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Granulosa Cell Tumor of the Testis: A Case Report of a Very Rare Tumor Treated with Sparing Surgery and Revision of the Literature

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Abstract

Introduction: The granulosa cell tumors (GCTs) belong to the sex cord stromal tumors of the gonads and they can affect both, the testis and the ovaries. GCTs of the testis arise from the epithelial elements of the sex cord and they are divided into 2 different groups: the juvenile type and the adult type. The adult type is a very rare tumor and to date only few cases have been described. We present a very rare case of a patient with an adult type of GCTs of the testis treated with sparing surgery.

Case study: A 32-years-old patient was admitted to our department with suspicion of tumor in the left testis. Alpha-FP, Beta-HCG and LDH were normal. The lesion was small, intra-parenchymal and easy to remove. Frozen sections were taken from the tumor and tumor bed as usual in our department before ablating the organ. The pathologist suspected a GCT, thereafter the surgeon decided to perform a sparing surgery.

Results: The definitive histology confirmed an adult type GCT; the lesion measured 15 mm and didn't show any infiltration. There were no evidence of angio-invasion, necrosis or severe nuclear atypia and very few mitoses. The computed tomography (CT) showed no metastases or enlarged lymph nodes. The patient underwent regular follow up: an ultrasound (US) of the testicles was done 3-6 and 12 months after surgery; after 1 year the patient shows no signs of disease on US and CT.

Conclusion: The adult type GCT is a rare entity; it can be malignant in 20-25% of the patients and occurs at any age after puberty with a limited number of reported cases: in the literature we have found 51 published cases, mostly case report. Testis sparing surgery could be offered by small, intra-parenchymal lesions, if frozen sections are suspicious for a GCT; by angio-invasion, necrosis, infiltrating margins, severe nuclear atypia or elevated mitotic count in the definitive histology a delayed radical orchiectomy can be performed. A personalized follow up is required.

Keywords: Granulosa cell tumors; Adult type; Testicular stromal tumors; Testis sparing surgery; Testicular tumors

Introduction

The granulosa cell tumors (GCTs) belong to the sex cord stromal tumors of the gonads and they can affect both, the testis and the ovaries. GCTs of the testis arise from the epithelial elements of the sex cord and they are divided into 2 different groups: The juvenile type and the adult type. The adult type is a very rare tumor and to date only few cases have been described. While most cases of adult GCTs are benign, some of these tumors show a malignant behavior with metastatic potential even after 10 years [1-2]. Due to the small number of reported cases, the tumor behavior is unpredictable, and the optimal management for the patients is difficult to establish. Here we present a case of a patient with an adult type of GCT of the testis, treated with testis sparing surgery.

Case Study

We describe a case of a 32-year-old man, who was admitted to our department with suspicion for tumor of the left testis. The ultrasonography showed a 1.5 cm hypo-echoic, vascularized lesion in the upper pole of the left testis. The patient had no history of cryptorchidism and his past medical history was not significant, including only a spontaneous pneumothorax, when he was 18-years-old. The patient denied decreased libido or erectile dysfunction in the months before. Alpha-fetoprotein (alfa-FP), human chorionic gonadotropin (HCG) and lactate dehydrogenase (LDH) were normal. The lesion was small, intra-parenchymal and simple to remove due to missing infiltration of the surrounding tissue. Frozen section was taken from the tumor and the tumor bed. The pathologist suspected a GCT and the surgeon decided to perform a testis sparing surgery, waiting for the definitive histology.

Results

The postoperative course was uneventful, and the patient was discharged from the hospital the day after the operation without complications. The definitive histology confirmed the previous report of adult-type GCT; the lesion measured 1.5 cm and did not show any margins infiltration. There was no evidence of angio-invasion or necrosis. The tumor was composed of clusters of cells with scanty cytoplasm in a predominantly micro-follicular pattern; very few mitoses could be seen (Figure 1). The immunohistochemical study showed positivity for calretinin, inhibin, CD-99 and beta-catenin while chromogranin, cytokeratin and melan-A were negative. No distant metastases or enlarged lymph nodes were seen in the computed tomography. The patient underwent regular follow up without any other surgery: an ultrasound of the testicles was done every 3 months for the first year

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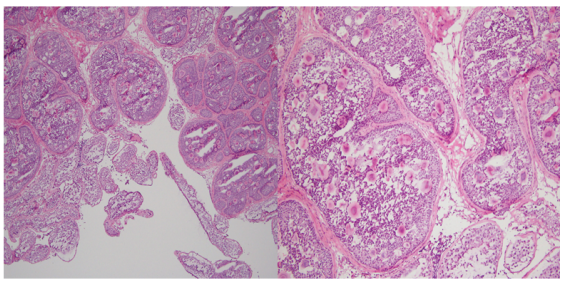

Figure 1: On the left side hematoxylin and eosin staining: In the bottom right corner the seminiferous tubules and above the neoplastic cells. On the right side 10x magnification of the neoplastic cells.

and then every 6 months. An abdominal ultrasound with a chest X-ray were performed 6 months after surgery and after one year a computed tomography, which showed absence of disease. Twenty months after the diagnosis the patient has done well and showed no signs of residual disease on testicular and abdominal ultrasound and on chest X-ray.

Discussion

GCTs of the testis belong to the sex-cord stromal tumors of the gonads, which include also Leydig and Sertoli cell tumor, fibromas and thecomas and represent 4-5% of all testicular tumors [1-3]. They were described for the first time in 1952 by Laskowski [4] in a 35-years-old patient and they can be divided in two distinct groups, the juvenile type and the adult type. Although the juvenile-type represents only 1-4% of pre-pubertal testicular tumors, it is the most common testis neoplasm in the first 6 months of life, it is typically benign, and a simple orchiectomy is enough for cure [2]. The adult type is instead extremely rare with only a limited number of reported cases to date: after a review of the literature we found only 52 published cases, mostly isolated as case reports [1-9]. It can occur at any age with a range from 12 to 83 [5-10] years in previous reports. In more than half of patients the clinical presentation is a painless and slow-growing testicular mass. Gynecomastia, erectile dysfunction and decreased libido may be present in 20-25% of the cases, due to hormonal or chromosomal abnormalities [8]. Although the clinical behavior is most commonly benign, approximately 20% of the adult type have been reported to be malignant with a metastatic potential even after 10 years [11] and poor prognosis. There are 5 reported patients with metastases at presentation [3-13] while almost 4 patients developed metastases between 12 and 121 months after diagnosis [11-15]. The retroperitoneal lymph nodes are the most common metastatic regions, but liver, lung and bone metastases have been described. The patients with retroperitoneal lymph node metastases show to have a relatively longer survival, if compared with patients with distant or multiple metastases [16]. Because of his low incidence and the lack of data, is difficult to establish the right management for these tumors: the identifications of some criteria to discriminate, which tumor can have a malignant behavior, can help to decide the type of surgical treatment and avoid an orchiectomy in patients with benign lesions. Previous studies have demonstrated that some histopathological findings can predict malignancy for Leydig cell tumors and that they can be used for all stromal tumors; tumor size >5 cm, positive margins, lymphovascular invasion, necrosis, cellular atypia and an increased number of mitoses per high-powered field (>3). Patients with 0 or 1 risk factors have a very low risk to develop disease recurrence or metastases [3-17]. Testis-sparing surgery could be offered in selected cases with small, intra-parenchymal lesions, if frozen sections are suspicious for a GCT while waiting for the definitive histologic diagnosis. Only in presence of 2 or more risk factors a delayed radical orchiectomy could be performed with retroperitoneal lymphadenectomy with suspicion for metastases. Our experience confirms the previous findings: in our patient no risk factors were found and 20 months after sparing surgery he shows no relapse or distant metastases. However, a personalized follow up is required: in a patient with 0 or 1 risk factor an ultrasound of the abdomen and testis can be sufficient every six months; in high risk patient it is suggested to perform US of the abdomen and testis in combination with a chest X-ray, alternated with CT of the abdomen and pelvis and US of the testis every 6 months, for more than 10 years as distant metastases can occur years after diagnosis [5-16].

Conclusion

The adult-type GCT is a rare entity, which can be malignant in approximately 20% of the patients. The reported case showed, that testis sparing surgery is safe in very selected cases, if frozen sections are available and if the definitive histology shows no risk factors. By presence of 2 or more risk factors more aggressive personalized treatments with surgery and eventually chemotherapy and radiotherapy are recommended.

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