Case Report

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Crossed testicular ectopia: Report of six cases

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ABSTRACT

Crossed testicular ectopia or transverse testicular ectopia is an extremely rare anomaly characterised by migration of one testis towards the opposite inguinal canal, usually associated with unilateral inguinal hernia. This report describes six cases of crossed ectopic testes, one of the largest series, and with unusual clinical histories.

Key words: Crossed testicular ectopia, transverse testicular ectopia, undescended Testis

INTRODUCTION

Crossed testicular ectopia or transverse testicular ectopia (TTE) is an extremely rare anomaly characterised by migration of one testis towards the opposite inguinal canal, usually associated with inguinal hernia. In most reported cases, the accurate diagnosis was not made before surgery. Treatment modalities include laparoscopic and surgical procedures. Unlike the undescended testis, which occurs in approximately 1% of male children, it has been noted that fewer than 100 cases have been reported in the world literature.[1] The typical presentation of crossed testicular ectopia is ipsilateral inguinal hernia and contralateral undescended testis.[2] In this manuscript, we report six cases to highlight the diagnosis of crossed ectopic testis in undescended testis cases with unusual clinical findings.

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CASE REPORTS

Case 1

A 5-year-old boy was admitted to our clinic with bilateral undescended testes and right inguinal hernia. On physical examination, both testes were palpated in the right groin region and ultrasound (US) supported this finding. Transverse incision of the right inguinal region visualized both testes at the level of the external inguinal ring [Figure 1]. After the hernia sac was prepared and tied, both testes were brought down sequentially into the ipsilateral and contralateral hemiscrotums with transseptal incision [Figure 2].

Case 2

A 6-year-old boy presented with bilateral undescended testes and left inguinal hernia. On physical examination, the left testis could be palpated at the inguinal region, but the right testis was impalpable and was not visualised by US. At surgery, a transverse incision of the left inguinal region was performed, and the left testis was found at the level of the external inguinal ring. During the preparation of the hernia sac, the right testis was also found in the sac. Similarly, both testes were brought down to both hemiscrotums with transseptal incision [Figure 3].

Case 3

An 8-year-old boy presented with a left impalpable testis and right inguinal bulging. On physical examination, the right testis was palpated in the swollen right inguinal region. The left testis was neither palpated nor visualised by US. At surgery, transverse incision of the right inguinal was made. The right testis was found at the level of the external inguinal ring, and the left testis was found in the hernia sac. In addition, the ductus deferens of the testes was fused proximally. After the sac was prepared and tied, the same surgical technique was successfully performed [Figure 4].

Case 4

A 2-year-old boy was admitted to the clinic with bilateral non-palpable testes and a previously repaired



Figure 1: Intraoperative appearance of Case 1. Right and left testes with funicular elements emerging from the right inguinal region

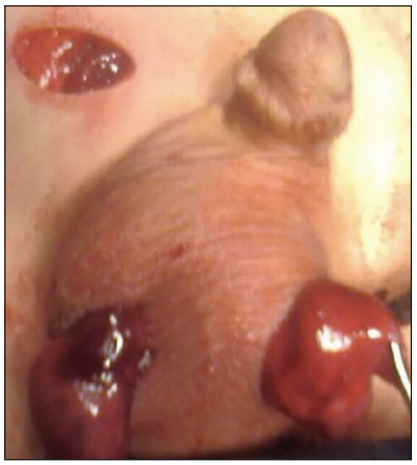


Figure 2: Intraoperative appearance of Case 1. Scrotum after transseptal orchiopexy

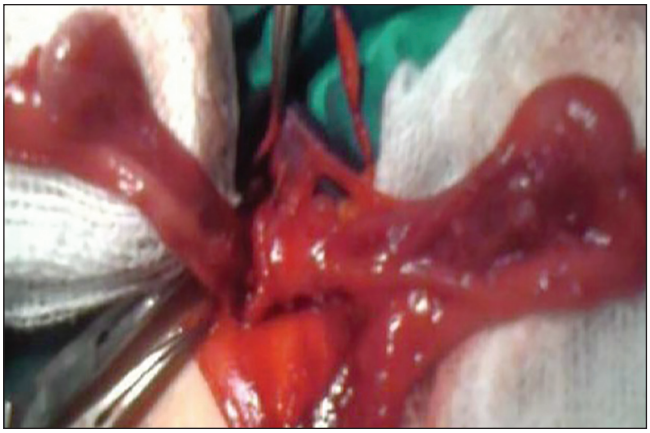


Figure 3: Intraoperative appearance of Case 2. Right and left testes with funicular elements emerging from the left inguinal region

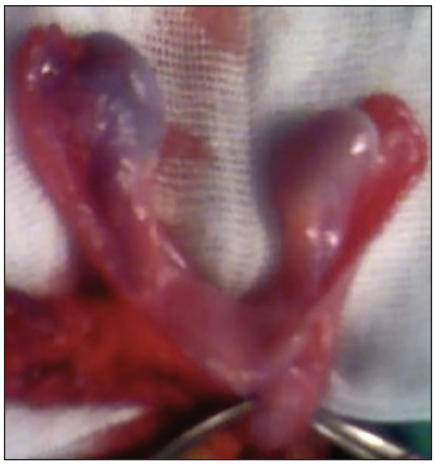


Figure 4: Intraoperative appearance of Case 3. Fused ductus deferens

left inguinal hernia. On physical examination, both testes were non-palpable, the scrotum was hypoplastic and there was a left inguinal transverse inguinal scar. The bilateral testes were visualised in the right inguinal canal entrance on US. During laparoscopy, both testes were visualised near the internal inguinal canal entrance in the abdomen. At operation, the right transverse incision was made and both testes were found. There was no cord abnormality or remaining Mullerian structures. Multiple cystic appearance was present on the surface of the testes and hence a testis biopsy was performed. After the hernia sac was tied, bilateral transseptal orchidopexy was done. Histological examination revealed testis tissue.

Case 5

A 3-year-old boy was admitted to the clinic with a left impalpable testis and a right hydrocele. On physical examination, the left testis was not palpable and there was an inguinal undescended testis with hydrocele. US confirmed the physical examination findings. During laparoscopy, the left testis was visualized close to the internal ring. The spermatic cord and vessels were stayed from the right internal orifice to the left, and connection anomalies between the testes and these structures were noted. Remaining Mullerian anomalies were not established, and a biopsy was not taken from the testes because visual inspection revealed completely normal testicular appearance in both testes. After the hernia sac was tied, a bilateral transseptal orchidopexy was done.

Case 6

A 3year-old boy was referred to the clinic with ambiguous genitalia and bilateral undescended testes. At the rightinguinal transverse incision, twotestes were found on the ipsilateral side, each having its own fused ductus deferens. exploration was performed laparoscopically and neither the left testis nor the left cord structure was found in the abdomen Transseptal orchidopexy was performed.

The medical records of all patients with the diagnosis of TTE were analysed with respect to age, side of TTE, mode of presentation, localisation of testes on physical examination, associated anomaly, localisation of testes on USG and at laparoscopy; and the operation performed are listed in Table 1.

DISCUSSION

The migration of the testes to an ectopic site, such as caudal to external ring, suprapubic, femoral, perineal and penile, has been reported. Crossed testicular ectopia is a rare anomaly of testicular migration wherein a testis descends into the opposite hemiscrotum. Typical clinical history is an inguinal hernia with contralateral undescended testis. Mean age at presentation is 4 years.[3] The patients in this study were much older than the usual presentation age with a mean of 5.2 years. This could be due to the low socioeconomic status of the region, which may have resulted in delayed awareness of the condition and delayed admission to the hospital.

The diagnosis of crossed testicular ectopia is often made intraoperatively during inguinal hernia repair. Only one out of six cases in this study was pre-operatively diagnosed TTE by ultrasonography (USG). The diagnostic clinical criteria consist of two testes located in the same hemiscrotum or inguinal canal with an empty contralateral hemiscrotum; however, as demonstrated by this present series, there are many different presentation modes.

Gauderer *et al*.[4] described a classification system for crossed testicular ectopia based on the presence of associated abnormalities: Type 1, the most common type (40-50%), is associated with inguinal hernia alone; Type 2 (30%) is associated with persistent or rudimentary Mullerian duct structures; and Type 3 (20%) is associated with other genitourinary abnormalities without Mullerian remnants. Mullerian duct remnants, duplication of ductus deferens and other anomalies have also been reported. Cases 1, 2, 4 and 5 can be classified as Type 1 and Cases 3 and 6 as Type 3, due to the presences of fused ductus deferentes. None of the cases was Type 2; therefore, contrary to current belief, Type 2 may account for less than the suggested 30% of all crossed ectopic testes.

Recently, USG, laparoscopy, and magnetic resonance imaging (MRI) have been used for the diagnosis of this condition.[5-7] However, none of the patients in this series underwent MRI. The aim of the treatment is fixation of the testes into the scrotum and exploration for ductus deferens anomalies, Mullerian remnants and subsequent malignancy. In one study, yolk sack tumour had developed on a patient who presented with transverse ectopic testes.[8] Transseptal orchiopexy was performed in all the patients in this present study, which is usually the preferred surgical option for fixing the testes into the scrotum. Laparoscopy may be beneficial whether Mullerian remnants and ductus deferens anomalies are present and may allow evaluation of contralateral hernia. Laparoscopic guide can be helpful, while performing the transseptal orchidopexy.[9] During exploration, two cases demonstrated ductus deferens anomaly, but this did not complicate the surgery. Mullerian duct remnants were not observed. However,

Table 1: The medical records of all patients with the diagnosis of TTE were analysed with respect to age, side of TTE, mode of presentation, localisation of testes on physical examination, associated anomaly, localisation of testes on USG and at laparoscopy; and the operation performed

Age	Side	Presentation	Localisation of testes (physical examination)	Associated anomaly	Localisation of testes (USG)	Localisation of testes (laparoscopy)	Operation
5	Right	Bilateral undescended testes	R: Right inguinal	None	R: Right inguinal	Not performed	Transseptal orchidopexy
		Right inguinal hernia	L: Right inguinal		L: Right inguinal		
6	Left	Bilateral undescended testis	R: Non-palpable	None	R: Not visualised	Not performed	Transseptal orchidopexy
		Left inguinal hernia	L: Left inguinal		L: Left inguinal		
8	Right	Right inguinal swollen	R: Right scrotum	Fused ductus deferens	R: Right scrotum	Not performed	Transseptal orchidopexy
		Left undescended testis	L: Non-palpable		L: Not visualised		
2	Right	Bilateral undescended	R: Non-palpable	None	R: Not visualised	Both testes at right internal ring	Laparoscopy assisted transseptal orchidopexy
		Left inguinal. Hernia	L: Non-palpable		L: Not visualised		
3	Left	Right hydrocele-recurrent	R: Right scrotum	Fused testis	R: Right scrotum	Left testes at internal ring	Laparoscopy assisted transseptal orchidopexy
		Left undescended	L: Non-palpable		L: Not visualised		
3	Right	Bilateral undescended	R: Right inguinal	Ambiguous genitalia, fused ductus deferens	R: Right inguinal	No left testes observed	Transseptal orchidopexy-inguinal exploration
		Right inguinal hernia	L: Non palpable		L: Not visualised		

TTE: Transverse testicular ectopia; USG: Ultrasonography

laparoscopy was performed on three of the six patients, which might explain why Mullerian remnants were not detected.

In the literature, the usual presentation of crossed testicular ectopia is ipsilateral inguinal hernia and contralateral undescended testis, whereas in this series, four out of the six patients admitted with bilateral undescended testes.[10] To the best of our knowledge, no more than 10 cases in the literature describe the two cases with cross-testicular ectopia and fused vas deferentes in this series.[11] Yıldız *et al*. in their study have reported a six-case series with TTE, which is the largest series showing this rare pathology up to our knowledge.[12]

CONCLUSION

The six-cases reported in this series had unusual clinical histories for crossed testicular ectopia, such as bilateral undescended testes with palpable or non-palpable testes, inguinal swelling instead of hernia and unsatisfactory impalpable testes treatment. However, the same surgical technique was performed on all cases, and impalpable testes were found in the hernia sacs of all patients. The main aim of the surgical therapy was the fixation of the testes, searching for Mullerian duct remnants and long-term follow-up for malignancy.[13] The fusion of ductus deferentes was determined in two cases, but accompanying Mullerian duct anomalies were not present in either patient. Surgical management might also be performed with laparoscopically assisted transseptal orchidopexy. It is recommended that the diagnosis of crossed ectopic testes should not be overlooked in undescended testis cases with unusual clinical findings.

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