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**Case Report**

# A rare presentation: duplication of testis

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## ABSTRACT

Objective of current study was to describe the clinical and embryological futures of polyorchidism and review of literature of similar cases. Polyorchidism is usually an incidental finding and an extremely rare congenital anomaly with less than 200 cases reported so far. Several theories regarding it but the exact explanations are still unknown. In majority of cases, patients are asymptomatic and have a painless inguinal or scrotal mass. While others may present with maldescended or cryptorchidism or incidentally discovered in association with indirect inguinal hernia, hydrocele, varicocele, epididymitis, infertility, testicular torsion, and extremely rare with testicular malignancy. Among polyorchidism, majority of cases are of triorchidism usually of left side. In present case report, triorchidism in its rarest form is diagnosed incidentally intraoperatively in an adolescent presented with left groin swelling and ipsilateral undescended testicle. The supernumerary testicle was intraoperatively found in peritoneal cavity and completely developed with separate cord structure and epididymis.

**Keywords:** Polyorchidism, Triorchidism, Supernumerary testis

## INTRODUCTION

Polyorchidism is an extremely rare congenital anomaly of urogenital system and refers to presence of more than two testicles. Less than 200 cases have been reported so far. This case thought to be unique as it describes the anomaly in its rarest presentation-Triorchidism with complete duplication of testis, epididymis and vas deferens. Hence reported to provide more information about this condition and to build up a basic management protocol.

## CASE REPORT

An 11 year old boy presented with left undescended testis since birth and a painless left single swelling at the neck of scrotum. The patient had no other medical history and clinical examination confirmed patients present complaints. Ultrasonography reveals left undescended testis. Laboratory conditions are within normal limits.

Surgical exploration planned for left orchidopexy on routine basis. Intraoperatively single undescended testicle in left inguinal canal reaching up to neck of scrotum observed. During its traction and repositioning in Dartos pouch another completely developed testicle with epididymis and vas deferens traced intra abdominally. Both testicles have separate well developed cord structures.

As the intra-abdominal testicle appears flabby and due to increased chances of torsion, devascularization and a possibility of malignancy in future, the intra-abdominal testicle removed and sent for histopathology which confirmed normal well developed testicle.

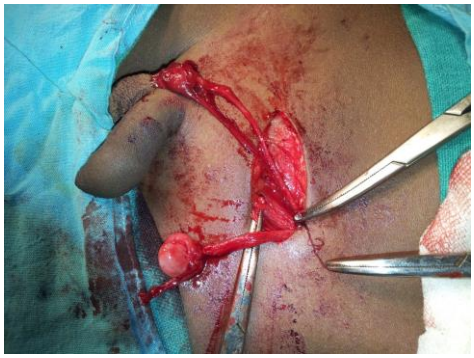

**Figure 1: Duplication of testis on left side.**

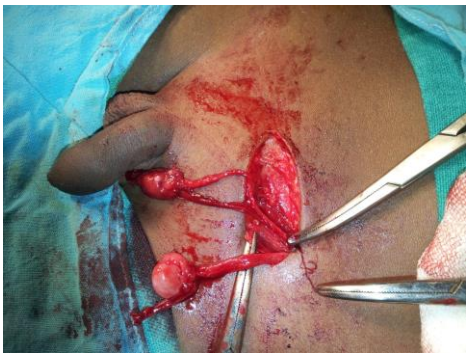

**Figure 2: Two completely developed testicle with separate vas deferens.**

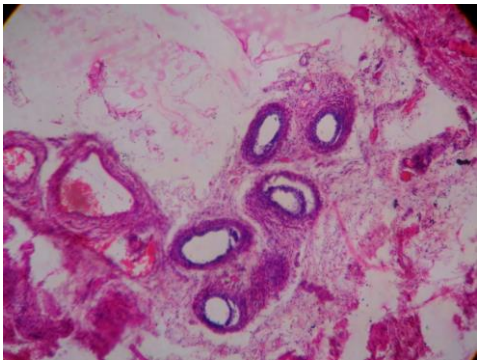

**Figure 3: Low power view showing well developed seminiferous tubules and intact vascular supply.**

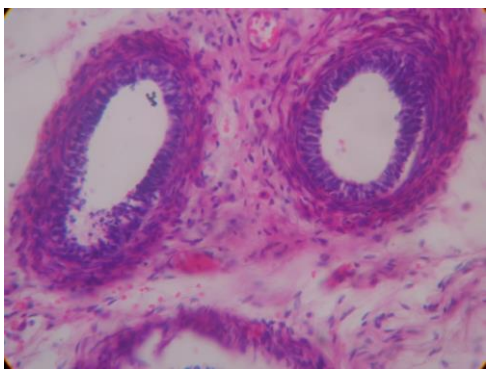

**Figure 4: High power view of seminiferous tubules.**

## DISCUSSION

### *Historical review*

Blasius[1] recorded the first case at routine autopsy in 1670. Ahlfeld[2] gave the first description in 1880. The first clinical case report was made by Lane[3] in 1895. Fewer less than 200 cases reported so far. Leung[4] described the anatomical variation on possible embryological basis (1988) which later on modified by Thum[8] (1991). Singer[5] (1992) suggested anatomical as well as functional classification.

### *Embryology and etiology*

In a normal embryo at about 6 weeks of embryonic life, the primordial testis develops from primitive genital ridge medial to mesonephric ducts.[6] At about 8 weeks primordial testis takes shape and epididymis and vas deferens arise from mesonephric duct.[4-6]

The exact mechanism for polyorchidism is unknown but few theories have been proposed on embryological basis. First is anomalous appropriation of cells in the genital ridge before 8 weeks of gestation. Second describes anomalous duplication, transverse or longitudinal division of genital ridge. Third is on the basis of incomplete degeneration of mesonephrones and development of peritoneal bands. No single theory can explain all forms of polyorchidism.[1,4-7]

In present case, there may be complete longitudinal division of genital ridge and mesonephric duct might be occurred resulting in complete duplication of testis, epididymis and vas deferens which is least common of all presentations.[4]

### *Classification*

On the basis of embryological development, Leung gave the first classification (1988) which later on modified by Thum (1991).[4,8]

Type A: The supernumerary testicle lacks an epididymis and vas deferens due to division at the tip of genital ridge not in contact with the mesonephric duct.

Type B: The supernumerary testicle has its own epididymis. Depending on the degree of division , it may be either connected longitudinally to the epididymis of normal testis and its vas deferens (B2) or it may lack any connections to the normal testis at all (B1). The division of genital ridge occurs transversely in the region where the primordial gonads are attached to the mesonephric ducts.

Type C: The supernumerary testicle has its own epididymis and both epididymis of the ipsilateral testis drain into single vas deferens. It occurs due to complete

transverse division of genital ridge as well as mesonephric duct [5,9,10].

Type D: Complete longitudinal duplication of genital ridge and mesonephric duct occurs resulting in complete duplication of testis, epididymis, and vas deferens.[11,12]

Singer et al. (1992) suggested classification based on reproductive potential of supernumerary testis.[5]

Type I: The supernumerary testis has reproductive potential because of attachment to draining epididymis and vas deferens. It includes Leung type B, C and D.

Type II: The supernumerary testis has no reproductive potential because of lack of draining system. It includes Leung type A.

Both these types are further subdivided into A and B depending on location of supernumerary testicle within scrotum or ectopic respectively.

***Clinical presentations, associated anomalies and complications***

In most cases of polyorchidism, a single supernumerary testis (Triorchidism) is present with left side being more frequent.[4] Bilateral supernumerary testis have also been reported.[9,13,14] The most common location is within the scrotum, superior or inferior to the ipsilateral testicle.[6] However the extra testicle may either be completely separated from a maldescended testicle or it may be maldescended itself with two testicles present in the scrotum.[15,16]

Most patients with polyorchidism identified incidentally, are adolescents or young adults with mean age of 15 to 25 years,[17] but it may widely vary from 4 to 75 years as well.[5,27]

In uncomplicated cases, patients usually present a painless inguinal or scrotal mass since long time. About 40% cases are associated with cryptorchidism and testicular torsion occurs in about 15%. Other associated anomalies are inguinal hernia (30%), hydrocele (9%), vericocele (<1%), hypospadiasis (<1%), anomalous urogenital union (<1%) and extremely rare is malignancy (<1%).[17-20] Malignant transformation may occur within the supernumerary testis; irrespective of its location and most common reported neoplasm are embryonic carcinoma, germ cell tumor and seminoma.[21,22]

Spermatogenesis in supernumerary testis is normal in about 50% of cases. However reduced or absent spermatogenesis have also been reported.[11,14,23,24]

Most patients with polyorchidism have a normal 46 XY karyotype but chromosomal abnormalities such as 46 XX karyotype with XY mosaicism and deletion of long arm of chromosome 21 have also been reported. Adult patients have normal secondary sexual characteristics in most of cases.[13]

Diagnosis can also be made as incidental finding on Ultrasonography of inguinal or scrotal mass that has an identical echo texture to that of ipsilateral testicle. USG finding can be confirmed on MRI, revealing a round or oval structure with homogenous intermediate signal intensity on $T_1$ weighted images and high signal intensity on $T_2$ weighted images, typical signal characteristics of testicular tissue.[25,26]

The only entity which may resemble polyorchidism sonographically is spleenogonadal fusion. It is a rare congenital anomaly in which there is fusion of spleen, gonad, epididymis or vas deferens occurs. Scrotal sonography reveals a mass of similar echogenicity and it may mimic a supernumerary testicle. It is differentiated on testicular sulfur colloid scan confirming presence of ectopic splenic tissue.[25,26]

***Management***

As the anomaly is extremely rare and usually diagnosed as an incidental finding during ultrasonography or intraoperatively, so there is no clear cut protocol regarding its management. Current treatment is conservative in the absence of any complications.[26-28]

Orchidectomy and biopsy of supernumerary testicle for diagnosis or follow up are not indicated. However in presence of coexisting conditions, such as testicular torsion, cryptorchidism, malignancy, surgical treatment is warranted.[29-30]

In our case, as there is high risk of malignancy in the intra-abdominal supernumerary testicle in future, so a decision of Orchiectomy taken along with orchidopexy and Dartos pouch for ipsilateral undescended testicle.[26-30]

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