Case Report

Polyorchidism; unilateral, one atrophic undescended double testicles

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Abstract

Polyorchidism is a very rare genitourinary anomaly defined with the presence of more than two testicles. Polyorchidism is associated up to 40% with undescended testicles. The present report is about an incidentally detected triorchidism case with unilateral, one atrophic undescended double testicles. A 4-year-old child, diagnosed with undescended left testicle revealed during orchiopexy polyorchidism with distinct epididymis and vas deferens. Whereas one of the testicles was in regular size; the other two were atrophic. Orchiectomy was conducted on the atrophic testicle and orchiopexy to the regular size testicle. The atrophic testicle excised was referred histopathological analysis and was diagnosed as atrophic testis. The patient discharged on the first postoperative day, was considered as normal during postoperative evaluation made on the third postoperative day. Polyorchidism is a rare genitourinary abnormality, and its management is still controversial. Yet, we believe that orchiectomy is to be conducted in atrophic testicle cases.

Key Words: Polyorchidism, supernumerary testicle, triorchidism, undescended double testicles

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INTRODUCTION

Polyorchidism, presence of more than two intra- or extra-scrotal testicles is a rare congenital anomaly. There are nearly 200 cases reported in the literature.[1]

Triorchidism is the most common type of polyorchidism. Triorchidism is the presence of three testicles and the extra testicle is called supernumerical testicle. Although its etiology is not completely known, it is asserted that it may be connected to the abnormal partition of the genital ridge during testicle embryogenesis.[2] The presence of triple testicles, triorchidism, is the most common polyorchidism type mostly located in the scrotum. Most of the patients apply with unpainful inguinal or scrotal masses, but they are generally asymptomatic.[3] Rarely undescended testicle may occur with clinical findings with an inguinal hernia and supernumeric testicle torsion.[4,5] Although ultrasonography (USG) and magnetic resonance

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imaging (MRI) facilitate supernumerary testicle diagnosis, the majority are asymptomatic and incidentally determined during surgical exploration. Polyorchidism is associated up to 40% with undescended testicles.[6] This report is about an incidentally detected triorchidism case with unilateral, one atrophic undescended double testicles.

CASE REPORT

A 4-year-old child, diagnosed with undescended left testicle revealed during orchiopexy polyorchidism with distinct epididymis and vas deferens. Whereas, one of the testicles was in regular size; the other two were atrophic [Figure 1]. Orchiectomy was conducted on the atrophic testicle and orchiopexy to the regular size testicle. The atrophic testicle excised was referred histopathological analysis and was diagnosed as atrophic testis [Figures 2 and 3]. The patient discharged on the first postoperative day, was considered as normal during postoperative evaluation made on the third postoperative day.

DISCUSSION

Polyorchidism is a rare congenital anomaly. At about 6 weeks of embryonic life, from the primitive genital ridge medial to the mesonephric ducts the primordial testis develops during a normal process. At 8 weeks, primordial testis takes shape, and the epididymis and vas deferens form from the mesonephric (wolffian) duct. Currently, exact mechanisms underlying polyorchidism are not known; however, longitudinal or transverse division of the genital ridge due to peritoneal bands development is suggested. Polyorchidism is most commonly observed as triorchidism, but cases such as four testicles (four cases) and five testicles (one case) were also defined in the literature.[1,6,7]

No consensus on polyorchidism classification is available. Leung, on one hand, described the first anatomical classification based on possible embryological differentiations: Type I: Supernumerary testis without epididymis or vas and attachment to the usual testis; Type II: The supernumerary testis draining into epididymis of usual testis sharing a common vas; Type III: The supernumerary testis with epididymis and both epididymis of the ipsilateral testes draining into one vas; Type IV: Complete duplication of testes, epididymis and vas.[8] On the other hand, Singer *et al.*, based on the reproductive potential of the supernumerary testicles, proposed an anatomical as well as functional classification: Type 1: Supernumerary testis with reproductive potential (Leung Types II, III, and IV); Type 2: Supernumerary testis without reproductive potential.[9] According to Leung, Type II is the most common type and with Type III they make up 90% of all polyorchidism cases.

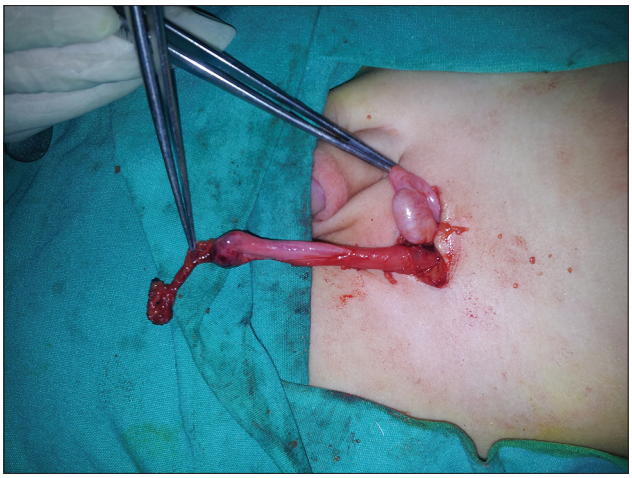

Figure 1: Intraoperative image

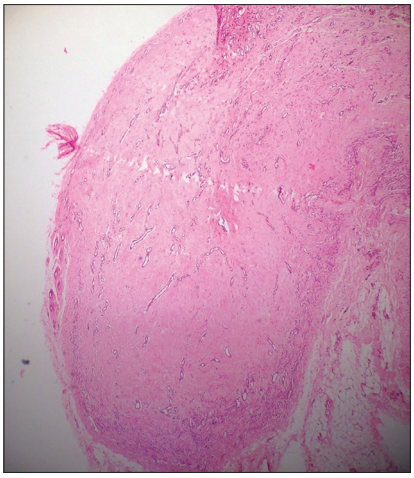

Figure 2: Completely fibrotic testis tissue with capsule (H and E, ×40)

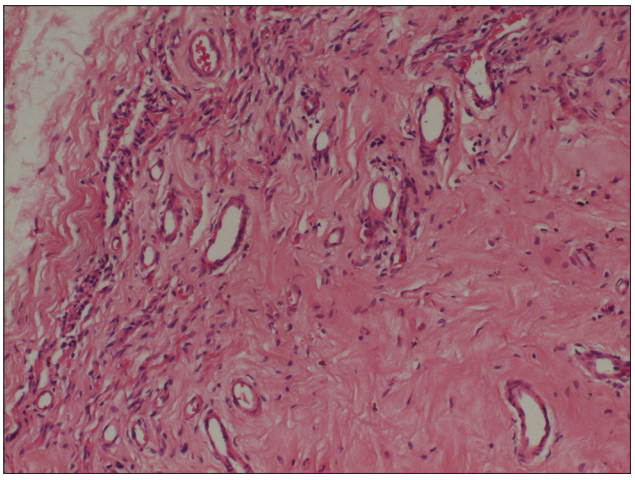

Figure 3: Completely fibrotic testis tissue (H and E, ×100)

The majority of polyorchidism cases are determined coincidentally while researching for the causes of other symptoms. Mal-descent (40%), hernia (30%), torsion (15%), hydrocele (9%), and malignancy (6%) are the most common

polyorchidism associated anomalies.[5,7,10] In most polyorchidism cases, a single supernumerary testicle (triorchidism) is present more frequently on the left side.[1] Bilateral supernumerary testicles have also been reported.[11] The scrotum (66%), inguinal (23%), and abdominal (9%) positions are the locations of supernumerary testicles.[5,7]

Although USG and MRI may bare effective in the diagnosis of polyorchidism, the majority of cases are asymptomatic and an incidentally determined during surgical exploration.[12]

In this case, during the surgical exploration for undescended left testicle, double separate epididymides and vas deferens were determined. This was Type IV in line with the classification made by Leung. However, quite extraordinarily, one of the testicles were normal the other atrophic.

CONCLUSION

Polyorchidism is a rare genitourinary abnormality, and its management is still controversial. Although USG is satisfactory in diagnosis in most cases, MRI may provide additional information in complicated cases. If polyorchidism management testicular malignity can be discarded, if supernumeric testicle is inside the scrotum and if there are no significant anomalies accompanying, it should be followed with USG conservatively at least two times a year, and surgical (orchiectomy) treatment should be done in the case of cryptorchidism, accompanying the disease, testicular torsion, and testicular malignity. Yet, we believe that orchiectomy is to be conducted in atrophic testicle cases.

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Conflicts of interest

There are no conflicts of interest.

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