

Case Report

Transverse Testicular Ectopia: A Rare Diagnosis during Herniotomy

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Abstract

Transverse testicular ectopia (TTE) is a condition characterized by migration of both testes towards the same hemiscrotum. The mean age of presentation is 4 years, with a typical presentation of a symptomatic inguinal hernia with contralateral impalpable testis. There is no reported predisposition to either left or right side. The diagnosis is usually intraoperative however, imaging modalities and diagnostic laparoscopy are useful in aiding pre-operative diagnosis. We present a case of a 5-year-old boy with a history of a painful swelling in the left inguinal region, with palpable left testis and contralateral empty scrotum. During the planned herniotomy in view of his pain, he was diagnosed with transverse testicular ectopia intra-operatively. Herniotomy on the left side with left testicular orchiopexy and transseptal fixation for the right testis was performed. He recovered well in the post-operative period. This rare congenital anomaly is often diagnosed intraoperatively as an incidental finding, and long-term follow up would be required due to the risk of malignant transformation.

Keywords: Hernia; Orchiopexy; Testicular ectopia; Undescended testes

1. Introduction

Transverse testicular ectopia (TTE) is a rare congenital anomaly where both testes migrates ipsilaterally towards the same hemiscrotum while the contralateral inguinal canal and hemiscrotum is empty. It was first reported by Von Lenhossek in 1886 as an autopsy finding [1]. Presentation is usually that of an inguinal hernia with ipsilateral palpable testis and an impalpable testis on the other side. Occasionally, two globular structures are palpable on the hernia side [2]. The classical description of TTE consists of two testes that are found on one side of the body while it is absent on the other side. They are identical in size and appearance, each with its own individual separate epididymis, vas deferens and testicular vessels. The vascular supply and the vas deferens of the ectopic testis is from

the appropriate side. On the side of the two testes, the processus vaginalis is patent while conversely, there is no hernia on the contralateral side [3]. The diagnosis of TTE is usually made intraoperatively as surgery is indicated for hernia and the rarity of TTE eludes a pre-operative diagnosis.

2. Case Presentation

A 5-year-old boy was referred by a general practitioner to the surgical outpatient department with history of a painful swelling in the left inguinal region since infancy. There were no other complaints related to this swelling. His younger brother was healthy and had no complaints. On examination of the inguinal region and the scrotum, there was positive cough impulse on the left side and the swelling was reducible. The left testis was palpable. The right side of the scrotum was underdeveloped and the testis was absent. Cough impulse on the right side was absent. With a clinical diagnosis of left inguinal hernia with right undescended testis, he was posted for a herniotomy in view of his pain. Intraoperatively, the hernia sac was identified and both testes unexpectedly delivered from the incisional wound. Each testis appeared normal with separate epididymis, vas deferens and testicular vessels (Figure 1). Both spermatic cords followed the left deep inguinal ring and entered the abdomen. The hernial sac was separated carefully and a herniotomy was performed. The left testis was secured in the left hemiscrotum. The right testis was fixed at the right subdartos pouch through the transseptal window using Vicryl 4/0 (Figure 2). There were no complications in the postoperative period and he was well on discharge. Ultrasound of the abdomen and pelvis done during the follow-up was normal.



Figure 1: Both Testis with separate cord structures. **Figure 2:** Both Testis fixed in each side of the scrotum.

3. Discussion

In utero, the descent of the retroperitoneal testis from the lumbar area (ventro-medial to the atrophied mesonephros) to the scrotum is attributed to the shortening of the gubernaculum. Cryptorchidism is defined as the failure of a testis to descend into its scrotal position, and this can be divided into acquired or congenital. Congenital cryptorchidism can be classified into either true undescended testes or ectopic testes. An ectopic testis descends through the external ring normally but is then diverted to an aberrant position. Testes that are situated in the superficial inguinal pouch are classified as ectopic but they should be considered as a variant of undescended testes [4]. Amongst the variants of ectopic testes, perineal ectopic testis is the most common. Other reported variants are femoral ectopic testis, pubopenile ectopic testis and TTE. TTE occurs when both testes migrate through a single inguinal canal towards a

common hemiscrotum. Besides TTE, the terms “transverse aberrant testicular maldescent, testicular pseudoduplication, and unilateral double testes” has been used in the literature to describe this similar condition [5]. Theories explaining the pathology of TTE include both testes being derived from the same germinal ridge, mechanical effect of persistent Mullerian duct structures preventing testicular descent or causing both testes to descend towards the same hemiscrotum, and defective gubernacular formation [6]. TTE is classified into three types according to the associated anomalies:

Type I: associated to inguinal hernia alone.

Type II: associated to persistent Mullerian remnants.

Type III: associated to other anomalies other than Mullerian remnants [2].

Persistent Mullerian duct syndrome with TTE has been reported in 20% to 49% of the cases [7]. Besides persistent Mullerian remnants, other associated anomalies that had been reported includes common vas deferens, seminal vesicle cyst and bilateral renal dysgenesis [5]. The ectopic testis carries an increased risk of infertility and a 5-10-fold increased lifetime risk of testicular neoplasia. It is typically characterized by a significant lack of germ cells, consequently infertility can often persist, even after early surgical intervention. It is interesting to note that there appears to be no impact on the testosterone levels despite the significant negative effect on fertility. Hence, the primary objective of early surgical management is to reduce the risk of testicular neoplasia as well as for cosmetic purposes [6].

Although most cases of TTE are diagnosed intra-operatively during a surgery for hernia or exploration for an undescended testis, pre-operative investigations such as ultrasonography (US), magnetic resonance imaging (MRI), arteriography, venography, computed topography [8] and diagnostic laparoscopy has been described to aid in the diagnosis. A pre-operative diagnosis allows assessment of the length of the spermatic vessels and the anatomy of the two vasa deferentia before committing to a particular mode of orchiopexy [7]. Arteriography and venography are invasive and requires general anaesthesia, a factor that needs to be taken into consideration when performed on paediatric patients. US has a sensitivity ranging from 82 to 88% in the detection of an impalpable testis [8]. A diagnostic laparoscopy has an additional benefit of allowing concurrent biopsies of gonads and Mullerian duct remnants to be taken [7]. Magnetic resonance venography (MRV) has a sensitivity of 100% with the main objective of localising the testicular vessels instead of imaging the testicular tissue. An approach in the work-up of the impalpable testis recommended by Lam et. al involves performing US as a first-line imaging modality due to its easy availability and lack of ionising radiation. If US findings are negative, MRI (which has a comparable sensitivity of 84%) is recommended as the next line of investigation, followed by MRV as the third line of investigation [8].

The main outline of the surgical management consists of fixation of the testes into the scrotum, searching for Mullerian duct remnants and other anomalies and long-term follow up to monitor for malignant changes. Surgical fixation can be accomplished by either a transseptal orchiopexy or extraperitoneal transposition of the testis. As Mullerian duct remnants rarely undergoes malignant degeneration, they should be managed conservatively if they

are encountered intra-operatively. However, there had been reported cases of malignant degeneration of Mullerian duct remnants [9, 10]. If the decision was made to remove the Mullerian duct remnants, extra precaution should be taken to ensure there is no vascular compromise of the vas deferens, which has a blood supply closely related to Mullerian derivatives. Apart from an open approach, either a fully laparoscopic approach or a laparoscopic-aided surgery had been reported to be successful in the management of TTE [7].

4. Conclusion

TTE is a rare congenital anomaly which should be suspected in a child during the clinical evaluation of an inguinal hernia with contralateral impalpable testis, as demonstrated in our case. TTE cases are incidental intra-operative findings during herniotomy. Surgical management which includes transseptal orchiopexy is indicated expediently to reduce the risk of testicular malignant changes, with long-term follow-up to monitor for such transformation.

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Conflict of Interest

The author(s) declared no potential conflicts of interest nor any funding involved in the publication of the article.

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