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Undescended Testis Revealing Triorchidism in a Child: A Case Report

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Abstract

Polyorchidism is a rare genitourinary congenital malformation, which diagnosis is generally fortuitous in children. We report the case of a 7-year-old boy admitted to the pediatric surgery service of Donka University Hospital in Conakry for an empty left scrotum. Initially, the diagnosis of left cryptorchidism had prompted orchidopexy by inguinal approach, whose intraoperative exploration revealed two undescended left testicles. The first,

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located at the deep ring of the inguinal canal, had normal volume, while the second, located in the inguinal canal, was hypotrophic. The orchiectomy of the latter was performed, associated with a dartos-pouch orchidopexy of the testicle of normal volume. After one year of follow-up, no complications were noted.

Keywords: Polyorchidism, Triorchidism, Undescended Testis, Child

Introduction

Polyorchidism, or the presence of three or more testicles, is a rare urogenital congenital malformation, first described in postmortem by Ashfeld in 1880 **(Sarr et al., 2015; Lustig et al., 2017)**. Since then, more than 200 cases have been reported in the literature **(Lustig et al., 2017; Mahmoudi et al., 2010)**. In children, polyorchidism is incidentally discovered during the surgical management of patent vaginalis, hydrocele, cyst of the spermatic cord, or most frequently, during orchidopexy indicated for undescended testis (UDT) **(Mahmoudi et al., 2010)**. We report a case of triorchidism in a 7-year-old boy, incidentally discovered during an orchidopexy indicated for left cryptorchidism. We discuss the epidemiological, diagnostic, and therapeutic aspects.

Observation

A 7-year-old boy was admitted to our pediatric surgery service at Donka University Hospital in Conakry (Guinea) for left scrotal vacuity noted by the parents since birth. The parents did not previously initiate any seeking for a medical solution. The mother's concern about the patient's future fertility motivated the present consultation. The mother did not report the use of contraceptive medication prior to the patient’s conception nor other hormonal drugs during the pregnancy. The patient was born at full term (39 weeks), and weighed 3200 grams. On admission, general physical examination was unremarkable. A left scrotal hypotrophy was noted, with an absence of the left testis within the scrotum. However, a gonad was palpated in the ipsilateral inguinal region without possibly being lowered to the scrotum. The right testicle was intrascrotal, with normal size. The remainder of the examination was unremarkable. Ultrasound (US) confirmed a left inguinal undescended testis, which had a normal size for age (15x8x9mm with a volume of 0.49 ml). Its margins were smooth and its echostructure normal, without focal or diffuse lesions. The diagnosis of left cryptorchidism was made, and after informed consent was obtained from the mother, we indicated a dartos-pouch orchidopexy.

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Method of operation

After a 3 cm left inguinal approach, surgical exploration revealed two testes: the first (of normal volume) at the level of the deep inguinal ring and the second (hypotrophic) in the left inguinal canal. The two testes were completely separated, each with its epididymis and vas **(Figure 1)**. The left vaginalis was patent concordant with Group D of Leung's Anatomical Classification of Polyorchidism **(Leung, 1988)**.

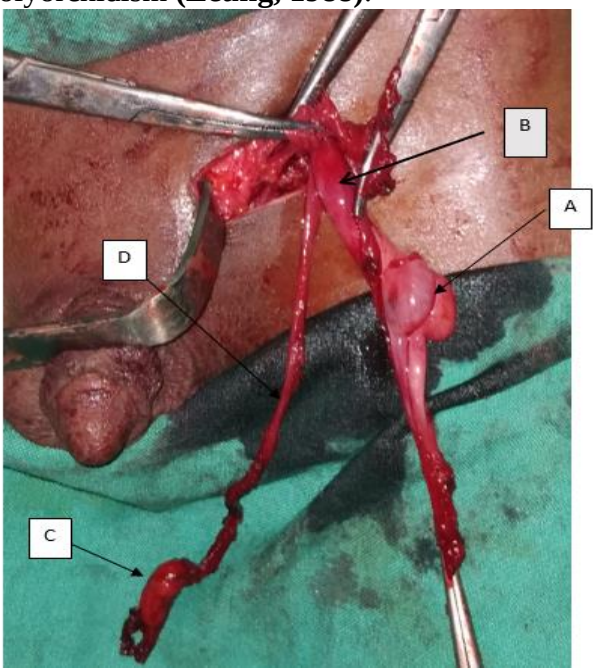

Figure 1. Intraoperative findings

Note duplication of the testis, epididymis, and vas deferens. The normal testis (A) had normal volume for age and eutrophic spermatic cord (B), while the supernumerary testis is hypotrophic (C), as its spermatic cord (D). We performed ligation of the patent vaginalis, with dartos-pouch orchidopexy of the healthy left cryptorchidic testis, along with orchiectomy of the hypotrophic supernumerary left cryptorchidic testis.

The patient received oral paracetamol (60 mg/kg QID) for seven days. The discharge occurred on a postoperative day 1. The surgical specimen (supernumerary left testis) was sent to pathology, which concluded sclerotic fibrosis of the hypotrophic testis and its epididymis and vas. After a 12-month follow-up, the left testis was palpable in the dartos-pouch, and the ultrasound showed normal structure and normal size for age.

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Discussion

Polyorchidism is a rare urogenital malformation; more than 200 cases have been reported **(Sarr et al., 2015; Lustig et al., 2017; Bahloul et al. 2011; Teklali et al; 2007)**. Some authors stated that its incidence is underestimated because polyorchidism can remain asymptomatic. Poor knowledge of this malformation and the rarity of pathology examination of inguinoscrotal masses could explain this.

Triorchidism is the most common presentation of polyorchidism, and the supernumerary testis is generally hypotrophic **(Lustig et al., 2017)**. The patients present at any age, with a wide variability **(Sarr et al., 2015; Mahmoudi et al., 2010; Bahloul et al., 2011)**. The supernumerary testes are usually located in the scrotum. However, they can be cryptorchidic in the inguinal region or the abdomen. They are often found on the left side, as in our case. The most common associated abnormalities are patent vaginalis, spermatic cord torsion, testicular malignancy, and undescended testis (UDT), as in our patient **(Sarr et al., 2015; Lustig et al., 2017; Naouar et al., 2007)**. Polyorchidism is commonly asymptomatic; thus, fortuitously discovered by palpating two testicles in a hemiscrotum during the physical examination for another condition. Sometimes inguinal pain or acute or non-acute scrotal swelling can happen **(Naouar et al., 2007)**. The diagnosis can be confirmed by ultrasound for non-cryptorchidic supernumerary testis or by magnetic resonance imaging **(Leung, 1988; Hatri et al., 2014)**. In our patient, the US identified a single cryptorchidic testis, overlooking the supernumerary hypotrophic testis, which was also undescended. This can be explained by the fact that the US is not specific in diagnosing UDT. In most cases, the discovery is incidental during surgical procedures of UDT or pathologies of the vaginalis **(Bahloul et al., 2011)**. This was the case in our patient, with incidental discovery during an open left orchidopexy.

The therapeutic approach to polyorchidism remains controversial **(Sarr et al., 2015; Lustig et al., 2017)**. The conservative management of the supernumerary testis is very controversial and considers the risk of subfertility and malignant degeneration **(Sarr et al., 2015; Mahmoudi et al., 2010; Teklali et al., 2007)**. Based on advances in imaging, some authors recommend conservative treatment of the supernumerary testicle, particularly for groups B and C of Leung's classification. This allows avoidance of epididymal and deferential lesions and thus improves the potential for fertility in young subjects. Additionally, a supernumerary testicular orchidopexy is made to avoid the torsion of its spermatic cord **(Sarr et al., 2015)**. Regular clinical and ultrasound follow-up is required **(Teklali et al, 2007; Naouar et al., 2007; Hatri et al., 2014)**. On the other hand, other authors favor orchiectomy of the supernumerary testis in case of an incidental discovery due to the risk of

malignancy, which happens in 1% to 7% **(Bahloul et al., 2011; Naouar et al., 2007; García et al., 2021)**. In our patient, an orchiectomy of the supernumerary testicle was performed, associated with dartos-pouch orchidopexy of the normal left testis. Our therapeutic approach is justified firstly by the fact that the supernumerary testicle was hypotrophic, then less helpful in reducing the risk of subfertility. Secondly, we aimed to reduce the double risk of malignant degeneration in our patient as both UDT and supernumerary testis are their risk factors.

Conclusion

Polyorchidism is a rare genitourinary congenital malformation, mainly discovered incidentally. Each pediatric surgeon should master its management principles so that when incidental discovery is made during orchidopexy or other surgeries of the inguinal region, the best decision would be taken in the patient's interest.

Consent for publication: We obtained written consent from the mother of our patient.

Conflict of interest: Authors declare that they have no competing interests.

Authors' Contribution: All mentioned authors contributed to this work and approved the latest version of the manuscript as submitted.

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