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Case Study

Zinner's Syndrome: Rare Urological Presentation In Adolescent.

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Abstract

Acute Urine retention in adolescent is uncommon presentation. Infection, trauma are the most common etiology on this age group. Simple work up and management can be concluded in family medicine clinic without urology reference.

Zinner's syndrome (ZN) is rare syndrome. It is presented usually with fertility problem and ejaculatory complaints on adults. Unusual early presentation of ZN in such age group with urological symptoms rather than fertility complaint is addressed in our case. Furthermore acute urine retention in ZN is not listed in literature.

Herein, the case is presented as urine retention first leading sign of syndrome diagnosis in such age. Endoscopic management was carried out with successful outcome.

Keywords : Zinner's, Urine retention, adolescent, ejaculatory ducts.

INTRODUCTION

On 1914, Zinner's syndrome was described as the classic triad of unilateral renal agenesis, ipsilateral ejaculatory duct obstruction, and ipsilateral seminal vesicle cyst [1]. An incidence of approximately 0.00214 % in newborns was reported [2]. Usually presentation was on fertility age group with ejaculatory or fertility concerns.

In our case, patient was adolescent. Presentation was atypical of this syndrome; he presented with acute urine retention.

The address of this condition is aimed to raise the attention to thoroughly investigation with urology reference when a strange presentation of this age group. More over to delight the endoscopic management that could relieves of unilateral ejaculatory obstruction and preserve future fertility and ejaculation.

CASE PRESENTATION

Boy 17-years old presented with acute urine retention with left testicular pain. The condition was not associated with any constitutional symptoms. He had normal vital signs. On first assessment differential diagnosis by the family physician was aimed to exclude Torsion, trauma and sever UTI. On clinical examination the testis were normal and bladder was palpable on supra-pubic region.

Later, laboratory tests did not support infection or any abnormalities including kidney function, inflammatory markers. Abdominal ultrasound showed the distended bladder and agenesis of left renal unit with a cystic swelling related to the base of the prostate compressing the bladder neck [**picture-1**]. Urinary catheter was inserted, with immediate passage of 600 cc of clear urine. Case was referred to urology for further assessment.

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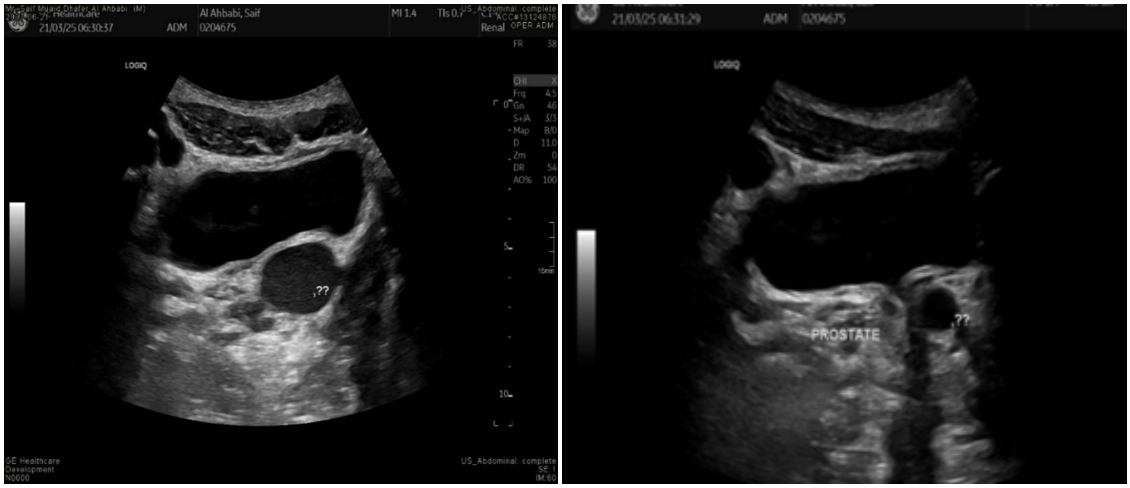
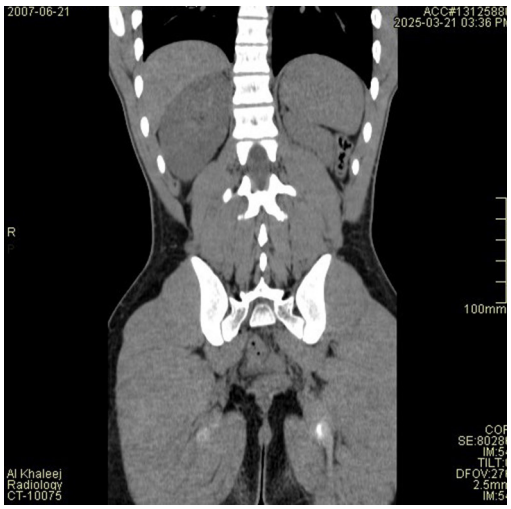
Picture 1. Pelvic Ultrasound shows full bladder, left dilated seminal vesicle



RADIOLOGY FINDINGS

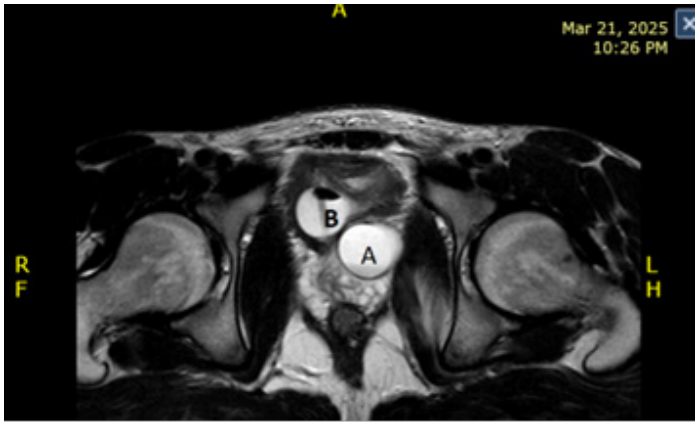
CT scan: Confirmed right renal agenesis and non-conclusive for pelvic pathology [**picture-2**]. Thereafter, an abdominopelvic MRI was conducted using a 1.5 Tesla, phased array coil, Axial T2 of lower abdomen and pelvis. Then high resolution images of the prostate were obtained using T2 in Sagittal, Axial and Coronal Planes.

Picture 2. Coronal non contrast CT scan shows solitary right kidney.

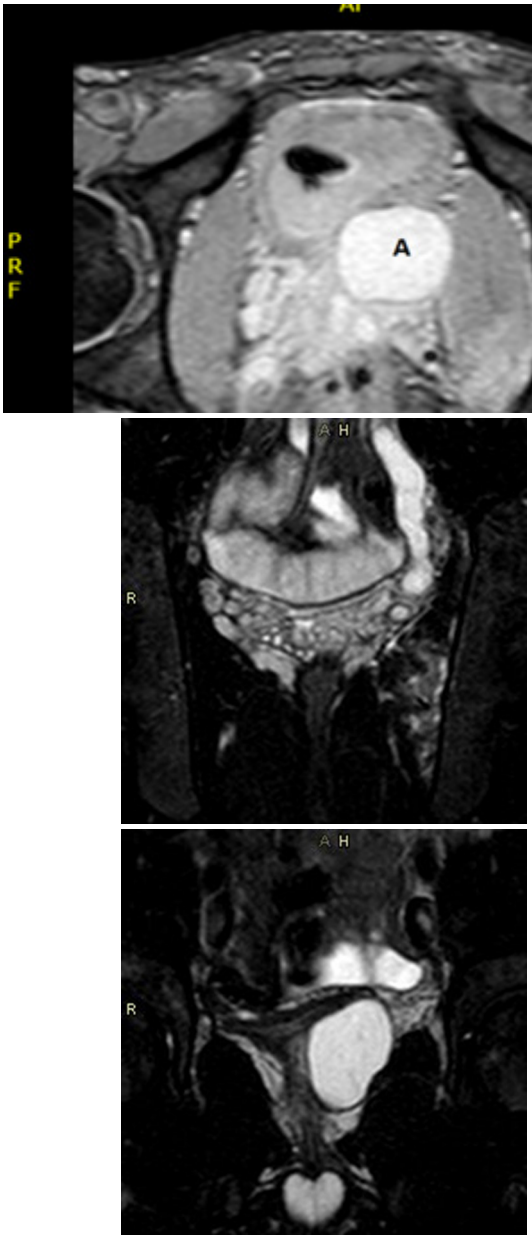


MRI images demonstrated agenesis of left renal unit with normal right renal unit. On Pelvic view a hyperintense left seminal vesicle cyst measuring 26 x 32 mm, communicating with seminal vesical ducts and ipsilateral ejaculatory duct dilatation (Obstruction signs). No solid components or signs of malignancy were seen. [**pictures-3 & 4**].The findings confirmed the diagnosis of ZN.

Picture 3. MRI of pelvis: axial view: A: left seminal vesicle, B: Balloon of urethral catheter.



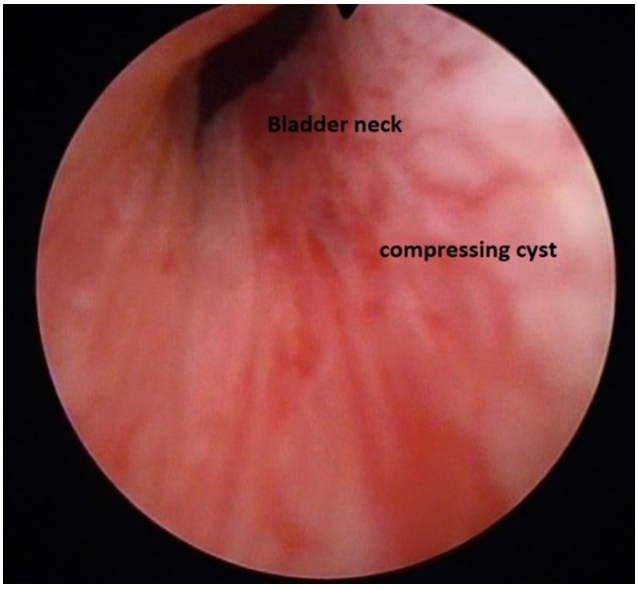
Picture 4. left seminal vesicle dilatation end to the cyst & compressing the ejaculatory duct.



SURGICAL FINDINGS

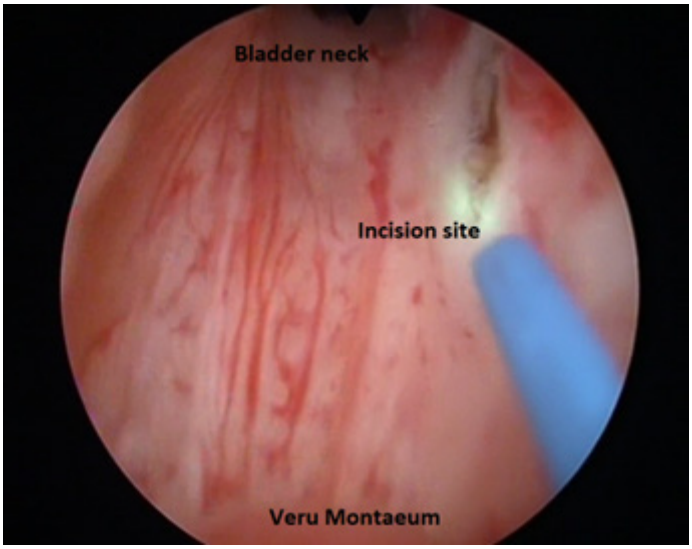
The patient and his family had counseled and consent was signed. Examination under anesthesia; large cystic bulge was felt on left prostate base. Diagnostic urethra-cystoscopy show normal anterior urethra. On prostatic urethra the lumen was deviated to right side and compressed with normal mucosa covering [**picture 5**]. It is compromised its diameter significantly. At the bladder neck there was a bulge on left side with hemi-trigon and right ureteric orifice was normally recognized.

Picture 5. Operative findings: Lumen is deviated and compressed to right side.

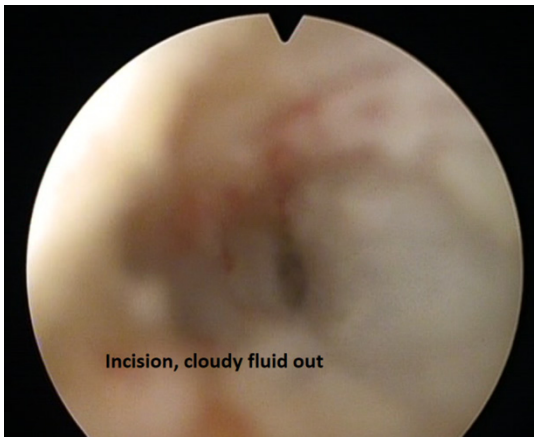


Using LASER fiber on cutting mode (Lumenis Pulse™ 120H Holmium Laser System with MOSES™ Technology), a small incision was created distal to bladder neck by approximate 1 cm and deepen gradually until cloudy content was expressed out. Manual augmented compression of the cyst rectally was carried out [**pictures 6, 7 & 8**]. Preserving the bladder neck is guard against retrograde ejaculation. Urinary catheter was removed after 3 days, patient voids spontaneously and satisfied. Residual urine was nil. No testicular pain. Later, pelvic ultrasound shows the cyst was markedly decompressed and wide communication orifice within urethra was observed [**picture 9**].

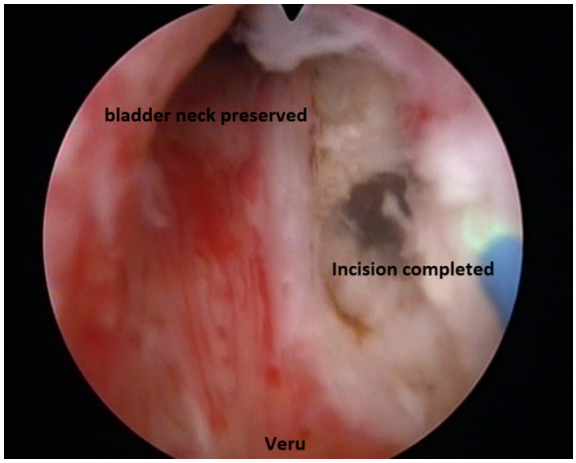
Picture 6. Operative findings: Incision started distal to bladder neck.



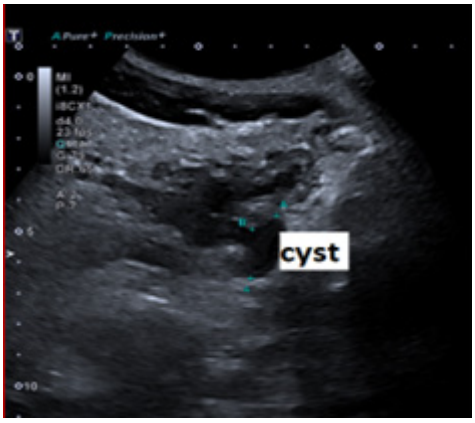
Picture 7. Operative findings: Cyst was opened and cloudy fluids started outflow.



Picture 8. Operative findings: Final results with preserved anatomy of urethra and bladder neck, Urethral lumen is patent without compression.



Picture 9. postoperative pelvic ultrasound : cyst is markedly reduced size



After one month, patient was called for semen analysis and further discussion of his fertility concerns.

DISCUSSION

Embryological, The mesonephric or wolfian ducts are present in human embryos. In males, this duct is transformed under the control of the testosterone and the anti-mullerian hormone into the hemitrigone, the bladder neck, proximal urethra (up to the external sphincter,), seminal vesicle, vas deferens, efferent ducts, epididymis, paradidymis, appendix epididymis, and more. The development of the kidney, ureter, seminal vesicle, and vas deferens can be adversely influenced by first trimester disturbances (4-8 week). The distal mesonephric duct is improperly developed which leads to atresia of the ejaculatory duct which becomes an obstruction that leads to cystic enlargement of the seminal vesicle. Abnormal development of ureter buds leads to renal agenesis or nephrodysplasia.

The obstruction occurring within the ejaculatory duct leads to a gradual accumulation of secretions in the seminal vesicle and eventually the formation of cysts. These problems can lead to azoospermia or oligospermia due to primary infertility. Also, depending on the size of the cysts, they may exert pressure on other structures which can cause pain in the perineum and the pelvic region [3].

Most patients with mesonephric duct anomalies are asymptomatic until the third or fourth decade of life and often manifest during the period of high sexual or reproductive activity. Commonly presented with perineal/ pelvic pain, painful ejaculation, dysuria, increased frequency of micturiton and oligospermia/azoospermia. In rare case hematospermia, uremia, epididymitis, abdominal hypogastric pain and infertility. [4, 5] Clinically 45 % of patients with Zinner syndrome may develop infertility [6, 7].

Radiological inevstaigations including ultrasonography and MRI, are most accurate for diagnosis. The ultrasound finding of the seminal vesicle cysts with ipsilateral renal agenesis are first-line diagnostic tool. Furthermore, MRI provides a more comprehensive evaluation of the pelvic anatomy, including the ejaculatory ducts [8]. In some cases, transrectal ultrasound with aspiration is also reported as an effective method for diagnosis [9].

Different surgical treatment options were discussed for treating infertility and relieve of ejaculatory duct obstruction [10, 11]. However this case - in the best of our knowledge- is first case report of urine retention as presenting symptom of ZN. Age presentation of the syndrome is always on fertile age group, and our case is the first diagnosed case on early adolescent. In addition to the Endoscopic urethral management and preserved ejaculatory function.

Our recommendation is: do not humble the simple sign in such age group, as it may manifest the hidden pathology. Rather than endoscopic minimal invasive surgical option could be a safe intervention and could relieve the obstruction of the ejaculatory duct by time.

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