

## Bilateral Polyorchidism Associated with Intraabdominal Cryptorchidism: A Case Report

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### Key Words

Testis · Polyorchidism management · Cryptorchidism

### Abstract

**Objective:** The aim of this study was to present a rare congenital anomaly of polyorchidism and to review the current management. **Clinical Presentation and Intervention:** We report the case of a 19-year-old man who was referred for the treatment of an indirect left inguinal hernia. The ultrasound demonstrated a testis on the right side with an additional mass. The preoperative laboratory studies were normal. At surgery, a solid mass consistent with an ectopic testis was removed. The histological evaluation showed the presence of a fourth testis without malignancy, confirming the diagnosis of bilateral, double testes associated with intraabdominal cryptorchidism, which was removed for histology. The postoperative endocrinological evaluation revealed hypergonadotrophic hypogonadism. The patient was followed up conservatively. **Conclusion:** Polyorchidism should be included in the differential diagnosis of a solid extratesticular mass, and its management should be conservative.

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### Introduction

Polyorchidism is an uncommon congenital anomaly with less than 100 cases reported in the literature, in which more than 2 testes are observed. Although the ex-

act mechanism for the manifestation of polyorchidism remains unknown, it has been suggested that the condition may be secondary to the division of the embryonic genital ridge [1]. Approximately 50% of the cases have been histologically confirmed. Bilateral polyorchidism has been reported only in 6 cases [2, 3]. Triorchidism is the commonest variety of polyorchidism [4]. Polyorchidism may be associated with cryptorchidism, mal descended testis, indirect inguinal hernia, testicular torsion, epididymitis, hydrocele or varicocele, malignancy and infertility [5]. The most common manifestations are painless groin and testicular masses, although a majority of patients is asymptomatic.

We report the case of a bilateral, double testis with an intraabdominal supernumerary testis and an indirect left-sided inguinal hernia.

### Case Report

A 19-year-old man was referred for the treatment of an indirect left inguinal hernia. The patient had no prior history of genitourinary complaints and was asymptomatic. On physical examination, his right hemiscrotum appeared to contain 2 separate masses. Both masses were of equal texture, similar to his normal testis. The ultrasound demonstrated a testis on the right side with an additional mass above, with equal echogenicity and size, consistent with a second testis on this side. The preoperative laboratory studies, including  $\alpha$ -fetoprotein and  $\beta$ -human chorionic gonadotropin levels, were normal.

The patient underwent repair of his left inguinal hernia with a synthetic mesh. At surgery, during the management of the in-

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**Fig. 1.** The left supernumerary testis.

ternal inguinal ring, we palpated a small, independent and solid mass consistent with an ectopic testis. The mass was removed and sent for a histological examination (fig. 1). His postoperative course was uneventful, and he was discharged on the first postoperative day.

Histological evaluation revealed the presence of a fourth testis and an epididymis without malignancy. Postoperative magnetic resonance imaging (MRI) of the scrotum confirmed the presence of a supernumerary testis on the right side. The hormone profile included follicle-stimulating hormone (9.1 IU/l; normal range 4–10 IU/l), luteinizing hormone (15.1 IU/l; normal range: 1–8 IU/l), prolactin (20.5 ng/ml; normal range: 2–15 ng/ml) and testosterone (0.82 ng/dl; normal range: 270–1,070 ng/dl). Two semen analyses showed a complete lack of spermatogenesis. As clinical and imaging evidence of the right supernumerary testis revealed no pathology, the patient was followed up conservatively.

## Discussion

The man presented had supernumerary testes which were asymptomatic, with painless groin or scrotal masses and azoospermia. In about 4–7% of supernumerary testes, a malignant degeneration or transformation occurs [6, 7], and in approximately 50–65% of supernumerary testes, there is a chance of active spermatogenesis [1]. Several embryological mechanisms have been proposed, including the degeneration of parts of the mesonephric components, the duplication or division of the genital ridge [2] or the accidental longitudinal or transverse division of the genital ridge, with or without the mesonephros, before the 8th week of gestation, either by a local ac-

cident or by the development of peritoneal bands [1]. There are several classification systems for polyorchidism based on anatomical arrangements of the testes and their outflow paths [8–10]. Four forms of polyorchidism have been described and classified by Leung [9], 2 of which, type II and III together, account for more than 90% of the cases of polyorchidism. The physical examination alone cannot accurately diagnose polyorchidism. Although biopsy is traditionally recommended for a definitive diagnosis, ultrasound can strongly suggest the diagnosis. On sonography, supernumerary testes demonstrate a second, freely mobile, well-defined ovoid structure with an echo texture and a vascular flow pattern similar to that of normal testes [11]. MRI can be helpful in clarifying the diagnosis, especially if sonographic findings are inconclusive [3].

## Management

The management of polyorchidism has been the subject of much debate in the literature. In the past it was common practice to remove the supernumerary testis. More recently, with advances in ultrasound and MRI technology, more conservative approaches have been advocated, as carried out in our case, in a normal, uncomplicated and orthotopic supernumerary testis [12]. Conversely, surgical exploration is mandatory, whenever the nature of the testicular mass remains unclear, with the added advantage of allowing the fixation of the testes to prevent torsion and the creation of a single testicular mass in certain cases, and biopsy to reveal the histological pattern [6]. Indications for a removal include malignant or dysplastic change on biopsy, and in those circumstances when the regular follow-up is unlikely to be reliable. The psychological and cosmetic considerations need to be taken into account as well. Therefore, the patient must participate in the decision-making.

## Conclusion

Polyorchidism should be included in the differential diagnosis of a solid extratesticular mass. With advances in imaging techniques, conservative management is advocated for benign cases, with a careful, yearly follow-up.

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