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CASE REPORT

Tubular ectasia of rete testis with obstructive azoospermia and infertility: A case report and literature review

Ahmed Al Saeedi[1] | **Ahmad Majzoub**[1,2] | **Haitham ElBardisi**[1,2,3] | **Mohamed Arafa**[1,2,4]

[1]Department of Urology, Hamad Medical Corporation, Doha, Qatar

[2]Department of Urology, Weill Cornell Medical-Qatar, Doha, Qatar

[3]Department of Urology, College of Medicine, Qatar University, Doha, Qatar

[4]Department of Andrology, Cairo University, Cairo, Egypt

Correspondence

Ahmed Al Saeedi
Email: FUTURE-DR@hotmail.com

Abstract

Background: Tubular ectasia of rete testis (TERT) is a benign condition due to dilatation of the rete testis as a consequence of multiple etiologies, including postinfectious, trauma, prior inguinal or scrotal surgeries, mechanical compression of extratesticular excretory ducts, and congenital malformation. TERT is a rare and underreported condition in the medical literature. Its association with infertility is lacking in the literature.

Case Presentation: We present a rare case of primary infertility and obstructive azoospermia associated with TERT. We performed extensive literature regarding the possible etiologies, associations, clinical diagnosis, and different management options of this entity. Our patient is a 33-year-old man presented with primary infertility. He was found to have obstructive azoospermia. During scrotal exploration, the diagnosis of intratesticular obstruction due to TERT was made. The couple was advised to undergo intracytoplasmic sperm injection using testicular sperms. This is the third report of TERT associated with azoospermia.

Conclusion: TERT is usually asymptomatic but can occasionally be associated with pain or abnormal semen analysis, but further studies are required to confirm its effect on fertility. Fortunately, it has a benign course and typically does not require surgical intervention. It is vital to differentiate it from neoplastic conditions.

KEYWORDS

azoospermia, male infertility, rete testis, tubular ectasia, ultrasound testes

1 | INTRODUCTION

Tubular ectasia of rete testis (TERT) is a benign condition due to dilatation of the rete testis as a consequence of multiple etiologies, including postinfectious, trauma, prior inguinal or scrotal surgeries, mechanical compression of extratesticular excretory ducts, and congenital malformation[1]. Its clinical significance comes from the fact that it may be misinterpreted as a malignant lesion, leading to unnecessary invasive treatment and psychological stress[2]. We present a 33-year-old man with primary infertility, TERT, and azoospermia. To the best of our knowledge, this is the third report of TERT with azoospermia reported in the literature.

2 | CASE PRESENTATION

We report a 33-year-old man who presented to the male infertility clinic with a 5-year history of primary infertility. He was otherwise healthy, with no history of scrotal pain, swelling, prior genital infections, or trauma. He had no significant past medical or surgical history of medical significance. On examination, both testes were

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of normal size (15 mL by Prader orchidometer) and consistency without palpable lesions. The epididymides were bilaterally bulky, with multiple cysts felt on the epididymal body and tail. Vasa deferentia were palpable bilaterally with no defects or nodularity. There was no clinical varicocele on either side.

Seminal fluid analysis was done twice and revealed normal volume (1.5 mL), alkaline pH, and positive fructose. Azoospermia was confirmed after centrifugation of the semen sample and examination of the centrifuged pellet. Serum hormones and tumor markers were all within normal limits (Table 1). Genetic workup showed normal male karyotype (46 XY) and no Y-chromosome microdeletions in any of the azoospermia factors (two multiplex polymerase chain reaction reactions with six Y-chromosome loci and ZFYX/Y and SRY internal controls. Set A: ZFX/Y, SRY, sY86, sY127, sY254; Set B: ZFX/Y, SRY, sY84, sY134, sY255). Scrotal ultrasound showed normal testicular volume (16 mL bilaterally) and echogenicity bilaterally. There was extensive cystic transformation of testicular parenchyma in the bilateral region of the rete testis, which was more extensive on the right side, extending into the adjacent parts of the epididymis (Figure 1) when compared with normal testicular ultrasound (Figure 2). Transrectal ultrasonography was performed and showed moderately enlarged prostate with obstructive changes at the level of the ejaculatory ducts leading to cystic dilatation of the seminal vesicles and vas deferens. Additionally, the ultrasound of the urinary tract was normal.

A diagnostic testicular needle biopsy was done, and normal spermatogenesis was observed, with 150 seminiferous tubules and more than 20 sperm per high-power field. The patient was, therefore, diagnosed with obstructive azoospermia and was counseled regarding an attempt at microsurgical Vasoepididymostomy after six weeks with lower chances of success.

TABLE 1 Serum hormones and tumor markers.

Test	Value	Normal limits
Hormones		
Estradiol	109 pmol/L	41–159 pmol/L
Prolactin	170 mIU/L	85–325 mIU/L
LH	4 IU/L	1.7–8.6 IU/L
FSH	5 IU/L	1.5–12.4 IU/L
Testosterone	18.7 nmol/L	10–31 nmol/L
Tumor markers		
Beta-HCG	<1 ng/mL	0–2 ng/mL
Alpha-fetoprotein	3 IU/mL	0–6 IU/mL
LDH	180 IU/L	135–225 IU/L

Abbreviations: FSH, follicle-stimulating hormone; HCG, human chorionic gonadotropin; LDH, lactate dehydrogenase; LH, luteinizing hormone.

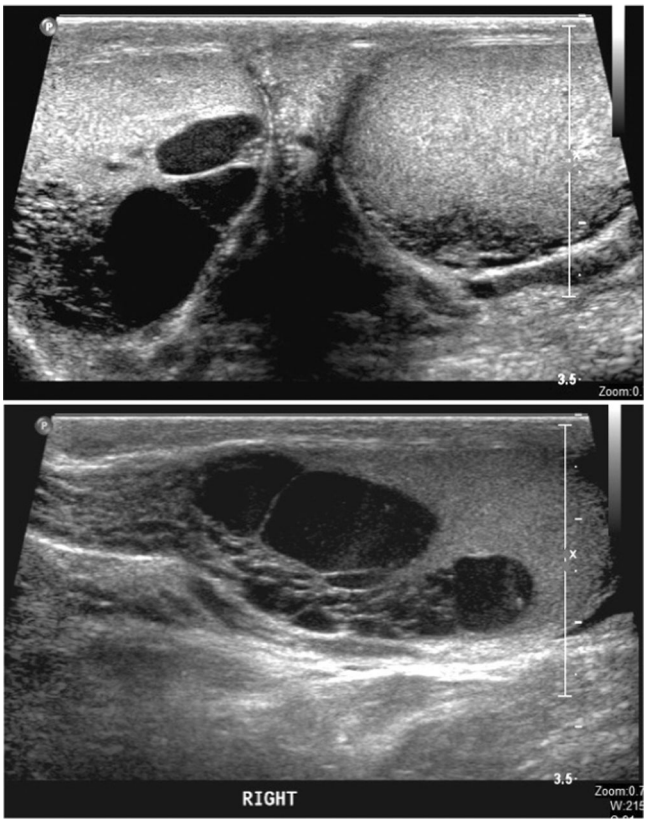

FIGURE 1 Testicular ultrasound demonstrating extensive cystic changes originating from the rete testis.

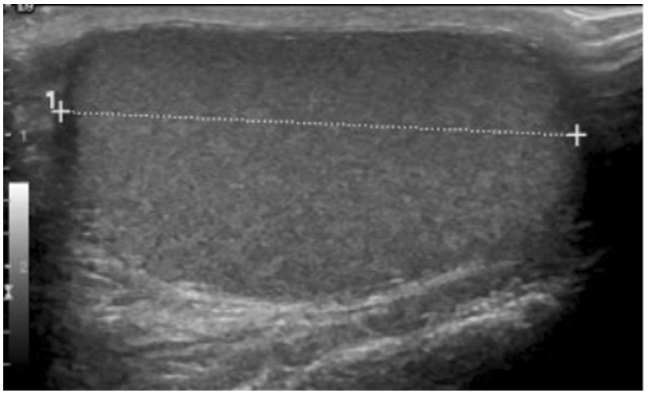

FIGURE 2 A normal testis ultrasound.

Intraoperatively, the epididymis demonstrated multiple cystic dilatations involving the entire epididymis without obvious dilated tubules (Figure 3). Vasography showed patent vasa without distal obstruction. Epididymal exploration with multiple aspirates from the cauda to the caput failed to reveal any sperm on microscopic examination. In contrast, testicular aspiration revealed many motile and immotile sperm, which were sent for cryopreservation for future use for intracytoplasmic sperm injection (ICSI). The diagnosis

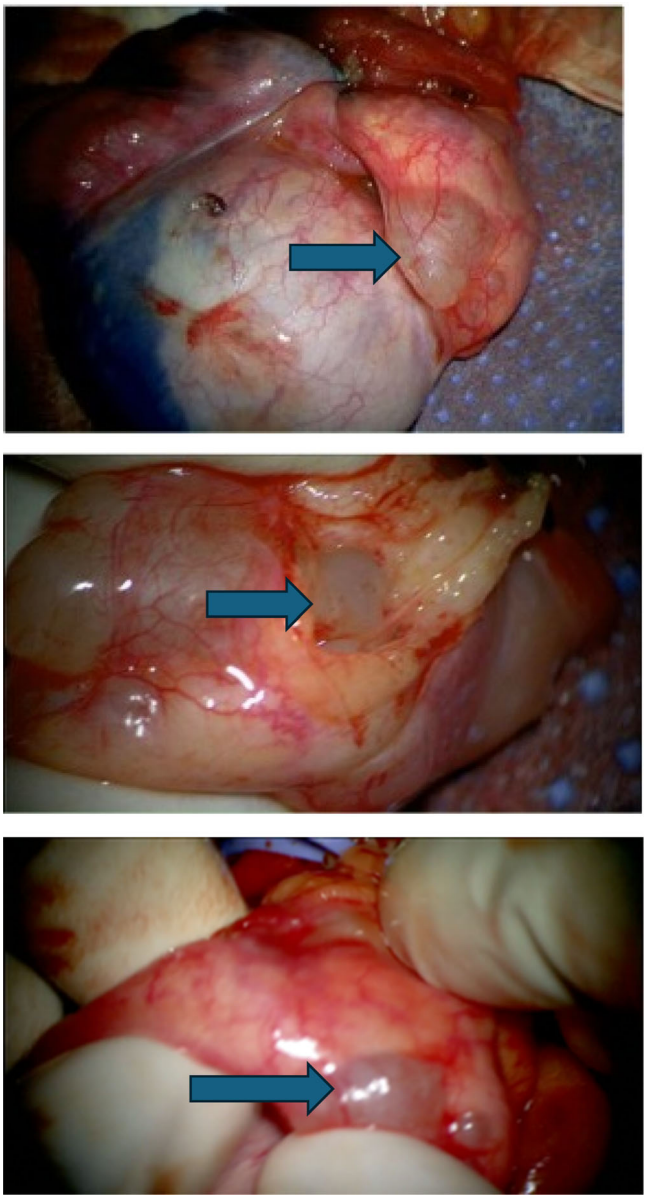

FIGURE 3 Intraoperative images showing multiple epididymal cysts (arrows).

of intratesticular obstruction was made, and the couple was advised to undergo ICSI using testicular sperm.

3 | DISCUSSION

The testis is composed of two main parts: the highly convoluted seminiferous tubules and the interstitial space[3]. The seminiferous epithelium is where spermatogenesis takes place, carried out by the germ cells. Sertoli cells, which are connected by tight junctions, provide structural and functional support for the developing germ cells and form the blood-testis barrier. The interstitial space comprises Leydig cells, blood vessels, macrophages, lymphocytes, connective tissue, and lymphatic vessels. The testis is composed of several hundred lobules separated by fibrous septa. Each lobule forms a straight tubule that converges at the mediastinum, forming the rete testis. Subsequently, sperm travels through the efferent ductules to the epididymis[3,4].

TERT is an uncommon benign condition, prevalent in approximately 1.64% of the general population and in 1.2% of men undergoing evaluation for infertility[3,5]. The median age of patients with TERT in the largest series in the literature was 62 years[6], compared to 33 years old in our case. Herrera et al. reported bilateral TERT in 42% of cases (8/19);[2] our patient also presented with bilateral involvement. Blockages at any point along the path of sperm could cause pressure to build up and lead to the formation of TERT[3]. Additionally, congenital abnormalities, such as testis-epididymis dissociation and arterial lesions, may also lead to ischemic atrophy of the epididymis and, eventually, TERT due to reduced perfusion from epididymal artery atherosclerosis[7]. Hormonal abnormalities, such as androgen-estrogen imbalance, could also be a factor as it increases the risk of structural and functional genital malformations because of epididymal duct atrophy[7,8]. Furthermore, past injuries, infections like epididymitis, and surgeries in the groin or scrotum are also potential causes[3].

TERT is typically found incidentally during testicular ultrasonography performed for unrelated reasons[1,9,10]. However, some patients may experience scrotal pain, which could be due to other conditions such as epididymal cysts, spermatoceles, or varicoceles[5,11]. TERT is commonly associated with epididymal pathologies. Burros et al. reported that 9 of 13 patients (69%) were found to have spermatoceles or epididymal cysts[12], while Brown et al. found epididymal abnormalities (cysts or epididymitis) in 34 of 40 patients (80%)[6]. Possible differential diagnoses of this entity include benign conditions (cystic dysplasia, intratesticular varicocele, epidermoid cyst, simple testicular cyst) and malignant etiologies (mixed teratoma and papillary adenocarcinoma of the rete testis)[3].

The diagnosis of TERT can be confirmed by clinical factors (such as age, presentation, and tumor markers) and characteristic ultrasound findings (anechoic cystic spaces in the mediastinum testis). Further assessment should be considered for young patients, palpable lesions, and solid components in the ultrasound[6,13]. Hamm et al. initially suggested that intratesticular cysts may arise from the rete testis, and all nonneoplastic cysts were impalpable. In their series, histological diagnosis identified 8 out of 13 cases (62%) as benign lesions, while the remaining cases were monitored for various durations without any signs of malignancy. They concluded that the diagnosis of intratesticular cysts could be established

based on clinical and radiological findings without the need for histological confirmation[10,12].

Rouvière et al. studied the evolution of TERT in 23 patients and 33 testes (10 patients had bilateral involvement) over a follow-up period of 8–19 months. They found that 7 TERT cases showed at least a 25% increase in size while preserving their typical characteristics. They concluded that even if the lesion size might enlarge, there is no need for further work-up if its sonographic appearance remains unchanged[14].

Only a few reports have investigated the relationship between TERT and male subfertility[5,15]. Jequier et al. conducted a study on the impact of TERT on fertility potential and found that 1.2% of the 749 men attending infertility clinics had TERT. Among these men, nine patients had TERT, seven of them had abnormal semen analysis results, and two had normal semen parameters. Three patients were azoospermic, with one having bilateral disease and two having unilateral involvement. Four patients had oligozoospermia, with one having bilateral involvement and the others having unilateral involvement. While infertility in these patients could be attributed to associated abnormalities, such as undescended testis, obesity, or varicocele, TERT could also contribute to abnormal semen parameters. The study suggests the need for further research in this patient group.

Ndluvo and Danso reported another case of bilateral TERT associated with azoospermia in a 46-year-old patient. The patient presented with infertility of 1-year duration and was found to have bilateral epididymal cysts and a left-sided varicocele. Unlike in our case, where normal spermatogenesis was documented by testicular biopsy, the cause of azoospermia was not clearly mentioned in their case[15]. Additionally, previously published cases of TERT with azoospermia were associated with other risk factors, such as undescended testis, varicocele, and morbid obesity. In contrast, our patient had none of these risk factors[5,16].

A conservative approach is generally proposed for the management of TERT without the need for a specific therapy, especially in asymptomatic patients with no suspicion of malignancy[1,16]. Surgical management may be required for the relief of chronic testicular pain. A spermatic cord block, followed by microsurgical denervation of the spermatic cord, can be offered to patients who experience symptom relief following the cord block[16]. Patients experiencing chronic testicular pain after cord block and denervation may need to consider orchiectomy. Interestingly, Ölçücü et al. reported a case of chronic orchialgia due to bilateral TERT that failed conservative management; the patient was treated with a combination of duloxetine (60 mg po) plus gabapentin (400 mg po) for 6 months, resulting in complete resolution of his pain[11]. In our case report, we present the third instance in the literature of TERT-associated infertility and azoospermia. This will provide more insight into this condition and its potential adverse effects on fertility.

Most studies published in the literature have focused on the clinical and sonographic features, potential causes of the condition, and possible differential diagnoses[1,2,9,10,12].

4 | CONCLUSION

TERT is a rare and underreported condition in the medical literature. It is usually asymptomatic but can occasionally present with pain. Diagnosis is made through thorough history taking and examination, supported by ultrasound characteristics that help differentiate it from neoplastic conditions. TERT can be associated with abnormal semen analysis, but further studies are needed to confirm its effect on fertility. Fortunately, it has a benign course and typically does not require surgical intervention.

AUTHOR CONTRIBUTIONS

Ahmed Al Saeedi and Haitham ElBardisi conceptualized the study, with Ahmed Al Saeedi drafting the original manuscript. Haitham ElBardisi, Mohamed Arafa, and Ahmad Majzoub contributed to the review and editing process, while Mohamed Arafa provided supervision.

CONFLICT OF INTEREST STATEMENT

The authors declare no conflict of interest.

ETHICS STATEMENT

This case report adheres to the highest standards of ethical conduct in research and publication. Patient information has been appropriately deidentified or excluded to protect privacy. This study conforms to the ethical guidelines of the Declaration of Helsinki and relevant institutional standards for the research and publication of anonymized patient data.

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