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Case Report

A curious case of toddler with hernia and much more

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ABSTRACT

Transverse testicular ectopia (TTE) with fused vas deferens is an extremely rare clinical entity. Herein, we presented a case of a 2 years old patient with left inguino-scrotal swelling associated with pain lasting for 3 days. Clinical examination revealed an empty right hemi-scrotum, a left-sided giant inguino-scrotal swelling. Laboratory tests were normal. Ultrasound imaging (US) of the scrotum demonstrated the presence of both testes in the same left hemi-scrotum with inguinal hernia and enterocele as content. Surgical intervention in suspicion of obstruction was carried out through inguinal incision. There was a congenital inguinal hernia with appendix (type 1-Amyand's hernia) and cecum as content with two well developed testes on the same side with separate epididymis and vas deferens. Both the testes were united by a mesorchium. Hence, reduction of contents with herniotomy and both the testes were anchored to inner aspect of left thigh one above the other. We reviewed the literature for rare diagnosis of TTE.

Keywords: Transverse testicular ectopia, Undescended testis, Amyand's hernia

INTRODUCTION

TTE is a rare congenital anomaly in which both testes descend on the same inguinal route ultimately lying on the same side of the scrotum. This pathologic condition mostly affects young males with a mean age of 4 years.[1-3]

CASE REPORT

A 2 years old patient was referred to our department complaining of 3 days history of left sided inguino-scrotal swelling with pain. His past medical history was remarkable for an absent right testis without any further clinical information. Upon physical examination, the patient presented with an empty and hypoplastic right hemi scrotum. The right testis was not palpable along the ipsilateral inguinal canal. On the contrary, a giant inguino-scrotal swelling was present on the left side which was tender to palpate. The patient also complained of inability to reduce the swelling over the left hemi scrotum. No signs of strangulation were present at the time. Clinically, a diagnosis of left obstructed inguinal hernia was made. Digital rectal examination was normal. Laboratory workup was insignificant. Color Doppler US revealed the presence of two normal testes in the same left hemi scrotum and inguinal with enterocele as the content. Each testis was paired by a normal epididymis but only one vas deferens could be identified. The arterial supply of both testes was normal. Subsequently, the patient underwent upper and lower abdominal CT with contrast administration which revealed cecum as the content of inguinal hernia and also normal urogenital tract but poorly visualized seminal vesicles. The CT findings excluded urogenital anomalies such as renal agenesis or malformation that are occasionally combined with testicular mal-descent and ectopy. On the other hand, patient underwent emergency exploration through an inguinal incision, hernial contents were visualised. There was a congenital inguinal hernia with appendix (type 1-Amyand's hernia) and cecum as content with two well developed testes on the same side with separate epididymis and vas deferens. Both the testes were united

by a mesorchium. Reconstruction and transseptal transfer of the ectopic testis to the contralateral side were deemed technically demanding, due to extensive testicular dissection and mobilization, both the testes were anchored to inner aspect of left thigh one above the other. Hence, reduction of contents with herniotomy and no trans-septal orchidopexy was performed. Parents were informed to bring the child for follow up in six months.

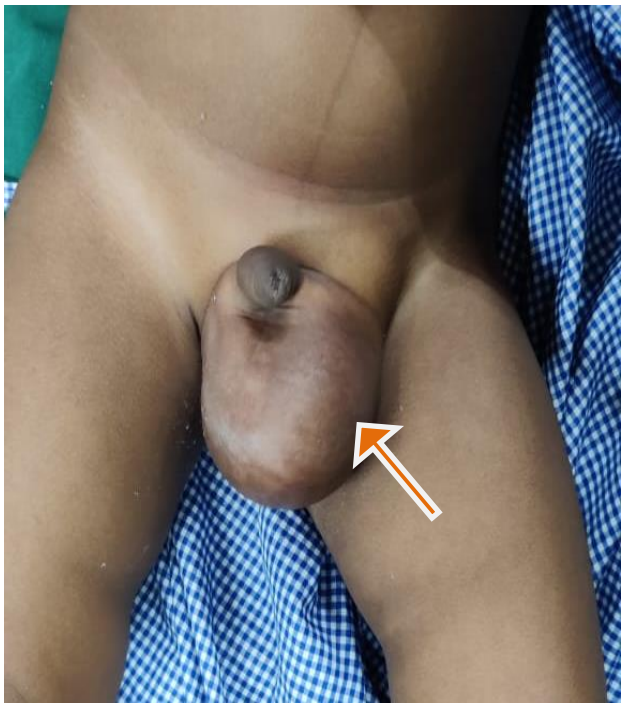

Figure 1: Left sided inguino-scrotal swelling.

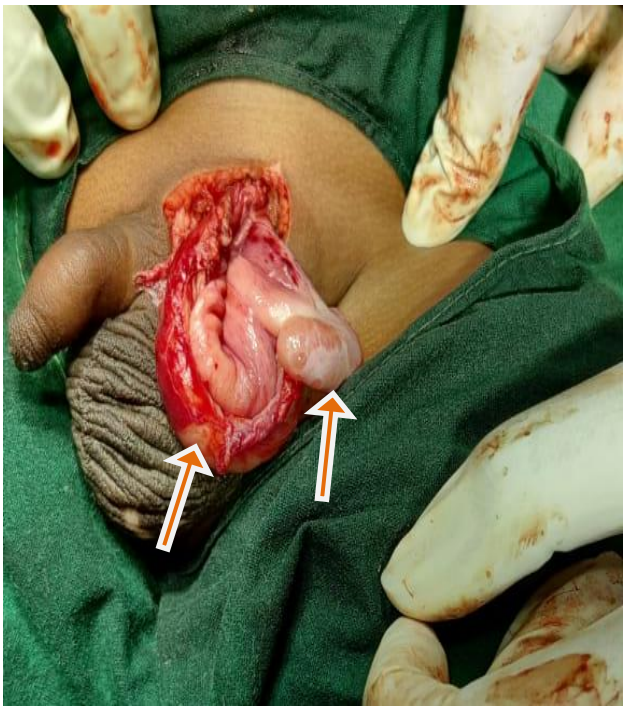

Figure 2: Both testes on the left side with mesorchium.

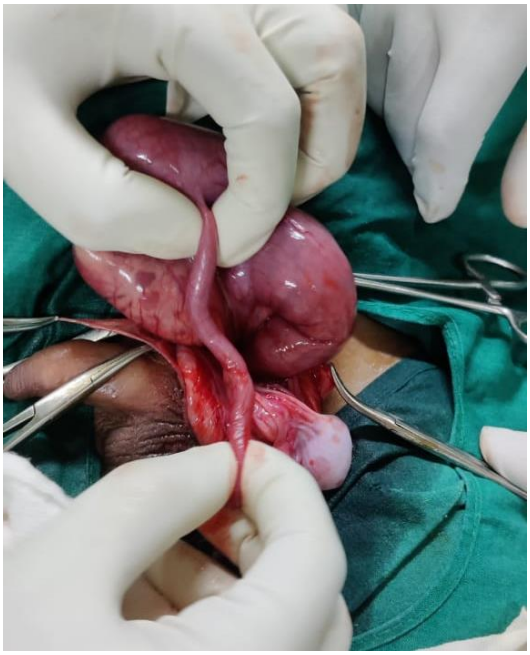

Figure 3: Appendix and cecum as the content of hernia with both testes.

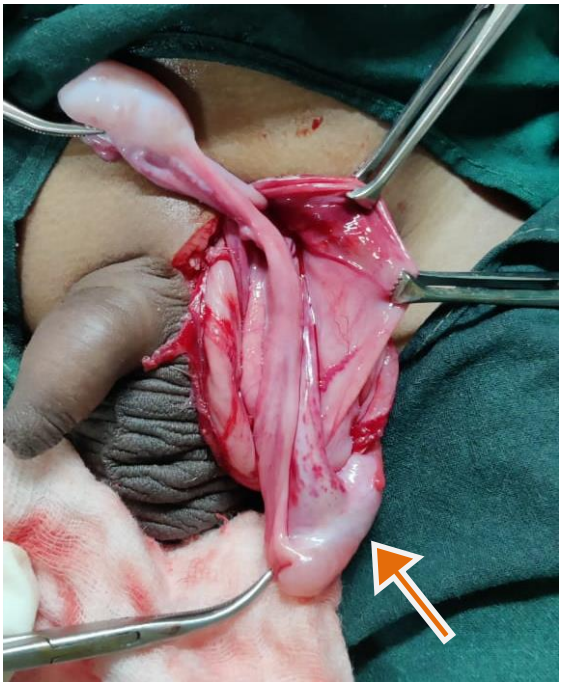

Figure 4: Both testes on the left side with separate epididymis and vas.

DISCUSSION

Testicular ectopia is an abnormally positioned testis that can lie anywhere along its theoretical descent route from the retroperitoneum down to the scrotum. In crossed or transverse ectopia the testis crosses the midline to land on the contralateral hemi-scrotum.[1-3] This pathologic condition was thoroughly described in children, with a mean age of 4 years at the time of diagnosis.[1,2,4,5] It was

first described in 1886 by Lenhossek on a 35 year old adult.[6] Associated clinical findings most commonly included ipsilateral inguinal hernia, hypospadias, pseudo-hermaphroditism and scrotal abnormalities or it can even present as part of the persistent Mullerian duct syndrome.[7-9] Based on the associated developmental anomalies, a classification system was proposed in 1982 replacing a former one that relied on the etiology of this pathologic entity.[10,11] According to Gauderer et al three types of transverse ectopia are recognised: type I, which is associated with an inguinal hernia and accounts for 40-50% of the cases, type II, which is accompanied by Mullerian duct remnants (30%) and type III (13-20%), which includes genitourinary anomalies other than persistent Mullerian duct such as hypospadias, pseudo-hermaphroditism, bifid scrotum, renal anomalies, seminal vesicle contralateral aplasia and seminal vesicle cysts.[11] In our case, we categorised it as type 1. Gray and Skandalakis postulated that in cases where two distinct vasa deferentia exist, the testes developed from two separate ipsilateral urogenital ridges and the crossing over occured during testicular migration.[12] According to Kimura who reviewed 11 cases of TTE, there was no true ectopia with an abnormal descent of the testi unless two separate vasa exist.[13] The investigation of crossed testicular ectopia included transabdominal US, MRI of abdomen and pelvis and contrast-enhanced CT to look for associated anomalies.[3,5,14] The role of MRI in the diagnostic approach of this pathologic entity was fundamental, as it can reliably distinguish the presence of two separate from a single fused vas deferens. To our knowledge, Akin et al and Yıldız et al have reported the two largest case-series of TTE so far, each one with six patients.[1,15] Most cases of TTE described in the literature are diagnosed before the age of 18 and management was targeted to protect fertility and reconstruct a normal anatomy by transferring the testis and repairing any associated anomalies such as inguinal hernias. In our case both testes were preserved and herniotomy was done. Gaur et al reported a successful transseptal orchidopexy in a 21 year old azoospermic adult with two equal-sized fused testes but with two distinct separate spermatic cords and normal vas.[16] Yanaral et al did not perform a surgical correction in a 19 year old azoospermic patient with fusion.[3] In 2015, Bascuna et al proposed a treatment algorithm which included extensive mobilization of the vas and the spermatic vessels to allow for transseptal fixation.[17] This was subsequently challenged by Raj et al in 2017 who argued for a less aggressive approach taking into consideration the length of the vas and even settling with a fixation in the same hemi-scrotum without jeopardizing striping the testis off it vascular supply.[18] In any case the aforementioned algorithm applied to young patients, whose testes were still developing and could be favored by a reconstruction. In our case, the patient was a toddler without associated anomalies, but with inguinal hernia (Amyand's hernia) where hernial contents reduced and hernitomy done. Testicular transfer was not performed for two reasons: firstly, because the need for excessive dissection and mobilization of vas deferens distorting the anatomy of the native testis; secondly, a paucity of data in the literature favouring such a management for restoring fertility in adults. Also it was uncertain whether TTE poses an independent risk factor for testicular malignancy but has been linked to seminomatous, non seminomatous germ cell tumours, and teratomas in published literature.[19,20] In summary, TTE was a rare congenital anomaly which should be included in the differential diagnosis of every young patient with an absent testis and fertility problems.

CONCLUSION

In this setting, clinicians should maintain a high index of clinical suspicion in every young patient with symptoms of testicular swelling with pain. If diagnosed, a thorough investigation should be employed for associated anomalies, taking into consideration the wide spectrum of associated conditions. Finally, it should be noted that the current classification system does not discriminate between a solitary and two distinct vasa deferentia, which might radically affect the therapeutic approach applied on patients exactly like the one presented here.

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REFERENCES

1. Akin M, Erginel B, Bilici S, Gedik S, Yıldız A, Karadağ CA, et al. Crossed testicular ectopia: report of six cases. Afr J Paediatr Surg. 2014;11(3):269-72.
2. 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. 2017;31:167-9.
3. Yanaral F, Yildirim ME. Testicular fusion in a patient with crossed testicular ectopia: a rare entity. Urol Int. 2013;90(1):123-4.
4. Naji H, Peristeris A, Stenman J, Svensson JF, Wester T. Transverse testicular ectopia: three additional cases and a review of the literature. Pediatr Surg Int. 2012;28(7):703-6.
5. Naouar S, Maazoun K, Sahnoun L, Jouini R, Ksia A, Elezzi O, et al. Transverse testicular ectopia: a three-case report and review of the literature. Urology. 2008;71(6):1070-3.

6. Lenhossek MV. Ectopia testes transverse. Anat Anz. 1886;1:376.
7. Esteves E, Pinus J, Maranh˜ao RF, Abib SC, Pinus J. Crossed testicular ectopia. Sao Paulo Med J. 1995;113(4):935-40.
8. Vaos G, Zavras N. Irreducible inguinal hernia due to crossed testicular ectopia in an infant. Hernia. 2004;8(4):397-8.
9. Kaul KN, Srivastava SM, Rehman R, Goel V, Yadav V. Persistent Mullerian duct syndrome with transverse testicular ectopia presenting as an incarcerated inguinal hernia. Hernia. 2011;15(6):701-4.
10. Thevathasan G. Transverse ectopia of the testis. ANZ J Surg. 11967;37(2):93-102.
11. Gauderer MWL, Grisoni ER, Stellato TA, Ponsky JL, Izant RJ. Transverse testicular ectopia. J Pediatr Surg. 1982;17(1):43-7.
12. Givel J, Merlini M, Clarke DB, Dusmet M. Surgery of the thymus. 1st ed. Berlin, Heidelberg: Springer Berlin Heidelberg; 1990.
13. Kimura T. Transverse ectopy of the testis with masculine uterus. Ann Surg. 1918;68(4):420-5.
14. Dahal P, Koirala R, Subedi N. Transverse testicular ectopia: a rare association with inguinal hernia. J Surg Case Rep. 2014;2014(10):97.
15. Yildiz M, Yi˘giter A, Bakan V. Transverse testicular ectopia. Pediatr Int. 2014;56(1):102-5.
16. Gaur DD, Purohit KC, Joshi AS, Gundeti MS. Crossed testicular ectopia: a rare case of knotting of the cords with testicular fusion. Br J Urol. 1998;81(6):924-5.
17. Bascuna R, Ha JY, Lee YS, Lee HY, Im YJ, Han SW. Transverse testis ectopia: diagnostic and management algorithm. Int J Urol. 2015;22(3):330-1.
18. Raj V, Redkar RG, Krishna S, Tewari S. Rare case of transverse testicular ectopia-case report and review of literature. Int J Surg Case Rep. 2017;41:407-10.
19. Eastham JA, McEvoy K, Sullivan R, Chandrasoma P. A case of simultaneous bilateral non-seminomatous testicular tumors in persistent Mullerian duct syndrome. J Urol. 1992;148(2):107-8.

    Zhu Y, Zhang S, Ye D, Shi G, Xiao W. Yolk sac tumor in a patient with transverse testicular ectopia. World J Surg Oncol. 2011;9(1):91.

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