Andrology and fertility

A case presentation with tranverse testicular ectopia

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ABSTRACT

Transverse testicular ectopia (TTE) is a rare congenital anomaly, in which both gonads migrate toward the same hemiscrotum with increased risk of testicular malignancy and associated with various abnormalities. Proper knowledge and imaging techniques are essential in diagnosis. Ultrasonography (USG) offer an easy, safe, and convenient alternative in assessment of TTE. We report a case of 41-year-male with infertility, who presented with left reducible inguinal hernia and right undescended testis. Final diagnosis was confirmed by USG and magnetic resonance imaging (MRI). The embryology, imaging features and USG evaluation of infertility are discussed with review of pertinent literature.

Introduction

TTE is a rare congenital anomaly with approximately 150 cases reported worldwide to date. It occurs when there is migration of both testes along the same inguinal canal toward the scrotum. The most common presentation of TTE is in young children at mean age of 4–5 years with cryptorchidism and contralateral inguinal hernia. Its detection is mostly incidental during surgical exploration or laparoscopy for a symptomatic hernia, although increasingly, a preoperative diagnosis can be made on USG, computerized tomography, MRI, or magnetic resonance venography. We present an interesting case of an adult with no obvious symptoms. Final diagnosis was made on clinical examination and imaging modalities.

Case presentation

A 41-year-old male patient, with history of primary infertility for 4 years, came to our hospital for consultation. No significant past history was present. On clinical examination, he was averagely built and well-nourished with well-developed secondary sexual characters. On genital examination, the right testes was not palpable in the scrotum and imaging modalities.

Per abdominal examination revealed a reducible lump in the left inguinal region measuring 30 cm × 20 cm × 20 cm. Impulse could be felt when patient coughed. USG screening for the abdomen, pelvis and scrotum was done and was suggestive of left-sided inguinal hernia, with the left side testes in the left hemiscrotum along with another structure of similar echogenicity on the same side with empty right hemiscrotum (Fig. 1). The upper one was 30.4 × 15.6 × 11.9mm(4.1ml) and the lower one was 38.9 × 22.1 × 18.2mm(11.0ml)(Fig. 2a). Both testicular echogenicity had separate epidyymis, vas deferens and a separate blood supply(Fig. 2b). An anechoic nodule with a size of 11.5 × 7.6 × 9.5mm was observed in the left upper epidyymis, with clear boundaries and regular shape, and no blood flow signal was detected. Echogenic omentum in size measuring 28.5 × 15.2 × 17.1mm was found within the inguinal canal with extension into the abdominal cavity. The clinical and USG diagnosis was right side TTE with left epidydymal cyst and left indirect inguinal hernia. On abdomen MRI, there was presence of left side testes in the left hemiscrotum with the presence of another ovoid mass attached to its one end. There was no finding of Mullerian structures in pelvic cavity(Movie 1). Final diagnosis of type 1 TTE was established. On nondestructive sperm analysis, the number of sperms was found to be 7.2 million/cc, the vitality of sperm was found to be 78% and normal sperms was found to be 71%. Further necrozoospermia and teratozoospermia were diagnosed.

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The patient refused any further investigation or treatment. Therefore, follow-up evaluation was planned on an annual basis including clinical examination, scrotal ultrasound and hormonal screening.
Discussion

TTE was first described by Von Lenhossek in 1886. It is a rare deformity in clinical practice. The diagnostic clinical criteria consist of two testes located in the same route of testicular descent with empty scrotum.¹

TTE should be considered in patients with cryptorchidism and contralateral inguinal hernia. Numerous hypotheses have been put forward to explain the evolutionary origin of the scrotum.¹,² However, there is still no unified understanding of the pathogenesis of TTE. From view of physical and mechanical effect, theories have been proposed as defective implantation of the contractile gubernaculum, congenital atresia of another annular anatomical structure and early adherence and fusion of the developing Wolffian ducts, which would either prevent testicular descent or stretch contralateral to follow. Except for physical and mechanical effect, Berg proposed the possibility of the development of both testes from the same genital ridge. Matsumoto considered sex chromosomal abnormality, inappropriate androgen secretion or abnormality of androgen receptor also contributing to the development of TTE.

According to the associated developmental anomalies, TTE has been classified into 3 types: Type 1, accompanied only by hernia (40%–50%); type 2, accompanied by persistent Mullerian duct structures (30%); type 3, accompanied by disorders others such as hypospadias (20%). According to the classification, our case was type 1 TTE. And the diagnosis was firstly established by USG, which has a high resolution transducer (>7.5 MHz) and offers the greatest accuracy in assessment of 100% of palpable and of 84% of non-palpable undescended testis (with a sensitivity of 76% and a specificity of 100%).² Patients with TTE are at increased risk of malignant transformation with the overall incidence of malignant transformation of gonads approximately 18%.¹ Hence, long term follow-up for tumors is required. USG is the preferred auxiliary examination method. Besides the early detection of undescended testis (UDT), USG is capable of assessing the position of the UDT, parenchymal structure and exclude malignant transformation or other congenital malformations.

Though reproductive problems have been discussed in TTE cases, most of infertility happened in those patients with persistent mullerian duct syndrome.³ The testicular volume has long been associated with testicular function and K. H. Tijani et al. found cut-off value of 18–20 ml as optimal peak testicular volume of spermatogenesis in sub-fertile

![Ultrasounography cross-view revealed empty right hemiscrotum compared with the left hemiscrotum.](image1)

![Longitudinal view of left scrotum found two separated testicular echoes, which were organized into top and bottom position. The echo pattern of the parenchyma was homogeneous. The volumes were calculated as 30.4 × 15.6 × 11.9 mm (4.1 ml, upper) and 38.9 × 22.1 × 18.2 mm (11.0 ml, lower). Both testicular echogenicity had blood supply.](image2)
Our patient had both testes lower than 18 ml. Besides, testicular atrophy index (TAI) was developed for assessment of the state of testicular development to help making a decision on continuing observation or performing orchiopexy and allows monitoring of the results of treatment. In boys, its value of 20% and more should be considered an indication for surgery. The threshold in adults has not been discussed. In our case, the TAI was 63%. We considered the possible cause of necrozoospermia and teratozoospermia in our patient was maldevelopment of bilateral testes despite there was no merger of other genitourinary abnormalities or mullerian remnants.

**Conclusion**

TTE is a rare genitourinary anomaly, the pathogenesis of which remains unclear. It should be included in the differential diagnosis for every adult patient complicated with an absent testis and fertility problems. USG can provide useful information for preoperative diagnosis of TTE. If diagnosed, a thorough investigation and long-term follow-up visit should be adopted for the possibility of merging a wide range of other anomalies.

**Availability of data and materials**

Not applicable.

**Authors’ contributions**

Jinzhou Wan, Yu Wang and Huizhen Song analyzed the patient data. Jinzhou Wan and Yu Wang wrote the preliminary draft, Ying Tang provided expertise and edited the manuscript. All authors read and approved the final manuscript.

**Ethics approval and consent to participate**

Not Applicable.

**Consent for publication**

Written informed consent was obtained from the patient for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor of this journal.

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**Declaration of competing interest**

The authors declare that they have no conflict of interest.

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**Abbreviations**

MRI  Magnetic resonance imaging  
TAI  Testicular atrophy index  
TTE  Transverse testicular ectopia  
UDT  Undescended testis  
USG  Ultrasounography

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