CASE REPORT

POLYORCHIDISM PRESENTING WITH INGUINAL HERNIA: A CASE REPORT AND A REVIEW OF LITERATURE

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ABSTRACT
Triorchidism is the commonest variety of polyorchidism, an entity with more than two testes is an extremely rare congenital anomaly of the testis. Although removal of the supernumerary testis is a safer alternative proposed, recent literature recommends more conservative approach in normal testes with vigilant regular follow up to screen for malignancy. This case presented as a left inguinal swelling diagnosed as indirect left inguinal hernia with left undescended testis. Intraoperatively indirect inguinal hernia was noted with supernumerary testis at deep ring in addition to normal left testis in hernia sac in the inguinal canal. The ectopic testis was small (2×2×1 cm) lacking epididymis and with short vas deferens. After performing herniotomy and orchidopexdy, supernumerary testis was excised for a possible malignancy. Histopathologicalexamination showed presence of immature testicular tissue.

Keywords: Polyorchidism, Triorchidism, Supernumerary Testis

1. INTRODUCTION
Polyorchidism is a very rare congenital anomaly of the urogenital system and is defined as the incidence of more than two testes[1]. Triorchidism is its most common presentation[2].The left side is mainly affected. Nearly 50% of the cases are identified between 15 and 25 years of age[3]. Commonly associated anomalies are testicular maldescent (40%), inguinal hernia (30%), testicular torsion (13%), hydrocele (9%), and hypospadias (1%)[4]. A higher tendency for malignant transformation in non-functional testis necessitates the radiological and functional assessment of polyorchid testes. We describe a case of triorchidism presenting with inguinal hernia in a ten-year-old male and briefly discuss its management.

2. CASE PRESENTATION
A ten-year-old boy was admitted with the complaint of left inguinal swelling for 3 years without associated history of trauma or pain. On physical examination, a reducible swelling in the left inguinal region with positive cough impulse and positive deep ring occlusion test was noted. The right testis was normal in size and palpated in the scrotum, whereas left testis was impalpable. So the diagnosis of left sided inguinal hernia with undescended testis was made. The family history was unremarkable. Other physical findings were normal. Ultrasound showed left testis in the inguinal canal whereas right testis in the scrotum. The patient was operated for left inguinal herniotomy. At the time of operation the left inguinal region was explored. Intraoperative findings of indirect inguinal hernia sac was present with small undescended supernumerary testis of size 2×2×1 cm at the deep inguinal ring having short vas deferens and a 2.5x2.5x1 left testis with epididymis and vas deferens in the hernia sac. (&2) Hernia sac with cord structures at the deep inguinal ring were identified. After performing high ligation and fixation of the normal testis into the scrotum, supernumerary testis was removed for a possible malignancyand anomalous anatomy. Histopathological evaluation revealed presence of immature testicular tissue. The postoperative period was uneventful.

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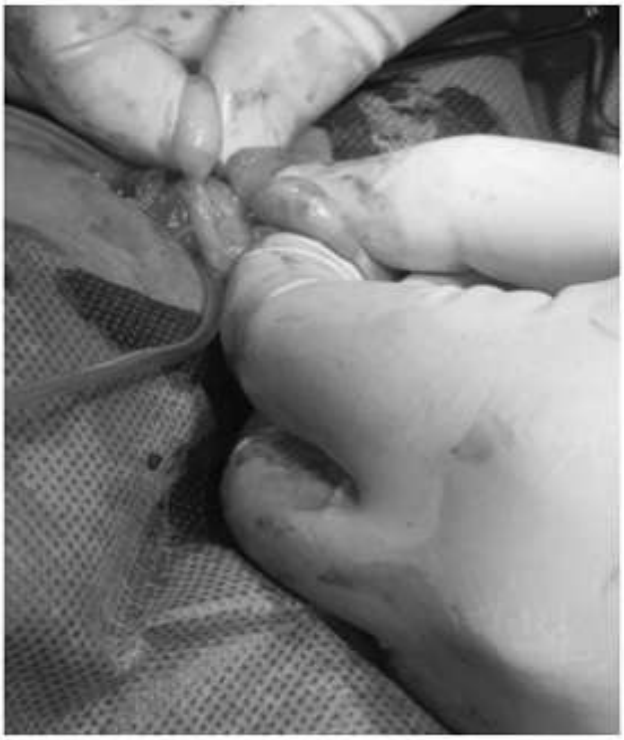

Figure-1 Both testes in the inguinal canal

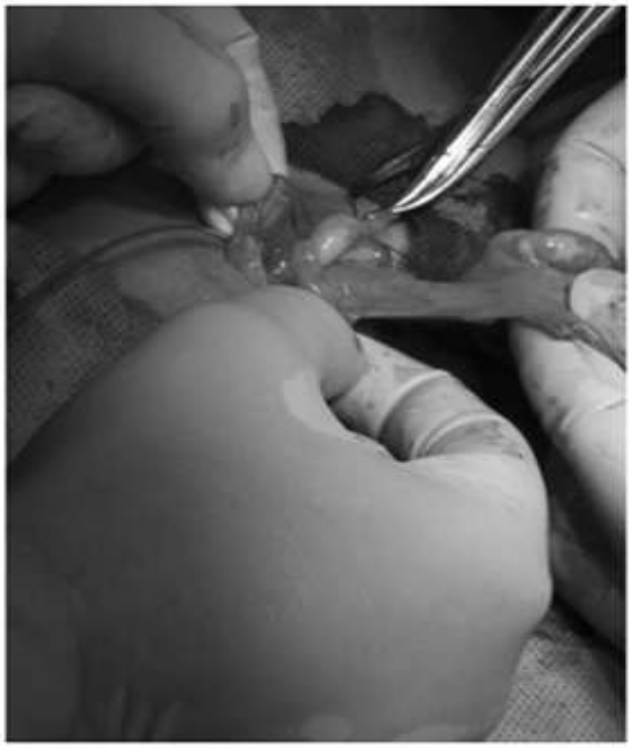

Figure-2 The small mass proximal to the left testicle proved to be a supernumerary testicle.

3. DISCUSSION

Polyorchidism is the incidence of more than two testes confirmed by histology. This condition is very rare, and nearly over a hundred cases were described in the literature. Although it can remain asymptomatic, it is often related to processus vaginalis anomalies and undescended testis in childhood. Supernumerary testes may be found in abdominal, inguinal or scrotal location; they are mostly on the left side, and they are usually smaller than both contralateral and ipsilateral testes. The exact cause of polyorchidism is not clear, though accidental division of genital ridge before 8 week of gestation could be a likely cause. There are theories suggesting the likely presence of multiple testes first being duplication of longitudinal genital ridge resulting into separate testes so total volume of testis surpasses of one. The second theory describes transverse division of genital ridge at different levels resulting into different combination of testes, vas deferens and epididymis.

Thum[8] proposed a functional classification of polyorchidism based on embryonic development (Table 1). According these classifications, our patient was considered as Type 1.

Table 1. Functional classification of polyorchidism based on embryonic development (table derived from Thum [8.]

Type I	The supernumerary testis lacks an epididymis and vas. The split-off part of the primordial gonad does not communicate with the mesonephric tubules from which the epididymis develops.
Type II	The supernumerary testis is linked to the regular testis by a common epididymis and shares a common vas with it. The division of the genital ridge occurs in the region where the primordial gonads are attached to the mesonephric ducts, although the latter are not divided.
Type III	The supernumerary testis has its own epididymis but shares the vas with the regular testis.

Polyorchidismusually presents itself in the 2nd to 3rd decade, with nearly 50% of cases reported between 15 to 25 years of age. The common presentations of polyorchidism manifests as maldescent (40%), hernia (30%), torsion (15%), hydrocele (9%), malignancy (6%), infertility and epididymitis.[1]

If a polyorchidism is suspected of palpable mass in the groin or scrotum, ultrasound is the effective, noninvasive investigation and preoperative assesment. On ultrasound, an accessory testis usually shows a fine granular echotexture similar to normal testis. Color Doppler ultrasound can give further information regarding blood flow pattern in the testis. MRI may provide confirmation when ultrasound is not conclusive[12].

The management of polyorchidism has been still controversial. In the past it was common practice to remove the supernumerary testicle with removal of the smaller mass [13]. More recently, with advances in ultrasound and magnetic resonance imaging technology, more conservative approaches have been recommended. Generally, authors have either supported surgical exploration or follow up with imaging investigations. Biopsy is a contentious issue and not routinely performed. A conservative approach needs magnetic resonance imaging, and high resolution sonography as an effective, noninvasive technique of correctly diagnosing polyorchidism. Several authors assert that conservative management is the suitable choice. They recommend that supernumerary testis, even in ectopic locations, must be saved if they look normal and are potentially functional. They believed that the absence of any concomitant disorder and if testicular tumour can be ruled out by sonography or magnetic resonance imaging, surgical exploration with biopsy could be unnecessary[4,8]. In contrast, surgical exploration has the benefit of allowing orchidopexy to prevent torsion and determining the testicular outflow tracts and estimating reproductive capacity[14]. Indications for removal include malignant or dysplastic change on biopsy, sonographic evidence of malignancy, and absent reproductive potential of the polyorchid testis which lacks an epididymis or vas. In our case, the supernumerary testis showed no reproductive capacity due to a lack of attachment to a cord structure. It was therefore removed because of anomalous anatomy and possible malignancy.

4. Conclusion

The diagnosis of polyorchidism is typically incidental. According to our opinion, if polyorchidism is associated with doubtful concomitant pathology, surgical exploration should be performed. But, in uncomplicated polyorchidism, conservative management with magnetic resonance imaging or ultrasonography observation should be recommended.

Consent

Written and informed consent was taken from the patient's parent for publication of this case report and images.

Conflict of Interests

No conflict of interests.

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