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Zinner Syndrome: Case report of atypical symptoms and literature

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ABSTRACT

Zinner Syndrome is a rare congenital anomaly of the urogenital tract, characterized by unilateral renal agenesis, ejaculatory duct obstruction and ipsilateral seminal vesicle cyst. Here, we present a case of a 54-year-old-male with anejaculation and frequent nocturnal emissions, denies fever, scrotal or perineal pain, hematospermia, hematuria, or lower urinary tract symptoms. Imaging studies revealed atypical findings of this syndrome. This study highlights the importance of recognizing atypical symptoms and appropriate management to relieve symptoms and improve quality of life.

1. Introduction

Zinner Syndrome, first described in 1914, is a rare congenital anomaly consists of the classic triad of unilateral renal agenesis, ipsilateral ejaculatory duct obstruction, and ipsilateral seminal vesicle cyst.[1] Since then, approximately 200 cases of this condition have been reported in the literature.[2] The accurate diagnosis can be easily achieved through a thorough physical examination and precise diagnostic imaging. Obstruction of the ejaculatory duct results in the accumulation of seminal fluid, which subsequently leads to the enlargement of the seminal vesicles. Symptoms are generally non-specific and primarily include pain, dysuria, frequent urination, perineal pain, epididymitis, and pain following ejaculation. Additionally, some studies have indicated that up to 45 % of patients with Zinner syndrome may develop infertility.[3,4]

The objective of this case report is to describe and analyze rare symptoms associated with Zinner syndrome that have not been previously documented in the medical literature.

2. Case description

The patient was a 54-year-old male with right renal agenesis from birth, diagnosed 20 years ago (Fig. 1). The patient first experienced symptoms in 2019 but did not seek medical evaluation due to the COVID-19 pandemic. He later presented with complaints of anejaculation and persistent nocturnal emissions. The patient denied having any associated symptoms, including fever, scrotal or perineal pain, hematospermia, hematuria, or lower urinary tract symptoms.

On physical examination, the patient's genitalia were found to be appropriate for his age. The penis was uncircumcised with no excoriations on the glans, and the meatus was patent and non-stenotic. Both testicles were well descended into the scrotal sacs without signs of calcifications, nodules, or indurations. Bilateral varicocele was present. Digital rectal examination revealed a 30 cc prostate that was smooth, non-fixed, and free of nodules, with a soft, fluctuant, and enlarged right lobe. The prostate was normothermic and non-tender upon palpation.

Laboratory evaluations were normal. Bladder ultrasonography measured a seminal vesicle associated cyst (Fig. 2a and b) (see Fig. 3).

An abdominopelvic MRI was conducted using a 1.5-T scanner. MRI showed seminal vesicle cysts near the right seminal vesicle associated with ipsilateral renal agenesis. These findings align with the characteristics typically observed in seminal vesicle cysts associated with Zinner syndrome.

Treatment consists in a laparoscopic management in bloc excision of the ipsilateral seminal vesicle cyst.

3. Discussion

Zinner syndrome is a rare congenital condition with an incidence of approximately 0.00214 % in newborns,[5] arising from abnormal development of the mesonephric duct during embryogenesis. This anomaly occurs when the ureteric bud fails to fuse with the mesonephric duct,

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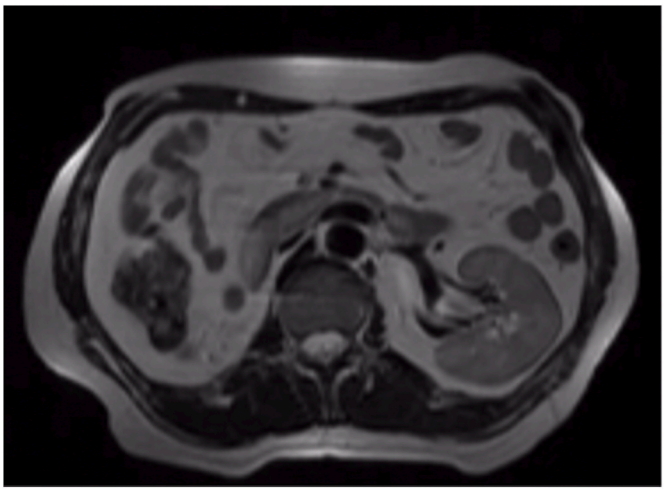

Fig. 1. MRI scan of a right renal agenesis (Axial T2-weighted image).

leading to ipsilateral renal agenesis. Simultaneously, incomplete differentiation of the Wolffian duct causes seminal vesicle cysts and ejaculatory duct obstruction. As a urogenital variant of renal malformations, Zinner syndrome predominantly affects the urinary and reproductive systems, leading to symptoms involving both.[6]

The typical onset of symptoms occurs during the third or fourth decade of life, likely due to the seminal vesicles reaching full functionality and increased sexual activity.[7] However, this case, presenting at 54 years of age, deviates from the norm, although the symptoms coincided with sexual activity.

Symptoms of Zinner syndrome can vary significantly, with case reports documenting testicular pain, infertility, urinary irritation, nocturia, scrotal swelling, fever, dysuria, and hematuria,[8] and are cases have also been reported as asymptomatic.[9] In this instance, the patient reported only anejaculation and frequent nocturnal emissions. Anejaculation can be attributed to ejaculatory duct obstruction caused by the seminal vesicle cyst. The frequent nocturnal emissions, while less commonly discussed in literature, could be linked to increased pressure in the seminal vesicles due to obstruction, although this remains speculative and warrants further investigation.

We present this case with atypical symptoms of altered ejaculatory dynamics in Zinner syndrome, emphasizing the need for additional research to elucidate the underlying mechanisms. Despite its rarity, early recognition and management of this syndrome are crucial, given its association with malignancies such as squamous cell carcinoma of the seminal vesicle.[10]

Diagnostic imaging, including ultrasonography and MRI, remains pivotal in diagnosing Zinner syndrome. While ultrasound is typically the first-line diagnostic tool and can reveal seminal vesicle cysts and ipsilateral renal agenesis, MRI provides a more comprehensive evaluation of the pelvic anatomy, including the ejaculatory ducts.[11] Transrectal ultrasound with aspiration is also reported as an effective method for diagnosis.[12]

Over 80 % of cases were asymptomatic during long-term follow-up. Consequently, conservative management of Zinner syndrome is generally endorsed for patients who are asymptomatic or have only mild symptoms, including occasional, transient, and non-specific issues such as urinary tract infections or orchiepididymitis.[13] The most robust and well-documented evidence for treating Zinner syndrome involves the use of transurethral resection of the ejaculatory duct. This surgical approach has been extensively studied and is supported by a significant body of research due to its effectiveness in alleviating symptoms

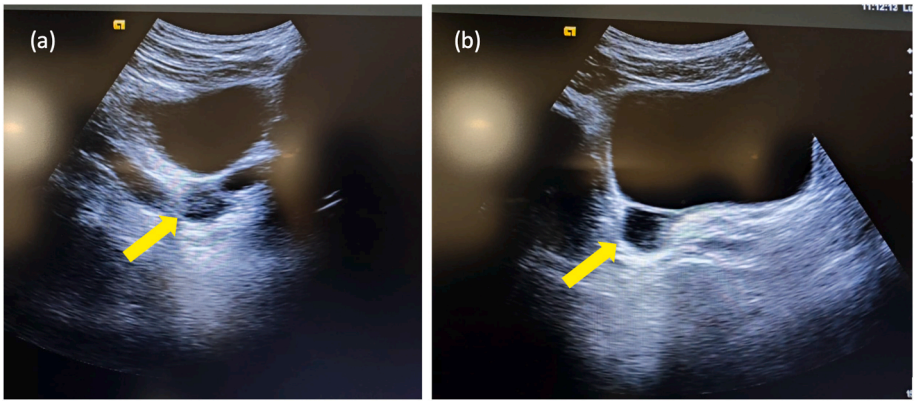

Fig. 2. (a) Axial section; (b) sagittal section. Bladder ultrasonography revealed a hypoechoic lesion located on the right dorsal aspect of the bladder (yellow arrow).

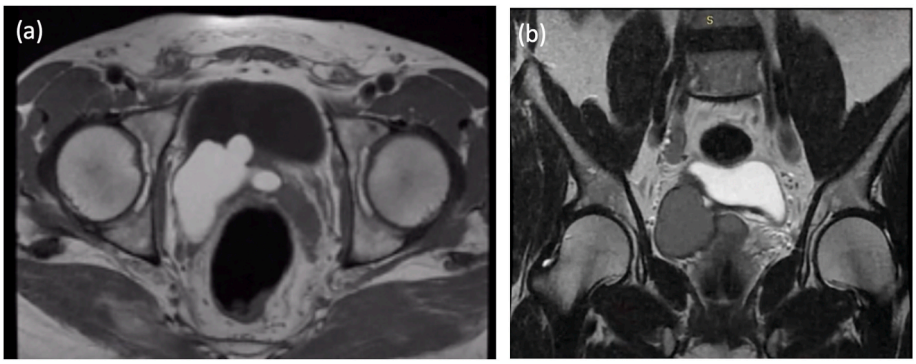

Fig. 3. (a) Axial T1-weighted MRI revealed a laterally located cystic lesion in the right seminal vesicle region. (b) Coronal T2-weighted image demonstrating a right seminal vesicle cyst with a remnant ureter.

associated with ejaculatory duct obstruction.[12]

CRediT authorship contribution statement

Alejandro Acuña-Pacheco: Writing – original draft. **Eduardo González-Rojas:** Writing – review & editing. **Pedro Iván Aguilar-Ordaz:** Writing – review & editing. **Joel Porfirio Rodelo-López:** Writing – review & editing. **Benjamin Bueno-Mendoza:** Writing – review & editing. **Israel Hernández-Rivera:** Writing – review & editing. **Jose Rene Jungfermann-Guzman:** Writing – review & editing. **Jesús Rodolfo Favela-Camacho:** Supervision, Writing – review & editing.

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Declaration of competing interest

The authors declare that they have no conflict of interest.

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