

Transverse testicular ectopia, a case report and review of literature

Transverse Hodenektomie, Fallbericht und Literaturübersicht

Abstract

Crossed testicular ectopia (CTE)/transverse testicular ectopia (TTE) is a rare but well known congenital anomaly, in which both gonads migrate toward the same hemiscrotum. It is usually associated with other abnormalities such as persistent Mullerian duct syndrome, true hermaphroditism, inguinal hernia, hypospadias, pseudohermaphroditism, and scrotal anomalies. About 100 cases of transverse testicular ectopia have been reported in published studies. We report a case of transverse testicular ectopia in an 8-month-old boy who presented with right inguinal hernia and nonpalpable left testis. On exploration, both testes were present in the right inguinal region. Bilateral orchiopexy was performed by crossing the left testis in the extra-peritoneal space and ipsilateral scrotal orchiopexy. The diagnosis could not be made preoperatively in most of reported cases.

Keywords: testis, undescended testis, cryptorchidism, testicular ectopia

Zusammenfassung

Gekreuzte Hodenektomie oder transverse Hodenektomie ist eine seltene, aber bekannte angeborene Anomalie, bei der beide Hoden in denselben Teil des Hodensackes (Hemiskrotum) wandern. Die Anomalie ist gewöhnlich mit anderen Fehlbildungen assoziiert wie z.B. persistierender Müller-Gang, echter Hermaphroditismus, Leistenhernie, Hypospadien, Pseudohermaphroditismus und Fehlbildungen am Hodensack. Etwa 100 Fälle mit gekreuzter Hodenektomie sind als Fallberichte publiziert worden.

Wir berichten über einen Fall von transverser Hodenektomie bei einem 8 Monate alten Jungen, der mit einer Leistenhernie rechts und einem nicht tastbaren linken Hoden vorgestellt wurde. Die Untersuchung ergab, dass beide Hoden in der rechten Leistenregion vorhanden waren. Eine bilaterale Orchiopexie mit Überkreuzung des linken Hodens im extraperitonealen Bereich und Fixation im seitengleichen Skrotum wurde durchgeführt. In den meisten berichteten Fällen konnte die genaue Diagnose präoperativ nicht gestellt werden.

Schlüsselwörter: Nicht abgestiegene Hoden, Kryptorchismus, Hodenektomie

Introduction

Transverse testicular ectopia (TTE) also named testicular pseudoduplication, unilateral double testis, and transverse aberrant testicular mal-descent, is a rare anomaly in which both testes descend or migrate through a single inguinal canal or hemiscrotum [1]. Often, the diagnosis is made during surgical exploration. In the literature more than 100 cases of TTE have been reported. We report a case of TTE discovered accidentally during surgery for right inguinal hernia and bilateral undescended testes.

Case presentation

The patient was a 7 months old infant, who admitted for the left nonpalpable testis, right undescended testis and right inguinal hernia. The patient was born with normal vaginal delivery, with a normal Apgar score. There was no history of illnesses or poor feeding or failure to thrive (FTT). General physical examination was unremarkable. Hematological examination and biochemistry lab data was normal. In external genitalia examination, the right

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Figure 1: Both testes are visible, protruding from right inguinal incision as TTE.

testis was palpable inguinally with an evident hernia and the left hemiscrotum was empty. The patient scheduled for synchronous bilateral orchiopexy and right inguinal herniotomy. Firstly, left inguinal incision was done, and no testicular tissue or even spermatic cord was found. Then, right inguinal incision was made. The right testis with its overlying tunica vaginalis was found at the deep inguinal ring. After opening of the tunica, the fluid inside of it drained and testis was found. At the proximal part of the cord another testis was found (Figure 1). The suspicion of left testis anorchia was changed to right side transverse testicular ectopia. Then, the plan changed to releasing of cords and bilateral orchiopexy. Two cords had common origin for 3-4 cm. The left cord was released at the site of its bifurcation, to the most proximal site, that distal to it releasing dissection was completed. Finally, the left testis was transferred with its cord to the left hemiscrotum easily and extra-peritoneally. Both testes were fixed in the sub-dartos pouch.

Discussion

TTE is a rare form of testicular ectopia. It was first reported by Von Lenhossek in 1886 [2]. More than 100 cases have been reported in the literature [3]. Several theories have been reported to explain the genesis of TTE. Berg [4] proposed the possibility of the development of both testes from the same genital ridge. Kimura [5] 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. Gupta and Das [6] 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.

An inguinal hernia is invariably present on the side to which the ectopic testis has migrated. On the basis of the presence of various associated anomalies, TTE has been classified into 3 types: Type 1, accompanied only by hernia (40% to 50%); type 2, accompanied by persistent or rudimentary Mullerian duct structures (30%); and type 3, associated with disorders other than persistent Mullerian remnants (inguinal hernia, hypospadias, pseudohermaphroditism, and scrotal abnormalities) (20%). According to that classification, our case was type 1 TTE. TTE associated with fused vas deferens is extremely rare. This condition may hinder the testis from being placed into the scrotum during orchiopexy [7].

The mean age at presentation is 4 years. The clinical presentation generally includes an inguinal hernia on one side and a contralateral or sometimes a bilateral cryptorchidism [8], [9]. Usually, the correct diagnosis is not made before surgical exploration, like our case, and it is revealed during herniotomy [9]. The diagnosis of TTE can be made preoperatively by using ultrasonography [10] by an experienced sonologist. Patients with TTE are at increased risk of malignant transformation. In fact, the overall incidence of malignant transformation of gonads

is 18% [11]. There have been reports of embryonal carcinoma [12], seminoma, yolk sac tumor [13], and teratoma [11]. Walsh et al. [14] in their study concluded that testicular cancer was nearly 6 times more likely to develop in cryptorchid cases whose operations were delayed until after age 10 to 11 years. Wood et al. [15] in their study showed that risk of malignancy in undescended testicles decreased if their orchiopexy performed before ages 10 to 12 years. In 2% to 97% of patients with crossed testicular ectopia, disorders of the upper and lower urinary tract system have been reported [16]. Once diagnosis of TTE is made, a conservative surgical approach in the form of orchiopexy is recommended for the preservation of fertility [9]. Laparoscopy is useful for both diagnosis and treatment of TTE and associated anomalies [17]. Management for testicular ectopia is either trans-septal or extra-peritoneal transposition orchiopexy [18], [19], a search for Mullerian remnants and other anomalies, and long-term postoperative follow-up. There were two options for left orchiopexy in our case: extra-peritoneal orchiopexy and trans-septal orchiopexy. In the extra-peritoneal technique the testis is brought to the contra-lateral hemiscrotum after its passing near the root of penis. In the trans-septal technique the testis should traverse the scrotal mediastinum to be fixed in it. In the case of fused vas deferens, unlike our case, a trans-septal orchiopexy is recommended. It may be misdiagnosed as an inguinal hernia and intersex [17] or present as an irreducible hernia, requiring urgent surgery [20].

Conclusion

TTE is a rare anomaly of which the pathogenesis remains unclear, although experimental evidence suggests that the gubernaculum may play an important role. The ectopic testis can lie in the hemiscrotum, in the inguinal canal, or at the deep inguinal ring. The diagnosis should be considered when unilateral hernia and concurrent cryptorchidism of the contralateral side are present. In suspected cases, laparoscopy and ultrasonographic evaluation may be helpful in diagnosing of this condition before surgery. Transseptal orchiopexy is highly recommended to manage TTE. Laparoscopy, at present, is useful for both diagnosis and management of TTE and associated anomalies.

Notes

Competing interests

The authors declare that they have no competing interests.

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Please cite as

Moslemi MK, Ebadzadeh MR, Al-Mousawi S. Transverse testicular ectopia, a case report and review of literature. *GMS Ger Med Sci*. 2011;9:Doc15.
DOI: 10.3205/000138, URN: urn:nbn:de:0183-0001386

This article is freely available from

<http://www.egms.de/en/journals/gms/2011-9/000138.shtml>

Received: 2011-03-02

Revised: 2011-06-06

Published: 2011-07-07

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