



Case Report

Polyorchidism in a Child with Bilateral Undescended Testis: How we have Done

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Abstract

Polyorchidism is defined as the presence of two or more testes. Currently less than 200 cases have been reported in the medical literature. The management problem in the child is due to intraoperative diagnosis, which requires an impromptu decision. We present the case of a three-year-old child, operated on for undescended testicles, with an occasional finding of triorchidism, with a double testicle on the left, the most typical side of the finding. In agreement with most of the authors, it was decided not to remove the supernumerary testis and to perform bilateral Shoemaker's orchidopexy.

Keywords: Orchidopexy; Polyorchidism; Undescended testis

Introduction

Polyorchidism represents a rare congenital abnormality of the genitourinary tract, characterised by the presence of an additional testicle. Generally found during evaluation for other conditions such as inguinal hernia, undescended testis and testicular torsion. Cryptorchidism appears to be the most important risk factor for malignancy in patients with supernumerary testes. Although it can remain asymptomatic, polyorchidism is often associated to processus vaginalis anomalies in childhood (hernia, hydrocele) and undescended testis. The review of the literature finds cases of polyorchidism revealed by testis torsion and an increased risk of malignancy and infertility. Currently less than 200 cases have been reported in the medical literature.

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2-year-old child, undergoing elective surgery for testicles retained in the middle third of the inguinal canal, since birth. The clinical and ultrasound examination did not reveal any suspicion about further morphological abnormalities of the didymes, which appeared to be of normal size. During surgery, after accessing the left inguinal canal, the presence of a supernumerary small testis with a shared common epididymis and vas deferens with the primary testicle. It was decided to solidify the two formations with

a thin resorbable suture, to avoid twisting of one relative to the other, and to practice orchidopexy in an extra-artic pocket with three anchor points on the larger didymus. Given the small size of the accessory didyme (about 5 mm), it was decided to postpone biopsies. The postoperative course was regular; the ultrasound examination three months after the operation made it possible to highlight both organs in the scrotum, with normal vascularization to the power doppler.

Discussion

The median age of presentation is between 15 and 25 years of age in 50% of the cases. The majority of patients are asymptomatic or present with a painless inguinal or scrotal mass, undescended testis and rarely torsion of the supernumerary testis [1]. The most common manifestation of polyorchidism is triorchidism where 3 testes are present. The supernumerary testis is most often located on the left side of the scrotum (Figure 1). Very few cases of triorchidism have been reported in children less than 2 years. The most common presentation of polyorchidism is triorchidism, and the left side is predominantly affected as in our case [2]. The majority of supernumerary testes are scrotal in origin (66%) followed by inguinal (23%) and abdominal (9%). Most cases were found during surgery for other symptoms including inguinal hernia, undescended testicle, testicular torsion and scrotal pain. Only 16% of patients complained of an accessory mass

without any symptoms. Embryologically, polyorchidism, or testicular duplication is thought to result from division of the urogenital ridge between the fourth and sixth week of embryological development. The exact explanation for the production 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, either through some local accident of development of peritoneal bands [1,3-6]. Various classifications of polyorchidism are found in the literature. Considering the embryologic development, Leung classified polyorchidism into 4 types [2], on the basis of relationship between testes, the epididymis and vas deferens (Table 1).

Figure 1: The supernumerary left testis with a shared common epididymis and vas deferens with the primary testicle.

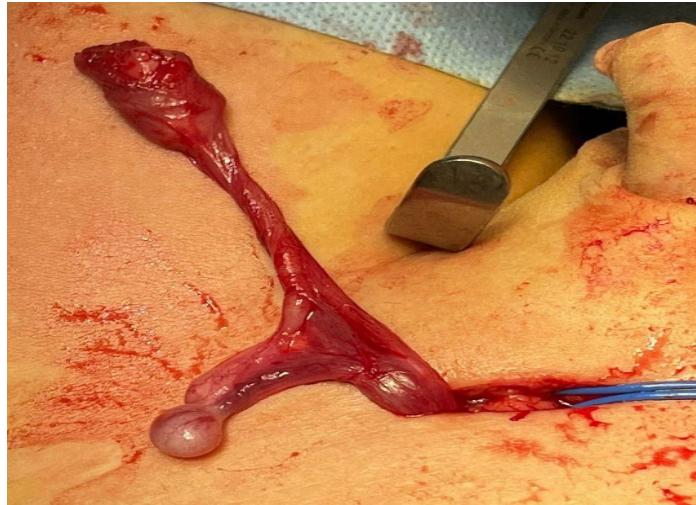


Table 1: Leung classification of polyorchidism

Type 1	Supernumerary testis without an epididymis or vas deferens
Type 2	Supernumerary testis with a shared common epididymis and vas deferens with the primary testicle
Type 3	Supernumerary testis has its own epididymis but shares a common vas deferens with the ipsilateral testicle
Type 4	There is complete duplication of testis, epididymis and vas

Instead, some years later, Bergholz classified this anomaly into 2 Types, based on the connection to the vas deferens and, therefore, the presence or absence of reproductive function [7]. Each of the 2 types is further divided into 3 subtype, on the basis of specific characteristics (Table 2).

Table 2. Bergholz classification of polyorchidism.

Type A	The supernumerary testicle is connected to a vas deferens. These testicles are usually reproductively functional.
<ul style="list-style-type: none"> • Type A1 • Type A2 • Type A3 	<p>Complete duplication of the testicle, epididymis and vas deferens.</p> <p>The supernumerary testicle has its own epididymis and shares a vas deferens</p> <p>The supernumerary testicle shares the epididymis and the vas deferens of the other testicles.</p>
Type B	The supernumerary testicle is not connected to a vas deferens and is therefore not reproductively functional
<ul style="list-style-type: none"> • Type B1 • Type B2 	<p>The supernumerary testicle has its own epididymis but is not connected to a vas deferens</p> <p>The supernumerary testicle consists only of testicular tissue</p>

Decision for surgery, biopsy or orchiectomy should be based on age of diagnosis, type, concomitant symptoms, cryptorchidism or suspected malignancy [8,9]. Since this is a rare occurrence and often a surprise during surgery, as in our case, decision-making problems about how to behave often arise. In these cases, there are at least three options to consider: a) removing the testis d'emblée; b) Perform biopsies, with the risk of damaging a small organ; c) Safeguard the supernumerary organ and follow up on the patient. Conservative treatment is advised in all uncomplicated cases. Complicated cases need a careful management, but several situations can be managed conservatively. In our opinion, in case of incidental diagnosis during operations for undescend testis, as in our case report, the most rational decision seems to be not to remove the supernumerary testicle, especially if, according to its anatomy, it is fertile.

Conclusion

In the management of this cases most of the authors agree in safeguarding the supernumerary testicle, guaranteeing its permanence in the scrotum and avoiding torsion, and monitoring the patient over time with serial clinical and ultrasound examinations. Our case report showed a type 2 polyorchidism, according to Leung, and a type A3 polyorchidism, according to Bergholz, so it was a functional reproductive testicle. In our opinion, this case, in particular due to the age of diagnosis, and because it was detected during surgery for non-descended testicles, was managed according to the recommendations in the literature, which provide for the safeguarding of the supernumerary testicle and orchidopexy.

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