

Leydig Cell Testicular Tumor Presenting as Bilateral Breast Masses: A Case Report

Abstract

Leydig cell tumors are rare but are the most common nongerm cell gonadal tumors. They are mostly benign but malignant variants have been reported. Leydig cells constitute the main androgen-synthesizing compartment in adult males but are also capable of estrogen production. This can manifest with clinical features of excessive hormone elaboration. We report a case of a 39-year-old man with abnormal bilateral breast development, reduced libido, and weak erection of 3 years' duration. He never noticed any testicular swelling before presentation. Examination revealed well-developed breasts bilaterally and a mass in the lower pole of the left testis. Scrotal ultrasound confirmed a hypochoic tumor measuring 2 × 3 cm in the lower pole of the left testis and hormonal evaluation revealed a markedly elevated estradiol level. A diagnosis of estrogen-secreting testicular tumor was made. He had a testis-sparing excision of the scrotal lesion as well as liposuction and excision of glandular tissues of the breasts. He had an uneventful postoperative recovery and was discharged a day after surgery. Histology of excised testicular lesion revealed a benign Leydig cell tumor. Four months following surgery, there was an improvement in libido, erection, and sperm concentration of the patient. The patient was also very satisfied with the cosmetic outcome of the excision of the bilateral gynecomastia. We recommend self-examination of testicles as an important step for early diagnosis of testicular tumors.

Keywords: Gynecomastia, Leydig cell tumor, testicular tumors

Introduction

Leydig cell tumors (LCTs) are rare tumors of the male testicular interstitium.^[1] They are uncommon and account for only 1%–3% of all testicular masses but are the most common nongerm cell gonadal tumors.^[2] Most of them are clinically benign, however, about 10% of the reported cases have been described as malignant.^[2] LCTs have been reported worldwide including in Africa and Nigeria but a thorough literature search did not reveal any report of LCT presenting with gynecomastia in Nigeria before this case.^[3–5]

LCTs of the testis are frequently functional or hormonally active, leading to either feminizing or virilizing syndromes. Almost all LCTs are functional, but a few are nonfunctional. In fact, LCTs are one of the most common causes of excess androgen production in males. They produce steroids, which can lead to disruptions in the endocrine system. Boys

with testosterone-secreting LCTs typically experience symptoms of early puberty, while excess androgens usually do not have significant effects on adults. Around 25% of LCTs produce estrogen, which can cause gynecomastia and sexual dysfunction as well as infertility in adults.^[1,6] LCTs have also been observed in prepubescent boys, who experience symptoms of both estrogen and androgen production, leading to the development of gynecomastia, and early puberty simultaneously.^[2] Our patient was an adult male who presented with bilateral gynecomastia as a result of excessive estrogen secretion by the tumor in the left testis.

Case Report

Mr. A.A. is a 39-year-old single man with a complaint of bilateral breast enlargement of 3 years' duration. Breast swellings were painless and progressively increased in size to the size of an adult female over about 6 months. There was no history of abnormal nipple discharge. He also complained of progressive weak erection

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Rufus Wale Ojewola, Rasheed Ayobami Aranmolate¹

Department of Surgery, Faculty of Clinical Sciences, College of Medicine of University of Lagos/Lagos University Teaching Hospital, Idi-araba, Surulere, Lagos, ¹Plastic and Reconstructive Unit, Grandville Medical Group, Off Adela, Surulere, Lagos, Nigeria

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Address for correspondence:

Dr. Rufus Wale Ojewola, Department of Surgery, College of Medicine of University of Lagos/Lagos University Teaching Hospital, PMB 12003, Idi-Araba, Surulere, Lagos, Nigeria.
E-mail: rwaleojewola@yahoo.com, rojewola@unilag.edu.ng

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with an International Index of Erectile Function (IIEF-5) score of 6 and progressive loss of libido, both noticed after the onset of breast enlargement. He had just lost a romantic relationship and was unwilling to start another because of these sexual symptoms. He presented to two peripheral centers before being referred for endocrinological evaluation. The endocrinologist requested luteinizing hormone, follicle-stimulating hormone, testosterone, prolactin, and estradiol. These hormones were within normal limits except for a markedly elevated estradiol level of 177.6 pg/mL (normal range in men = 12–34 pg/mL). Magnetic resonance imaging (MRI) of the brain was also essentially normal. He was then referred for urological review on account of the reduced libido and weak erection.

A general physical examination revealed a healthy young man with male facial and pubic hair distribution but with well-developed breasts bilaterally [Figure 1]. Abdominal examination was essentially normal, but scrotal examination revealed a firm to hard swelling in the lower pole of the left testis measuring approximately 2.0 × 2.5 cm in size. The right testis was essentially normal. A diagnosis of a suspected estrogen-secreting left testicular tumor with bilateral gynecomastia was made. Testicular tumor markers, namely, beta-human chorionic gonadotropin (hCG), alpha-fetoprotein (AFP), and lactate dehydrogenase (LDH) were within the normal level. The hormone profile repeated still showed markedly elevated estradiol of 179.9 pg/mL. Seminal fluid analysis (SFA) also showed severe oligospermia of 2.65 million/mL and total motility of 24%. Scrotal ultrasound confirmed a hypoechoic tumor measuring 2 × 3 cm in the lower pole of the left testis.

He had bilateral liposuction and excision of the glandular tissue of both breasts which yielded about 1.8 kg of tissues as well approximately 1,100 mL of liposuction effluent [Figure 2] as well as scrotal exploration at the same sitting. Scrotal exploration through a linear incision on the tunical albuginea revealed a completely intratunical and well-encapsulated mass in the lower pole of the left testis



Figure 1: Bilaterally well-developed breast in a man

[Figure 3]. He had a testis-sparing excision during which the lesion was enucleated nicely from adjacent unaffected testicular tissue. Hemostasis was secured and the tunical albuginea closed using Vicryl 3/0 suture. Histology of the mass revealed a benign LCT. He had an uneventful postoperative recovery and 3 months postoperatively, estradiol returned to a normal level (16.1 pg/mL). There was marked improvement in libido while erection also improved with a new IIEF-5 score of 17 four months after surgery. Furthermore, SFA also improved significantly with a sperm concentration of 13.8 million/mL and total motility of 58%. The patient was also satisfied with the cosmetic outcome of the gynecomastia treatment [Figure 4].

Discussion

Testicular cancer is one of the less common cancers and tends to mostly affect men between the ages of 15 and



Figure 2: Specimen of the excised lesion and liposuction effluent



Figure 3: Well-encapsulated mass in the lower pole of the left testis



Figure 4: Breast sizes 4 months postoperatively

49 years. It is the most common cancer among people assigned male at birth aged 15–35.^[3] Of all primary testicular tumors, more than 90% are germ-cell tumors like seminoma and nonseminoma, while the remainders are nongerminoma neoplasms like tumors of the Leydig cells and Sertoli cells as well as gonadoblastoma.^[3] Leydig cells constitute the main androgen-synthesizing compartment in adult males but are also capable of estrogen production.^[7] LCTs are rare neoplasms arising from gonadal stroma, accounting for 1%–3% of all testicular tumors in adults and 4% in prepubertal children.^[2,8]

Although LCTs can arise at any age, they affect mostly men between 20 and 60 years as seen in this index patient who was 39 years old at presentation which corresponds to the age group with the highest incidence. LCTs are usually unilateral and can affect any of the testes. Few cases of bilateral LCTs have however been reported.^[9]

LCTs can present in varied ways. Testicular tumors generally present with noticeable testicular mass and or features of the manifestation of hormones emanating from the tumor. It may present as a painless lump in the testis or the mass may be an incidental finding on scrotal ultrasonography performed for other conditions like evaluation of infertile men.^[10] Though the lump is present in most cases, it may never be noticed by the patient as it is usually painless.^[11] In this patient, the lump was painless and was never noticed by the patient until he was examined. Lump associated with pain is a common reason for early presentation in a few cases.^[3]

The most common hormones termed testicular tumor markers, namely, beta-hCG, AFP, and LDH are usually secreted in germ-cell testicular tumors and do not manifest with any clinical symptom.^[3] However, for estrogen-secreting tumors, presentation is usually feminizing features like abnormal breast enlargement, declining erection, and reduced libido in males as seen in this patient.^[12] And these may be the first symptoms before the lump is felt. In

fact, our patient never felt this mass until after a scrotal examination. Azoospermia or oligospermia and infertility are uncommon manifestations and if present may be reversed after treatment.^[13,14] In the index case, the estradiol level regressed dramatically to normal within 3 months of excision. It is also noteworthy that erection and seminal fluid parameters also improved after surgery as his IIEF-5 score improved to 17.

It may be a diagnostic dilemma locating the underlying cause of recent onset gynecomastia. Our patient had visited two general practitioners (GP) who had no idea of the cause of gynecomastia. The second GP referred him to an endocrinologist for evaluation. Expectedly, an MRI of the brain was done but revealed no abnormality. Pituitary gland hormones were also essentially normal. The association of reduced libido and weak erection necessitated a referral for urologic evaluation, after which a testicular lump was discovered. This finding suggests the importance of a thorough examination including a scrotal examination in every case of adult-onset gynecomastia.

Gynecomastia usually results from an imbalance between testosterone and estrogen. Estrogen causes breast tissue to grow.^[15] In a testicular neoplasm, gynecomastia is caused by estrogen elaboration from the neoplasm or by increased aromatase activity due to the stimulus of the b-HCG by the tumor.^[16] Rarely, hyperprolactinemia may lead to gynecomastia by its effects on the hypothalamus to cause central hypogonadism.^[17,18] The testicular differential diagnosis of LCT associated with gynecomastia includes estrogen-secreting seminoma and large-cell calcifying Sertoli cell tumors which can also result in symptoms such as gynecomastia and decreased libido in addition to testicular mass as seen in this index patient.^[19,20]

Once gynecomastia has been diagnosed, treatment of the underlying cause is warranted. Gynecomastia developing from the testicular tumor will almost never regress on its own and therefore requires treatment in addition to the removal of the testicular mass.^[15] Treatment includes liposuction only but additional excision of glandular tissue is necessary in patients with big glands like in the index case. A small incision for breast parenchymal removal in gynecomastia assisted by liposuction showed a good technical approach for consistent improvement in the quality of life as in this index patient.^[21] Excision of the testicular mass tumor is indicated in all cases. Enucleation is sufficient in well-delineated cases like the index case, whereas partial or radical orchiectomy may be required in some cases, especially in malignant or large tumors.^[13] This treatment is usually followed with improvement in the symptoms of hormone manifestation of the tumor. This was the case with our patient.

The impact of gynecomastia on mental health and quality of life is enormous. Various psychosocial and psychological consequences of gynecomastia include but

are not limited to depression, anxiety, disordered eating, body dissatisfaction, reduced self-esteem and many more.^[22] In fact, our patient lost a heterosexual relationship because of abnormal breast development which did not go well with his partner. Associated decreasing libido and weak erection also contributed to the eventual separation.^[23]

Conclusion

LCTs are uncommon neoplasms arising from the gonadal stroma. It is critical for physicians to remember and not overlook the possibility of this rare tumor. Gynecomastia in an adult is a symptom of an underlying endocrine problem. This underscores the importance of examination of the testes in a male patient who presents with this symptom. The self-examination of testicles appears to be a very important step for early diagnosis of testicular tumors.

Authors' contributions

- Rufus Wale Ojewola—concept, data collection, and initial and final write-up.
- Rasheed Ayobami Aranmolate—Data collection and review of the final write-up.

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Conflicts of interest

There are no conflicts of interest.

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