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Understanding Transverse Testicular Ectopia: Lessons from a Retrospective Analysis of 14 Pediatric Cases

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Abstract

Objectives: To comprehend the etiopathogenesis, clinical presentation and abnormalities associated with this rare congenital defect along with diagnostic modalities and effective management techniques for the treatment of TTE in children by conducting a retrospective analysis using clinical information from 14 cases over a period of 10 years.

Methods: Children with TTE were enrolled in this study that underwent treatment between January 2014 to January 2024 at the Institute of Mother & Child Health (IM&CH) PUMHSW, Shaheed Benazirabad, Nawabshah, Pakistan. Clinical data including age, clinical manifestations, TTE-associated anomalies, ultrasonographic findings, and surgical procedures was collected from medical records of all patients. Follow up was conducted up to a period of 6 months.

Results: All 14 cases were treated successfully using laparoscopic surgery. Most of the cases presented with Inguinal Hernia along with TTE and the diagnosis was made intraoperatively except for four cases out of which one was diagnosed during clinical examination of abdominally palpable testes, while ultrasonography was conducted as a baseline modality for investigating TTE. Follow up of all the cases was achieved for not more than 6 months.

Conclusion: TTE must be suspected in patients presenting with nonpalpable undescended testis and contralateral inguinal hernia. The contralateral processus vaginalis sac is found in all instances of TTE. Ultrasonography is necessary for a prompt diagnosis of TTE. Laparoscopic surgery is a secure, efficient, and minimally invasive treatment modality for successful correction of TTE.

Introduction

Transverse testicular ectopia (TTE) or crossed testicular ectopia (CTE) is a very rare form of urogenital anomaly in which both testes migrate and descend through a solitary inguinal canal. Both testes or one of them can be found ectopically in the abdominal cavity, inguinal canal, or descend into the hemi-scrotum, leaving the other hemi-scrotum empty [1]. It has been estimated that the rate of incidence is 1 in 4 million [2]. The etiology of TTE is yet to be defined. However, anatomical factors have been proposed as potential

causes or contributing factors to the failure of testicular descent, such as defective implantation, rupture or tearing of the gubernaculum, internal inguinal ring obstruction, formation of adhesions between the testis and surrounding structures, and delayed closure of the umbilical ring [3]. Clinically, it is usually described by the presence of an inguinal hernia on one side with either contralateral or bilateral cryptorchidism [4]. TTE usually occurs in association with other abnormalities such as Persistent Müllerian Duct Syndrome (PMDS), Disorders of Sexual Differentiation (DSD), inguinal hernia, hypospadias and scrotal anomalies [5]. On the basis of the presence of these associated abnormalities, TTE is classified into three types, Type 1– associated only with Inguinal Hernia (40% to 50%), Type 2 – associated with Persistent or Rudimentary Müllerian duct structures (30%), and Type 3 – associated with other abnormalities e.g. hypospadias, DSD and scrotal abnormalities (20%) [6]. Transverse Testicular Ectopia (TTE) gives rise to complications such as inguinal hernias, testicular torsion, infertility, and testicular tumors [7]. Patients with TTE are thus at an increased risk of developing gonadal malignancies such as embryonal carcinoma, seminoma, yolk sac tumor, and teratoma [4]. Often, the diagnosis of TTE is made during surgical procedures like orchiopexy, herniorrhaphy, or herniotomy [8]. Non-operative imaging methods, i.e. ultrasound (US), computed tomography (CT), emission computed tomography (ECT), magnetic resonance imaging (MRI), MR venography (MRV), and arteriography, are used to locate the undescended testis in patients who present with empty scrotum or impalpable testis [9]. The primary objectives of TTE treatment are to maintain fertility, reduce the risk of testicular neoplasia, treat congenital anomalies, hernias, and to perform orchiopexy. The surgery may be conducted by laparotomy, an inguinal method, a laparoscopy-assisted inguinal method, or laparoscopy [10]. TTE is surgically managed with herniotomy with trans-septal testicular fixation (Ombredanne's technique), contralateral fixation through a suprapubic subcutaneous tunnel, and staged procedure [11]. Postoperative follow-up of testicular position and growth is critical in TTE patients. In those with residual Müllerian structures, close follow-up is mandatory to avoid risk of development of malignancy [12].

Methodology

This retrospective study includes data gathered from cases of TTE treated at the Institute of Mother & Child Health (IM&CH), Shaheed Benazirabad, Nawabshah, Pakistan from January 2014 to January 2024. The patients mostly presented with Undescended Testes or Inguinal Hernia. Diagnosis was made intraoperatively in most cases but ultrasound was conducted as a baseline investigative modality in each case. The patients underwent laparoscopic orchiopexy or laparoscopy-assisted transseptal orchidopexy along with inguinal exploration depending on location of testes and viable tissue. A total of 14 cases were included in this study. The clinically relevant data i.e. age of patient, associated anomalies, diagnostic modalities, management and follow-up, was duly collected. Statistical analysis was performed with SPSS 25.0 for Windows to analyze this data.

Results

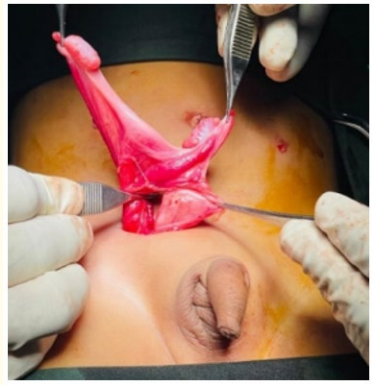

Figure 1: TTE with PMDS.

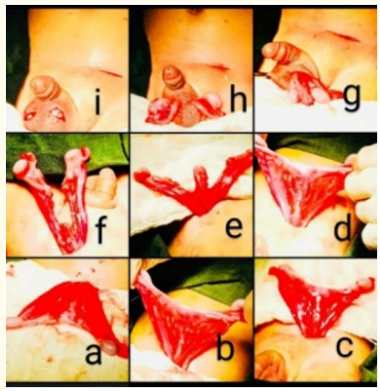

Figure 2: (a) finding at inguinal exploration. (b) \view of TTE with PMDS. (c, d, e, f) defines how dissection was done with the excision of PMDS. (g) both testes are taken down to the same side of scrotum. (h) trans septal orchiopexy (h) final appearance in their respective scrotal pouches.

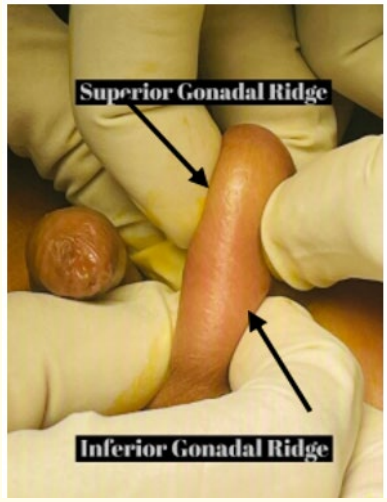

Figure 3: Palpable superior gonadal ridge and inferior gonadal ridge.

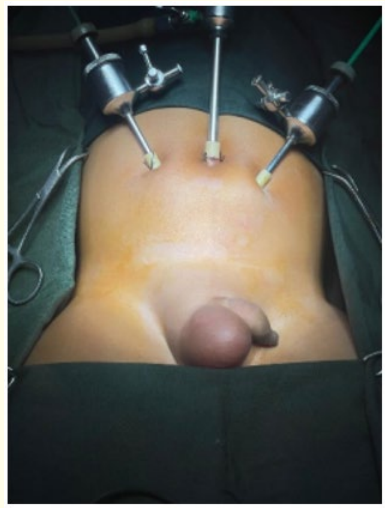

Figure 4: Laparoscopic Assisted Orchiopexy in a patient presenting with impalpable testes on left side and right sided inguinal hernia.

Case	*Age (months)*	*Side*	*Associated Anomalies*	*Diagnostic Modalities*	*Surgical intervention (Open/Laparoscopic)*	*Follow-up (months)*
1.	6	Right	Inguinal Hernia	Preoperative	Laparoscopic Transseptal Orchiopexy	2
2.	6	Left	Inguinal Hernia	Intraoperative	Inguinal Exploration for Hernia, Laparoscopic Transseptal Orchiopexy	3
3.	60	Right	Absent Testes	Intraoperative	Laparoscopic Orchiopexy	4
4.	8	Left	Inguinal Hernia	Intraoperative	Inguinal Exploration for Hernia, Laparoscopic Transseptal Orchiopexy	6
5.	9	Right	Inguinal Hernia	Intraoperative	Inguinal Exploration for Hernia, Laparoscopic Transseptal Orchiopexy	3
6.	48	Right	Absent Testes	Preoperative	Laparoscopic Orchiopexy	5
7.	12	Left	Inguinal Hernia with PMDS	Intraoperative	Inguinal Exploration for Hernia, Laparoscopic Transseptal Orchiopexy, and Laparoscopic Excision of PMDS	6
8.	12	Right	Inguinal Hernia	Preoperative	Inguinal Exploration for Hernia, Laparoscopic Transseptal Orchiopexy	6

9.	18	Right	Undescended Testes with Hypospadias	Intraoperative	Laparoscopic assisted Transseptal Orchiopexy	4
10.	20	Left	Undescended Testes with Hypospadias	Preoperative (on clinical examination)	Laparoscopic assisted Transseptal Orchiopexy	6
11.	23	Right	Undescended Testes with Hypospadias	Intraoperative	Laparoscopic Orchiopexy	6
12.	24	Left	Absent Testes with Micropenis	Intraoperative	Hormone Replacement Therapy, Laparoscopic Orchiopexy	6
13.	14	Right	Inguinal Hernia with PMDS	Intraoperative	Inguinal Exploration for Hernia, Laparoscopic Transseptal Orchiopexy, and Laparoscopic Excision of PMDS	5
14.	7	Right	Inguinal Hernia	Intraoperative	Inguinal Exploration for Hernia, Laparoscopic Transseptal Orchiopexy	4
TTE: Transverse Testicular Ectopia, PMDS: Persistent Mullerian Duct Syndrome.						

Table 1: The brief review of medical records of all cases of children with TTE.

Among the 14 patients, 8 cases (57.1%) were associated with inguinal hernia, out of which 5 cases of hernia (62.5%) were right-sided while the rest 3 cases (37.5%) presented on the left side. The median age of the patients was 13 months (IQR: 8 months, 23 months). A total of 14 boys were suspected of having TTE (100%) by physical examination and preoperative ultrasonography from which 10 of them (71.4%) were confirmed intraoperatively during laparoscopic exploration, 3 of them (21.4%) were confirmed via ultrasound while 1(7.1%) was confirmed by physical examination during abdominal palpation. The details of all 14 cases are listed in Table 1. All of our patients were referred to pediatric surgery department for undescended testis or a contralateral inguinal hernia. 8 cases presented with inguinal hernia (57.1%) and their confirmatory diagnosis was made during laparoscopic exploration of inguinal hernia. From these 8 patients that presented with hernia, 2 of the (25.0%) were found to be associated with PMDS. None of the patients in our study had a previous history of related operations. Out of the 14 patients included in this study,1 was diagnosed upon clinical examination and was found to have 2 testis like masses in unilateral inguinal scrotum and contralateral empty hemi scrotum upon palpation(as shown in Fig:3). All of the cases accepted Ultrasound (US) as preoperative assessment, which confirmed the position of testes in 3 cases. The definitive diagnosis was made intraoperatively in 10 (71.4%) cases. 8 cases out of 14 (57.1%) presented with inguinal hernia from which 2 (25.0%) were associated with PMDS. 3 cases (21.4%) were associated with hypospadias and 1(7.1%) case was associated with Micropenis. Diagnostic laparoscopy played a major role in all cases. The contralateral inguinal hernia was identified and repaired in the 8 cases that presented with inguinal hernia along with TTE. All of the cases underwent surgical treatment, using laparoscopic orchidopexy or laparoscopic assisted transseptal orchidopexy-inguinal exploration. Laparoscopic excision was used to correct PMDS in 2 cases. The post-operative follow-up period ranged from 2 to 6 months. 13 patients were found to have had testes in orthotopic position, bilaterally equal in size upon physical examination with normal vascular flow confirmed by US. Only in 1 case reduction in volume during follow-up was observed. No TTE-associated complications were found. Moreover, no tumor was found by urogenital and pelvic system US in all patients.

Discussion

TTE was first described by Von Lenhossek in 1886 [13]. This rare condition may present with a variety of clinical manifestations, usually inguinal hernia as well as associated anomalies such as PMDS, DSDs, and hypospadias [7]. TTE with PMDS was first described by Jordan in 1895 which is an uncommon form of internal male sex development in which there are Mullerian residues in an individual with a male phenotype [14]. The specific causative factors of TTE still remain unclear although few theories have attempted to explain its embryological background [10].

The contributing factors for this condition include the development of both testes from the same germinal ridge, early fusion of the developing Mesonephric (Wolffian) Ducts, adherence of testicles to Müllerian structures, and obstruction of the inguinal ring, which may result in failure of testicle to descend properly on the same side [14]. Additionally, experimental studies have proven the importance of the gubernaculum in the descent of the testes, where the abnormal attachment of the gubernaculum seems to be the widely accepted theory [5].

The mean age of presentation of TTE of the patients included in our study was 19 months, which is almost the same as the mean age from the cases in our literature review (16 months) [10]. It has been found that timely diagnosis and early detection of the associated anomalies is imperative in treating TTE effectively.

The most commonly observed clinical presentation of TTE in this study was Inguinal Hernia (57.1%) which aligns with findings reported by Gauderer (40-50%). Furthermore, the percentage of cases of TTE associated with PMDS in our data was 14.2% while the common predisposition for this condition according to the classification is 30%. This suggests that better understanding of the condition is required in order to treat cases of PMDS that occur in association with TTE. Three cases presented with undescended testes with hypospadias in our cohort which corresponds with the percentage for this type of TTE in the Gauderer classification, which is 21.4% and 20% respectively. In only 1 case out of the 14 included in this study, a patient presented with micropenis along with TTE which was treated with neo-adjuvant Hormone Replacement Therapy (HRT) because it aids in improving fertility outcomes, pubic hair growth induction and penile growth [15].

The vast majority of the cases included in this study (71.4%) were diagnosed intraoperatively. Ultrasound was used as a baseline screening test in all cases. One case from our study however, was diagnosed via Clinical Examination where the patient presented with empty scrotum/undescended testes and both testes were palpable abdominally on the same side (Figure 3). Thus, diagnosis of TTE can be made using physical methods, non-invasive investigations such as ultrasound and surgical methods such as laparoscopy.

The principles of treatment of TTE are oriented towards preserving fertility, correction of associated anomalies, repairing hernia, orchiopexy and reducing any possible risk of development of neoplasia. Surgical management may include laparoscopic transseptal orchiopexy-inguinal repair, laparoscopically assisted transseptal orchiopexy and laparoscopic excision of PMDS [16].

Laparoscopy plays a vital role in both diagnosis [17] and management of TTE along with its associated abnormalities. It facilitates the identification of persistent Mullerian duct structures, mobilization of testicles in case of orchiopexy and allows us to perform laparoscopic orchidectomy in case of intra-abdominal testicular atrophy [5]. It also allows evaluation of the shape, vas, and side of testes as well as assessment of testicular vessels of for length [10]. All of the cases included in this study were managed successfully via Laparoscopic surgical intervention.

Post-operative follow-up in TTE is highly imperative in keeping track of the location and development of the testes, especially in cases of TTE that are associated with PMDS due to increased risk of development of malignancy10,18. In our study however, a follow-up period of greater than 6 months could not be achieved for any patient due to lack of education and awareness [18, 19]. It is however, recommended that patients of TTE must adhere to lifelong follow-up which usually shows orthotopic, proportionate testes along with normal vascular flow. Case 10 from our study however, revealed reduced testicular volume after laparoscopic assisted transseptal

orchiopexy.

We acknowledge the limitations of this study by stating that follow-up was lost after 6 months because this cohort includes patients from a rural area in Pakistan, a developing country, where a vast majority of the population is deprived of basic education and does not have access to quality healthcare which is also the reason why our study included such a small number of cases. Nonetheless, the healthcare community in our country is doing its best to ensure that these patients are provided with the best quality of care by maximizing the use of available resources.

Ethical Approval

Ethical considerations for this study were granted by the Ethical Review Committee (ERC) of the Institute of Mother & Child Health (IM&CH) Shaheed Benazirabad, Nawabshah.

Conflict of Interest Statement

The authors declare that they have no known competing financial or personal interests that might influence the.

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