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Transverse testicular ectopia with scrotal hypospadias but without inguinal hernia – Case report of a rare association



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ABSTRACT

Transverse testicular ectopia is an extremely rare anomaly, in which both the testis migrate towards the same hemiscrotum through the same inguinal canal. It is usually associated with other abnormalities such as persistent Mullerian duct syndrome, true hermaphroditism, inguinal hernia, pseudohermaphroditism, and scrotal anomalies; the association with inguinal hernia being the commonest. We, here, report a case of transverse testicular ectopia in a 12 years old boy having the ectopic testis in contralateral hemiscrotum without any inguinal hernia but having scrotal hypospadias with severe chordee. The diagnosis was made on clinical examination and confirmed on ultrasonography. Trans-septal orchiopexy was done for ectopic testis and hypospadias and chordee were managed by staged repair.

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1. Introduction

Transverse testicular ectopia (TTE), which is also called crossed testicular ectopia (CTE), is an uncommon anomaly of testicular migration characterized by the descent of both testis through the same inguinal canal, towards the same hemiscrotum [1,2]. It can be associated with other anomalies like persistent Mullerian duct syndrome (PMDS), true hermaphroditism, inguinal hernia, hypospadias, pseudohermaphroditism, seminal vesicle cysts, renal agenesis and scrotal anomalies [3–6]. An inguinal hernia is invariably present on the side to which the ectopic testis has migrated. We, recently, managed a rare case of transverse testicular ectopia associated with scrotal hypospadias with severe chordee but without any inguinal hernia in our institute which is a government tertiary care centre.

2. Case report

A 2 years old male child, son of a migratory labourer presented to us around 10 years ago with the complaint of both the testis lying in the same scrotal sac and abnormally located external urethral meatus since birth. There was history of ventral bending of the

penis on erection since birth. On local examination, the right scrotal sac was absent and both the testis were lying in the left scrotal sac with the urethra opening in the scrotal region by the side of the left scrotal sac. The penis had chordee even in flaccid state. Both the testes could be palpated well separately in the left scrotal sac and were having equal size and consistency (Fig. 1). So, a clinical diagnosis of transverse testicular ectopia with scrotal hypospadias with chordee was made. The ultrasound of the bilateral inguinoscrotal region corroborated the clinical findings as both the testis were confirmed to be in left scrotal sac and no other testis like structure could be located in right inguinal canal, at right deep inguinal ring or intra-abdominally and there was no hernial sac on the left side. Both the testis were enclosed in the same tunica vaginalis sac and all of their anatomical elements (vas deferens, epididymis, vascular supply) were present and normal (Fig. 2). Abdominal computed tomography and magnetic resonance imaging (MRI) of the pelvis showed no evidence of urinary system abnormalities or rudimentary Müllerian duct structures (uterus, fallopian tubes and ovary). The patient was planned for a staged repair for hypospadias and underwent chordee correction at the age of 3 years. He was lost to follow up after that and had urethroplasty done at the age of 8 years at some private hospital in his native state. He reported to our department again at the age of 12 years and trans-septal orchiopexy by Ombredanne's technique was done followed by fistula closure (Fig. 3)

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Fig. 1. Clinical photograph showing both the testes separately in the left scrotal sac with no scrotal sac on right side.

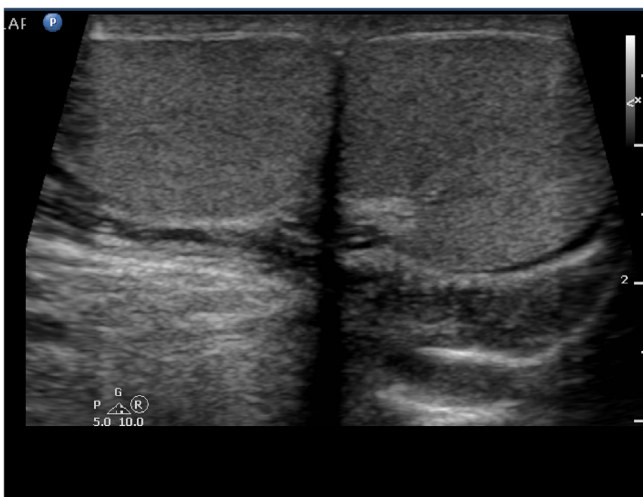


Fig. 2. Ultrasonographic image showing both the testes enclosed in the same tunica vaginalis sac.



Fig. 3. Clinical photograph showing external urethral meatus in scrotal region, absent right scrotal sac and dilator in the newly fashioned urethral tube.

3. Discussion

Transverse testicular ectopia (TTE) is also known as crossed testicular ectopia, testicular pseudo-duplication, unilateral double testes and transverse aberrant testicular mal-descent. It is the migration of the testis to the opposite side where both testes pass through the same inguinal canal. The ectopic testis may lie in opposite hemiscrotum, inguinal canal or at the deep inguinal ring. The first description is usually attributed to Lenhossek who in 1886 described this form of ectopia as a part of an autopsy performed by his father. The first case published in English literature was in 1907 by Halstead and followed by 100 documented cases in the published literature [2,7–9]. The patients usually present with uniform symptoms of an inguinal hernia on one side and an impalpable testis on the other side. Each testis has a corresponding spermatic cord, but in most cases, the two cords fuse to form an inseparable thick-walled structure that is several centimeters proximal to the testes. Each testis has its own blood supply from the appropriate side. When passing into the internal inguinal ring, the spermatic vessels of the ectopic testis cross the midline and collide with the contralateral internal ring, along with the spermatic cord of the other side. Both testes usually share a single patent processus vaginalis [10,11]. Fusion of the two spermatic cords in the inguinal canal is common and is usually inseparable [10,12]. Familial occurrences have been reported, and it can be associated with other anomalies like persistent Mullerian duct syndrome (PMDS), true hermaphroditism, inguinal hernia, hypospadias, pseudohermaphroditism, seminal vesicle cysts, renal agenesis and scrotal anomalies [3–6]. Ipsilateral inguinal hernia is invariably present in these cases but in our patient there was no evidence of inguinal hernia neither on clinical examination nor on ultrasonography of the inguinoscrotal region.

On the basis of the presence of various associated anomalies, TTE has been classified into 3 types [2]:

- Type 1 - Accompanied only by hernia (40% to 50%)
- Type 2- Accompanied by persistent or rudimentary Mullerian duct structures (30%)
- Type 3- Associated with disorders other than persistent Mullerian remnants (hypospadias, pseudohermaphroditism, and scrotal abnormalities) (20%).

So, our patient belonged to type 3 TTE. Several embryological theories explaining the origin of TTE have been proposed. There is a direct relationship between testicular ectopia and the development of the gubernacula. The gubernacula are divided into five slips, one going to each ectopic site. During testicular descent, testes follow the bulk of the gubernacula in the scrotum. When one of the other four branches contains most of the gubernacula, the testes are determined to be in an ectopic location [13]. Campbell suggested that TTE is, in fact, a unilateral development of two testes, and the contralateral testis also develops but is retained in the abdomen [14]. The superficial inguinal pouch, lateral to the external inguinal ring, is the most common location of ectopic testes. TTE is the rarest of all types of aberrant descent but probably shares some of the same mechanisms. Kimura defined true TTE as present only if there are two distinct deferent ducts and a common duct revealing the development of the testes from one genital ridge [15]. PMDS might result from the failure to synthesize or release Mullerian duct inhibitory factor (MIF) or to respond to MIF, as well as the timing of the release of MIF. It seems possible that the mechanical effect of the PMDS prevents testicular descent or causes both testicles to descend toward the same hemiscrotum, resulting in TTE.

Usually the correct diagnosis is made during surgical exploration. However, preoperative diagnosis may be established using ultrasonography, computed tomography, magnetic reso-

nance imaging and laparoscopy. Surgical management of TTE is either trans-septal orchiopexy, also known as Ombredanne's technique or extraperitoneal transposition orchiopexy. Long-term follow up is required as infertility and risk of malignancy are known complications of TTE.

4. Conclusion

TTE associated with scrotal hypospadias but without inguinal hernia is an extremely rare anomaly and possibly has never been reported in literature. The diagnosis of TTE is usually considered when unilateral hernia and concurrent cryptorchidism of the contralateral side are present but it can also present without inguinal hernia as in our case. So, presence of inguinal hernia should never be considered a prerequisite for a clinical diagnosis of TTE. In suspected cases, ultrasonographic evaluation, MRI of the pelvis and laparoscopy may be helpful in diagnosing this condition before surgery. Management is either trans-septal orchiopexy or extraperitoneal transposition orchiopexy.

We state that the work has been reported in line with the SCARE criteria [16].

We also declare that there are no conflicts of interest amongst the authors.

Conflicts of interest

There is no conflict of interest amongst the authors.

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There was no source of funding for our research.

Ethical approval

Not applicable as it is a case report.

Consent

The consent has been taken from the parents of the child for publication of this case report.

Author contribution

Pradeep Kajal – Operated upon the patient and wrote the article.

Kamal N Rattan – Supervised the article and did the final editing.

Namita Bhutani – Reviewed the literature and gave important inputs regarding the management of the case.

Vijender Sangwan – managed the investigative part including the ultrasonography and provided the images.

Guarantor

Kamal N. Rattan.

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