

Giant Inguinoscrotal Hydrocele with Crossed Testis Diagnosed in Old Age—A Case Report

Case Report

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Abstract

Crossed testis is one of the rare congenital anomalies which is usually diagnosed before puberty. It involves testicular migration in which a testis descends into the opposite hemiscrotum. Typical history at the time of presentation is an inguinal hernia with contralateral impalpable testis. The patient in this study was much older than the usual presentation age. The main objective of treatment for crossed testis includes preservation of fertility, repair of associated congenital anomalies, inguinal hernia repair, orchidopexy, frequent follow ups because of higher risk of malignancy. Here we present a 60-year-old infertile male patient with complaint of non-tender huge left sided inguino-scrotal swelling.

Keywords: Crossed testis; Hydrocele; Laparoscopy; Orchidopexy.

Introduction

Crossed testis, also known as Transverse Testicular Ectopia/ Testicular Pseudo duplication/Transverse aberrant testicular mal-descent/unilateral double testes, is an extremely rare anomaly where both the testes migrate through a single inguinal canal or hemi-scrotum. It is such a condition which is often diagnosed during surgical exploration [1], while treating inguinal hernia patients with contralateral undescended testis [2]. There are five types of testicular ectopia: superficial inguinal (interstitial), femoral (crural), perineal, pubopenile, and crossed [3].

The proper diagnosis of the condition is aided by pre-operative ultrasonography and diagnostic laparoscopy. Standard mainstay of treatment is surgery, which includes inguinal hernia repair, transseptal orchidopexy, and repair of congenital anomalies [4].

Our case stands amongst the very few cases of crossed testis which has been diagnosed in the geriatric patient, who comes with huge left sided inguino-scrotal hernia with right impalpable testis and accidentally discovered to have crossed testis during surgery.

Case report presentation

A 60-year-old man with low socio-economic status presented to surgical department with complaint of huge swelling in the

left inguino-scrotal region for 10 years which was insidious in onset, progressive nature, no aggravating or relieving factors, not associated with pain. Patient is a known case of epilepsy who is on regular medications. Patient is infertile yet never sought medical attention because of low socio-economic status. His secondary male phenotype, hormonal profile and sexual performance were normal. On physical examination, there was a nontender cystic mass, the upper limits of which could not be palpated, along the inguino-scrotal region. The right testis was impalpable (Figure 1). A solitary, pyriform-shaped, non-tender swelling in the inguino-scrotal region, measuring 20cm in length and 7cm in width, extending from the midpoint of the inguinal ligament to the base of the scrotum, was palpable. The area above the swelling could not be palpated, confirming the diagnosis of inguino-scrotal swelling. Swelling was not reducible with a positive cough impulse. After examining the external genitalia, the examination was concluded. The phallus was flaccid, but the external urethral meatus was sufficient. The right testis could not be felt. A resonant note was heard on percussion, but no specific auscultatory findings were discovered.

Intra-operative findings

We approached the massive swelling through a left inguino-scrotal incision, and an indirect hernia originating from the deep inguinal ring was observed. The spermatic cord and indirect sac

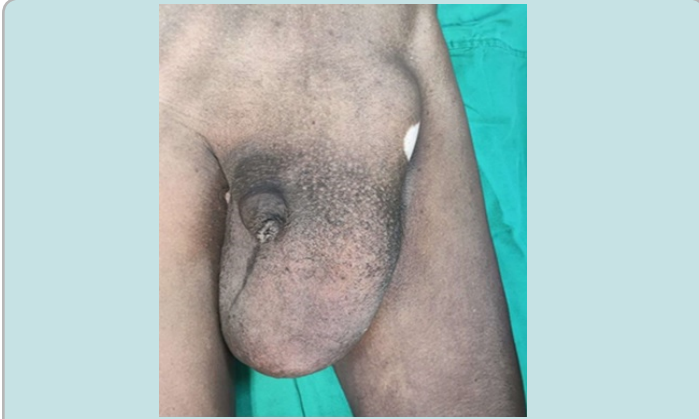


Figure 1: Pre-operative image showing huge inguino-scrotal hydrocele.

were found, and the sac extended all the way to the root of the scrotum. A separate fluid-filled sac was found to be a hydrocele, the fluid was drained, and both testes were found in the left hemi-scrotum. The excessive hydrocele sac was removed. Both testes had separate epididymis that emerged into cord structures. A soft tissue web was found between both testes. Since the right testis was found to be atrophied, a simple orchidectomy was performed. Orchidopexy was performed on the left testes. Transfixation was performed after opening the indirect hernia sac and reducing the contents. The Lichtenstein tension-free mesh repair was done (Figure 2 & 3).

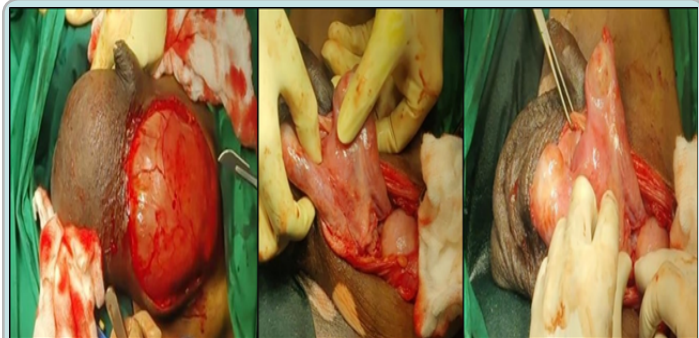


Figure 2: Pre-operative image showing huge inguino-scrotal hydrocele.

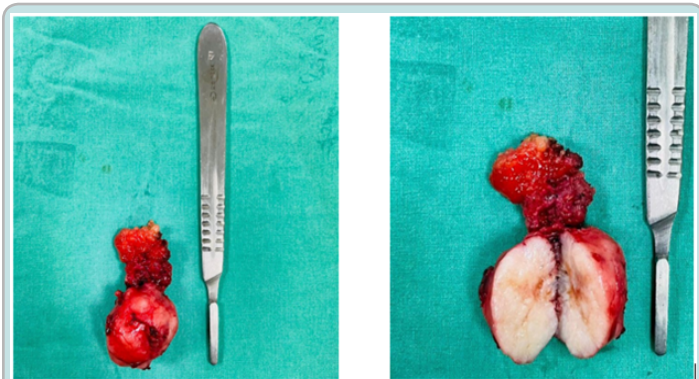


Figure 3: Right atrophied testis with its cut section.

Post-operative findings—Surgery went uneventful with no post-operative complications. Patient was doing well in the frequent follow ups which included physical examination and scrotal Doppler findings being normal. The left testis which was fixed in left hemi-scrotum was hemodynamically normal in scrotal Doppler ultrasonography.

Discussion

Crossed testis is a rare anomaly of testicular migration in which a testis descends into the opposite hemiscrotum. An inguinal hernia with a contralateral impalpable testis is a common history at the time of presentation [5]. Mean age at presentation is 4 years [2]. The patient in this study was much older than the usual presentation age. This could be due to the low socioeconomic status, which may have resulted in delayed awareness of the condition and delayed admission to the hospital. In 1886, Von Lenhossek was the one who first described this condition in an autopsy performed by his father [4]. Incidence of this condition is 1 in 4 million worldwide in children [3].

Crossed testis is usually asymptomatic [6]. The most common presentations are the presence of two scrotal masses, inguinal swelling, an undescended testis, cryptorchidism, [7] and torsion of the supernumerary testis [8]. The cryptorchidism which results in the abnormal appearance of the primary spermatocytes which leads to infertility and testicular tumour development [9]. Crossed testis may also be an incidental finding most of the times encountered during surgery [6]. 50% of patients have been diagnosed between the age group of 15 and 25 years [10]. Just like the case report described by Yigitler C et al, in our case, the impalpable testis in the right inguinoscrotal region and the contralateral double testis indicated that a ductal fusion abnormality might be present, leading to the transverse ectopia of the right testis bringing down the left hemiscrotum [6]. In our patient, we removed the atrophied right testis fixing the left testis, as malignancy potential was more and no additional benefit to fertility was possible.

Many theories have been reported which postulates the genesis of crossed testis [1]. Berg proposed a theory which states that there is the possibility of the development of both testes from the same genital ridge [11]. Kimura in his study concluded that if both vasa deferentia arose from one side, there had been unilateral origin but if there was bilateral origin, one testis had crossed over [12]. Gupta and Das postulated in their study that adherence and fusion of the developing Wolffian ducts took place early, and that descent of one testis caused the second one to follow [13]. Whereas, Esteves E et al believes in mechanical causes such as internal inguinal obstruction, mesorchia adhesions, absent peritoneum vaginalis process, absent gubernaculum and duct or gonads fusion [3].

Persistent paramesonephric (Muller's) duct remnant—tubes, rudimentary uterus, hemi uterus occurred in 38% of crossed testis cases [3]. Persistence of mullerian remnant in phenotypically normal males represents a recessive trait with male sex restriction [14, 3] in Chromosome 19, consisting of abnormal mullerian inhibiting substance (MIS) receptors, or inactive forms of MIS or even inadequate synthesis of MIS, by the fetal testis [15]. In these cases, the testis would initially aggregate the mullerian remnant and would be carried to the opposite side. Inversely, it is likely that crossed testis could be the cause, not the consequence, of some cases of Muller's duct remnant. Considering that the ectopic

testis has already migrated to the other side before 8–9th week, the period of the Muller duct's sensitivity to MIS. Due to the lack of enough MIS concentration on the original side, the ipsilateral mullerian structures wouldn't degenerate. Usually, the remnants are hemiuterus, tubes or mixed structures associated with an abnormal ectopic testis [3].

Inguinal hernia will obviously be present on the side of crossed testis. On the basis of various anomalies associated with crossed testis, Gauderer et al. [16] has postulated a classification system for crossed testis which includes 3 types: Type 1–crossed testis associated with only hernia (40–50%); Type 2–crossed testis associated with persistent or rudimentary mullerian duct structures (30%); Type 3–crossed testis associated with disorders other than persistent Mullerian remnants (inguinal hernia, hypospadias, pseudohermaphroditism and scrotal abnormalities) (20%). According to this classification, our case belongs to Type 1 crossed testis. Patients with this anomaly have higher risk of developing into malignancy. Total incidence of gonads undergoing malignant transformation is 18% according to the study conducted by F. Berkmen [9]. Malignant changes which can be encountered are embryonal carcinoma [17], seminoma, yolk sac tumour [18], and teratoma [9]. Testicular carcinoma was 6 times more likely to occur in cryptorchid cases whose surgeries were postponed until after the age of 10 to 11 years, according to Walsh et al. [19]. Wood et al. in their study showed that risk of malignancy in undescended testicles decreased if their orchidopexy performed before ages 10 to 12 years [20]. In 2% to 97% of patients with crossed testis, disorders of the upper and lower urinary tract system have been reported [21].

Investigations for crossed testis must include ultrasonography of abdomen and scrotum, MRI abdomen and pelvis and contrast enhanced CT to look for any other associated anomalies. MRI has its role in this pathology to distinguish between the presence of two separate from a single fused vas deferens [22].

The fundamental principle of treatment for crossed testis includes preservation of fertility, repair of associated congenital anomalies, inguinal hernia repair, orchidopexy, frequent follow ups because of higher risk of malignancy. Surgery can be performed with inguinal approach, laparoscopy, laparoscopy assisted inguinal approach or laparotomy. Exploration by laparoscopy will help in extensive visualization of abdomen which help in diagnosing of anomalies of mullerian structures, vas deferens etc. [4].

The various surgical techniques used in the treatment of crossed testis are transseptal orchidopexy and transperitoneal orchidopexy. The ectopic testis is relocated to the other side using the transseptal orchidopexy procedure from the aperture made in the scrotal septum [4]. In the transperitoneal orchidopexy method, the ectopic testis is placed to the extraperitoneal area by crossing the root of the penis and fixed into the other side of the hemiscrotum [23]. Vas deferens and testicular vessels must be long enough to use this technique. The preferred mode of surgical treatment for crossed testis is the transseptal orchidopexy technique (Ombredanne's technique) [4]. Bascuna et al. has designed the treatment algorithm for this technique based on the length of the funicular elements [24], and this algorithm was modified by Raj et al. [25]. Unlike Bascuna et al., Raj et al. argued that extensive dissection should not be performed in order to understand the anatomical associations between vas deferens and testicular vessels in this algorithm [25]. Additionally, when transseptal orchidopexy was not an ideal solution, they recommended fixing

both testicles into the same hemiscrotum [4, 25]. Contralateral transseptal orchidopexy method should be applied in cases where funicular structures (vas deferens, testicular vessels) are short, and transseptal orchidopexy is not suitable for the ectopic testis (Modified Ombredanne's Operation) [4]. For instance, right-sided ectopic testis with short funicular elements should be placed into the right hemiscrotum. However, it should be placed into the left hemiscrotum through the hole in the scrotal septum if this ectopic testis has long funicular elements [26]. If the funicular structures of both testicles are short, and none of the transseptal or contralateral transseptal orchidopexy techniques cannot be applied, both testicles can be placed into the same hemiscrotum [25].

Conclusion

There is more risk for malignancy in the people where this condition is diagnosed after puberty along with low fertility rate. Preserving fertility and maintaining of normal anatomy along with the repair of any associated congenital anomalies should be the mainstay of treatment. Highly opted treatment is transseptal orchidopexy, yet in this era of laparoscopy, for both diagnosis and management laparoscopy remain beneficial.

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None.

Conflict of interest

The authors declared no potential conflicts of interests with respect to authorship and publication of this article.

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