Where is the testis? The role of ultrasound and diagnostic laparoscopy for Crossed Testicular Ectopia (CTE):
Case report and review of literature

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Where is the testis? The role of ultrasound and diagnostic laparoscopy for Crossed Testicular Ectopia (CTE): Case report and review of literature

Crossed Testicular Ectopia (CTE) or transverse testicular ectopia is an anecdotic urogenital anomaly in which both testes are located on the same side, generally associated with a patent processus vaginalis (PPV). The condition can be detected by ultrasound. Nevertheless, the diagnosis is often missed preoperatively and CTE is recognized intraoperatively. Controversy exists regarding management and the role of diagnostic laparoscopy. The surgical technique depends on the anatomy of vas, vessels and testis found on surgical exploration. Diagnostic laparoscopy can be useful to *rule out a vanishing testis and detect Müllerian remnants. We present the case of 8-months infant with no palpable testis on the right side and no signs of inguinal hernia, reporting the management and reviewing the scarce existing literature in this regarding.*

KEY WORDS: Crossed Testicular Ectopia, Laparoscopy, Ectopia, Testis, Transverse Testicular Ectopia, Urogenital Abnormalities

Introduction

Crossed testicular ectopia (CTE) refers to a congenital urogenital anomaly in which both testes are located on the same side. It is also known as transverse testicular ectopia, testicular pseudoduplication, unilateral double testes and transverse aberrant testicular mal descent. The first comprehensive description of the condition is attributed to von Lenhossek in 1886. The testes can be retained in the abdomen or descend via a single inguinal canal in the inguinal region, or in one hemi-scrotum. The processus vaginalis is almost invariably patent on the side of the CTE and other genital anomalies can be present including persistent Müllerian duct remnants, anomalies of sexual differentiation, hypospadias, scrotal anomalies, seminal vesicle cysts and renal agenesis [1,2]. Unlike the undescended testis, which occurs in approximately 1% of male children at 1 year of age [3], CTE is extremely rare and currently there are less than 150 cases reported in literature [4]. The diagnosis can be suspected on clinical examination and corroborated by preoperative ultrasound. The use of CT scan and MRI has been reported. Controversy exists regarding management and the role of diagnostic laparoscopy. Nevertheless, the diagnosis is often missed preoperatively and CTE is recognized intraoperatively [5].

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ABBREVIATIONS

CTE: Crossed testicular ectopia
PPV: Patent Processus vaginalis

Herein we present a case of export and describe the diagnostic work-up and management at our centre in comparison to the existing literature, particularly regarding the role of diagnostic laparoscopy.

Case report

An 8 months-old boy presented in the clinic for bilateral undescended testes. At clinical examination, the left testis was palpable in the left inguinal region whilst the right one was unpalpable. External genitalia were otherwise normal, and no bulging suggestive of inguinal hernia was observed. The patient had already undergone two US with contrasting results. In one the right testis could not be found, in the other both testes were described on the left side, one in the inguinal canal and the other distal to it. However, since the right ectopic testis was not palpable, a diagnostic laparoscopy was performed at 14 months of age. Intraoperatively (Fig. 1), no Müllerian remnants were found, the right inguinal ring was closed without vessels or vas getting into it, the left inguinal ring was patent and both vases entered it. Both testes were visible within the inguinal canal. A left inguinal exploration was performed. The two testes appeared connected through the fusion of the connective tissue above the head of their epididymis (Fig. 2). The processus vaginalis was separated from the gonads and cord and ligated at the internal inguinal ring (Fig. 3). We observed two deferential ducts fused in a Y shaped at their origin. The testes had a diminished texture, and the right one was also considerably smaller, hypotrophic, with tiny vessels. Considering the length of the right spermatic cord, shorter than usual, we opted for an ipsilateral orchiopexy placing sequentially the right testis in its hemi-scrotum through a trans-septal incision. No post-operative complications occurred, and the patient was discharged after 2 days. Follow-up was uneventful till the present day. The gonads were stable regarding size and location.

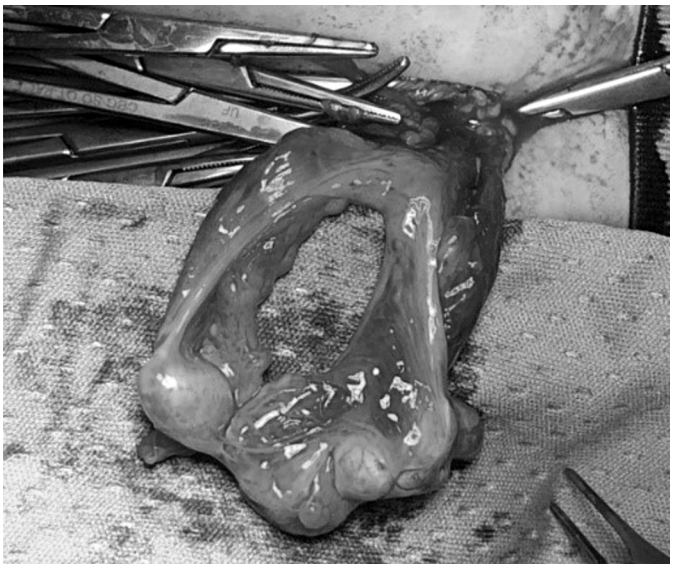

Fig. 2: The testes appeared connected above the head of their epididymis.

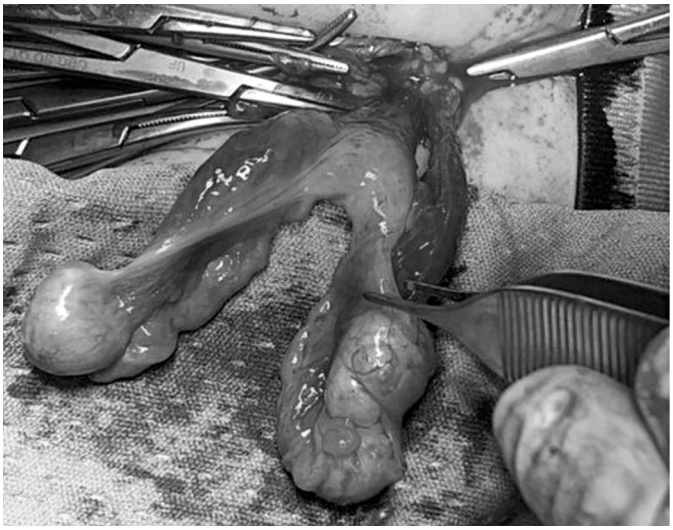

Fig. 3: Processus vaginalis was separated from the structures and ligated at the internal inguinal ring.

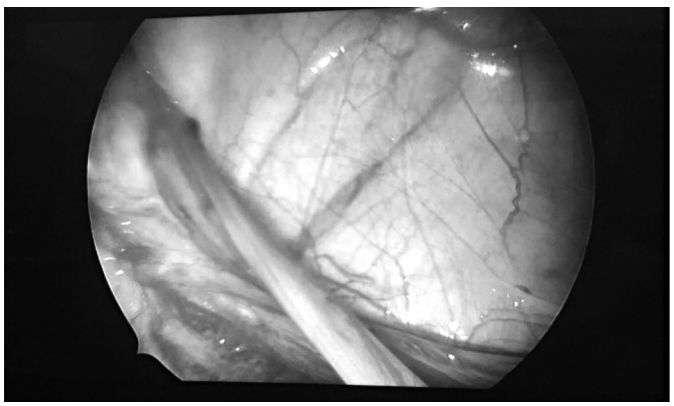

Fig. 1: Intraoperative laparoscopic view: The left inguinal ring was patent and both vases entered it.

Discussion

CTE is an extremely rare but well-recognised urogenital anomaly in which both gonads are located on the same side. The aetiology remains unclear. Several theories have

been proposed. Berg proposed the possibility of the development of both testes from the same genital ridge [6]. Gupta and Das, instead, thought that the adherence and fusion of developing Wolffian ducts happened early and that the descent of one testis caused the second one to follow [7].
Others suggested a defective implantation of the gubernaculum testis or an obstruction of the inguinal ring preventing the descent of the testis on the ipsilateral side [8]. According to Kimura, a real crossed ectopia occurs only in presence of 2 distinct deferent ducts, whilst a common duct suggests a unilateral origin [9]. This theory fits with the presence of fused vas deferens but does not explain the migration towards either side. Another option is that both testes lie in the same processus vaginalis before descent, which occurs when the testes are bound together or if the vasa are bounded to Müllerian remnants [8]. About associated anomalies, CTE can be classified into 3 categories: type 1, associated only with a PPV (40-50%); type 2, associated to Müllerian remnants (persistent Müllerian duct syndrome) (30%); type 3, paired with other disorders such as hypospadias, disorders of sexual differentiation, and others (20%) [10].
The most of the cases are diagnosed intraoperatively during surgical exploration [11]. The diagnosis can be suspected preoperatively if both testes are palpable on the same side and the contralateral hemi-scrotum is empty. In such circumstances, a pre-operative ultrasound or an MRI can be helpful [11,12].
The main problem with ultrasound is its operator dependency, and it might be difficult on little and not compliant patients. MRI is more detailed, but its prohibitive costs may limit his use [13]. CT scan is not recommended in paediatric patients for the radiation exposure [12]. Therefore, whenever in doubt, like in nonpalpable unilateral testes, explorative laparoscopy is the procedure which provide more detailed information [14,15]. Due to the different presentations, various surgical treatments may be. The surgical goals are to preserve fertility and place the testes in the hemiscrotum.
Surveillance for potential malignant development, higher than in simple undescended testis, has a crucial role [16].
The options used and reported in literature are inguinal exploration and orchidopexy, laparoscopy exploration and orchidopexy (inguinal or laparoscopic-assisted), laparoscopy exploration and transseptal ipsilateral and/or contralateral orchidopexy (inguinal or laparoscopic-assisted), orchiectomy (inguinal or laparoscopic) [16]. Fixing both testes in the same hemi-scrotum is also an option, whenever transseptal orchidopexy is not feasible [17].
A review of the contemporary literature regarding the role of laparoscopy in management of pediatric patients with CTE of the last 20 years (January 2002 - June 2022, English language) was performed using PubMed. The following Mesh search headings were used: "crossed testicular ectopia", "transverse testicular ectopia", "laparoscopy". Forty patients were managed with laparoscopy and diagnostic laparoscopy was realized in all [15,18,27-35,19-26]. However, only 16 (40%) were treated laparoscopically [20,23,24,28,30-32,34]. Indeed, the inguinal access still has its place [15,18,29,30,33-35,19-22,24-27].
In one case an inguinal orchiectomy was performed [26].
Diagnostic laparoscopy also allows investigating associated conditions, such as Müllerian remnants.
The persistent Müllerian remnants vary among individuals and alter the normal anatomy, thus may complicate management [20]. Fourteen (35%) of the patients of our review showed Müllerian remnants at diagnostic laparoscopy [20,21,25,30,32-34]. The excision of the Müllerian remnants was realized in 7 cases (50%) [25,30,32-34], in three laparoscopically [32,34]. The indications for resection were inability to perform orchiopexy [25,33,34] and surgeon's preference [30,32]. The management for the CTE associated with the remnants remains controversial. The arguments against resection are various.
Although testicular malignancy risk is increased in these cases, justifying orchiopexy or orchiectomy for a non-mobilizable testis, no malignant degeneration of persistent Müllerian structures has been reported [20].
Furthermore, any surgical excision of uterus or fallopian tubes risks damage to vasa deferentia and the deferential blood supply to the testis [20,21,25]. Instead, for other authors the management of remnants is exclusively excision in order to prevent any possible risk of transformation [30,32].

Conclusion

On the base of our experience and the reviewed literature, we support the role of laparoscopy in management of pediatric patients with CTE, as for other causes of nonpalpable undescended testis. Minimally invasive surgery may confirm the diagnosis, clarify the anatomy and define the surgical plan.

Riassunto

L'ectopia testicolare crociata CTE) o l'ectopia testicolare trasversa sono anomalie urogenitali episodiche nelle quali entrambi i testicoli sono localizzati sullo stesso lato, generalmente associati alla permanenza del processo vaginale pervio (PPV). Questa situazione può essere scoperta con l'ecografia. Cionondimeno la diagnosi preoperatoria può spesso mancare e l'anomalia viene scoperta intraoperatoriamente. Vi sono controversie a riguardo del trattamento da adottare e il ruolo della laparoscopia.
La tecnica chirurgica dipende dall'anatomia del deferente, dei vasi sanguigni e del testicolo individuate nel corso dell'esplorazione chirurgica. La laparoscopia diagnostica può essere utile per escludere l'agenesia testicolare e per l'individuazione dei residui Mulleriani.
Presentiamo il caso di un infant di otto mesi con assen-

za di testicolo palpabile dal lato destro senza segni di ernia inguinale, per riferire del trattamento adottato e per riferire della scarsa letteratura esistente a questo riguardo.

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