Cross Testicular Ectopia with Torsion: A Diagnostic Challenge

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Abstract
Cross testicular ectopia is a rare entity. Although torsion in a undescended or ectopic testis is known, patient presenting with torsion of both orthotopic and cross ectopic testes has not been reported earlier. We present this report of a nine month old child presenting with cross ectopic testis with torsion of both testes. The child underwent detorsion and transseptal orchidopexy of the ectopic testis along with herniotomy.

Keywords: Cross testicular ectopia; Ectopic testis; Testicular torsion

Introduction
Cross testicular ectopia or transverse testicular ectopia is a rare anomaly in which both the testis are found in single hemiscrotum. The ectopic testis cross the midline and descend through the contralateral inguino-scrotal route. Although torsion of an ectopic testis is a possibility, it has not been reported in a case of cross ectopia in the English literature. We present this report of an infant presenting with torsion of both an orthotopic and cross ectopia testis. This made the pre-operative diagnosis difficult, hence the referral and surgery got delayed.

Case Presentation
RB, 9 month old male child, presented with one day history of pain and swelling of right groin and scrotum. He had no fever, dysuria or other symptoms. His left testis was absent in the scrotum since birth. There was no history of swelling on right groin or scrotum prior to the present symptoms. The child was plump and very active. The left hemiscrotum was underdeveloped and the left testis was not palpable in scrotum or inguinal region. The right inguinoscrotal region was swollen, it was tender and the contents could not be reduced. The testis was swollen with a fluid filled sac inside the scrotum. Ultrasonography suggested a right inguinal hernia and left undescended testis, which could not be located. Right inguinal exploration was done; the cord structure was bulky and two vas deferens could be identified. The scrotal content was delivered with difficulty. Both the testes were found in the right scrotal sac, both testes were swollen and looked dark because of torsion. A common mesorchium of about 6 cm from deep inguinal ring was noted and distal to that both cord structures were twisted (Figure 1). On detorsion, colour in both the testes improved but did not regain normal colour. An empty hernia sac was identified and herniotomy was performed. One testis was placed on the left scrotal sac making a hole in the septum and the other was placed in the right scrotum. The child had uneventful recovery. A Doppler scan was done 15 days later which showed improved vascularity in both testes (Figure 2).

Discussion
Cross Testicular Ectopia (CTE) is a rare congenital anomaly occurring in 1 in 4 million male children, in which both testes migrate and descend through a single inguinal canal. Von Lenhossek first reported the anomaly in 1886 and since then less than 150 cases have been reported in the English literature [1-3]. An inguinal hernia is
always present and depending on other associated anomalies, cross testicular ectopia is classified into three types: type 1 is the commonest type seen in half of the cases in which only inguinal hernia is present [4]. In type 2 mullerian duct structures are seen along with hernia and are seen in 30% of the cases. Jordan first reported a type 2 case in 1895 [5]. Type 3 is seen 20% cases, in which other anomalies such as, hypospadias, scrotal anomalies and pseudohermaphroditism are seen along with hernia [6]. In all types, the patient is male with 46XY genotype. Testicular torsion of both the ectopic and the normally descended orthotopic testis was not reported earlier. One report mentions about knotting of the cords with testicular fusion [7].

Several theories have been postulated for cross testicular ectopia. Lenhossek ascribed the cause to either a faulty testis, a faulty testicular descent or to an abnormal gubernaculum. Linser supported this view [8]. Berg proposed the possibility of developing both testes from the same genital ridge [9]. Kimura reviewed eleven cases of cross ectopia and held that faulty development of the Wolffian duct may result in cross ectopia [10]. Gupta and Das postulated that adherence and fusion of the developing Wolffian ducts took place early, and that descent of one testis caused the second one to follow [9,11]. Gray and Skandalakis postulated that in cases where two distinct vasa deferentia exist, the testes develop from two separate ipsilateral urogenital ridges and the crossing over occurs during testicular migration [12].

Diagnosis of cross testicular ectopia rest primarily on strong clinical suspicion in children with unilateral inguinal hernia with contralateral nonpalpable undescended testis [13]. A pre-operative ultrasonography usually confirms the diagnosis. However, in inconclusive ultrasound report, CT scan or MRI may be advisable.

Most cases of crossed ectopia in the literature present before the age of 18 years and the treatment was targeted to preserve normal anatomy and fertility and to correct associated anomalies. Gaur et al. [7] performed a transseptal orchidopexy in an azoospermic young adult who had two equal sized fused testes and normal looking spermatic cords with vas deferens. Bascuna et al. [14] suggested extensive mobilization of the vas and spermatic vessels to perform a transseptal orchidopexy. Raj et al. [15] proposed a less aggressive procedure to preserve vascular supply to the testis. In type 2 CTE, excision of mullerian structure without injuring the vas and epididymis is important [16].

Children with cross testicular ectopia need long-term follow up as they have fertility issues and risk of developing malignancy in the mullerian structures has been reported [6,16]. Azoospermia is common in cross testicular ectopia due to intrinsic testicular problem [17].

References