SUPERNUMERARY TESTICLE: CASE REPORT AND BIBLIOGRAPHIC REVIEW

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Summary.- OBJECTIVE: We report a case of supernumerary testis, a rare anomaly with only around 100 cases reported in the literature.

METHODS: We describe the case of a 26-year-old man who consulted for a left paratesticular tumor. Physical examination and ultrasound showed a 2-cm nodular lesion over the left epididymis. The lesion was confirmed as supernumerary testis by surgical examination and biopsy and was subsequently excised, given the malignancy potential.

RESULTS: The supernumerary testis was evaluated using two standard classifications, one assessing function and embryological development, and the other assessing topography, anatomy, and reproductive potential.

CONCLUSIONS: The differential diagnosis for an intrascrotal mass should include the possibility of a supernumerary testes; hence, surgical examination and biopsy are necessary. Supernumerary testes should be excised in the case of pain, dysplasia, or in situ carcinoma, or whenever the biopsy is inconclusive.

Keywords: Supernumerary testis. Polyorchism. Polyorchidism

Resumen.- OBJETIVO: Presentar un caso de teste supernumerario, rara anomalía de la que existen descritos y fundamentados aproximadamente 120 casos.

MÉTODOS: Presentamos el caso de un varón de 26 años que consultó por presentar tumoración paratesticular izquierda. La exploración física y ecográfica evidenciaron una lesión nodular de aproximadamente 2cm sobre epidídimo izquierdo que se confirmaría posteriormente durante la exploración quirúrgica y biopsia. Dada la posibilidad de malignización se decidió extirpar el teste supernumerario.

RESULTADOS: Para evaluar un teste supernumerario disponemos de varias clasificaciones que evalúan la funcionalidad y el desarrollo embriológico del mismo, así como la topografía, la anatomía y su potencial reproductivo.

CONCLUSIONES: Ante una masa intraescrotal debemos tener en cuenta a la hora de realizar el diagnóstico diferencial la presencia de un teste supernumerario, siendo fundamental la exploración quirúrgica y biopsia. Realizaremos exéresis del teste supernumerario siempre que sea origen de dolor, displasia, carcinoma in situ, o la biopsia arroje dudas.


INTRODUCTION

Polyorchism or polyorchidism is a rare congenital anomaly defined as the incidence of one or more extra testes. It was first described postmortem by Blasius in 1670, and the first histologic evidence was provided by Lane in 1895 (1). The extra testis may be intrascrotal or extrascrotal, and a left location tends to
predominate. It is important to study the presentation of the supernumerary testis because it can be associated with urogenital disorders. The urologic anomaly is extremely rare, with fewer than 100 cases described in the literature (2).

**CASE REPORT**

A 26-year-old man with no relevant medical history was referred by his primary care physician to our outpatient clinic for a left paratesticular tumor that had been slowly but steadily growing. The tumor was not associated with any local trauma or other symptoms.

The physical examination revealed a normal penis, and mobile right and left testes of normal size and consistency. A palpable nodule, painless to the touch and measuring some 2 cm in diameter, was detected over the left vas deferens, independent of the testis on the same side.

Tumor marker tests were normal. A Doppler ultrasound of the testis revealed the presence of an extratesticular lesion, measuring 1.3 cm in diameter. The lesion was adjacent to the head of the epididymis, isoechogenic to the testis, and of similar Doppler pattern, consistent with an adenomatoid tumor of the epididymis (Figure 1).

Based on the clinical evaluation, surgery to examine and resect the lesion was scheduled. A left scrotectomy was performed under spinal anesthesia. Subsequent dissection by planes as far as the left-sided tunica albuginea revealed a mass supplied by the seminal vesicles at the level of the head of the epididymis. This mass, measuring approximately 2 cm in diameter, was suggestive of a supernumerary testis (Figure 2). The mass was excised, and a pathology study confirmed the initial diagnosis.

**DISCUSSION**

Around the fourth to sixth week of embryonic development, the primitive urogenital ridge develops from the intermediate mesodermal, eventually going on to form the testicular primordium (3,4); the epididymis and vas deferens develop from the Wolffian (mesonephric) duct (4,5).

Polyorchidism results from an abnormal division of the urogenital ridge, with or without the mesonephros, before the eighth week of gestation (3). Depending on where the division occurs, a supernumerary testis will develop with either shared or individual vas deferens and epididymis (6,7,8).

No chromosomal disorder has been demonstrated for polyorchidism (7).

The anomaly is usually diagnosed in childhood or adolescence when the patient presents with mild, intermittent scrotal pain associated with swelling, as well as a hard mass in the scrotal, inguinal, or abdominal region.

Complications associated with the condition include cryptorchidism (40%), hernia (30%), torsion (15%), hydrocele (9%), and malignancy (6%) in the form of teratoma, rhabdomyosarcoma, seminoma, or embryonal carcinoma (9). Cases of infertility as well as cases of postvasectomy fertility have also been described. All these complications tend to develop on the same side as the supernumerary testis (9,10).

The most common finding is triorchidism, typically developing on the left side (60%), proximal and anterior to the normal testis. The right side tends to be favored when there is cryptorchidism or malignancy.
Presentation with bilateral duplication is very rare, and fewer than ten cases have been reported (12).

Two classifications are described in the literature. Thum's classification assesses function and embryological development, and Singer's classification is based on topography, anatomy, and the reproductive potential of the testis (13).

Thum's classification refers to three types of supernumerary testis: in type I, the supernumerary testis has no epididymis or vas deferens, in type II, it is linked to the normal testis by a common epididymis and shares the vas deferens, and in type III, it has its own epididymis but shares the vas deferens with the normal testis.

Singer's classification is based on two supernumerary testis types, called type I and type II, with and without attachment to a draining epididymis and vas deferens, respectively, and with no reproductive potential in the latter. These two types are further classified as A if the position is intrascrotal, and B if the position is ectopic, whether inguinal or abdominal (13).

Our case was a Thum type II or a Singer type IA.

As to the sperm production potential of supernumerary testes, around half are active, although the testes in many cases have no vas deferens (14).

The differential diagnosis for this entity includes disorders such as hydrocele, spermatocele, epididymal cyst, aberrant epididymal tissue, and spermatic cord cysts (Nuck cysts), which can simulate an intrascrotal mass (10).

Fundamental to diagnosis is Doppler ultrasound, which indicates the presence or absence of blood flow, echogenic homogeneity, and the presence of tumor masses (15). Magnetic resonance imaging has the advantage of multiplanar imaging, with high soft-tissue resolution and contrast; furthermore, the technique provides greater anatomic detail and the same results regardless of operator, making it ideal for identifying supernumerary testes. Scintigraphy may be of only limited use when torsion is suspected or the testis is small (14,16).

In our patient, the Doppler ultrasound revealed a suspicious mass subsequently confirmed as an ectopic testis.

Other complementary methods can be used to assess malignancy or the likelihood of malignancy, for example, assessment of follicle-stimulating hormone, testosterone, alpha-fetoprotein, and beta-human chorionic gonadotropin, semen analysis, and finally, a biopsy to evaluate spermatogenesis and the existence of foci of dysplasia (7,10,11).

Until 1980, patients with polyorchidism were almost invariably treated by orchidectomy of the additional testis (17). Recent studies indicate the avoidance of surgery (and preservation of future fertility) for patients who are asymptomatic and whose extra testis is normal according to clinical criteria and radiologic and additional tests. In patients with a normal extra testis, resection should not be performed even in the case of pain, although the testis should be fixated to avoid possible torsion (2).

The fact that a supernumerary testis may eventually become malignant is in itself sufficient indication for excision for some authors. Although histology studies often show atrophy and germ cell aplasia, there is little evidence of dysplastic changes (18).

The broad consensus is that Singer type IB, IIA, and IIB ectopic testes should be excised, whereas type IA testes should only be resected in the following circumstances: obvious dysplastic changes in the biopsy, aspermatogenesis, radiologic evidence of malignancy, patient preference, or impossibility of regular patient follow-up (13,18,19,20). An intraabdominal location is also sufficient reason for excision according to some authors (10).

CONCLUSIONS

Although polyorchidism is rare, it should be considered in the differential diagnosis for intrascrotal masses. Sonography and magnetic resonance imaging are both useful in identifying the condition.

Surgical examination enables an assessment of supernumerary testis location, anatomy, and relationship with ipsilateral anatomic features as well as histologic analysis of malignancy potential and spermatogenic function of the additional testis. Depending on the results, the supernumerary testis may be either excised or monitored periodically. In either case, the patient should be informed of the treatment options and agree on the approach with his physician.

REFERENCES AND RECOMMENDED READINGS

(*of special interest, **of outstanding interest)


