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## CASE REPORT

# Crossed testicular ectopia – A rare anomaly: Case report and review of the literature

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### Abstract

A relatively uncommon urogenital anomaly known as crossed testicular ectopia (CTE) can be defined by the emigration of one testis into the opposing inguinal canal, which could possibly be ectopic in the abdominal cavity, inguinal region, or even descend to the opposing hemi-scrotum. Following a review of the literature, the most common classification is to subdivide CTE into three categories based on associations: (a) which tends to be associated with inguinal hernia alone; (b) which is predominantly associated with persistent mullerian remnants; and (c) which is most frequently associated with other anomalies without mullerian remnants. We describe a rare CTE case in a 2-year-old kid who presented with a nonpalpable right testis, an empty right hemiscrotum, and a MRI that showed both testicles were located in the left inguinal canal. Laparoscopy was used to make the final diagnosis, which was followed by a left inguinoscrotomy with transseptal tension-free orchiopexy. The cornerstone of surgical care consists of proper testicular fixation into the scrotum, a search for other anomalies such as mullerian duct remnants, and long-term follow-up due to the potential risk of malignancy.

**Keywords:** Orchiopexy, Testicular ectopia, Undescended testis

## 1. Introduction

Approximately 10 % of extrascrotal testes are ectopic, given that the testes are misdirected during their final descent and end up in an unusual place [1].

This extremely uncommon condition is also reported as unilateral double testis, testicular pseudoduplication, unilateral transverse testicular ectopia, and transverse aberrant testicular maldescent in the literature.

Although the most common ectopic location is the superficial inguinal pouch (interstitial) [2], several forms of testicular ectopia are known: femoral (crural), perineal, pubopenile, and crossing [3,4].

The average age at which testicular ectopia reveals itself is 4 years [5]. And, most of the time, this condition gets discovered at the time of a herniotomy or during surgery for an undescended testis [6,7].

We presented a 2-year-old kid who had been referred to us at Al-Galaa teaching hospital with an

empty right hemi-scrotum, and a MRI results revealed that both testicles were within the left inguinal canal.

## 2. Case report

### 2.1. Patient information

A 2-year-old male kid presented by his parents to the pediatric surgery clinic with a referral report from the outpatient clinic as a case of an ectopic right testicle within the left inguinal canal documented by MRI report revealed that the two testicles are within the left inguinal canal (Fig. 1), despite the fact that MRI is not routinely performed in such cases in our unit.

### 2.2. Clinical finding

The general examination revealed no signs of systemic abnormalities. The right hemiscrotum was empty, and an examination of the right inguinal

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Fig. 1. MRI finding tow testicles are within the left inguinal canal.

area revealed no palpable testis. Only one palpable testis was detected in the left scrotum upon examination. There had been no previous history of bulging, indicating a left oblique inguinal hernia. The external genitalia were of the male type, and circumcision had not been performed (Fig. 2). The previous medical and surgical history was negative.

### 2.3. Diagnostic assessment

The patient was referred for an ultrasound, which revealed an empty right scrotum, the left testis within the left scrotal sac, and the right testis at the proximal end of the left inguinal canal, with both testicles measuring  $15.6 \times 6.5$  and  $15.5 \times 6.8$  mm.

### 2.4. Intervention

First, a laparoscopy was performed, and the right side was completely obliterated with no vas or



Fig. 2. Normal left scrotum and empty right scrotum.

gubernaculum. The posterior abdominal wall did not contain the right testis. On the left side, there was a hernia sac with two testes of identical size separated by epididymides and vas deferens (Fig. 3).

The left inguinoscopy was then performed by a left transverse inguinal incision in the lower abdominal crease. The left testicle was discovered with its vas deferens and blood vessels flowing from the left side after adequate dissection. The blood vessels and the vas of the right testicle originated on the right side and crossed the bladder anteriorly till the left inguinal ring, with no gubernaculum attached to the ectopic testis (Fig. 4).

The left testis was fixed in the left scrotum pouch. However, during dissection, the right testicular spermatic cord was discovered to be long enough to allow transseptal orchiopexy without tension. As a result, the ectopic right testis was fixed to the contralateral hemiscrotum via a transseptal route (Fig. 5), incisions were closed, and recovery went off without issues.

### 2.5. Follow-up and outcomes

The patient's postoperative recovery is uneventful, with no postoperative complications, and he is discharged home at night.

## 3. Discussion

Crossed testicular ectopia (CTE) which is a form of congenital testicular ectopia is an incredibly uncommon anomaly and affects only one in every four million children [8].

In the first description, Von Lenhossek 1886 identified anomalous gubernaculum of the testis as a reason, and since then, around 260 cases of CTE have been observed in the literature [9,10].

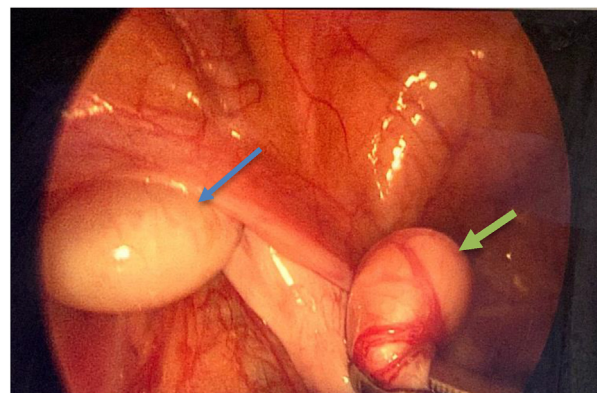


Fig. 3. Laparoscopic finding, the blue arrow pointing to the left testicle, and the green arrow pointing to the ectopic right testicle.

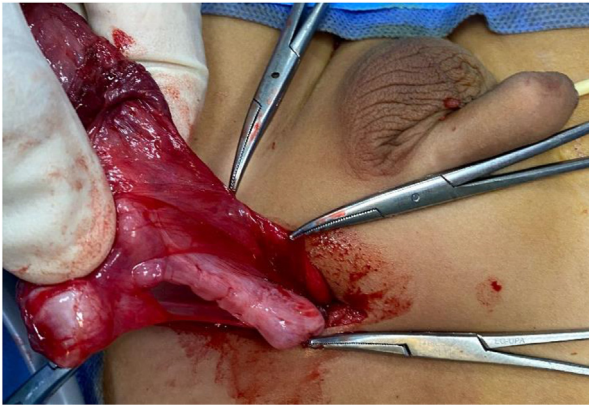


Fig. 4. Surgical finding from left inguinoscrotal hernia repair.

We found three types of CTE after reviewing the literature: type 1: presents with hernia only (40–50 %), type 2: presents with CTE and persistent müllerian duct remnants (30 %), and type 3: presents with CTE and other disorders such as hypospadias, true hermaphroditism, pseudohermaphroditism, and scrotal anomaly [11].

In one study done by Yöldöz and colleagues reported six cases of CTE within the period of October 2001 to January 2008. All of them presented as a case of inguinal hernia and four of them diagnosed at the time of surgery. To the best of the information we have, this is considered one of the largest data sets exhibiting this rare phenomenon [12].

Although several hypotheses have been developed to explain the initial cause of CTE, there is no general agreement on the causation. CTE is thought to be developed because of defects in gubernacular development, as well as putative variables such as testicular adhesion to müllerian structures or a fusion of Wolffian ducts [13].

Some cases, particularly if accompanied with other congenital anomalies or ambiguous genitalia, karyotyping may be required; in cases of true CTE, the karyotype is always 46XY [14].



Fig. 5. The left scrotum with both testes before fixation in each hemi-scrotum.

Laparoscopy has been used for both the evaluation and management of these instances, and even a combined approach of an inguinal technique assisted by laparoscopy, as we did, has been documented [15].

Both testes were found in the same inguinal canal in our case. Other researchers have discovered them in the same hemiscrotum. The most frequently finding is ductus deferens fusion. However, duct duplication has also been documented. The primary goal of surgical management is to fix the testes into the scrotum, search for müllerian duct remnants and other anomalies, and provide long-term follow-up due to the risk of malignancy.

Patients who have CTE are more likely to develop malignancy. The overall incidence of malignant transformation is 18 % [16], which is higher than the risk of undescended testicular cancer, which tends to vary with gonad location: 1 % with inguinal testes and 5 % with abdominal testes [17,18]. They are also at risk for fertility problems that require long-term follow up [11].

For CTE surgical management, there were two options: extraperitoneal orchiopexy or transseptal orchiopexy, which we carried out in our case in point. A long transseptal orchiopexy (Ombredanne's technique), is the preferred surgical technique for treating CTE. The blood flow to the vas deferens and testicles must be maintained as part of the therapeutic intervention strategy for this technique [19,20]. Bascuna et al. [21] established the scheme for the above technique, which Raj et al. [22] modified.

### 3.1. Conclusion

CTE is an extremely uncommon anomaly with no confirmed cause. Multiple related anomalies are evident as scrotal problems and hypospadias, müllerian duct remnant, and karyotyping is a crucial clue to exclude in these patients. It is anticipated that keeping this uncommon clinical entity in mind will be advantageous in cases of bilateral undescended testis or unilateral impalpable undescended testis, as in our case. When there is a unilateral hernia and a concurrent contralateral impalpable undescended testis, the diagnosis of CTE should be suspected. In possible scenarios preoperative imaging studies such as ultrasonography performed by qualified personnel can provide as much detail as regarding the location of both testicles, as well as an evaluation to rule out other potential anomalies and determine the best surgical treatment option. Laparoscopy is useful for both evaluation and management of CTE and related anomalies. However, if the spermatic cord of the ectopic testicle has been determined to be long

enough to allow transseptal orchiopexy without tension, transseptal orchiopexy is strongly advised to manage that CTE. Karyotyping may be required in some situations, particularly if it is coupled with other congenital anomalies of the genitalia. Patients frequently require long-term follow-up since they may have future problems with fertility and are at a greater risk of developing testicular malignancy.

### Patient consent

An informed written consent was taken from the family for operating, reporting this rare case, and the accompanying images.

### Author's contribution

1- Mohammed R. Balaswad: corresponding author, admitted and followed the patient, operated on, reporting the case, and the patient data recording, drafting the manuscript and Reviewing the literatures.

2- Wael Ghorab: co-author, operating on the patient, as a head of clinical team with clinical supervision.

3- Shrief Mebed: co-author, operating on the patient, clinical supervision, and guidance.

All authors have read and approved the manuscript.

### Conflicts of interest

There are no conflicts of interest.

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