Urology Case Reports 35 (2021) 101520

Contents lists available at ScienceDirect

Urology Case Reports

journal homepage: http://www.elsevier.com/locate/eucr

Oncology

Testicular leydig cell tumor revealed by hydrocele

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ARTICLE INFO

Keywords:
Leydig cell tumor
Testicular tumor
Particularities

ABSTRACT

Leydig cell tumor (LCT) is a rare testicular tumor with a low incidence accounting 3% of all testicule neoplasms. Due to its rarity, the natural history of LCT is poorly understood. Patients can present with atypical symptoms and endocrine disorders. The diagnosis of LCT is based on histological and immunohistochemical examination. We report a new case of leydig cell tumor in a 61-year-old man presenting with a left testis hydrocele. The patient underwent a left orchidectomy and the diagnosis of LCT was established. Even in front of a benign pathology such as hydrocele, exploration is necessary to detect any testicle tumor.

Introduction

Leydig cell tumor (LCT) is a rare non-germ testicular tumor with a low incidence accounting 3% of all testicule neoplasms.[1] Although most behave benignly, 10% are malignant with metastatic forms.[2] The common clinical presentation is a painless testicular mass with or without endocrine manifestations. Due to the difficulties in diagnosis, some clinicopathological features have been described as putative signs of malignancy. Surgical resection remains the standard treatment of both benign and malignant LCT. We describe here a new case of LCT of the left testis occurring in a 61-year-old male. The aim of this report was to describe the characteristics of LCT with a review of literature.

Case report

In Juin 2020, a 61 year-old man, presented to the department of the urology with a painless swelling of the left testicle. On physical examination, the left testis was increased in size, indurated especially in its upper pole with an irregular surface and a hydrocele of great abundance. The patient has also unilateral gynecomastia. The routine laboratory data including complete blood cell count, renal function and liver function tests were negative. The serum germ cell tumor markers such as alphafeotoprotein (AFP) and β human chorionic gonadotropin (β -HCG) were normal. Oestrogen and oestradiol levels were increased. Scrotal ultrasonography showed an increased hypoechogenic left testicle mesuring 90 mm. The patient underwent left orchidectomy. Pathological study showed a well circumscribed intratesticular mass mesuring 7*4*5 cm (Fig. 1,a). Microscopically, the tumor cells were hexagonal with abundant eosinophilic cytoplasm containing Reinke crystalloids. High mitotic figures, nuclear atypia and necrosis were observed. The adjacent testicular tissu, spermatic cord and surgical margin were free of malignancy (Fig. 1b and c,d). At immunohistochemical examination, the tumor cells were positive for inhibin, calcretine, and MelanA. Therefore, the diagnosis of a Lydig cell tumor with histological signs of malignancy was established. A body computed tomography (CT) was normal. The patient was discharged. Two months after surgery, we noted an improvement of gynecomastia with normalization of the oestrogen and oestradiol levels with complete remission.

Discussion

Testicular tumors are extremely rare, representing 1–1.5% of all tumors in men.[3] There are two types of primary testicule tumor: germ cell tumors and sex/cord stromal tumors.[4] Among the sex/cord stromal tumors, leydig cell tumor (LCT) is the most common histological subtype, accounting for approximately 3% of all testicular neoplasms.[1] It has been first described by Sacci in 1895. An increase in the incidence of Leydig cell tumors has been observed over the last decades because of better imaging techniques.[1] There are 2 age peaks in the incidence of LCT: it is more frequent in adults between the third and the sixth decade, such as our patient, but it can occurs also in child aged between 3 and 9 years.[3] Children may suffer from precocious pseudopuberty. However,

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https://doi.org/10.1016/j.eucr.2020.101520
Received 22 November 2020; Accepted 1 December 2020
Available online 5 December 2020

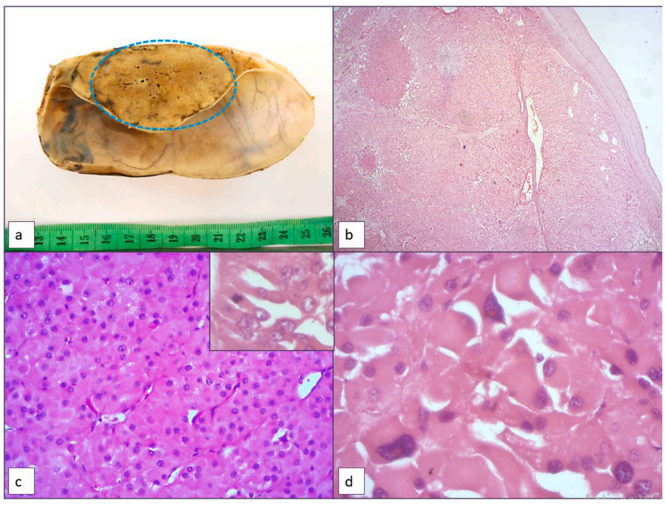

Fig. 1. a: Gross appearance: a solid lobulated intra testicular tumour (blue circle) with yellow-tan cut surface surrounded by a hydrocele.
b, c, d: Histologic features: (b) a sheet-like growth pattern Leydig cell-tumour with scattered necrotic areas, HE x 40 (c) large polygonal monomorphic tumour cells with abundant eosinophilic cytoplasm, round nuclei, conspicuous nucleoli, HE x 100 [inset: a mitotic figure, HE x 400], (d) and focal nuclear atypia, HE x 400. (For interpretation of the references to colour in this figure legend, the reader is referred to the Web version of this article.)

adults can present with atypical symptoms of testicular masses and endocrine disorders or stay completely asymptomatic and discovered incidentally by ultra-sound.[3] Gynecomastia is an infrequent symptom; its prevalence is less than10%.[2] In the current report, the patient presented a gynecomastia on examination but the proeminent symptom leading him to consult was the painless swelling of the testis which was related to hydrocele. For this reason, even in front of a benign pathology such as hydrocele, exploration is necessary to detect any testicle tumor. In up to 80% of cases, hormonal abnormalities with high oestrogen and oestradiol levels, low testosterone, and increased levels of LH and FSH are reported.[5] In our case, AFP, HCG, LDH were normal which concord with the pathological study. The diagnosis of LCT is based on histological and immunohistochemical findings. Histologically, LCT is defined by the proliferation of large polygonal tumor cells with granular eosinophilic cytoplasms and prominent nucleoli arranged in sheets pattern. Immunologically LCT is associated with a positive expression for inhibin and Melan-A, but the expression of calcretinin and vimentin varies. Distinction between benign and malignant forms, is very difficult. Although metastasis is the only reliable criterion of malignancy, the authors identified clinicopathological features as presumptive signs of malignancy, including: Large size (>5cm), old age [more than 40 year], mitotic activity (>3 per 10 high-power fields), vascular invasion, cytologic atypia, necrosis, infiltrating edges, extratesticular extension, and aneuploidy.[4] Those criteria were also significantly associated with metastatic behavior in Leydig cell tumors. In our case report, 5 features were consistent with the above criteria: patient's age, tumor size, cytologic atypia, high mitotic activity and the presence of necrosis. The standard treatment for LCT is radical orchidectomy.[3] In our case report, due to the negative resection margins and absence of distant metastases, no added management was necessary. Survival is poor in metastatic LCT, with a low reponse for systemic chemotherapy or radiation.[2]

Conclusion

Clinicians should be aware about the necessity of testicles examination in case of gynecomastia. Even in front of a benign pathology such as hydrocele, exploration is necessary to detect any testicle tumor.

Declaration of competing interest

None.

References

1. Gheorghisan-Galateanu AA. Leydig cell tumors of the testis: a case report. *BMC Res Notes*. 2014;7:656.
2. Muheilan MM, Shomaf M, Tarawneh E, Murshidi MM, Al-Sayyed MR, Murshidi MM. Leydig cell tumor in grey zone: a case report. *International Journal of Surgery Case Reports*. 2017;35:12–16.
3. Zhu J, Luan Y, Li H. Management of testicular Leydig cell tumor: a case report. *Medicine (Baltimore)*. 2018;97(25):e11158.
4. Tahaineh S, Mughli RA, Fallatah M. Giant mixed Sertoli-Leydig- Granulosa sex cord tumor of the testis; clinical, histopathological, and radiological features: a case report. *Pan Afr Med J*. 2017;27:51.
5. Vukina J, Chism DD, Sharpless JL, et al. Metachronous bilateral testicular Leydig-like tumors leading to the diagnosis of congenital adrenal hyperplasia (adrenogenital syndrome). *Case Rep Pathol*. 2015;2015:459318.