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Case Report

Triorchidism in a Pediatric Patient: A Case Report and a Literature Review

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Abstract

Testicular duplication, supernumerary testis or most commonly known as polyorchidism constitutes an especially rare congenital anomaly of the genitourinary tract. We report a 10-year-old male patient presented to our clinic with a visible as well as palpable left scrotal mass. With the view to determining whether the surgical excision of the additional testis or its monitoring by imaging is more preferable, we took many factors into consideration, including patient's comorbidities, testicular biopsy's outcome as well as the classification of the extra testes. The diagnosis of the additional testis was confirmed by histopathology and underline the need to maintain a high index of suspicion.

Keywords: Testicular Duplication; Triorchidism; Polyorchidism; Pediatric; Accessory Testis; Genitourinary Tract

Introduction

Testicular duplication, supernumerary testis or most commonly known as polyorchidism constitutes an especially rare congenital anomaly of the genitourinary tract, with a few cases in the international literature [1]. In the vast majority of cases, it is presented as triorchidism [2]; in other words, the presence of an additional testis either inside (66%) or outside the scrotum. Regarding the extra scrotal sites of detection, these may be inguinal (23%) and abdominal (9%) [3]. The side which is affected the most is the left hemiscrotum (66%) [4,5], which is also the case with our patient. Since triorchidism can be rather unlikely to occur, amplitude of other syndromes come to mind and rank the highest places in the differential diagnosis, leading to a misdiagnosis of the former. Amongst them, we may find varicocele, hydrocele, inguinal hernia, cryptorchidism, epididymitis, testicular neoplasm [3,6] to name only a few. In the current case report, we present the imaging findings in triorchidism along with the contribution of histopathological analysis to the diagnosis, without omitting to mention the treatment of choice.

Case Presentation

A 10-year-old male patient presented to our clinic with a visible as well as palpable left scrotal mass. The lump had appeared approximately a year prior to the clinical examination, without any complaint of discomfort or even pain on behalf of the patient.

Furthermore, the size of the mass maintained a stable course since its detection. The boy had not a relevant past medical history and had not undergone any previous abdominal or inguinal surgery. During the clinical exam, we did not encounter any sign of edema or haemorrhage on the left side and any pathological findings on his right hemiscrotum. Moreover, there was no evidence of hernia or lymphadenopathy in inguinal examination. The patient did not report lack of appetite or presence of fever, nor did his parents mention any alteration of his bowel or urinary habits. In the first place, his laboratory findings did not correspond with an active inflammation since all relevant markers were within the normal range, eliminating epididymitis as a possible underlying cause. The values of Hct and Hgb which fluctuated just below their lowest normal values merely set the suspicion for a mild type of anemia , whereas the perfectly normal value of platelets did not predict any haemorrhagic predisposition. In addition, all cancer biomarkers were normal thus, the possibility of a malignancy was significantly low in the differential diagnosis. Thereafter, an ultrasound scan was performed on the patient, which showed the presence of what was presumed to be a left epididymal cyst. Nevertheless, histopathological analysis is nowadays the most sufficient and definitive diagnostic tool so we proceeded with a biopsy with the prospect of identifying the consistency of this engorgement (Figures 1,2).

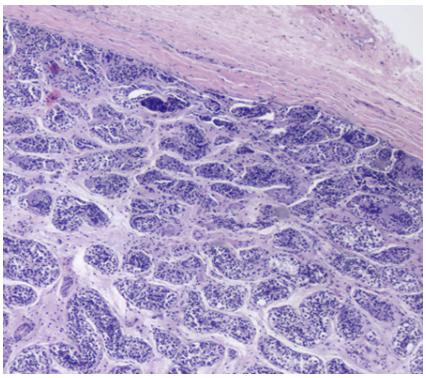

Figure 1: The tubular diameter of the seminiferous tubules is smaller than normal. There is slight increase of intratubular fibrosis.

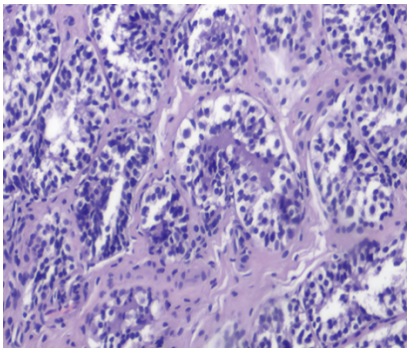

Figure 2: There is slight increase of intratubular fibrosis. Increased number of Sertoli cells in the seminiferous tubules.

The results of the biopsy indicated macroscopically a grey small tumour with a diameter of 0.4 cm and microscopically, with the consistency of an atrophic testis. As it was anticipated, the tumour was smaller than the normal unilateral one, both of which were composed of identical testicular tissue. Hence, its coevaluation along with genetic, imaging and clinical assessment of the patient's genitourinary system was suggested. For every one of these procedures, an informed consent by the parents was mandatory. As far as the management strategy is concerned, we opted for the surgical removal of the accessory testis, as depicted in Figures 3 and 4, since the latter had its own epididymis but shared a common vas deferens with its neigh-boring testis. There were no postoperative complications and the incision healed quickly and with a satisfying cosmetic appearance.

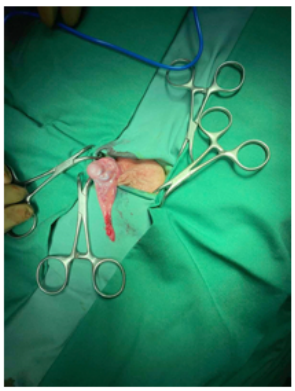

Figure 3

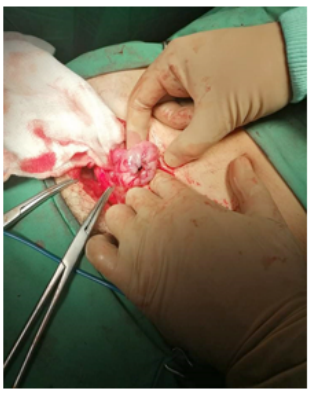

Figure 4

Figures 3&4: Intraoperative pictures. Left triorchidism. Excision of the additional testis in the left hemiscrotum with independent epididymis and common vas deferens.

To this day, the patient has been discharged from our clinic and has not encountered any derivative inconvenience 1 year follow up.

Discussion

Testicular duplicity or polyorchidism is one of the most challenging conditions to acknowledge despite the leaps of progress on behalf of the scientific community. The pathogenesis remains to be unraveled but certain theories have been more promoted than others; there is the explanation of an impediment during the embryological development of the primitive testis from the medial part of the mesonephric genital ridge which leads to a transversal or less often a longitudinal division of the ridge [6,7,8]. Alternatively, a degradation of various mesonephric accessories is feasible whilst not so popular with the majority [8,9]. Triorchidism is the most typical image of the condition in question. Patients experiencing triorchidism may face from zero symptoms [3,10] with a possible palpable mass inside or outside of the scrotum to dysuria and pain near the affected area [3], although the latter constitutes an exception. About 50% of the cases refer to male patients between 15 and 25 years of age [7,11,12]. The diagnosis of triorchidism should not be depending solely on physical examination as there are a number of other pathologies that may mimic it including multiple types of hernia, testicular neoplasms, inflammatory diseases, teratoma [13] etc. Therefore, the algorithm of treating triorchidism can be the following:

i. Record of the patient's medical and family history and attention to the reported symptoms and signs

ii. Careful and meticulous physical examination

iii. Additional questions to the guardians for clarification of the situation, when the patient is not so descriptive due to his age, mental status, character etc.

iv. Ultrasound scan for detection of its location and/or number (3 or more testes)

v. NMRI if available

vi. Biopsy of the atrophic testis and histopathological exam

vii. Standardized management that can either be surveillance, orchidopexy or even surgical excision.

Concerning the last step of the algorithm (vi), it goes without saying that neither of these practices is a panacea or outclasses the others, for it is still partially considered to be terra incognita. Triorchidism can be associated with a number of complications such as testicular torsion (13%), indirect inguinal hernia (30%), cryptorchidism (50%) and last but not least, testicular malignancy (up to 7%) [5]. Therefore, it is our duty to weigh cautiously the situation and decide upon a treatment that is best suited for the patient. For this purpose, we used the Leung [14], Hancock [9] and Wolf's [15] classification according to which: -Polyorchidism can be divided into 4 types with corresponding subtypes based on the anatomy of the testes and their adjacent structures, as listed below [7] (Figure 5):

Type A - testis being drained by a deferent duct:

- A1: The drained additional testis has its own epididymis and vas deferens.
- A2: The drained additional testis share a common deferent duct with its neighbour but separate epididymis
- A3: The drained additional testis can share both epididymis and deferent duct with its neighbour

Type B- testis not being drained by a deferent duct:

- B1: The undrained supernumerary testis has its own epididymis
- B2: The undrained supernumerary testis does not have its own epididymis and as a consequence, it consists of testicular tissue only.

Taking these parameters into account, and the biopsy's results, we categorized the patient's triorchidism as Type A2 as the two unilateral testes had the same duct but individual epididymides. We concluded to its surgical excision fearing of the unforeseen possibility of the malignancy in the proximal or distal future, whilst understanding the matter of controversy around such management. To contradict the alternative solution of orchidopexy, we felt that since we deal with an atrophic testis, it most likely has minor to none capability of spermatogenesis. Finally, we proposed regular monitoring of the patient for ensuring the maximum control of the situation.

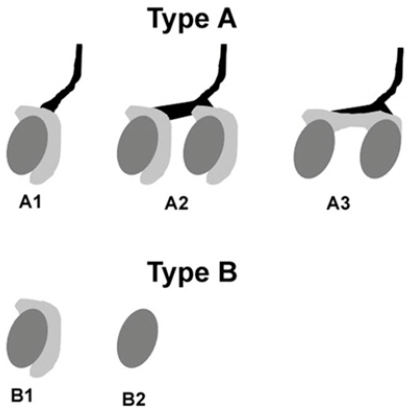

Figure 5: Classification of polyorchidism [6].

Conclusion

The present case report approaches the issue of a histologically confirmed triorchidism as an incidental finding by the patient's parents and later on, by the clinical practitioner during a physical exploration. With the view to determining whether the surgical excision of the additional testis or its monitoring by imaging is more preferable, we took many factors into consideration, including patient's comorbidities, testicular biopsy's outcome as well as the classification of the extra testes. All of the aforementioned data proved that the removal of the mass will not impair in any way the patient's reproductive function. That said, its presence is clearly not indispensable and in point of fact, surgery can diminish the future peril of testicular cancer if the atrophic testes were to be preserved.

Declaration of competing interest: There is no conflict of interest.

Consent: Written informed consent was obtained from the patient's guardians for publication of this case report and the accompanying images of the latter.

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