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Polyorchidism with Oligospermia: A Rare Case Report with the Current Review

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Abstract

Polyorchidism means the presence of more than two histologically proven testes in the scrotum. A 28-year-old man with polyorchidism with oligospermia and infertility presented in surgery OPD. In our case, the supernumerary testis was present in the left scrotum, superior to the left testicle, along with varicocele. The reported incidence of polyorchidism with varicocele is relatively low (1.4%), and only a few cases have been reported in the literature. Scrotal colour doppler, ultrasonography and magnetic resonance imaging confirmed the presence of double testes. Microscopic varicocele ligation was done to treat oligospermia. Semen quality and quantity improved during follow up.

Keywords: polyorchidism; supernumerary testis; oligospermia; varicocele

Introduction

Polyorchidism is an uncommon congenital anomaly. It is defined as the presence of more than 2 histologically proven testes. Most of the time, only three testes are present, but as many as five testes have been reported in the literature.[1] In1670, the first clinical case was reported, but in 1895, the first histologically confirmed case was described.[2]The supernumerary testis can be accompanied by male testicular descent (22%–40%), hernia (24%–30%), painless scrotal or inguinal swelling (16%), torsion (15%), hydrocele (9%), pain (7%), malignancy (6%), and varicocele (1.4%)[3]Most common site is left half of the scrotum. In our case, the extra testicle was also present in the left scrotum. Supernumerary testicles are mostly within the scrotum(75%) but can also be found in the inguinal region(20%) or the abdomen(5%).[4]So far, about 200 cases have been reported in the literature.[5,6] As per the literature, our case is third in the number who has presented with varicocele and polyorchidism).[7]

Case Report

A 28-year-old man presented to our outpatient clinic with a history of left scrotal mass and infertility. On palpation, the right testis was normal. On examination, two almost equal-sized masses were palpable in the left scrotum with grade three varicocele (Fig-1).

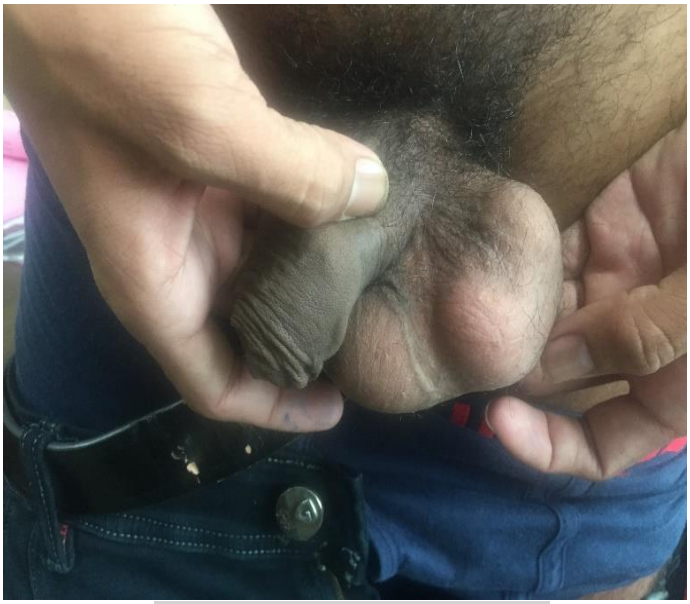

Fig-1 showing a photo of three testes

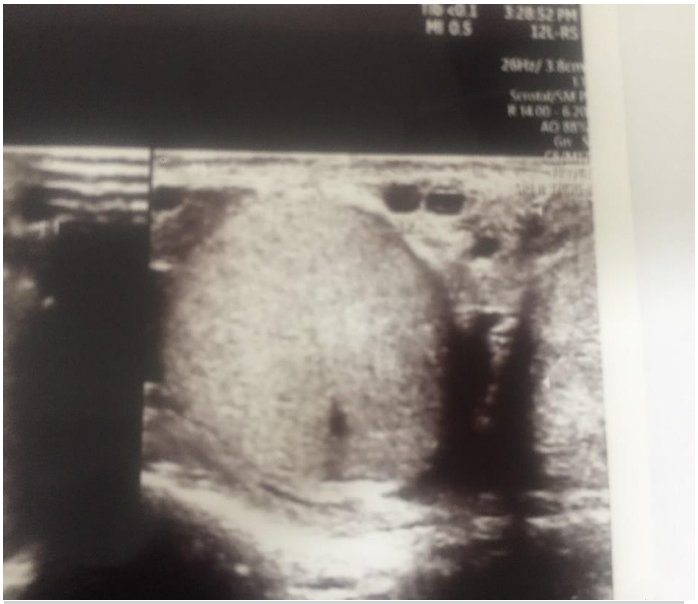

Fig-3 U/S showing normal testis in the right half of the scrotum

Follicle-stimulating hormone, testosterone, bHCG, and LDH were within normal limits. Sperm analysis showed oligoasthenospermia. The mean values on two semen evaluations in 15 days showed moderate oligoasthenospermia. Semen analysis one revealed less than 2 million sperm per ml, 50% motility [1% progressive, 49% nonprogressive], immotile 50%, 20 % normal morphology and 80% abnormal morphology and 0-1 pus cell/HPF, 3.0 ml volume.Semen analysis two revealed>2 million sperm per ml,40% mortality[2% progressive,38%nonprogressive],60%immotile,30%normal morphology,70%abnormal,nil pus cells/HPF, 2 ml volume.Abdominal ultrasonography and chest radiography were normal. Scrotal ultrasound revealed two testes on the left half and normal size testis in the right half of the scrotum (Fig-2Fig-3).

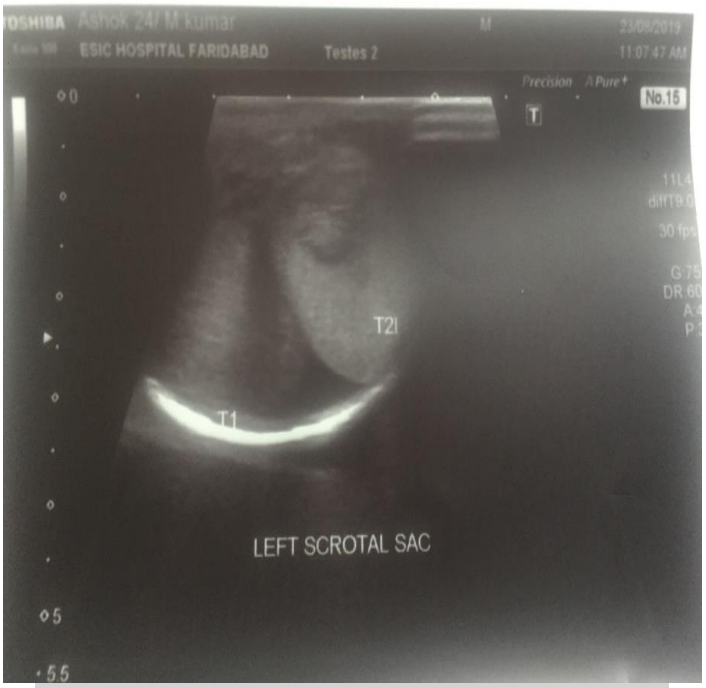

Fig-2 U/S showing two testes in the left half of the scrotum

Color Doppler ultrasonography of scrotum showed polyorchidism and grade IV varicocele on the left side with minimal hydrocoele (Fig-4). The echogenicity of both testes was normal, thus ruled out malignancy. The Colour doppler study also revealed normal flow and waveform noted bilaterally. Both the testes in the left scrotal sac showed normal vascularity. Magnetic Resonance Imaging of the scrotum confirmed two separate testes along with two separate epididymides and the single spermatic cord in the left hemiscrotum(Fig-5).

The main reason for surgery was the presence of grade IV varicocele & oligospermia. After taking consent for surgery, Exploration of the left inguinal region was done under spinal anaesthesia. The inguinal canal was opened, and testes were delivered. Two testes surrounded by a common tunica vaginalis were found. The distal testis was measured as 3.5 x 3 x2.5 cm, and the proximal testis 3x 2.5 x 2 .5cm. Two left testes have a common spermatic cord. Biopsies were done in both testes, which revealed normal spermatogonial proliferation. There were no signs of malignancy. Microscopic varicocele ligation was done. After 9 months, the mean values on two semen evaluations in 15 days showed improvement. (3.2 ml volume, > 15 million sperm per ml, 60% motility). After one year of follow up, scrotal ultrasonography and physical examination were normal.

Discussion

Polyorchidism is an uncommon testicular congenital anomaly. Triorchidism is the commonest occurrence, and the supernumerary testis is often located in the left half of the scrotum. Even the presence of four testes has reported in the literature.[8,9] The supernumerary testis's most common site is within the scrotum, superior or inferior to the ipsilateral testicle.[5] In our case, the supernumerary testis was located in the left scrotum, superior to the left testicle along with varicocele. The reported incidence of polyorchidism with varicocele is relatively low (1.4%), and only a few cases have been reported in the literature.

In most cases, polyorchidism diagnosis is incidental. Commonconcomitant disorders associated with supernumerary

testis are testicular maldescent (22%–40%), hernia (24%–30%), painless scrotal or inguinal swelling (16%), torsion (15%), hydrocele (9%), pain (7%), malignancy (6%), and varicocele (1.4%). [3,10] Our case presented with infertility and oligospermia due to left side polyorchidism with grade III varicocele.

The exact cause of polyorchidism is not clear. This may occur due to defective testicular development. The urogenital ridge develops into the testis. Mesonephric ducts form epididymis and vas deferens. The urogenital union of the urogenital ridge and mesonephric ducts begins after 3 months, and they undergo transverse division.[11]

According to embryological theories, there is a duplication of the genital ridge or division of the genital ridge to form supernumerary testis.[12-15] The most likely is that polyorchidism results from the transverse division of the urogenital ridge between the fourth and sixth weeks. The theory of genital ridge duplication only explains some of the variations found. If a more medial duplicated ridge existed, it would be less likely to communicate with the mesonephric tubules. Therefore, it would lack an outflow path, which only occurs sometimes with polyorchidism.[13,16]This leaves only division of the genital ridge that accounts for all variations. As the primitive gonadal ridge is an elongated structure, transverse fragmentation could occur at any level and extend onto the mesonephric duct.[13,16]This would explain the inequality of the two testes.[17] If the separation were located caudal to the persisting group of mesonephric tubules, the superior gonad would develop spermatogenically active tubules, and the caudal gonad would lack an outflow path.[12,15]The duplication of the testes with a single epididymis and vas deferens is more common.[17]The more common anomalies associated with polyorchidism are maldescent of one of the supernumerary testes(15–50%), inguinal hernia (30%) and testicular torsion(13%).[3,18] Other associations include hydrocele, epididymitis, varicocele, infertility, cryptorchidism and malignancy.[18] Abdominal polyorchidism has also been reported in the literature.[19]

Leung classification of polyorchidism is based on testis embryology: 1) polyorchidism without vas deferens or epididymis in the supernumerary testis, 2) testes sharing these two structures with the ipsilateral testicle, 3) testes with their epididymis and sharing the vas deferens, and 4) supernumerary testes with their annexes.[20] Our patient was considered to be in group 3.

Singer et al. [15] performed a polyorchidism classification according to the localization of the extra-testis and the patient's fertility status. Our patient was infertile. In a meta-analysis study done by Bergholz and Wenke, it was found that the majority of cases were present on the left side (65%) as in our case [21]

Based on embryologic development, Thum classified polyorchidism into three types. Type 1: The supernumerary testis without an epididymis and vas. Type 2: The supernumerary testis is linked to the normal testis by a common epididymis and shares a common vas with it. Type 3: The supernumerary testis has its epididymis but shares the vas with the normal testis.[14] In our case, it was type3 according to Thum's classification.

Bergholz classified based on the supernumerary testis' anatomy and reproductive potential as shown in table 1, and our patient had Type A2 polyorchidism.

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	

Table 1: is showing the Bergholz classification of polyorchidism. [22]

The extra testicle usually presents as a scrotal mass predominantly left-sided. In our case, the extra testicle was also present in the left scrotum. Most of the time, the right side testis is undescended, but polyorchidism is often seen on the left side. The larger size of the left testis might subdivide more readily than the right testis during development. [23]

There are typical sonographic features of polyorchidism, and the diagnosis is made based on sonography findings [7,24]. Magnetic resonance imaging can be used for confirmation but is more useful in cases complicated by cryptorchidism or neoplasia.

Treatment of polyorchidism depends upon the site of the testis and on ultrasound findings. If the supernumerary testis is intrascrotal and the patient is fertile with a healthy testis, do a close follow-up to preserve spermatogenesis. Whereas others recommend excision of the supernumerary testis due to the risk of malignancy.[25] Thus the management of polyorchidism depends upon two critical factors. The first is the reproductive power of polyorchid, and the second being the risk of malignancy in the polyorchid testicle. Thus patients with polyorchidism can be divided into two major groups; -

1-Simple polyorchidism: Supernumerary testes are in an orthotopic position without any malignancy signs and any association with other abnormalities. Conservative management is a choice in this group.

2-Complicated Polyorchidism: Supernumerary testes are associated with other abnormalities. This group was subdivided:

2a)-Polyorchidism plus signs of malignancy (Ultrasonographic Features, Elevated Tumor Marker Levels). They were advised to perform radical orchiectomy of all testes on the suspicious side.

2b-Polyorchidism plus torsion. Then orchidopexy if the testis is not necrotic; in another situation, orchiectomy.

2c-Polyorchidism plus cryptorchidism. If the patient is young, then orchidopexy is advised. In contrast, in adults, a supernumerary testis in the ectopic position means an unnecessary risk of malignancy. Hence, then excision of the supernumerary functional ectopic testis is done in adults.[26] Polyorchidism may be associated with varicocele with primary infertility (as in our case). If infertility is associated with this condition, then both testes are preserved.[27] Microscopic varicocele ligation along with fixation of the testis is the treatment of choice in such cases.

Our case presented with polyorchidism with oligospermia and varicocele. Despite no malignancy risk (due to the intrascrotal site, healthy testis on U/S and normal markers), we operated on our patient to treat oligospermia with infertility. Microscopy

varicocele ligation was done to improve the quality and quantity of semen. Both testes were preserved. Postoperative follow up showed improved semen quantity as well as quality.

Conclusion

Polyorchidism or supernumerary testis is a rare congenital anomaly. No surgical exploration is needed if there is no associated disorder and on imaging testis is healthy. If Supernumerary testis is found incidentally during a groin exploration, then frozen section examination should be done to rule out malignancy. Orchidopexy should be performed to minimize the chances of torsion. Psychological considerations must be kept in mind as most of these patients are young adult.

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