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INCIDENTALLY DIAGNOSED TRIORCHIDISM IN AN ELDERLY PATIENT PRESENTING WITH HYDROCOELE

Urology

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ABSTRACT

A supernumerary gonad is a rare finding and can be encountered incidentally in a variety of clinical scenarios like inguinal hernioplasty, scrotal exploration for testicular torsion, or simply on palpation of the external genitalia as part of routine examination. Till date, approximately 190 cases have been reported regarding this rare anomaly. Herein we report one such case who presented to us with left hydrocele and incidentally detected to have a palpable nubbin of tissue at the base of the scrotum, which later turned out to be a supernumerary testis. The location of the supernumerary testis in our case was caudal to the orthotopic testis, and this particular location, to the best of our knowledge, has not been reported in medical literature so far.

KEYWORDS

Triorchidism, Polyorchidism, Supernumerary, Hydrocoele

BACKGROUND:
Polyorchidism is a rare congenital anomaly defined as the presence of more than 2 testes. As a urologist, we come across various types of scrotal swellings, the most common ones being sebaceous cysts, hydroceles and testicular malignancies. Hence the differential diagnosis of a supernumerary gonad presenting as a scrotal swelling, comes last on our list. Nevertheless, this anomaly has fascinated urologists, radiologists and other medical personnel alike, ever since it has been discovered.

Case Presentation:
A 63-year-old gentleman presented to us with a chief complaint of a left-sided scrotal swelling which had been gradually increasing in size for the past few years. On physical examination, a cystic transilluminating swelling of size 10 x 8 x 6 cm was palpable in the left hemiscrotum. Another firm swelling of size 2 x 1.5 x 1.5 cm was noted at the base of the scrotum which was separate from the left hydrocele sac. The right testis was normal on examination. Provisional diagnosis of a left hydrocoele was made and decision was taken to go ahead with hydrocelectomy.

USG scrotum showed bilateral hydrocele (left >> right) along with a well-defined structure of size 2 x 1.5 x 1.5 cm at the base of scrotum with echogenicity & morphology similar to testis. The left testis measured 3.2 x 2 cm, and the right testis measured 3 x 2 cm. A possibility of right supernumerary testis was considered based on ultrasound imaging. Pre-operatively, on palpation, since the mass was firm and non-fluctuant, a diagnosis of a sebaceous cyst with inspissated thick contents was kept. The other working diagnosis was that of a small chronic walled-off abscess arising from an initial episode of folliculitis. Also, a lipoma was kept as a differential diagnosis. A rare possibility of supernumerary testis was also kept.

DISCUSSION:
Historically, the first reported case of polyorchidism can be traced back to the 14th century [1]. One of the earliest findings of polyorchidism were recorded in post-mortem reports documented by Blassius in 1660. The first histologic description of polyorchidism was given by Ahlfeld, in 1880 [2]. The first case of surgical excision of an extra testicle was reported by Lane in 1895 [3]. The pre-operative diagnosis of polyorchidism is difficult, but sonography is of value since most of the extra testes are located in the scrotum. The exact mechanism for occurrence of polyorchidism is not known, but longitudinal or transverse division of the genital ridge, possibly by development of peritoneal bands, has been proposed [4-6].

On the basis of the embryologic development, Leung [5] classified polyorchidism into 4 types, as shown in Figure No.4. In type A, the supernumerary testis lacks an epididymis and vas deferens. In type B, the supernumerary testis has its own epididymis. Depending on the degree of division, the supernumerary testis may be connected longitudinally to the epididymis of the normal testis and its vas deferens (B2), or it may lack any connections to the normal testis (B1). In type C, the supernumerary testis has its own epididymis and shares the vas deferens with the regular testis in a parallel fashion. This variant results from incomplete longitudinal division of the genital ridge and the proximal portion of the mesonephric duct. In type D, complete longitudinal duplication of the genital ridge and mesonephric duct occur, with resultant complete duplication of testes, epididymides, and vas deferens. This type may be associated with an ipsilateral duplicated ureter and is the least common. Our index case belongs to type A of Leung classification, as the supernumerary testis lacked its own epididymis and vas deferens. However, one surprizing fact was that, the supernumerary testis was located caudal to the normal testis, unlike what has been depicted in Leung classification (Figure 4).

Singer et al [6] proposed a classification based on reproductive potential of the supernumerary testis. In most cases of polyorchidism, a single supernumerary testis (triorchidism) is present, with the left side being more frequent [5]. However, in our case the supernumerary testis was located on the right side. The most common location of the supernumerary testis is within the scrotum [4]. Malignant transformation may occur within the supernumerary testis, irrespective of its location [7,8]. The major sonographic feature of polyorchidism is a scrotal mass that has an echotexture identical to that of the ipsilateral testicle.

CONCLUSIONS:
Polyorchidism is a rare congenital anomaly and may be encountered incidentally as a painless scrotal mass or an incidental sonographic finding in most of the cases. Surgical intervention is needed only in symptomatic cases. Asymptomatic cases may be followed-up. Although no guidelines exist regarding the protocol for follow-up evaluation of such cases, it is reasonable to suggest that such patients be followed on a yearly basis with scrotal ultrasonography.

List of Abbreviations:
USG – Ultrasonography

Declarations:
1. **Ethical Approval:** Not applicable
2. **Competing interests:** None
3. **Authors' contributions:** Dr. RSD was the primary operating surgeon. Dr. SKD wrote the manuscript. Dr. UKM was an observer in this case.
4. **Funding:** No funding was received.
5. **Availability of data and materials:** Not applicable

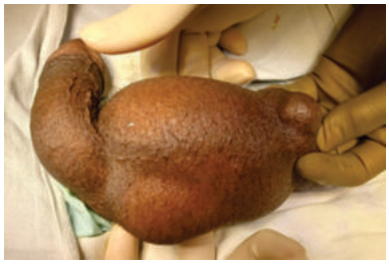

Fig.1: Pre-operative picture showing the relationship of the supernumerary testicle (marked with arrow), on inspection, to the right testis, and the left hydrocele.

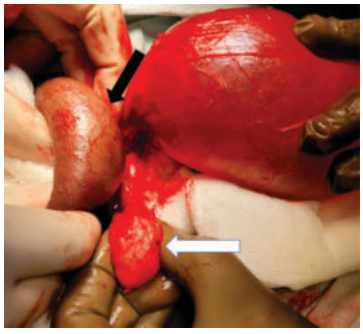

Fig. 2: Intra-operative picture depicting the 3 testes; isolation of the nubbin of tissue (white arrow) situated at the base of the scrotum. On exploring adjacent structures, no apparent vas deferens or cord structures could be traced. (Hydrocele depicted by arrowhead; right testis depicted by black arrow)

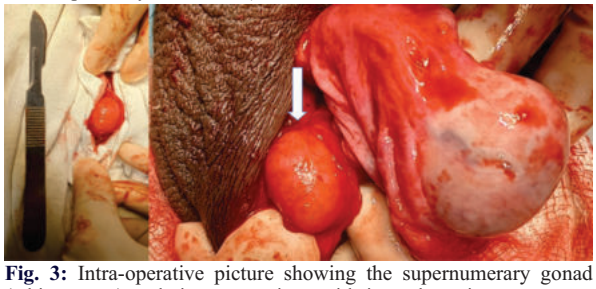

Fig. 3: Intra-operative picture showing the supernumerary gonad (white arrow) and size comparison with its orthotopic counterpart, after the hydrocele was repaired.

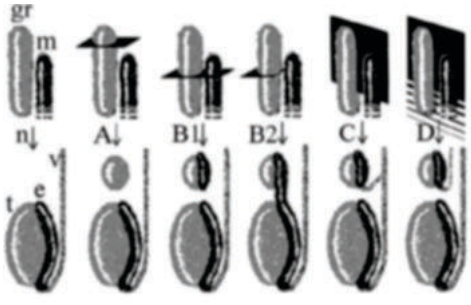

Fig. 4: Illustrative diagram showing the planes of division in the various types of supernumerary testicles, according to Leung classification. n=normal embryo, gr=gonadal ridge, m=mesonephric duct, t=primordial testis, e=epididymis, v=vas deferens.

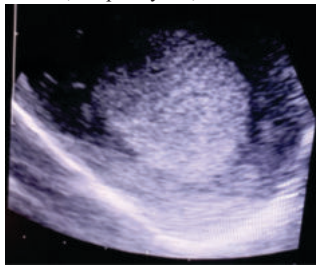

Fig. 5: Ultrasound image showing the supernumerary testis, exhibiting similar echogenicity and morphological characteristics as the orthotopic testes.

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