Case Repot

Crossed Testicular Ectopia

Awad R. Abdalla*, Eltaib A. Saad

Department of Surgery, Faculty of Medicine, University of Khartoum.

Summary

Crossed testicular ectopia (CTE) is an extremely rare congenital anomaly, in which both testes migrate towards the same hemiscrotum through the same inguinal canal. Herein we report a case of a crossed testicular ectopia in a 6- month-old boy. The diagnosis was suggested clinically and supported by ultrasound findings. On groin exploration, both testes were found in the right inguinal canal each one had its own vas deferens and vascular pedicle. A trans-septal orchiopexy was performed by advancing the left testis via the midline scrotal septum into the left subdarots pouch. Literature was reviewed, and mechanisms that are postulated for this anomaly are presented.

***Corresponding author:** Department of Surgery , Faculty of Medicine, University of Khartoum.*
Email:awadrahama68@yahoo.com

Introduction:

Crossed testicular ectopia (CTE) or transverse testicular ectopia (TTE), is a rare congenital anomaly in which both testes migrate towards the same hemiscrotum via the same inguinal canal[1,2]. A few cases have been reported in the English medical literature (about 147 cases) since the first description by Von Lenhossek[3,4]. Many mechanisms have been postulated for this extremely rare gonadal anomaly[5-7]. However, the exact cause is still poorly understood[1]. Because of the rarity of this clinical occurrence; a definitive diagnosis can't be made preoperatively in most of the cases and the condition is usually encountered intra-operatively during groin exploration for an indirect inguinal hernia or undescended testis with an empty contralateral hemi-scrotum. We present a case of crossed testicular ectopia (CTE) in a 6-month-old boy who was brought by his parents with a left empty scrotum and a right groin swelling. Examination revealed an empty left hemiscrotum and two rounded, soft and mobile inguinal swellings which were consistent with testes in the right inguinal canal. A presumptive diagnosis of CTE was made and supported with ultrasound findings. Operative findings confirmed the diagnosis. He underwent a trans-septal orchidopexy. We reviewed the literature and described various mechanisms that are postulated for this anomaly.

Case presentation:

A 6- month- old Sudanese male was brought by his parents to the Pediatric Surgery Unit at Soba University Hospital because they noticed that he had a left empty scrotum and a right groin swelling during bathing. He was a product of a normal vaginal delivery at a rural hospital where there was no routine neonatal examination performed for him. He passed through normal milestones up to presentation time. Past medical history was significantly unremarkable. There was no history of undescended testis in the family.

On general examination, he was a well- looking boy with both weight and height within normal centiles according to his age and sex. The left hemiscrotum was well- developed but empty and no testis was palpable in the left inguinal canal. There were no other palpable swellings in the left lower abdomen, root of penis or the left upper medial thigh. Right groin examination revealed two separate, small (2X4 cm), rounded, soft and mobile inguinal swellings over the superficial inguinal ring with a testicular texture. Additionally,

a normally thickened spermatic cord was felt easily in the right groin. The findings were very consistent with a right palpable undescended testis (UDT) and a left crossed ectopic testis (CET). No cough impulses were noticed over both groins. A distal coronal hypospadias was also noticed. Systemic examination was essentially normal. A presumptive diagnosis of a crossed testicular ectopia with coronal hypospadias was made.

Ultrasound (US) examination revealed an empty left hemi-scrotum with no testis-like shadow in the left inguinal canal or intra-abdominally. No Mullerian duct remnants were seen in the left side. Right groin US showed two testes-like shadows in the right inguinal canal. No indirect inguinal hernias were seen in both sides. The child was booked for an elective trans-septal orchiodopexy after appropriate preoperative investigations.

He underwent right groin exploration under general anesthesia. A right inguinal incision was made and the right inguinal canal was opened. The right spermatic cord was delivered out and two, viable and normally-sized testes were found. Both testes had a (common) meso-orchium proximally, but each one had a separate vasa deferenes and its own vascular pedicle (Figure 1). They were almost equal in size. There was no associated right-sided indirect inguinal hernia. No Mullerian duct tissue remnants were found.A meticulous dissection was done to separate the two testicular tissues with preservation of their vasa deferentia and vascular pedicles. The right testis was fixed into the right subdarots pouch with Vicryl 5/0, while the left one was advanced through the midline trans-septum into the left subdarots pouch, under no tension, as the length of the left spermatic cord was fortunately enough to be advanced (Figure 2). The latter was fixed into the left subdarots pouch with Vicryl 5/0. The postoperative recovery was uneventful and the operative findings were explained to the parents.

Discussion:

Crossed testicular ectopia (CTE) is a rare gonadal anomaly in which both testes migrate towards the same hemi-scrotum via the same inguinal canal [1,2] It is considered as one of the rarest types of testicular ectopia[8]. The crossed ectopic testis can be found at different locations including contralateral hemi-scrotum, contralateral inguinal canal (as in the current case) and contralateral femoral triangle[9]. It usually occurs in the right side (as in the present case), however, left sided cases were also reported[10].

Several theories have been postulated to explain embryological basis of this anomaly[5- 7]; most of them were linked to the abnormalities of normal testicular descent with a special emphasis on the role of gubernaculum[11]. Berg had suggested that both testes arose from the same genital ridge[5]. In the same context, Gupta and Das[6] postulated that adherence and fusion of the developing Wolffian ducts takes place early on and that descent of one testis causes the other one to follow it toward the same hemiscrotum[6]. However, Gray and Skandalakis[7] suggested that each testis is arising from the ipsilateral genital ridge and the crossing-over occurs during testicular descent. This theory is supported by the observation that, in most of the cases, each testis has its own vas deferent and vascular pedicle[7]. Nevertheless, the exact mechanism is still unknown[1].

The condition is associated with a wide variety of congenital anomalies. Accordingly, a classification system has been proposed in the literature[12] and it was based on the type of the associated anomalies[12]. Type I is the most common type (40-50%) and it is associated with indirect inguinal hernia; Type II (30%) is associated with persistent Mullerian duct tissues; and Type III (20%) is associated with hypospadias, pseudo-hermaphroditism or scrotal anomalies[12]. Our reported case belongs to the latter type as it was associated with a distal coronal hypospadias.

Most cases of CTE are encountered incidentally during groin exploration for undescended testes or repair of indirect inguinal hernias with an empty contralateral hemiscrotum[10,13]. The clinical presentation is generally with unilateral (or bilateral)

cryptorchidism or indirect inguinal hernia in the vast majority of cases[13]. A very rare presentation with an incarcerated irreducible indirect inguinal hernia was reported in the literature[14]. Nevertheless, the diagnosis can be barely established on clinical examination alone as there is always a wide range of differential diagnoses that include: testicular duplication; hydrocele of the cord; spermatocele ; Morgagni cyst; and possibly testicular tumors[15].

Ultra-sonography is a good initial diagnostic investigation that can be used to assess the condition[15]; however, MRI is more superior in delineating the anatomy for preoperative localization of the testes and detection of Mullerian tissue remnants (if they are present), but it is rarely used[15]. Laparoscopic search is considered the gold standard for both diagnosis and management of this condition[16].

The aim of the surgical management of CTE is fixation of testes into the scrotum and to search for Mullerian duct remnants[15]. In addition, long-term follow up is advised by many authors due to the increased risk of associated testicular cancers[10] as testicular malignancy has been reported in some cases, especially when cryptorchidism was an association[15]. A variety of procedures have been described, including a staged- surgery to bring the ectopic testis into its correct canal, trans-septal orchidopexy and extra-peritoneal transposition[15].

In conclusion, crossed testicular ectopia is an extremely rare anomaly that is encountered during the surgical exploration for an indirect inguinal hernia or undescended testis with an empty contralateral hemiscrotum. Various associated anomalies have been reported and a classification system is established based on the type of these associated anomalies. Trans-septal orchidopexy is a common approach for the management of this condition while the laparoscopic search is the gold standard for both diagnosis as well as management.

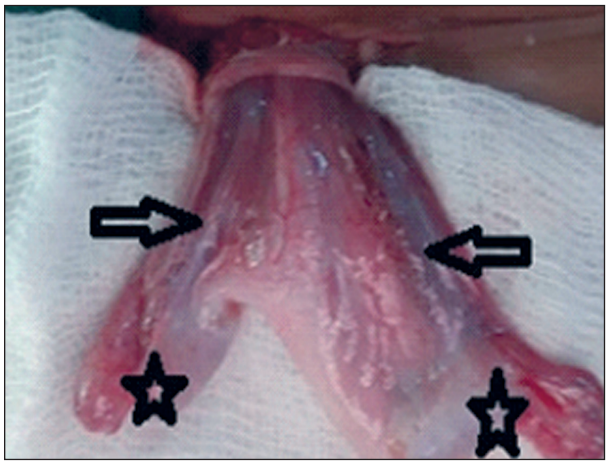

Figure 1. The right spermatic cord delivered out with two testes (stars) and their vas deferenti and vascular pedicles (horizental arrows).

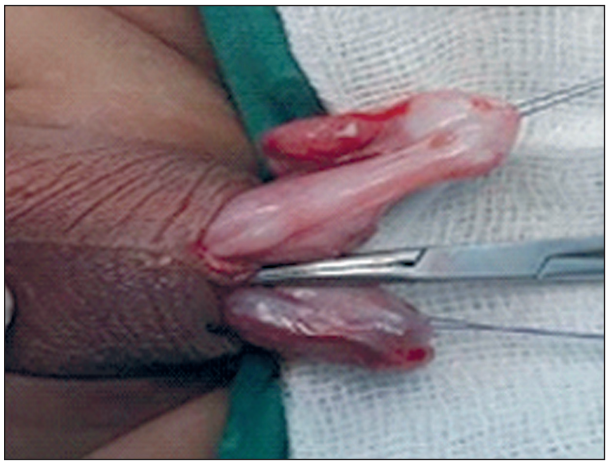

Figure 2. Showing fixation of both testes into their corrsponding subdarots pocuhes with advancement of the left one throught the midline scrotal septum.

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