Contents lists available at ScienceDirect

Urology Case Reports

journal homepage: www.elsevier.com/locate/eucr





Pediatric Surgery Department, Hospital Habib Thameur, Tunis, Tunisia

ARTICLE INFO

Keywords: Persistent mullerian duct syndrome Transverse testicular ectopia Nonpalpable testis Surgery

ABSTRACT

Transverse testicular ectopia(TTE) is a rare anomaly defined by the presence of both testis in the same hemiscrotum or in the same inguinal canal.The main treatment of TTE is surgical intervention. Here we report a case of type 2 transverse testicular ectopia in an eight months old child. The boy had a right side inguinal hernia and a non-palpable testis on the left side. Ultrasonography confirmed the presence of both testis at the right inguinal region. Our case was associated with the persistence of Mullerian duct. After the excision of the Mullerian duct remnant, bilateral orchiopexy was performed.

1. Introduction

Transverse testicular ectopia (TTE) is a rare anomaly characterized by the presence of both testis in the same hemiscrotum or both testes are migrated and descend through a single inguinal canal. This congenital anomaly of undescended testes is reported in less than 150 cases in literature.It can be associated with other anomalies like persistent Müllerian duct syndrome (PMDS), true hermaphroditism, inguinal hernia, hypospadias, pseudohermaphroditism, seminal vesicle cysts, renal agenesis and scrotal anomalies.¹ TTE is usually found incidentally in patients operated for inguinal hernia or undescended testicles. The main treatment of TTE is surgical intervention with hernia repair, if present.

In this case study, we report a case of type 2 transverse testicular ectopia in child.

2. Case report

An eight-month-old boy patient was referred to our pediatric surgery consultation for right side inguinal hernia. According to medical history: he was born at term, and there was no medical problem during the postnatal period.

Physical examination showed an uncircumcised penis, the left testis was impalpable and left hemiscrotum was empty. The right testis was palpable at the scrotum with evidence of right congenital hernia.No other anomalies were present (Fig. 1).

Bilateral inguinal ultrasound revealed that both testes are present within the content of the hernia sac at the right inguinal region. Exploration of the right groin confirmed the presence of both testis in the same side one above the other. Each testis had vas deferenses, vascular supplies and epididymis. The two gonads are connected to each other via a unique uterus like structure, suggesting persistent müllerian duct remnant, located midline(Fig. 2).

We have noticed that the uterus like structure limit the placement of the testis in the contralateral hemiscrotum. So, we proceed to the remove of this structure since there was a good dissection plan between the two vas deferens and the structure.

After herniotomy, the left testis was brought to the contra-lateral left hemiscrotum using *trans*-septal orchiopexy, also the right testis was fixed in the sub-dartos pouch(Fig. 3).

The child discharged at the same operative day. Follow up for a period of 1 month showed no postoperative complication. Karyotyping showed a male 46 XY pattern.

Anatomopathological examination confirmed the persistence of Mullerian duct structure.

3. Discussion

TTE is also known as crossed testicular ectopia, testicular pseudoduplication, unilateral double testes and transverse aberrant testicular mal-descent. TTE has been classified into 3 types:type 1 accompanied only by hernia (40%–50%),type 2 accompanied by persistent or rudimentary Mullerian duct structures (30%),type 3 associated with disorders other than persistent Mullerian remnants (hypospadias, pseudohermaphroditism, and scrotal abnormalities)(20%).So, our patient belonged to type 2 TTE.

* Corresponding author. E-mail address: fatma.bchini@etudiant-fmt.utm.tn (F. Bchini).

https://doi.org/10.1016/j.eucr.2021.101909

Received 21 September 2021; Received in revised form 13 October 2021; Accepted 19 October 2021 Available online 20 October 2021 2214-4420/© 2021 The Authors. Published by Elsevier Inc. This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/by-nc-nd/4.0/).







Fig. 1. Empty left hemiscrotum with inguinal right side hernia and palpable right testis at the scrotum.



Fig. 2. Testis are related to each other by an uterus like structure suggesting persistent müllerian duct remnant.



Fig. 3. Left orchiopexy was performed after passing the left testis through *trans-septal* window of the scrotum followed by ipsilateral right scrotal orchiopexy.

Female internal genitalia, including uterus, fallopian tubes and the upper two-thirds of a vagina, are observed in PMDS males because Müllerian duct regression does not occur.²This disorder of male sex development (DSD) is caused by Mullerian Inhibitory Factor(MIF)deficiency secreted from the fetal Sertoli cells (46 XY DSD).

Usually the correct diagnosis is made during surgical exploration. However, preoperative diagnosis may be established using ultrasonography, computed tomography, magnetic resonance imaging and laparoscopy.¹

Due to its varied presenting scenarios, surgical management of TTE had many options. It consists on inguinal exploration and orchiopexy, diagnostic laparoscopy and orchiopexy, diagnostic laparoscopy and *trans*-septal orchiopexy, diagnostic laparoscopy and *trans*-septal contralateral orchiopexy with congenital anomalies repair at the end. Some authors suggested fixing both testicles into the same hemiscrotum in cases where transseptal orchiopexy was not feasible.

Onur et al., recommended excision of Mullerian duct remnants without risking the vas deferens.³ However,Morgan et al. recommended leaving it if they don't produce symptoms, as surgical excision of these structures can be difficult.⁴In our case, we preferred to excise the Mullerian structures because of the increased risk of malignant transformation in the long term.⁵

Karyotyping may be required in some cases particularly if associated with other congenital anomalies of the genitalia or in case of ambiguous genitalia. 2

4. Conclusion

TTE is a rare condition that should be considered in cases with undescended testis.

Patients can be presented with inguinal hernia on one side and cryptorchidism on the other side or with impalpable testis without any associated hernia and the anomaly may be discovered incidentally during diagnostic laparoscopy for cryptorchidism.

Karyotyping is recommended for cases of TTE with PMDS.

Patients usually need long term follow up as such patients may have future fertility problems and there is an increased risk of malignant transformation in the long term..

CRediT author statement

Fatma Bchini: Writing – original draft preparation. Malek Boughdir: Conceptualization. Aida Daib: Validation. Sameh Tlili: Visualization, investigation. Youssef Hellal and Nejib Kaabar: Supervision.

References

- Kajal P, Rattan KN, Bhutani N, Sangwan V. Transverse testicular ectopia with scrotal hypospadias but without inguinal hernia – case report of a rare association. Int. J. Surg. Case Rep. 1 janv 2017;31:167–169. Disponible sur: https://www.sciencedirect. com/science/article/pii/S2210261217300470.
- Jabali SS, Mohammed AA. Crossed testicular ectopia: case report with review of literature. Int J Surg Case Rep. 2020;75:189–192.
- Telli O, Gökçe Mİ, Haciyev P, Soygür T, Burgu B. Transverse testicular ectopia: a rare presentation with persistent müllerian duct syndrome. J Clin Res Pediatr Endocrino. sept. 2014;6(3):180–182. Disponible sur: https://www.ncbi.nlm.nih.gov/pmc/artic les/PMC4293649/.
- Morgan RJ, Williams DI, Pryor JP. Müllerian duct remnants in the male. British Journal of Urology. déc. 1979;51(6):488–492. Disponible sur: https://onlinelibrary.wil ey.com/doi/10.1111/j.1464-410X.1979.tb03584.x.
- Farikullah J, Ehtisham S, Nappo S, Patel L, Hennayake S. Persistent Müllerian duct syndrome: lessons learned from managing a series of eight patients over a 10-year period and review of literature regarding malignant risk from the Müllerian remnants: persistent müllerian duct syndrome. BJU International. déc. 2012;110(11c). E1084-E1089. Disponible sur: https://onlinelibrary.wiley.com/doi/10.1111/j.146 4-410X.2012.11184.x.