UroToday International Journal®

www.urotodayinternationaljournal.com
Volume 1 - December 2008

Late-Presentation Cross-Ectopia Testes in an Infertile Man 31-Years Old

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Submitted on 6 June 2008 - Accepted for Publication on 26 September 2008

ABSTRACT

We present a 31-year-old man with transverse testicular ectopia of the left testis. He presented to our surgical outpatient department with right scrotal swelling, originally diagnosed as an obstructed hernia, and an impalpable testis in the left scrotum. We performed a right herniotomy on the patient and found a large lobulated left testis and normal right testis in the right scrotum. Diagnosis of transverse testicular ectopia was made on the operation table, as the man originally came for repair of obstructed right inguinal hernia. This is the first case of transverse testicular ectopia we have come across since 1966.

KEYWORDS: Transverse testicular ectopia, Inguinal hernia, Urogenital anomalies

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INTRODUCTION

Transverse testicular ectopia is an extremely rare but well recognized entity in which both gonads migrate towards the same hemiscrotum [1-3]. The clinical finding is usually symptomatic inguinal hernia on the side to which the ectopic gonad has migrated and an impalpable testis on the other. In most reported cases, the correct diagnosis is not made preoperatively but on the operation table, as the patient is operated on for repair of inguinal hernia [4]. The patient usually presents to the hospital because of cryptorchism on one side and inguinal hernia on the other. Patients are usually very young, under 1 or 2 years of age.

The aim of presenting this case is that the patient had multiple genitourinary system developmental deformities and longstanding crossed testicular ectopia that underwent malignant changes. The diagnosis and management was extremely delayed because of social reasons.

CASE REPORT

A 31-year-old male patient presented to our hospital with primary infertility, right inguinal hernia obstruction, and left undescended testis. On clinical examination, the left scrotum was empty, and no testis was palpable on the left side. The obstructed right inguinal hernia was confirmed, and the left testis was palpable in the right scrotum.

A hemoglobinopathy (Hb) test revealed 12.5 gm/dl of hemoglobin, and his erythrocyte sedimentation rate (ESR) was 60 mm/l. Ordinary ultrasonography and transrectal ultrasonography (TRUS) revealed presence of both testes in the right scrotum and no testis in the left side (fig. 1). Intravenous urography revealed both kidneys on the right side, and cross kidney ectopia was also diagnosed (fig. 2). General hormonal assay was normal apart from prolactin, which was slightly elevated to 8.5ng/ml (normal male range in our lab is 0.9-7.5ng/ml). General seminal fluid examination (GSFE) revealed a low ejaculate volume (0.8 mL) and sever oligospermia on repeated occasions and at different intervals. The possibility of cross ectopia testes was hypothesized on clinical and sonographic grounds.

During operation, the right inguinal exploration revealed a large lobulated left testis within the right scrotum associated with an indirect inguinal hernia (fig. 3).

doi:10.3834/uij.1939-4810.2008.11.02

ISSN 1944-5792 (print), ISSN1944-5784 (online)

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case observation

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During dissection, the left testis was encountered in the right inguinal canal. Each testis was noted to have its corresponding spermatic cord, and each mass had a seperate vas deferens (fig. 4). The left testis was of a large size and identical in appearance. Each had its own vascular pedicle. The left testis had an extraordinarily long spermatic cord and a hard and possibly tumorous mass in the upper pole. Orchiectomy was performed after the right inguinal herniotomy.

DISCUSSION

Normally, the testes are located in the scrotum at birth. Transverse testicular ectopia, an extremely rare anomaly, is a deviation of testicular descent resulting in unilateral location of both testes in one hemiscrotum. The spermatic cord of the ectopic testes originates from the appropriate side, but this is usually associated with an inguinal hernia [5,6,7]. In most reported cases, the correct diagnosis is not made preoperatively.

Ectopic testes have been reported at different sites, including the superficial inguinal pouch; suprapubic, femoral, and perianal areas; and at the base of the penis [4]. Migration of the testis to the opposite side where both testes pass through the same inguinal canal is known as transverse testicular ectopia. Over one hundred cases of transverse testicular ectopia have been reported in the literature [5,6]. The first description of the entity is usually attributed to Von Lenhossek [7], who in 1886 described this form of ectopia as part of an autopsy performed by his father twenty years earlier. Subsequently, Jordan [8] reported the case of an 8-year-old boy operated on for left inguinal hernia. The first case published in English literature was reported in 1907 by Halstead [9] and has since been followed by hundreds of other cases.

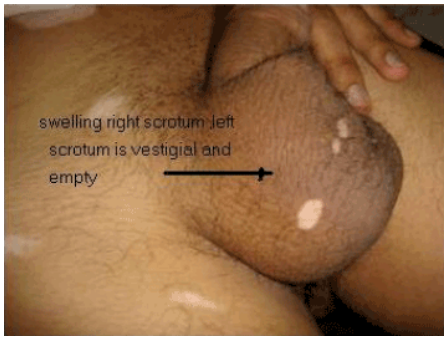

Figure 1. Right scrotum swelling; left scrotum vestigial and empty
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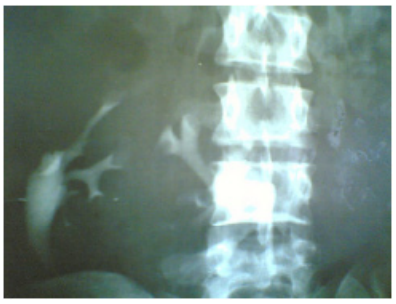
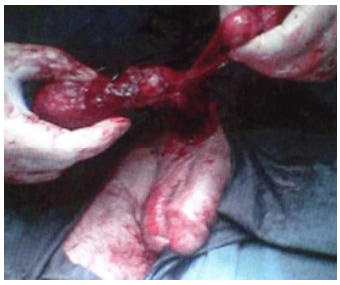
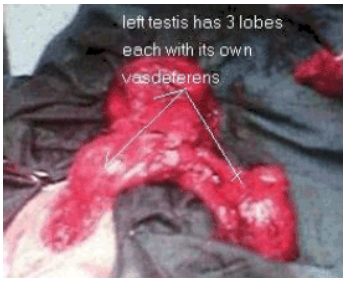


Figure 2. Intravenous urography showed cross-ectopia kidneys on the same side of cross-ectopia testes
doi: 10.3834/uij.1939-4810.2008.11.02.f2

A number of theories have been proposed to explain the etiology of ectopic testes. The first serious explanation was the multiple insertion theory provided by Lockwood [10] when he reported that the gubernaculums testis terminates in 5 tails attached to the bottom of the scrotum, the front of the pubis, the perineum, the scarpa, triangle in the thigh, and the region of the inguinal ligament just medial to the anterior superior iliac spine [10,11]. Gupta and Das [12] postulated that adherence and fusion of the developing Wolffian ducts takes place early and descent of one testis causes the second testis to follow it. Gray and Skandalakis [13] felt that since both ducts are separate in most cases, a crossing over must have occurred later. Kimura [14] suggested that if fusion of the ducts is present, it can be assumed that the two testes arose from the





left: Figure 3. Left testis looked lobulated in to 3 masses with hard nodule in the upper pole
doi: 10.3834/uij.1939-4810.2008.11.02.f3

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right: Figure 4. Complete separation of the left testis, united into one at the right inguinal canal
doi: 10.3834/uij.1939-4810.2008.11.02.f4

same genital ridge, and true crossing of the testis occurred only when a separate ductus deferens reached each testis.

Based on the presence of various associated anomalies, transverse testicular ectopia has been classified into three types: (1) associated with inguinal hernia alone (40-50%), (2) associated with persistent Müllerian duct structures (30%), and (3) associated with other anomalies without Müllerian remnants (inguinal hernia, hypospadias, pseudohermaphroditism, and scrotal abnormalities) (20%).

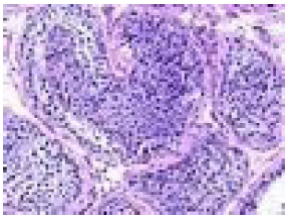
Results of histopathology of the right testis showed a typical histological picture of non-fertile testis (fig. 5 and fig. 6), while histopathology of the left testis beside a non-fertile testis showed evidence of preinvasive carcinoma in situ (fig. 7).

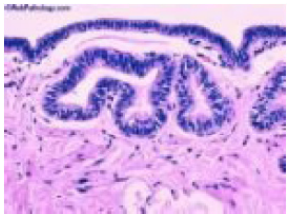
Polyorchism testes are prone to trauma, torsion, inguinal hernia (10-20% of cases), and sterility. Additionally, they are associated with a 5-50x increased risk of testicular carcinoma and may cause cancer in normal descended testes. Biopsies of both testes are suggested to detect intratubular germ cell neoplasia. A positive biopsy indicates a 50% chance that the testis will develop a germ cell tumor at 5 years versus a minimal chance if there is a negative biopsy. Exploration should be done early in life to reduce risk of malignancy. Deficient spermatogenesis persists in 10-60% of cases [15].

Testicular ectopia usually comes to the surgeon's attention because of a symptomatic inguinal hernia on the side to which the ectopic testis has migrated. In most of the reported cases, the diagnosis was only made during operation and not preoperatively. Recently, MRI has been suggested for preoperative location of impalpable testis [16]. Little attention has been focused on the treatment. When the transverse ectopic testis lies in the inguinal canal or at the external ring, it should be separated from the hernia and moved into the scrotum with its supplying cord structures lying alongside those of the ipsilateral testis.

ACKNOWLEDGEMENTS

We wish to thank Professor Shawqi Gazala (FRCS) for acting as a consultant urologist, Rezgary Teaching Hospital, and Ms. Chelan for her secretarial assistance.





left: Figure 5. Right side testicular biopsy showing the architecture of the typically infertile left testis
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right: Figure 6. Left testicular histology showing scanty of sperm inside the tubules with areas of fibrosis
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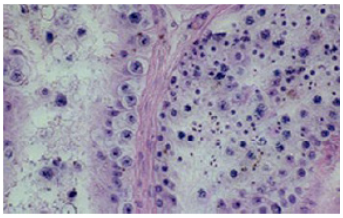

Figure 7. Hematoxyllin-eosin staining showing general features of the histologic pattern. CIS cells visualized immunohistochemically with an anti-PLAP antibody.
doi: 10.3834/uij.1939-4810.2008.11.02.f7

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TO CITE THIS ARTICLE: Zangana AM. Late-Presentation Cross-Ectopia Testes in an Infertile Man 31-Years Old. UIJ. 2008 Dec;1(5). doi:10.3834/uij.1939-4810.2008.11.02