National Board of Examination - Journal of Medical Sciences
Volume 2, Issue 3, Pages 270–277, March 2024
DOI 10.61770/NBEJMS.2024.v02.i03.010

CASE REPORT

Cryptorchidism with Duplicate Vas Deferens and Epididymis: A Rare Case with Literary Review

Anil Kumar Nallabothula,[1] Anirudh Suseel Nalumaru[2,*], Anupam Bhargava[2], Ashish Kumar Jayant[2] and Harsha Vardhana Varma Mudunuri[2]

[1]*Professor and HOD, Department of Urology, Sri Venkateswara Institute of medical sciences (SVIMS), Tirupati, Andhra Pradesh, India*
[2]*Assistant Professor, Department of Urology, Sri Venkateswara Institute of medical sciences (SVIMS), Tirupati, Andhra Pradesh, India.*

Accepted: 18-January-2024 / Published Online 27-February-2024

Abstract

Background: Incidence and recognition of the congenital anomalies involving vas (or) ductus deferens, also called as sperm duct, are low, particularly when duplication of the sperm duct and epididymis occurs together. These anomalies, although of rare incidence, should be considered while performing surgeries related to the spermatic cord, to prevent inadvertent damage.

Case presentation: We, here, present a case of a seventeen year old boy with undescended testis on left side, where laparoscopic evaluation revealed polyorchidism. The accessory testis was removed, and histo-pathological examination confirmed duplication of the sperm duct and the epididymis.

Conclusion: This case scenario highlights the importance of recognizing and managing rare urogenital anomalies during procedures involving handling of spermatic cord structures.

Keywords: Cryptorchidism, polyorchidism, duplicate vas deferens, duplicate epididymis, vanishing testis

*Corresponding author: Anirudh Suseel Nalumaru
Email: sudarsansrikanth.1991@gmail.com

Background

Cryptorchidism is not an uncommon presentation. But rare is polyorchidism with an estimate of less than 200 cases reported in the medical literature. Prevalence of sperm duct anomalies is quite less and estimated at less than 0.05 % of the general population. Varieties of anatomical variations were recognized in the sperm duct and epididymis but are quite rare [1]. Duplication is such a rarer entity. It is defined as the identification of a second sperm duct in the spermatic cord contents, and it should not be mixed-up with double vas deferens, an entity which describes ipsilateral agenesis of the kidney with a blind ureter that ends in ejaculatory system. It has been encountered in various surgical procedures involving spermatic cord, which include inguinal hernia management, orchidopexy, varicocoelectomy, vasectomy and radical prostatectomy. We operated a case of undescended testis found to be polyorchidism following laparoscopic examination and inguinal exploration. Histo-pathological examination revealed the presence of duplication of the sperm duct and the epididymis with possible vanishing testis. Here, we present our case scenario with reference to the pre-existing literature.

Case Presentation

History and clinical findings: A seventeen year old male with otherwise no known comorbidities came to our out-patient clinic with the complaint of absent left testis. He had no genito-urinary or abdominal complaints. Surgical, medical, and family histories were non-contributory. He was a term baby delivered through normal vaginal delivery with uneventful perinatal period. On examination his right testis was present in normal position. Left hemi-scrotum was empty, but well developed with good rugosities. Left testis could be identified as a bulge in the left inguinal region more towards deep ring with preserved testicular sensation. On duplex ultrasound examination, the position of left testis was confirmed and the size and vascularity were comparable with the right testis. Basic preoperative workup, which included baseline investigations, cardiac and anaesthesia consultation were done and found to be normal.

Therefore, we proceeded with laparoscopy followed by inguinal exploration and orchidopexy.

Operative findings: Laparoscopic examination was normal and no abdominal cryptorchidism or complete urogenital non-union was noted and fibrous adhesions were released around the cord and vessels till they entered deep ring to provide adequate length for scrotal fixation of testis. A solitary spermatic cord was noted entering the left deep inguinal ring. Then inguinal incision was given and layers dissected. Spermatic cord was identified; internal spermatic fascia was laid open. We could see two testes in the spermatic cord (Figure 1). Proximal testis, which was normal in size for the age, was connected to spermatic cord which contained testicular vessels whereas distal small testis had connection with a second spermatic cord which had only a sperm duct (Figures 2 and 3). We removed the distal testis along with its vas deferens because of its smaller size as compared to the proximal testis and absence of it's to the blood vessels. The proximal testis was mobilised into scrotum and fixed in the sub-dartos pouch. Wounds were closed with absorbable sutures, procedure was uneventful and scars healed well (Figure 4).

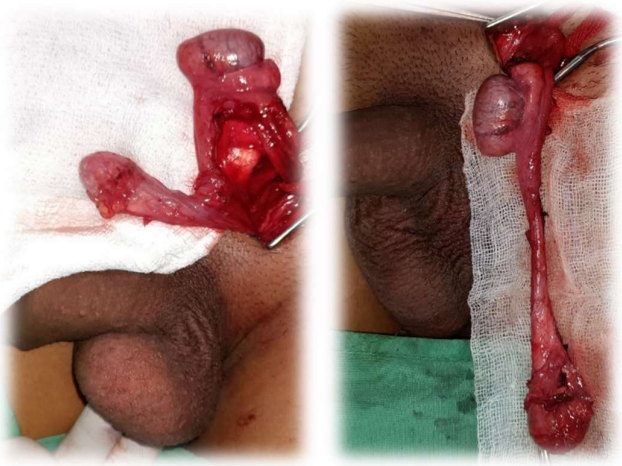

Figure 1. Intra op –Duplication of left testis with normal looking proximal testis and abnormal distal accessory testis

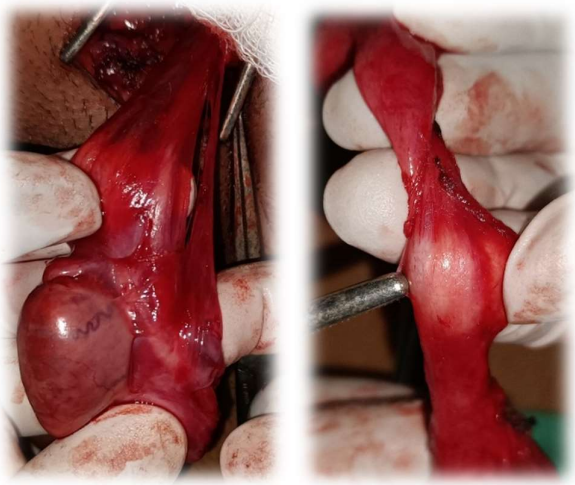

Figure 2. Intra op – normal proximal testis and its vas

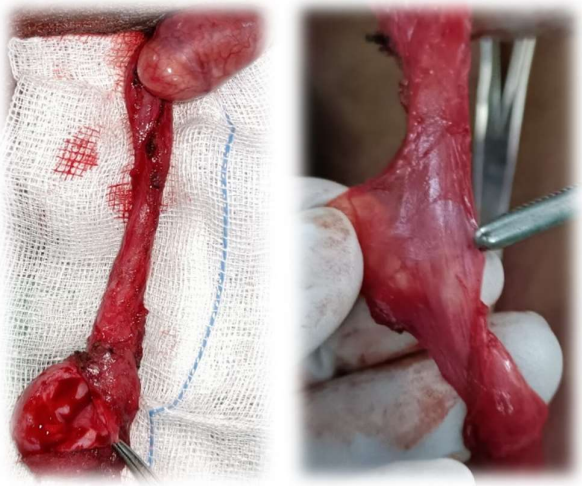

Figure 3. Intra op – accessory distal testis and its vas

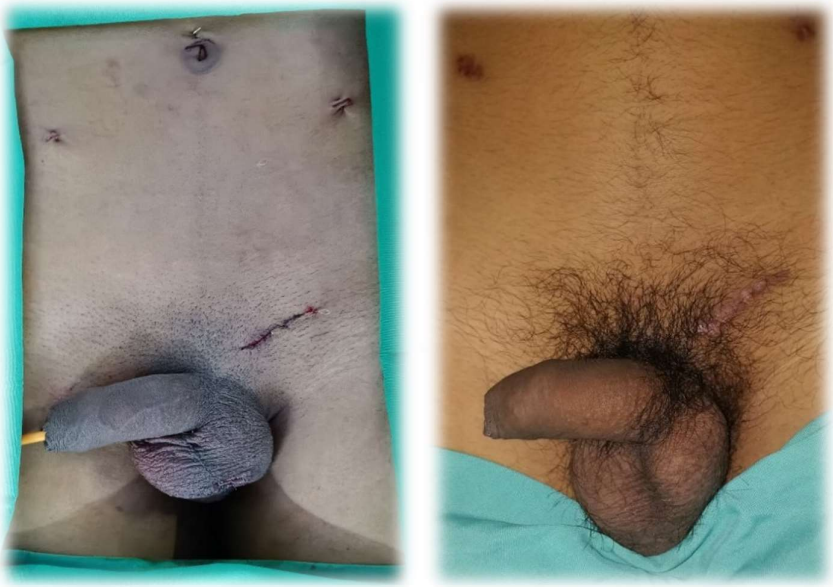

Figure 4. Post op day zero & after 6 weeks

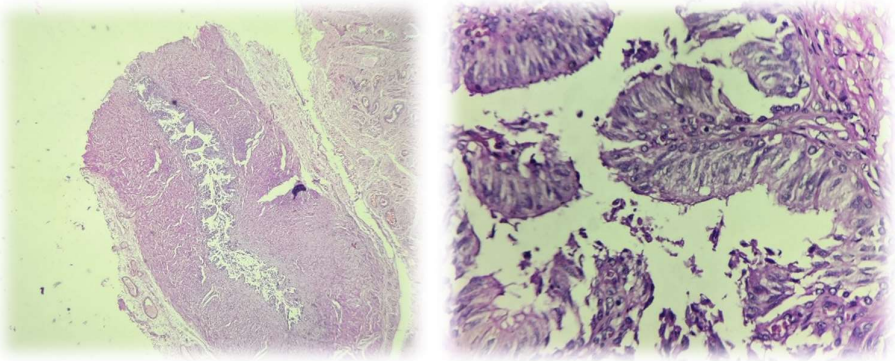

Figure 5: A - Vas deferens 40 x, B - Vas deferens 400 x

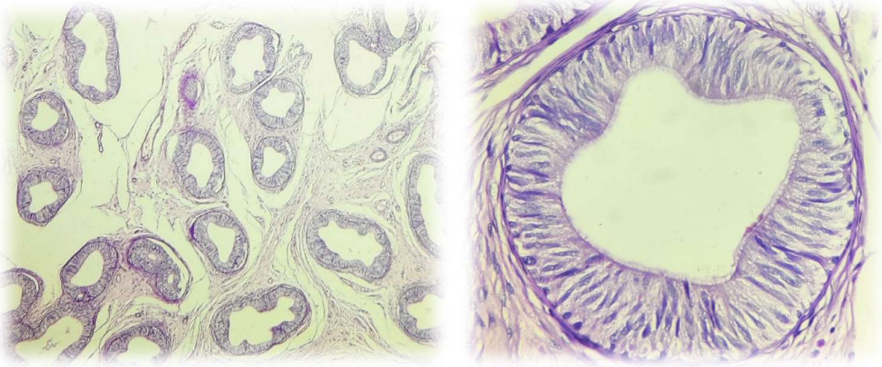

Figure 6: A - Epididymis 40 x, B - Epididymis 400 x

Histo-Pathological examination findings: The resected distal testis showed only fibrous tissue was found and it had no seminiferous tubules, sertoli cells or sperm cells. But normal epididymal and vas deferens micro-anatomy was identified (Figures 5 and 6).

Discussion

The case presented here involved the presence of a duplicate sperm duct and epididymis presenting as polyorchid testis in a cryptorchid patient. The primary sperm duct in communication with the proximal testis was normal. This duct was seen arising directly from the epididymis, ran through the inguinal canal straight into the left deep inguinal ring. Distal supposedly "duplicate testis" had a sperm duct which was connected to the primary sperm duct at the deep inguinal ring proximally. This accessory testis was found to have only epididymis along the tunica vaginalis.

There is no exact explanation for the formation of polyorchidism, but various theories were proposed in the literature.

Leung, in 1988, described the variations in the anatomy (Table 1 and Figure 7) by a possible embryological origin. According to him, the type II variation is most common and types II and III clubbed together account for almost more than 90 % of cases of polyorchidism [2].

Singer et al., in 1992, suggested a different classification of polyorchidism which was based on both anatomy and function. [3].

- **I:** The supernumerary testes are attached to the epididymis and vas deferens with proper drainage and have reproductive potential (Similar to Leung Types II, III and IV).
- **II:** Testes with lack of such contiguity or attachment, hence they have no reproductive potential (Similar to Leung Type I).

Bergholz et al. presented an anatomical classification system of polyorchidism according to testicular reproductive function (Table 2 and Figure 8). Here anomalies of the sperm duct and the epididymis were grouped as anomalies in number, like absence or duplication, variation in location, like ectopia, abnormality in continuity, like segmental hypoplasia, and aberrations in integrity, like a diverticulum [1].

Liang et al. developed a classification system exclusively for poly-vasa deferentia [4].

- **I** - Duplicate vas deferens in the spermatic cord, but no polyorchidism.
- **II** - Multiple vas with presence of polyorchidism.
- **III** - False poly-vasa deferentia. Here an ectopic ureter is noted which drains into ejaculatory system.

Based on the above classification by Liang and his team, our patient belongs to either Type I poly-vasa deferentia or Type II poly-vasa deferentia with vanishing testis.

Table 1: The Leung classification of polyorchidism [2]

- **Type-I:** supernumerary testis lacks an epididymis or vas and has got no attachment to the usual testis. (Division of genital ridge only).
- **Type-II:** the supernumerary testis drains into epididymis of usual testis and they share a common vas. (Division of genital ridge occurs in the region where the primordial gonads are attached to the metanephric ducts, although the mesonephros and metanephric ducts are not divided, i.e.-incomplete division).
- **Type-III:** the supernumerary testis has its own epididymis and both epididymis of the ipsilateral testes draining into one vas. (Complete transverse division of mesonephros as well as genital ridge).
- **Type-IV:** complete duplication of testes, epididymis and vas. (Vertical division of genital ridge and mesonephros).

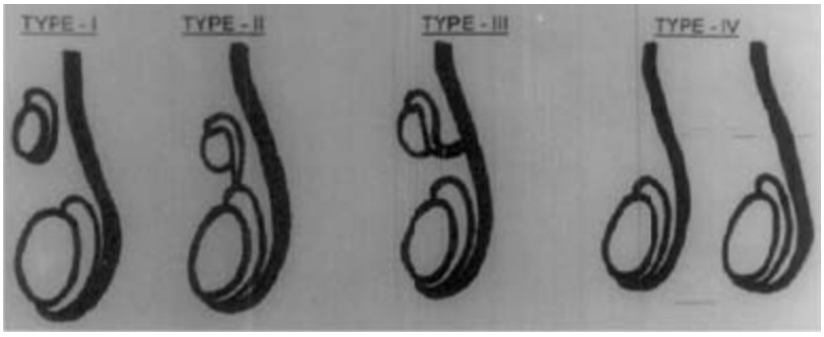

Figure 7. The Leung classification of polyorchidism [2]

Table 2: The Bergholz classification of polyorchidism [1]

Type A	Type B
Testis drained by outflow path (vas deferens)	Testis not drained by outflow path
A1 - Testis with its own epididymis and vas deferens	B1 - Undrained testis with its own epididymis
A2 - Testis with its own epididymis but shares a vas deferens with neighboring testis	B2 - Undrained testis with no epididymis
A3 - Testis shares a common epididymis and vas deferens with neighboring testes	
A4 - Testis has its own vas deferens but shares a common epididymis with neighboring testis	

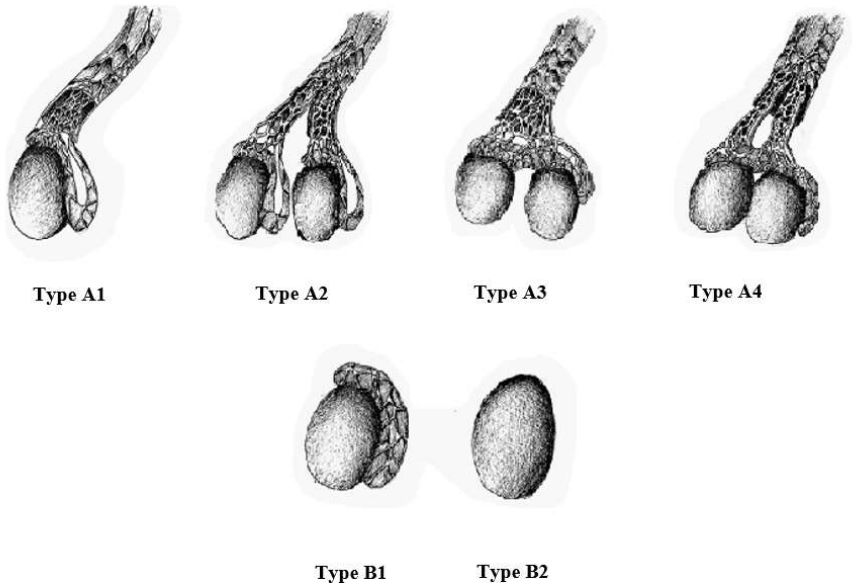

Figure 8. The Bergholz classification of polyorchidism [1]

Patients with cryptorchidism may have frequent association with anomalies of the vas deferens and epididymis. To our knowledge, there are very few reported cases of duplication of sperm duct and epididymis. One such case scenario is of an infant with left cryptorchidism who underwent left orchidopexy at an age of twelve months and intraoperatively they found two testes within the spermatic cord. In this child, the distal testis was connected to the testicular vessels along with a sperm duct whereas the proximal testis was communicating with only a sperm duct. The distal testis was preserved and orchidopexy was done. The other (proximal) testis was removed and sent for histo-pathology, which revealed only grouped tubes covered with a simple columnar epithelium. This was surrounded by spindle cells with no identifiable testicular tissue [5]. The other documented case is that of a four year old boy whose left testis was impalpable. He underwent laparoscopy which revealed, not only the sperm duct entering the internal inguinal ring, but also a small intra-abdominal testis which was supplied by the testicular artery and vein. Inguinal canal, on exploration, revealed that the vas terminated in a nubbin of tissue. Histology showed epididymal structures in the areas both adjacent to the testis as well as in the terminal nubbin of the sperm duct. This case scenario is an example of the urogenital non-union. A blind-ending sperm duct found intra-operatively on exploration of inguinal canal might be considered as an evidence of vanishing testis syndrome [6].

We assume that the aetiology of our case is polyorchidism with left duplex testis accompanied by vanishing distal accessory testes. This is supported by the presence of well-developed rugosities of left hemi-scrotum which may be because of the distal testis which later atrophied. Urogenital non-union has an embryological explanation and it is frequently associated with a cryptorchid testis. Hence, we suggest that patients with complete urogenital non-union should be

evaluated further by diagnostic laparoscopy, to either identify the presence of an intra-abdominal testis or confirm total absence of the testis. Our patient did not have a complete urogenital non-union.

Conclusion

Here, we report one of the rarest cases – A patient with undescended testis found to be poly orchid during surgery but confirmed as duplication of vas and epididymis on histo-pathological examination. Surgeons must be aware of this abnormality while doing surgeries involving inguinal exploration or handling of spermatic cord in order to avoid any iatrogenic injury to cord contents.

Conflicts of interest

The authors declares that they do not have conflict of interest.

Funding

No funding was received for conducting this study.

References

1. Bergholz R, Koch B, Spieker T, Lohse K. Polyorchidism: a case report and classification. J Pediatr Surg. 2007;42(11):1933–5. http://dx.doi.org/10.1016/j.jpedsurg.2007.07.031
2. Leung AK. Polyorchidism. Am Fam Physician. 1988;38(3):153–6. https://pubmed.ncbi.nlm.nih.gov/3046267/
3. Singer BR, Donaldson JG, Jackson DS. Polyorchidism: functional classification and management strategy. Urology. 1992;39(4):384–8. http://dx.doi.org/10.1016/0090-4295(92)90220-q
4. Liang MK, Subramanian A, Weedin J, Griffith DP, Awad SS. True duplication of the vas deferens: a case report and review of literature. Int Urol Nephrol. 2012;44(2):385–91. http://dx.doi.org/10.1007/s11255-011-0042-7
5. Terawaki K, Satake R, Takano N, Yonekawa H, Ikebukuro K, Komuro H, et al. A rare case of duplicated vas deferens and epididymis. J Pediatr Surg Case Rep. 2014;2(12):541–3. http://dx.doi.org/10.1016/j.epsc.2014.11.005
6. Foley PT, Sparnon AL, Lipsett J. Urogenital nonunion--the case for laparoscopy for the impalpable testis. Pediatr Surg Int. 2005;21(8):655–6. http://dx.doi.org/10.1007/s00383-005-1396-7