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CASE REPORT

Bilateral gynaecomastia – a diagnostic indicator of malignant testicular mass: case report and review of literature

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Gynaecomastia is the most common benign breast disorder in men. Though uncommon, testicular malignancies are known to cause bilateral gynaecomastia. Consequently, a high index of suspicion is required when there is a coexistence of testicular mass. The case presents a 48-year-old male with a nine-month history of left testicular swelling and a history of bilateral gynaecomastia. The patient had no abnormality on clinical or biochemical examination (LDH 192 U/L [100–190], ß-hCG < 1 IU/L, α-FP < 0.6 µg/L [0.0–7.0]). Ultrasound imaging revealed a hypoechoic mass on the lower pole of the left testis, and the histology of the radical orchidectomy specimen revealed a Leydig cell tumour (LCT). This tumour histology is commonly associated with gynaecomastia and beta-human chorionic gonadotropin (ß-hCG)-secreting tumours, such as choriocarcinomas, with a strong differential diagnosis. This report underlines the importance of a clinical breast examination in patients being evaluated for a testicular mass, as this may indicate an unusual malignancy.

Keywords: gynaecomastia, malignancy, testicular mass

Introduction

Over the last three decades, there has been a heightened incidence of testicular malignancies warranting public awareness, thereby increasing the number of men who seek urological care on account of symptoms associated with testicular pathology.[1] Many patients have had confirmation on high-resolution ultrasound scans of the testis and prior clinical examinations. However, few have had a clinical breast examination at presentation.

The presentation of a functioning testicular mass with an endocrine abnormality (such as gynaecomastia) is rare in men. Leydig cell tumours (LCTs) account for 1–3% of all testicular malignancies.[2,3] The peak incidence at presentation is a testicular mass or an endocrine abnormality in the fourth decade of life. Due to the secretion of oestrogens and testosterone from the tumour cells, a LCT is associated with male sexual precocity and gynaecomastia in children and adults, respectively.[4]

Interestingly, gynaecomastia may precede the evidence of a LCT on clinical examination. We report a case in which bilateral gynaecomastia was caused by a testicular LCT. This finding may appear uncommon, but it reiterates the importance of a detailed physical and biochemical examination of any patient with a testicular mass, especially when coexisting with unexplained gynaecomastia, to exclude a testicular tumour.

Case report

A 48-year-old male with a nine-month history of testicular swelling was referred by a private urologist. On examination, the patient had a non-tender, hard swelling on the lower pole of the left testis, with surrounding cystic areas. A testicular ultrasound revealed a solid lower pole mass on the left testis with hydrocele. The right testis was normal.

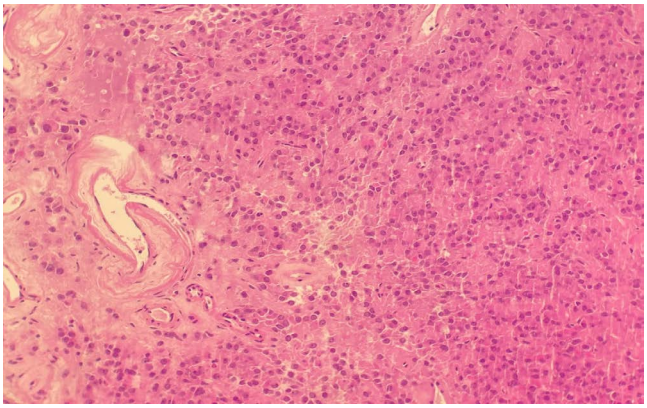

Figure 1: Low-power frozen section taken intraoperatively

A hypoechoic mass was present within the lower pole of the left testis with a maximal diameter of 25 mm. There was internal multifocal calcification. Scrotal Doppler revealed a highly vascular mass with no extratesticular extension. A repeat scrotal ultrasound at our facility revealed a right testicle, 22 × 19 × 31 mm (TRV × AP × CC), a left testicle, 26 × 21 × 34 mm, and a heterogeneous, poorly defined left intratesticular mass with associated calcifications. There was increased flow within the intratesticular mass, with associated hydrocele and debris in the left scrotum. The right testicle was normal and homogenous with no abnormality. Non-benign lesion serum tumour markers were within normal limits (LDH 192 U/L [100–190], ß-hCG < 1 IU/L, α-FP < 0.6 µg/L [0.0–7.0]).

The patient was admitted for urgent left radical orchidectomy. However, a frozen section biopsy was performed due to the clinical history and normal tumour marker values. The frozen section revealed a LCT, warranting left radical orchidectomy as planned (Figure 1). On-table examination of the chest revealed Tanner stage 4 bilateral gynaecomastia, to which the patient alluded that it preceded the testicular symptoms and he had been on weight

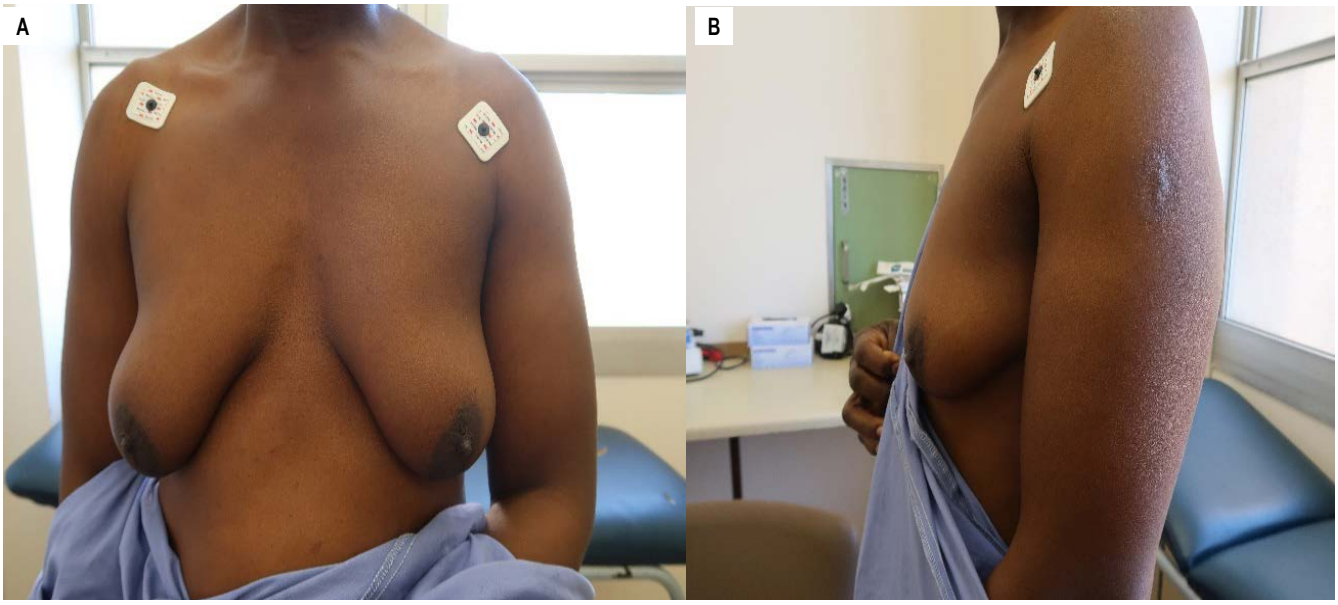

Figure 2: (A) Anterior-posterior and (B) lateral views of the patient (patient provided consent)

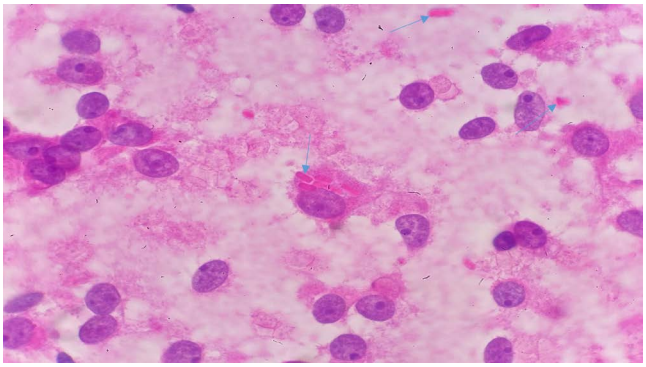

Figure 3: High-power touch imprint cytology showing Reinke crystals or crystalloids* . Eosinophilic(red) crystalline inclusions-intracytoplasmic,intranuclear or extracellular (arrows showing Reinke crystals)

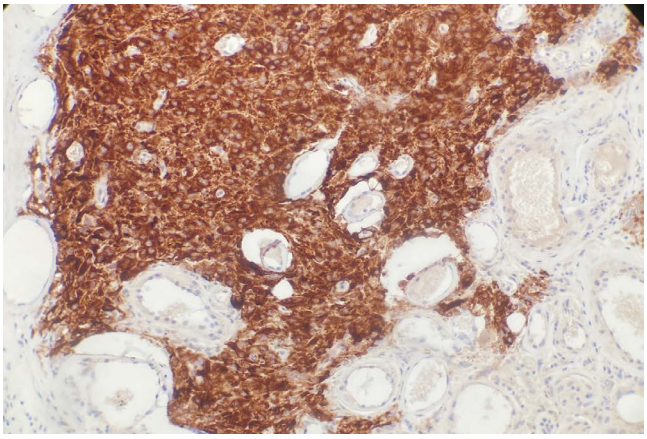

Figure 4: Inhibin immunostaining

reduction regimen to reduce the manifestation (Figure 2). There was no prior staging computed tomography.

Intraoperatively, frozen section revealed sheets of epithelioid cells with prominent nuclei, nuclear hyperchromasia, abundant granular eosinophilic cytoplasm, and Reinke crystals.** Formal sections showed a LCT consisting of polygonal cells with abundant eosinophilic granular cytoplasm and round nuclei with prominent central nuclei. Scattered Reinke crystals are seen (Figure 3).

There is no evidence of increased mitosis, necrosis, or lymphovascular invasion. Immunohistochemistry revealed diffuse inhibin cytoplasmic positivity.

The patient declined further management of the gynaecomastia. However, written informed consent was obtained from the patient to publish the case report.

Discussion

There is a rising incidence of testicular cancer worldwide.[1] Most of these tumours are palpable, warranting confirmation of diagnosis on ultrasonography.[5] About 15–25% of patients with LCTs have associated gynaecomastia which accounts for 3% of testicular malignancies.[6] This report reiterates the importance of a thorough clinical examination to exclude gynaecomastia in a healthy man with or without scrotal mass pathology.

LCTs may occur in patients of any age with a bimodal peak incidence in the prepubertal age group and patients aged between 20 and 50 years. Histopathological features typically include a spherical and lobulated shape with a tan to brown colour. In approximately 50% of cases on microscopy, sheets and nests of Leydig cells are visible, usually with an eosinophilic cytoplasm separated by fibrous septae and Reinke crystalloids.[7,8]

LCT diagnosis poses a great challenge to surgeons, though in children, the clinical presentation invariably manifests as isosexual precocious puberty.[9] The presentation of gynaecomastia or impotence occurs in about 20% of adults with LCTs. However, our patient had no history of impotence despite a preceding history of gynaecomastia, for which he had not sought treatment.

* Touch imprint cytology was more predictive than wet preparation, especially regarding testicular pathological specimens.
** Reinke crystals are structures pathognomonic of Leydig cells, which are essential in the function of testosterone production and vital for men's reproductive health.

Only 10% of LCTs are bilateral, with 7–10% known to metastasise. This is worrisome because it is the obvious feature of malignancy.[10] In older patients, it is associated with a poor prognosis because of its resistance to chemotherapy and radiation.

Gynaecomastia is defined as a generalised enlargement of the male breast, and it is the most common benign breast condition seen in men. It may be unilateral or bilateral, occurring in about 40–65% of males. The common aetiologies are age, diseased states, drugs, or idiopathic with a history of an elevated oestrogen/androgen circulating ratio.[11]

The diagnosis of gynaecomastia demands appropriate investigation and prompt treatment despite the uncommon aetiology of malignant testicular origin and its life-threatening route. Many researchers have attributed it to a reduced testosterone/17β-estradiol (E2) ratio due to an increased production of E2 or an enhanced aromatase production by the testicular tumour.[12]

We recommend that all patients with gynaecomastia have their testes carefully examined, in addition to hormonal profiling, and vice versa, depending on which presents first. Gynaecomastia before a readily palpable testis tumour is a recognised problem from previously published literature. Nonetheless, there is a need to reiterate a holistic examination of all patients to exclude these ominous signs that could warrant further investigation and evaluation.[13]

Conclusion

This case report aimed to alert physicians to the significance of a physical examination of patients with a testicular mass. It also highlights the importance of repeated testicular self-examination in men with breast enlargement, as it may further assist in the evaluation of the patient.

Conflict of interest

The authors declare no conflict of interest.

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