Supernumerary testis or polyorchidism is a rare congenital anomaly. It is often associated with processus vaginalis anomalies and with increased risk of malignancy and infertility. The approach to management has changed over time, with improvements in imaging techniques allowing surveillance to replace surgical excision or exploration and biopsy. In this study, two patients were managed with orchidopexy and have had a close follow-up of 2 years.

**Keywords:** Infertility, malignancy, orchidopexy, polyorchidism, supernumerary

**INTRODUCTION**

Polyorchidism is a congenital malformation of the male genital system characterized by the presence of more than two testes in the same individual. Although the existence of three testes is the most common presentation, four testes have been reported in six patients and five in one case. The age at diagnosis varies from newborn to 74 years, with a mean of 17 years. Testicular duplication is usually an incidental finding during surgery for inguinal hernia, cryptorchidism, or testicular torsion. We present two cases of polyorchidism and review literature and management protocols.

**Case Report**

Two patients, 5 years and 4 years old, respectively, presented with the complaint of absent testis in the left hemiscrotum since birth.

In both these patients, the testes were palpable in the superficial inguinal pouch. Both patients underwent inguinal exploration.

**Intraoperative findings**

Patient 1: 1 cm \( \times \) 1.5-cm testis was found in the inguinal canal along with another testicular structure sized 0.75 cm \( \times \) 0.75 cm with separate vas deferens and epididymis [Figure 1].

Patient 2: 1 cm \( \times \) 1.2-cm testis along with a second testicular structure measuring 1 cm \( \times \) 1 cm with separate vas deferens and epididymis found in the inguinal canal [Figure 2].

Subdartos pouch orchidopexy was done for both testes. In both patients, viability of supernumerary testis was confirmed with Doppler in the postoperative study. Ultrasound abdomen was done in the postoperative period showing no Mullerian structures. Follow-up scan showed both testes in the scrotum [Figure 3].

Both patients are doing well in a close follow-up of 2 years.

**Discussion**

Polyorchidism, or supernumerary testis, is a rare entity with <200 reported cases. Blasius described the first case of polyorchidism in 1670, Ahlfeld is credited for the first histologically proven case found at autopsy in 1880, and Lane reported the first histologically proven case of surgical intervention in 1895. Polyorchidism is thought to result from abnormal division of genital ridge embryologically. The supernumerary testes are in the intrascrotal location in approximately 75% of cases. Nearly 20% of supernumerary testes are inguinal, with the remaining retroperitoneal in location. About 75% of cases have been left sided, as were both our patients. The attachment of the supernumerary testis of its vessels and draining structures varies. Although there is no definite statistics in the literature, it appears that duplication of the testes with a single epididymis and vas deferens is more common.

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The exact explanation for the development of polyorchidism is not known, although several theories have been proposed including anomalous appropriation of cells, initial longitudinal duplication of the genital ridge, and transverse division of the genital ridge. According to Skandalakis et al., supernumerary testis occurs because of an anteroposterior division of the genital ridge, and it is suggested that lateral pairing of the upper end of the Wolffian duct with fusion of the two branches at the lower level is the cause of two spermatic cords. On the basis of embryologic development, Leung classified polyorchidism into four types [Table 1].

Type B is the most common, with Types B and C constituting approximately 90% of cases. Type D is very rare. Both our cases seem to fit Type D most appropriately.

Testicular duplications are generally asymptomatic, and are an incidental finding during surgical exploration. The most common associated anomalies reported are maldescent or cryptorchidism (40%), inguinal hernia (30%), torsion (15%), hydrocele (9%), and malignancy (6%).

Polyorchidism can be diagnosed by color Doppler ultrasonography and magnetic resonance imaging (MRI). The supernumerary testis has echotexture and vascular flow similar to a normal testis. MRI is very helpful if ultrasound diagnosis is not certain. The MRI characteristics of supernumerary and normal testes are similar (intermediate signal intensity on T1-weighted ages and high signal intensity on T2-weighted images). In our case, the diagnosis of polyorchidism was not entertained preoperatively based on physical examination, so radiological investigations were not done.

There appears no clear consensus on the appropriate management of polyorchid patients. Reported cases have considered and recommended three principal approaches: surgical excision, exploration, and biopsy or surveillance using physical examination, imaging, and serological markers to screen for malignancy. The two primary objectives remain preservation of reproductive potential and minimization of the risk of malignancy within the supernumerary testis. Secondary concerns include the likelihood of compliance with surveillance, parental preference, and cosmesis. Some authors argue for excision of all accessory testes on the basis that spermatogenesis would be expected to be impaired and

| Table 1: Leung’s classification of polyorchidism |
|-----------------------------------------------|
| Type | Description                                |
|------|--------------------------------------------|
| Type A | Supernumerary testis lacks an epididymis and vas deferens |
| Type B | Supernumerary testis has its own epididymis |
| Type C | Supernumerary testis has its own epididymis and shares the vas deferens with regular testis in a parallel fashion |
| Type D | Supernumerary testis has its own epididymis and vas deferens |
the testes are therefore unlikely to contribute positively to fertility. In contrast, several reports suggest that as many as half to two-thirds of supernumerary testes have a normal histology.[5] The risk of malignancy, its magnitude, and its implications for management seem controversial. In addition, some authors describe an increased risk of testicular torsion in polyorchid patients, affecting either the normal or the accessory testis. For this reason, orchidopexy of all testes, including the supernumerary testis, has been recommended, accepting the inherent risk of trauma to a normally located testis.

Our patients seemed to have had triorchidism, with a Leung Type D supernumerary testis on the left side. In our cases, both the testes were brought down into the scrotum. Two years of follow-up is uneventful.

We conclude that supernumerary testis is an extremely rare congenital anomaly and is mostly found incidentally during correction of processus vaginalis anomalies. Encountering polyorchidism warrants the decision of whether to perform additional surgery or biopsy on asymptomatic patients or following up with imaging. The reproductive potential of the supernumerary testis, as defined by its draining structures, plays a pivotal role. Meticulous and careful inguinal exploration is needed during surgery and in all patients with normal-appearing gonads, orchidopexy should be performed to minimize the chances of torsion, malignancy, and infertility. These patients need to be followed up, clinically and radiologically, over a period of time.

Declaration of patient consent
The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest
There are no conflicts of interest.

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