Transverse Testicular Ectopia Presenting as Primary Infertility in Adulthood: A Case Report

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Abstract

Background
Transverse testicular ectopia (TTE), also known as crossed testicular ectopia, is a rare congenital anomaly in which both testes migrate to the same side of the body. TTE is typically diagnosed during the childhood; however, on rare occasions, it may present or be identified in adulthood.
Case presentation
We report a case of a 35-year-old male, married for three years, who presented with primary infertility. Clinical examination revealed an empty scrotum, with neither testis palpable in the scrotum nor the inguinal canals. Semen analysis revealed azoospermia. Ultrasonography demonstrated the presence of both testes in the right iliac fossa in relation to the external iliac vessels, and the diagnosis was subsequently confirmed on magnetic resonance imaging (MRI). Although most cases of TTE are associated with additional anomalies—most commonly inguinal hernia—our patient had no associated anomalies which may have contributed to the late presentation in adulthood, as medical attention for undescended testes was not sought earlier by the patient.
Conclusions
TTE is an uncommon condition and is most frequently diagnosed in childhood. Presentation in adulthood with infertility is exceptionally rare. This case highlights a delayed presentation of TTE with infertility. This case report further emphasizes the role of ultrasound and MRI in establishing an accurate preoperative diagnosis.

Keywords: Transverse Testicular Ectopia, Undescended Testes, TTE, Infertility, Azoospermia.

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Background

Transverse testicular ectopia (TTE) is a rare congenital anomaly also known as crossed testicular ectopia. It is frequently associated with other abnormalities including persistent Müllerian duct syndrome (PMDS), true hermaphroditism, inguinal hernia, hypospadias, pseudo hermaphroditism, and scrotal anomalies (1, 2). TTE is an uncommon developmental condition, occurring in approximately one out of every four million patients (1). There is scarce data available on the incidence of TTE with intraabdominal cryptorchidism due to the rarity of combined pathology (3). TTE most commonly presents in childhood with an empty or hypoplastic scrotum and undescended testes with or without associated inguinal hernia. The majority of cases are identified before 4-years of age (4). A recent literature review of 129 cases of TTE reported a median age at presentation of 24 months, with inguinal hernia present in approximately 71% of patients (5). Patients may occasionally present with acute complications such as an obstructed inguinal hernia, necessitating emer-

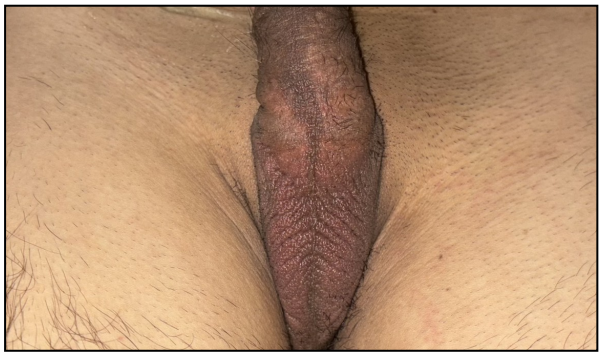

Figure 1: Clinical photograph showing an empty scrotal sac.

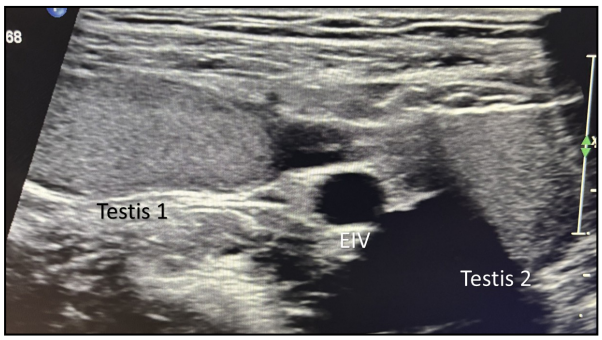

Figure 2: High-resolution ultrasonography of the right iliac fossa showing both testes (red arrows). One testis is located lateral to the external iliac vessels, while the other is located medial to the external iliac vessels.

gency surgical intervention. Presentation in adulthood as primary infertility, as seen in our patient, is exceptionally uncommon. Previously, the diagnosis of TTE was usually made intraoperatively. However, with the advent of advanced imaging modalities, the diagnosis is now commonly established preoperatively, most often using ultrasonography (USG) or magnetic resonance imaging (MRI) (5). USG is the first line imaging tool but has limitations in visualizing intra-abdominal testes. MRI is the imaging modality of choice in the diagnosis of TTE with a reported sensitivity of 82.4% and specificity of 100% (6). Untreated TTE is associated with complications which include testicular atrophy, infection, and malignancy (7). Therefore, neglecting or delaying the treatment can increase the risk of infertility with corresponding increase in chance of malignancy (8).

Case report

A 35-year-old male, married for three years, presented with primary infertility. There was no history of prior surgery or trauma. General physical examination was unremarkable. On local examination, the scrotal sac was empty (Figure 1). No testes were palpable within the scrotum or in either inguinal canal. There was no evidence of inguinal hernia on either side.
Semen analysis revealed azoospermia. Serum testosterone level was 7.74 ng/mL (reference range: 2.49–8.36 ng/mL). Baseline investigations including complete blood count, liver function tests, and kidney function tests were within normal limits.
A provisional diagnosis of bilateral undescended testes was made, and the patient was referred to the radiology department for further evaluation.
USG demonstrated both testes located in the right iliac fossa, in relation to the external iliac vessels (Figure 2). One testis was located anteromedial to the external iliac vessels, while the other was located antero-

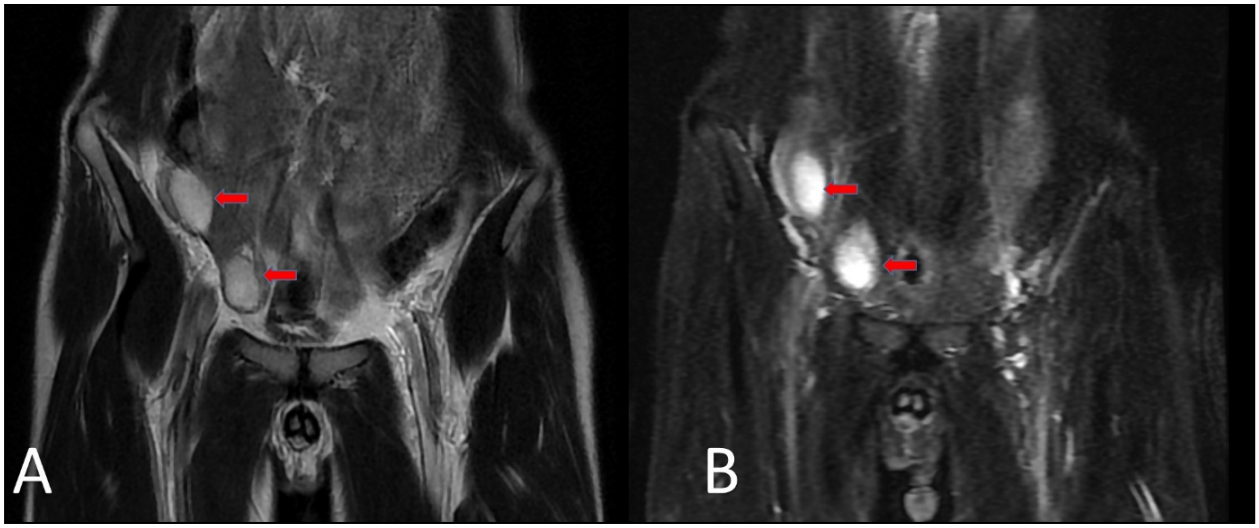

Figure 3: Coronal T2-weighted (A) and short tau inversion recovery (STIR) (B) MRI sequences demonstrating both testes within the right iliac fossa, indicated by red arrows.

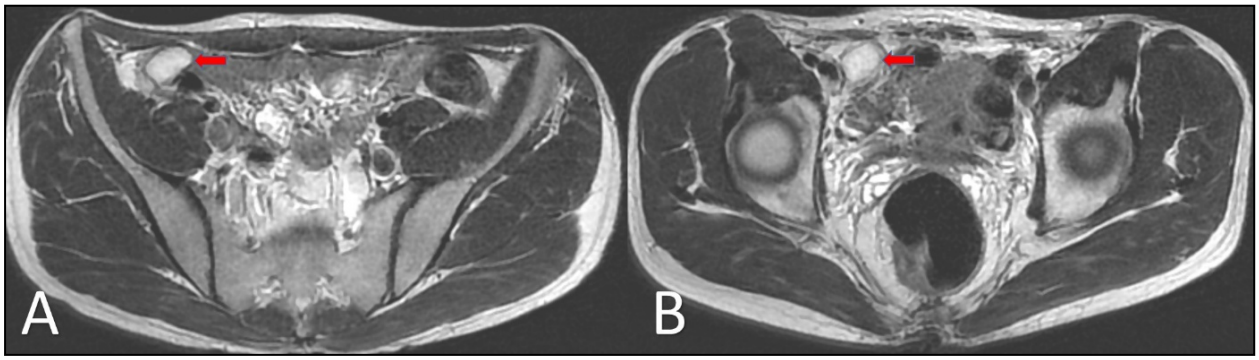

Figure 4: Axial T2-weighted MRI images of the lower abdomen showing two testes in the right iliac fossa. One testis is located anterolateral to the external iliac vessels, and the other is located medial to the external iliac vessels, indicated by red arrows.

lateral to them. The testicular volumes were reduced measuring 5.5 cc and 6.6 cc, respectively. Both testes showed normal echo texture, with no focal mass lesions identified. MRI was advised for further anatomical delineation which confirmed the presence of both testes in the right iliac fossa with similar anatomical relationships to the external iliac vessels (Figure 3, 4).
The urinary bladder, prostate, and seminal vesicles were normal. Both kidneys were normal. No persistent Müllerian duct structures were identified. Based on these findings, a diagnosis of TTE with bilateral undescended testes located in the right iliac fossa was made and patient was planned for surgery.

Discussion

TTE is a rare congenital anomaly with a reported prevalence of approximately 1 in 4 million (1). Embryologically, both testes develop intra-abdominally and descend into the scrotum. In TTE, one testis crosses the midline and descends through the contralateral inguinal canal, resulting in both testes being located on the same side, usually within one inguinal canal or hemiscrotum. The exact embryological mechanism underlying TTE is not fully known. However, a slew of proposed explanations have been put forth. It has been proposed that due to faulty development or fusion of the Wolffian ducts or owing to malformations of gubernacular or development of abnormal adhesions between the testes or deep inguinal ring obstruction, or displacement caused by persistent Müllerian duct structures both testes migrate to same side (8, 9). PMDS is thought to result from impaired formation or release of Müllerian inhibiting factor (MIF), resistance of target tissues to MIF, or deviation in the normal timing of MIF secretion. It seems plausible that the mechanical effect of the persistent Müllerian duct structures prevents the descent of testis or causes the both testes to migrate towards the same side producing TTE (5, 6). TTE is frequently associated with other congenital anomalies such as inguinal hernia, PMDS, and hypospadias. However, none of these associations were present in our patient.

The ectopic testis may be located in the contralateral hemiscrotum, within the inguinal canal, or at the level of the deep inguinal ring. An inguinal hernia is commonly present on the side to which the ectopic testis has descended. TTE on the basis of the presence of various other associated anomalies can be classified into 3 types (4, 5). Type 1 is accompanied only by inguinal hernia and accounts for approximately 40% to 50% of cases. Type 2 is accompanied by rudimentary or persistent Müllerian duct structures and accounts for approximately 30% of cases. Type 3 is less common, accounting for approximately 20% of cases and is associated with other genitourinary anomalies like hypospadias, pseudo hermaphroditism, and scrotal abnormalities.

Diagnosis of TTE is typically established through a combination of history, clinical examination and imaging, with USG and MRI playing a crucial role in precise localization and assessment of associated anomalies. The presentation in our case is exceptionally rare, as both testes were undescended and intra-abdominal, located in the same iliac fossa. The majority of cases of TTE are associated with additional anomalies, with inguinal hernia reported in approximately 71% of cases. However, our patient had no associated anomalies, which may have contributed to the delayed presentation, as

medical attention was not sought for the undescended testes by the patient.

The treatment of choice for TTE is surgical management, which includes mobilization and fixation of the testes (orchidopexy) along with correction of any associated inguinal hernia (9, 10). Surgery may be performed via an open inguinal approach, laparoscopy, laparoscopy-assisted inguinal approach, or laparotomy, depending on the location of the testes. Orchidopexy can be achieved using either a transseptal or transperitoneal approach. Transseptal orchidopexy also known as Ombredanne's technique is considered the preferred modality and is usually performed in conjunction with repair of hernia and correction of associated anomalies (10, 11). This surgical technique comprises of separating the ectopic testis from the cord structures and hernia sac, followed by transfer of testis to the contralateral hemiscrotum through the scrotal septum. In contrast, the transperitoneal approach involves placement of ectopic testis into the extraperitoneal space by crossing the root of the penis and fixing it in the opposite hemiscrotum (11). The ultimate goal of surgery is to position the testes in the most anatomically and functionally optimal location to avert any future complications. Surgical outcomes depend on the age at presentation, location of the testes, and degree of testicular parenchymal degeneration. Delayed or neglected treatment increases the risk of complications, including infertility and malignant transformation. It is reported that undescended testes carry an approximately six-fold increased risk of malignancy if left untreated. However, early orchidopexy particularly if performed before 12 years of age drastically reduces the risk of these complications (9, 10). But optimal outcomes are most often achieved when surgery is performed within the first year of life (5). Therefore, surgery should be performed as early as possible to optimize outcomes.

Conclusion

The present case highlights the delayed presentation of TTE in adulthood as primary infertility, an exceptionally rare manifestation of a condition that is most commonly present in pediatric age group. The diagnosis was initially suspected on clinical examination, established on USG, and subsequently confirmed by MRI, which located both testes in the right iliac fossa, thereby emphasizing the pivotal role of imaging in establishing an accurate preoperative diagnosis.

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Declarations

Consent for publication: The author clarifies that written informed consent was obtained and the anonymity of the patient was ensured. This study submitted to Swiss J. Rad. Nucl. Med. has been conducted in accordance with the Declaration of Helsinki and according to requirements of all applicable local and international standards. All authors contributed to the conception and design of the manuscript, participated in drafting and revising the content critically for important intellectual input, and approved the final version for publication. Each author agrees to be accountable for all aspects of the work, ensuring its accuracy and integrity.

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