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Testis sparing surgery for Leydig cell tumors: New three cases and review of the current literature

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Abstract
Introduction: Leydig cells tumors of the testis are uncommon, representing between 1 and 3% of the testicular tumors and for which the natural history and therapy are debated between radical orchitectomy and organ-sparing surgery.
Subjects and methods: We report three new cases of Leydig cells tumors, treated in our department and we discuss the clinical, diagnostic and therapeutic aspects of this uncommon tumor.
Results: The mean patient age was 29 years (23–37 years). Medical referral was for a testicular pain in two and gynecomastia in one case. All patients were treated surgically, through an inguinal incision and the procedure included clamping of the spermatic cord. During surgery, frozen section were analyzed in two cases and the tumor enucleation with organ-sparing surgery was performed. The other patient had an unilateral orchidectomy and controlateral tumorectomy. The mean follow-up was 40 months with no local recurrence.
Conclusion: Patients diagnosed with Leydig cells tumors have a good prognosis; this study shows the safety of conservative surgery treatment, provided it is subsequently followed by close surveillance, as it preserves maximum fertility, and these tumors usually have a favorable prognosis.

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Introduction

Leydig cell tumor (LCT) is the most frequent interstitial neoplasms of the testis [1], accounting for 0.8–3% of all testicular tumors [2] and 4–9% of testis tumors in prepubertal males [3]. The recent surgical approach to treat these tumors is conservative [4].

Here we report three new cases of Leydig cell tumors treated with organ-sparing surgery and we discuss the clinical, diagnostic and follow-up of this uncommon tumor.

Case reports

Since 2005, we have diagnosed three new cases of Leydig cell tumor of the testis, in patients A, B and C (aged 23, 28 and 37 years, respectively). Patient A with history of precocious puberty due to congenital adrenal hyperplasia was referred for bilateral testicular pain. Patient B consulted for mild left testicular pain and swelling of several months' duration. Patient C presented with a one-year history of symptomatic left gynecomastia, which on ultrasound examination had the appearance of simple gynecomastia (confirmed by fine needle aspiration). Physical examination of the external genitalia revealed a mildly enlarged and sensitive bilateral testes in patient A, and left higher testicular pole in patients B and C. In all patients, testicular ultrasound showed a homogeneous, hypoechogenic nodule within the testis and increased vascular perfusion (Figs. 1 and 2). Laboratory findings of AFP, β-hCG were within normal ranges, except for patient C who has increased level. Chest X-ray was normal in all cases.

Patient A was treated by unilateral orchidectomy and controlateral tumorectomy. Patients B and C underwent tumor enucleation and frozen sections were analyzed during surgery (Fig. 3). There were no complications during or after surgery.

In all patients the diagnosis of LCT was confirmed by the definitive pathological report. Histopathological examination revealed

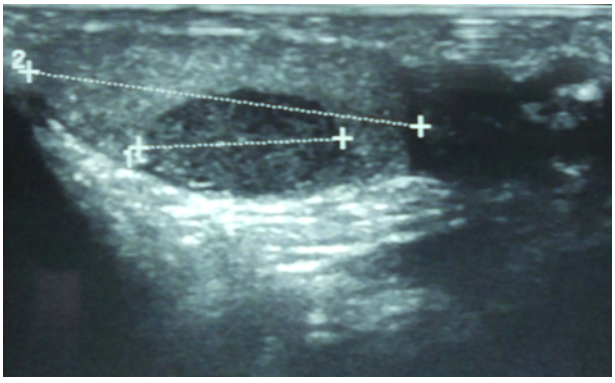

Figure 1 Ultrasound showed a homogeneous, hypoechogenic nodule within the left testis.

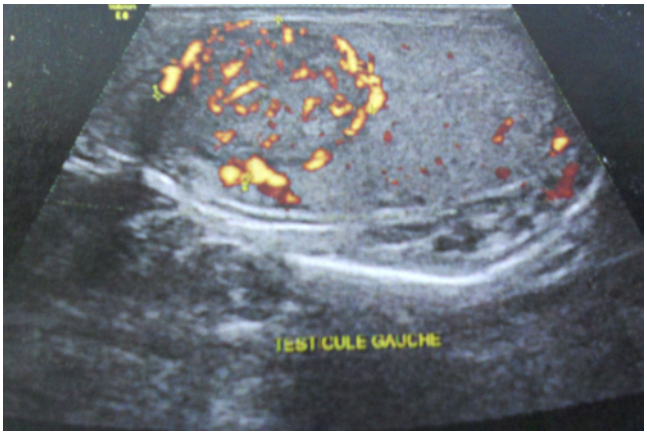

Figure 2 Doppler sonography showing high vascularity of Leydig cell tumor with a prominent peripheral blood flow.

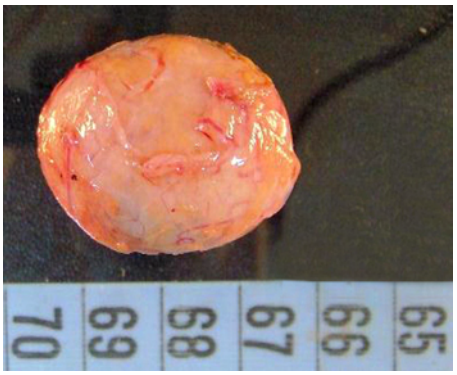

Figure 3 Macroscopic appearance of testicular Leydig cell tumor is brown and well demarcated mass.

cells which had a round nucleus, often with a moderate sized central nucleolus, exhibiting moderate variation in size and an abundant eosinophilic cytoplasm. Numerous rod-shaped intracytoplasmic crystals, Reinke's crystalloids, were observed.

An exceptional mitotic figure was present. Necrosis and hemorrhage was not seen (Fig. 4). Inhibin-alpha and vimentine were positive. The mean follow-up was 40 months (10–56 months). No patient died from the disease during the follow-up and none developed local nor distant recurrence.

Discussion

Leydig cell tumors of the testis are rare and benign in most cases [5]. Symptoms at presentation are rarely suspicious for the presence of LCT of the testis. The presenting features are enlarged testis, gynecomastia and sexual dysfunction. In prepubertal cases, presenting manifestations are usually those of isosexual precocity. Asymptomatic tumors may be detected incidentally in testicular sonography [6].

Hormonal activity is found in 20% of the cases, including increased estradiol and decreased testosterone serum levels, leading to adult feminization and children masculinization, respectively.

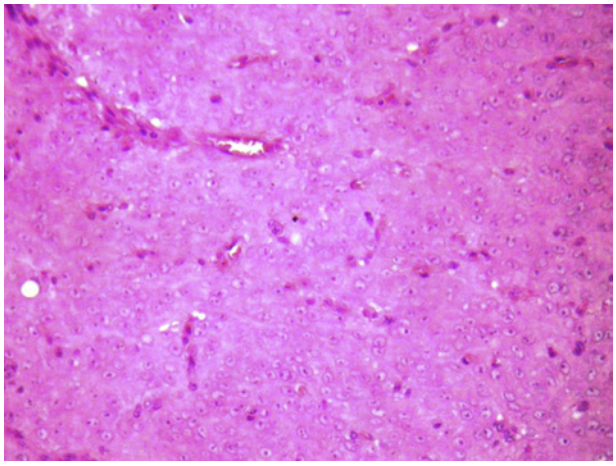

Figure 4 Histologic section shows cells with round nucleus and abundant eosinophilic cystoplasm.

Table 1 Some studies reported in the literature and their follow-up.

First author	Number of cases	Mean age (years)	Recurrence	Follow-up (months)
Carmignani (2007) [5]	22	35 (5–61)	No	41 (1–230)
Giannarini (2007) [9]	17	41.6 (28–55)	No	91 (12–192)
Suardi (2009) [8]	29	–	No	4.6 (0.6–16.2)
Loeser (2009) [7]	8	34 (18–49)	One metachronous in the spermatic cord	41 (1–86)
Bozzini (2013) [4]	22	35 (5–61)	No	180 (77–290)
Our study	3	29 (23–37)	No	40 (10–56)

The endocrinologic manifestation may precede the palpable testis tumors [6].

Radiologically, LCT of the testis typically has a homogeneous hypoechoic sonographic appearance, with peripheral hypervascularization.

To the best of our knowledge, testis sparing surgery was performed for Leydig cell tumors with good results [4]. It allows the preservation of testicular parenchyma and avoids the psychosocial consequences related to orchiectomy [5]. We opted to this conservative approach, as several studies reported in Table 1 and no patient has developed a local or distant recurrence.

The prognosis for benign Leydig cell tumors is excellent. The clinical and hormonal manifestations remit after orchidectomy or tumorectomy in 90% of the cases. But, the persistence of virilizing and feminizing features is not necessarily an indication of malignancy and these features may be to some extent irreversible [6].

Conclusion

Although many teams prefer total orchidectomy because of the diagnostic difficulty associated with malignant forms, simple tumorectomy should become the first-line treatment, provided it is subsequently followed by close surveillance, as it preserves maximum fertility, and these tumors usually have a favorable prognosis.

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