Crossed testicular ectopia: a case report and review of the literature

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Crossed testicular ectopia (CTE) is an extremely rare anomaly in which deviation of testicular descent results in unilateral location of both testes. It is usually associated with an inguinal hernia, with the spermatic cord of the ectopic testis originating from the appropriate side. Most often the diagnosis of CTE is not made until surgical exploration. We report a case of CTE in a 2-year-old boy who presented with left-sided inguinal hernia with concurrent nonpalpable right testis. Diagnosis was made during diagnostic laparoscopy followed by left open inguinal exploration during which high ligation of the left hernia and trans-septal orchiopexy, contralateral transposition tension-free for each testis was done. *Ann*

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Introduction

There are five known types of testicular ectopia: superficial, inguinal (interstitial), pubopenile, perineal femoral (crural), and crossed. Crossed testicular ectopia (CTE) is a rare congenital anomaly and is also referred to as unilateral double testis, testicular pseudoduplication, and transverse aberrant testicular maldescent. The testis may fail to descend completely along its normal path from the retroperitoneum to the scrotum (cryptorchidism). The abnormalities associated with CTE are persistent Müllerian duct syndrome, true hermaphrodites, inguinal hernia, hypospadias, pseudohermaphroditism, and scrotal anomalies. Herein, we present a 2-year-old boy with left inguinal hernia and contralateral nonpalpable testis, and review literature.

Case report

A 2-year-old boy presented with a swelling in the left groin and an empty scrotum on the right side. Family history was insignificant and examination of other systems proved normal. Physical signs showed left inguinoscrotal hernia with easily reducible content. External genitalia were of male type, and circumcision had not been performed. The right hemiscrotum was empty with normal-appearing urethral meatus. The left testis was palpated in the scrotum. The right testis was not visualized by ultrasonography. During laparoscopy, the right side was completely obliterated with no vas or gubernaculums. The right testis was not found in the posterior abdominal wall. On the left side there was a hernia that was exaggerated by the pneumoperitoneum with a testicle inside; however, its blood supply was coming from the right side. The testicle was reduced into the abdominal cavity and it was clearly originating from the right side; next, external pressure was applied to the hernia and the left testicle comes in view (Fig. 1). CTE was subsequently diagnosed (Fig. 2).

Biopsies were taken from a tissue that resembled a rudimentary uterus between the testicles. The two testes were of good size and were identical in appearance. Each had its own vascular pedicle and vas deferens. A biopsy was taken from the testes following herniotomy and high ligation (Fig. 3). The ectopic gonad was fixed to the opposite hemiscrotum through a trans-septal route (Fig. 4). As the cord structures had adequate length, no difficulty was encountered in the procedure. The patient made an uneventful recovery.

Discussion

CTE is an extremely rare congenital anomaly of testicular ectopia. More than 100 cases have been reported in the literature [1], as it was first described by Von Lenhossek [2], who suggested abnormal gubernaculum testis as a cause.

The mean age at presentation is 4 years. The clinical presentation generally includes an inguinal hernia on one side and a contralateral or sometimes a bilateral cryptorchidism [3,4] (Fig. 2). However, presentation with an obstructed inguinal hernia that cannot be reduced is known [5,6].Thus, it will be beneficial to keep this rare clinical entity in mind, in cases of incarcerated inguinal hernia with contralateral undescended testis.

Usually, the correct diagnosis is not made before surgical exploration, and in cases in which laparoscopic search is not the initial procedure the diagnosis is revealed during open herniotomy [4]. Preoperative diagnosis is possible using ultrasound [7], and the value of an experienced sonologist cannot be overstated. MRI is useful when the testis is not detected by ultrasound [8]; the only problem is the high cost.

Several authors have explored different theories to understand the development of CTE. One theory proposed the possibility of the development of both testes from the same genital ridge [9]; however, Kimura [10] stated that true CTE occurs only if there are two distinct deferent ducts, and a common duct suggests the development of the testis from one

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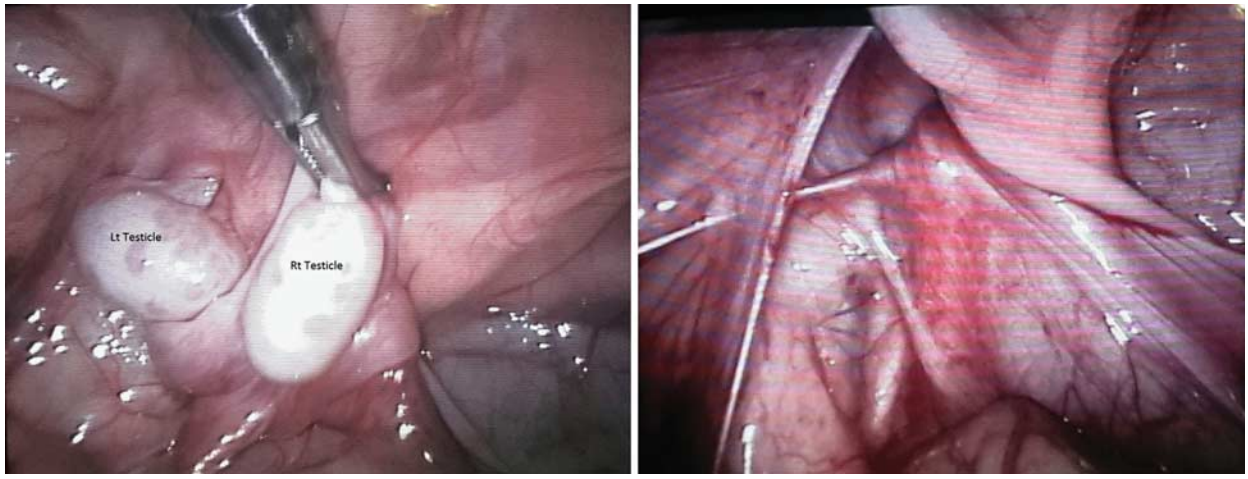
Fig. 1



Laparoscopic view of the crossed ectopic testis.

Fig. 2

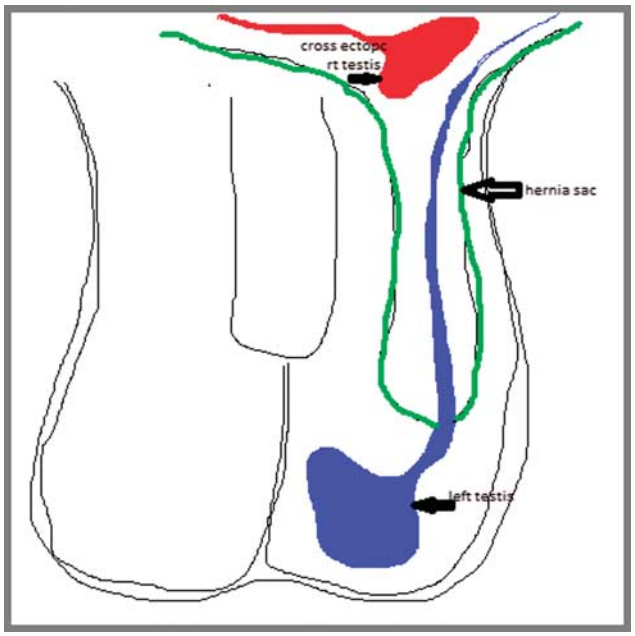
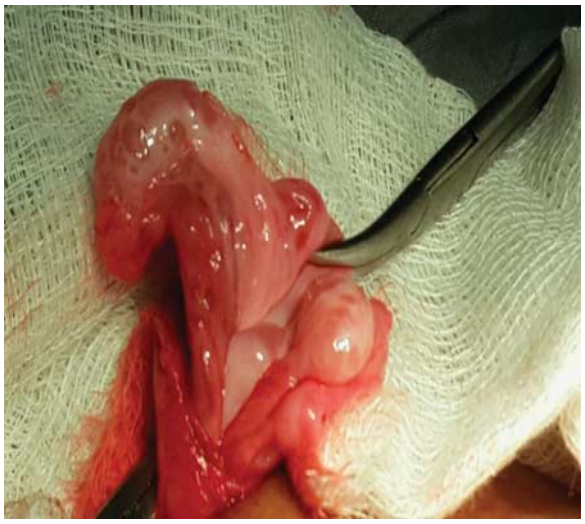


Illustration showing crossed ectopic testis.

Fig. 3



Showing the two testis after conversion to open left inguinal exploration.

genital ridge. Gupta and Das [11] postulated that earlier adherence and fusion of the developing Wolffian ducts might lead to the descent of one testis causing the second one to follow. Another theory states that in cases that present persistent Müllerian duct syndrome the crossed ectopia is caused by the traction exercised on the testis by the Müllerian duct. Frey and Rajfer [12] noted that defective ipsilateral gubernacular development might predispose to CTE.

CTE has been classified into three types: type 1, accompanied only by hernia (40–50%); type 2, accompanied by persistent or rudimentary Müllerian duct structures (30%); and type 3, associated with disorders other than persistent Müllerian remnants (inguinal hernia, hypospadias, pseudohermaphroditism, and scrotal abnormalities) (20%). According to that classification, our case was type 1 CTE. On extremely rare occasions, the vas deferens are fused, a situation that may hinder the testis from being placed into the scrotum during orchiopexy [13]. The CTE and testicular duplication should be differentiated. In case of duplication, both testicles arise from the same genital ridge, and they have a common deferent duct and blood supply [8].

In 2–97% of patients with CTE, disorders of the upper and lower urinary tract system have been reported [14]. Laparoscopy is useful for both diagnosis and treatment of CTE and associated anomalies [15]. Patients with CTE are at an increased risk of malignant transformation. The overall incidence of malignant transformation is 18% [16]. There have been reports of embryonal carcinoma [17], seminoma, yolk sac tumor [18], and teratoma [16].

The treatment of transverse testicular ectopia is focused on the detection of associated congenital abnormalities and

Fig. 4

Postoperative view of trans-septal orchiopexy.

placement of ectopic testicles into anatomical positions. This preserves fertility and allows monitoring for the development of malignancy. If two gonads come into view during exploration of one inguinal side, intraoperative intra-abdominal evaluation via mini laparotomy allows for the detection of Müllerian structures and genitourinary congenital abnormalities. A biopsy should be taken from tissue remnants between the cord structures or from the abdomen. There is no report of malignancy arising from the retained Müllerian structures. Therefore, routine hysterectomy is not recommended in patients who have obvious uterus and fallopian tubes. Extensive dissection of vas deferens and excision of persistent Müllerian duct structures should be avoided to prevent injury [19].

There were two options for orchiopexy – extraperitoneal orchiopexy and trans-septal orchiopexy – in the papers that reported 147 cases, which was the highest number reported [20,21]. In the extraperitoneal technique, the testis is brought to the contralateral hemiscrotum after its passing near the root of the penis. In the trans-septal technique, the testis should traverse the scrotal mediastinum to be fixed in it (Fig. 5).

Conclusion

CTE is a rare anomaly for which the pathogenesis remains unclear. The diagnosis should be suspected when there is unilateral hernia and concurrent contralateral cryptorchidism. In suspected cases, an appropriate ultrasonographic evaluation to rule out other potential abnormalities is indicated. Laparoscopy is valuable for both diagnosis and management of CTE and associated anomalies. Trans-septal orchiopexy is highly recommended to manage CTE.

Fig. 5

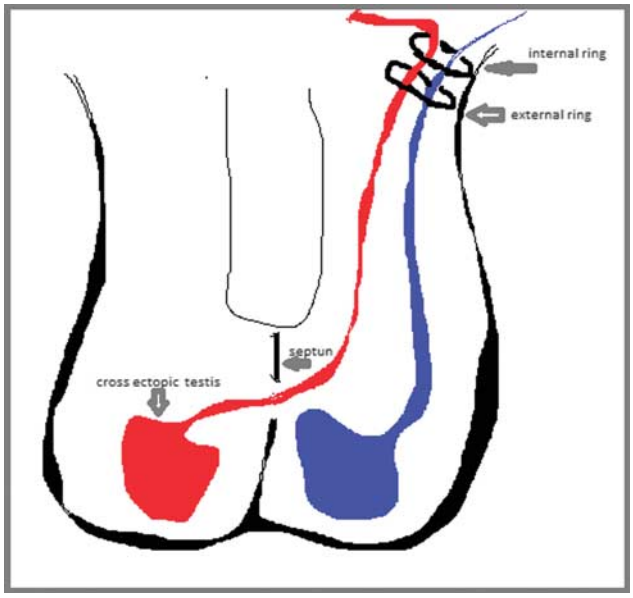


Illustration of trans-septal orchiopexy.

Acknowledgements

Conflicts of interest

There are no conflicts of interest.

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