosis and timely intervention can enable partial or complete recovery of reproductive function.

Urethrocystoscopy revealed normal anatomy of both anterior and posterior urethra. The ureteral

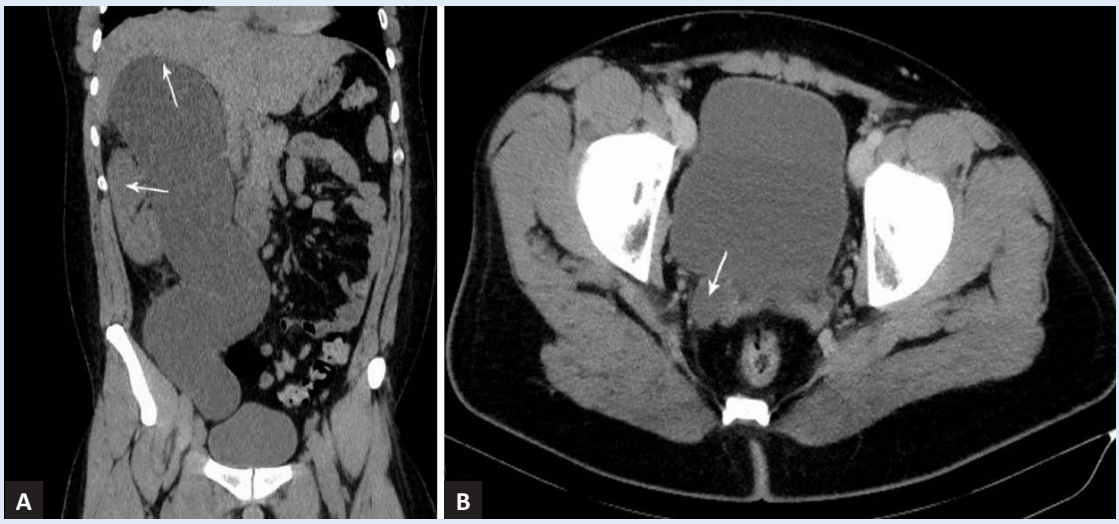

Figure 1. CT scan of abdomen and pelvis in its coronal (A) and axial planes (B) showing gigantic cyst in the right hemiabdomen. Right kidney is displaced laterally and liver cranially (arrows). The cyst is arising from the right seminal vesicle which is also enlarged (arrow).

orifices, one on each side, were in their normal anatomical positions, and the trigone was normally developed. Right retrograde ureteropyelography revealed tortuotic ureter and malrotated pyelon. The right ureter was intubated with double J stent to prevent obstructive nephropathy and to facilitate eventual future surgery. Transrectal ultrasonography revealed no abnormalities in the prostate gland anatomy; however, a giant cyst arising from the right seminal vesicle was observed, suggesting a diagnosis of a large seminal vesicle cyst (Figure 2). Subsequently, the cyst was percutaneously drained using a Malecot catheter inserted in the posterior axillary line. Brownish fluid was analyzed, and the fluid creatinine was similar to serum creatinine values. Cytological examination revealed numerous neutrophils and fibrin, with no evidence of spermatozoa. Streptococcus mitis was isolated from the cyst aspirate. Urine and blood cultures were negative.

The patient's condition improved following antibiotic therapy and the drainage of nearly 4000 mL of cyst content. He no longer reported abdominal pain, and his body temperature normalized. Five days after drain insertion, ultrasonography demonstrated no residual fluid collection. Prior to hospital discharge, the drain was removed, and a Double-J stent was left in situ. C-reactive protein (CRP) was 13 mg/L, and leukocyte levels were within normal limits. Further office evaluation included measurement of LH, FSH, testosterone, and prolactin levels, as well as semen analysis. Blood tests were within normal range, but semen analysis revealed low sperm count ($5{,}4x10^6$/mL) and poor sperm motility (total motility 23%, progressive motility 19%). After a month, the symptoms of abdominal discomfort recurred. An MRI of the abdomen and pelvis revealed a cyst of the same size as before drainage. The patient was scheduled for laparoscopic exploration and cyst excision and was referred to sperm cryopreservation. The laparoscopic approach was not successful due to the compromised anatomy caused by the large retroperitoneal mass. Conversion to an open procedure revealed a large cyst attached to the parietal peritoneum. The cyst extended medially to the inferior vena cava, cranially to the liver and caudally to the minor pelvis. One part of the cyst was attached to the upper pole of the right kidney and following resection it was clear that the cyst represented duplicated, dilated upper pole collecting system and megaureter ectopically inserted into the right seminal vesicle (Figure 3). Postoperative recovery was uneventful, double J stent was removed, and the patient was discharged from hospital after seven days. Histopathology confirmed the urothelial lining of the cyst and chronic inflammation with foci of ulceration and hemorrhage.

Three months after surgery all the parameters of semen analysis were within normal range (sperm concentration $62x10^6$/mL, total motility 41%, progressive motility 34%).

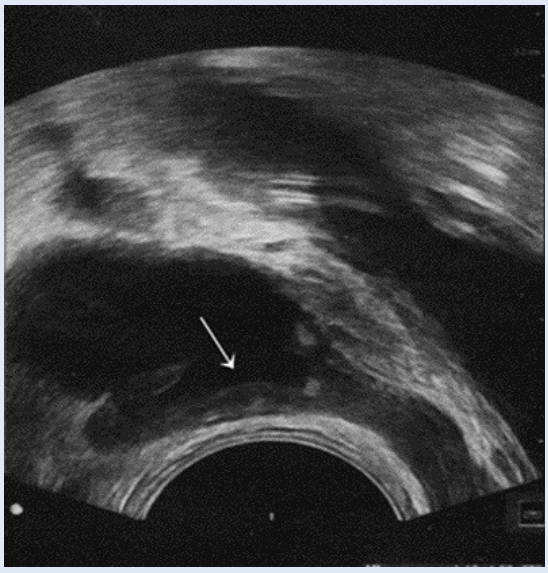

Figure 2. Transrectal prostate ultrasonography showing a giant cyst arising from the right seminal vesicle (arrow)

DISCUSSION

The urogenital system has the same embryological origin – intermediate mesoderm. All of three kidney generations (pronephros, mesonephros and metanephros) develop from intermediate mesoderm but definitive kidney could not develop if there is no contact with ureteric bud which arises from caudal part of Wolffian duct. Nephrons develop from metanephrogenic mesenchyme but collecting ducts, renal pelvicalyceal system and ureter from the ureteric bud[1]. If the ureteric bud branches early during development,

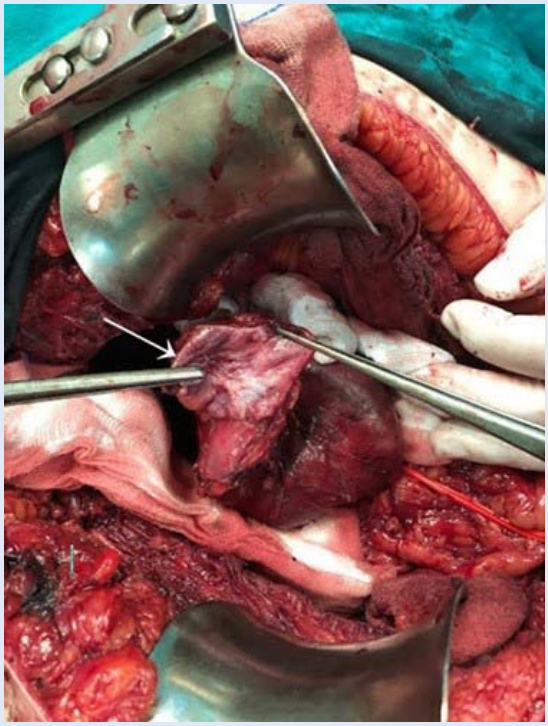

Figure 3. Intraoperative image showing epithelial lining of the cyst around the right kidney upper pole (arrow).

it then results in bifid ureter formation while an accessory ureteric bud results in complete ureteral duplication. Sometimes, when the accessory ureteric bud is more cranially placed on mesonephric duct it ends more distally (for example vesical neck, posterior urethra, seminal vesicle, vas deferens or epididymis). Ectopic ureteral insertion may result in maldevelopment of an ipsilateral kidney[2]. Seminal vesicle cysts can be congenital or acquired. Congenital seminal vesicle cysts occur because of maldevelopment of distal part of mesonephric duct. Therefore, those cysts are often combined with ejaculatory duct atresia, ipsilateral ureteral maldevelopment, ectopic ureteral insertion and ipsilateral kidney maldevelopment. Acquired seminal vesicle cysts are predominantly associated with inflammation and secondary ejaculatory duct obstruction. Seminal vesicles are the second most common site of ectopic ureteral insertion[3]. Clinical presentation depends on seminal vesicle cyst size. The cysts which are smaller in size (<5 cm) are usually asymptomatic[4]. Larger cysts are symptomatic, especially between 2nd and 4th decade of life when seminal fluid accumulates in the vesicles[5]. Patients often complain of perineal, pelvic, abdominal or flank pain, ejaculation related discomfort, urethral discharge, lower urinary tract symptoms and reproductive health problems[6]. Imaging of seminal vesicle cysts includes transabdominal ultrasound and transrectal ultrasound. To evaluate possible abnormalities of the kidney development it is necessary to perform the imaging of the upper urinary tract as well[7]. In large pelvic and retroperitoneal cysts, CT and MRI of the abdomen are useful in determining the origin and relations of cysts to other pelvic and abdominal structures. Cystoscopy is necessary to evaluate trigone morphology and morphology of ureteral orifices.

Asymptomatic seminal vesicle cysts should not be treated while symptomatic cysts should be treated either by transurethral unroofing or by laparoscopic, robotic, or open surgical approach[8]. Ejaculatory duct obstruction caused by cyst oppression may result in inadequate semen transfer, low sperm volume, oligoasthenozoospermia and even azoospermia. Some evidence suggest that long term semen stasis may lead to seminal vesicle inflammation and thus further worsen semen quality[9]. Furthermore, inflammatory cytokines and reactive oxygen species (ROS) may also have an impact on the sperm quality. The presence of proinflammatory cytokines, TNF-α, IL-1α and IL-1β may have certain physiological function in the male reproductive tract. When the levels of these cytokines are higher than normal, like in inflammation, they become very harmful to sperm production. Moreover, inflammation is also associated with increased production of ROS which are well known to have a negative impact on sperm quality and thus infertility[10].

Only a few published articles presented data on fertility in patients with seminal vesicle cysts. In our case, it is unlikely that congenital malformation of the genitourinary tract is the only cause of abnormal semen analysis. Sperm stasis due to cyst obstruction, local and systemic effect of proinflammatory cytokines, and excessive ROS production probably played a significant role in patient's low sperm quality.

CONCLUSION

Seminal vesicle cysts often present with lower urinary tract symptoms, sexual problems (ejaculatory pain, hematospermia, scrotal pain) and symptoms of genitourinary tract infections. Frequently, seminal vesicle cysts are also associated with other urogenital abnormalities and therefore a broad spectre of diagnostic and imaging tools should be used to establish correct diagnosis. Probably the mere presence of a cyst is not the only reason for semen abnormalities, but a combination of obstructive and inflammatory factors.

Conflicts of Interest: Authors declare no conflicts of interest.

A rare congenital anomaly involving an afunctional, duplicated upper pole of the right kidney with an ectopic ureter inserting into the ipsilateral seminal vesicle can result in the formation of a giant retroperitoneal cyst and male infertility. Surgical excision may resolve symptoms and lead to complete normalization of semen parameters, restoring fertility.

REFERENCES

1. Park JM. Embryology of the genitourinary tract. Campbell – Walsh Urology 11th edition. Philadelphia, Elsevier; 2016, p. 2823.
2. Dorko F, Tokarčík J, Výborná E. Congenital malformations of the ureter: anatomical studies. Anat Sci Int 2016; 91(3):290-4.
3. Patel B, Gujral S, Jefferson K, Evans S, Persad R. Seminal vesicle cysts and associated anomalies. BJU Int 2002; 90:265-71.
4. Livingston L, Larsen CR. Seminal vesicle cyst with ipsilateral renal agenesis. Am J Roentgenol 2000;175:177-80.
5. Coşkun B, Dalkiliç A, Sönmez NC, Arısan S, Ofluoğlu Y, Ergenekon E. A case of seminal vesicle cyst associated with ipsilateral renal agenesis diagnosed during an investigation of urinary incontinence. Turk J Urol 2013;39(1):53-5.
6. Farrelly JS, Cleary MA, Weiss RM, Morotti R, Stitelman DH. Acquired seminal vesicle cyst in a teenager with a urethral stricture of unknown etiology. J Pediatr Surg Case Reports 2016;7:8-12.
7. Carvalho HA, Paiva JLB, Santos VHV, Andrade M, Galvao-Teles A. Ultrasonic recognition of a cystic seminal vesicle with ipsilateral renal agenesis. J Urology 1986;135(6): 1267-8.
8. Celigoj FA, Costabile RA. Surgery of the scrotum and seminal vesicles. Campbell – Walsh Urology 11th edition. Philadelphia, Elsevier;2016.p.946.
9. Cheng G, Liu B, Song Z, Xu A, Song N, Wang Z. A novel surgical management for male infertility secondary to midline prostatic cyst. BMC Urol 2015;15:18.
10. Azenabor A, Ekun AO, Akinloye O. Impact of inflammation on male reproductive tract. J Reprod Infertil 2015;16(3):123-9.