Dear Editor,

Sertoli cell tumors (SCTs) account for less than 1% of all testicular neoplasms and are included in the group of sex cord tumors together with Leydig cell, granulosa, and mixed tumors.

We report the case of a 69-year-old male patient with an unremarkable history who attended our office reporting intermittent pain in the left testis for the past several months, but no voiding symptoms. Localized pain was reported in the upper pole of left testis, in which no irregularities, masses or abnormal swellings were palpated. An ultrasound examination showed a hypoechogenic, homogeneous lesion, 1.5 cm × 1 cm in size, located in the upper pole of left testis and surrounded by a thin capsule. The lesion was not related to epididymis or other testicular structures (an epidermoid cyst was suspected). Both the rest of the testis and the contralateral testis were normal. Tumor markers (AFP, ß-hCG, and LDH) were negative. Based on clinical and radiographic data, tumor resection was performed with intraoperative biopsy, based on which a lesion of benign characteristics with a totally normal adjacent pulp was reported. Conservative surgery was therefore decided. The final pathological study reported a clearly outlined, non-encapsulated tumor surrounded by seminiferous tubules with complete spermatogenesis. The specimen showed a diffuse growth pattern consisting of solid nests (fig. 1) with tubular and retiform structures. No cell atypia or mitosis were seen. Stroma was markedly hyalinized, and tumor cells were immunoreactive to vimentin, CD 99, and synaptophysin (fig. 2).
SCTs are part of the group of sex cord tumors together with Leydig cell, granulosa, and mixed tumors, and are considered an uncommon disease because they account for only 1% of all cases diagnosed. They may occur at any age, but a majority of them are found at 15-80 years of age, with a mean of 45 years.

Tumors are usually asymptomatic, with negative markers, and most often appear as a chance finding in an imaging study done for other reasons. However, although they are not as metabolically active as Leydig tumors, they may cause gynecomastia or sexual precocity related to excess hormone production in approximately 20%-30% of patients. Unlike germ cell tumors, they are not related to cryptorchism, but are usually associated to syndromes such as the Peutz Jeguers or Carney syndromes. In the reported case, testicular pain occurred in a 69-year-old man with no signs or symptoms of excess hormone production and not associated to other concomitant diseases.

Study was completed with a CT scan of the abdomen and pelvis, which was completely normal, thus ruling out potential retroperitoneal adenopathies.

After 12 months of follow-up, the patient is currently asymptomatic, with fully normal markers, testicular ultrasound, and physical examination.

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Three histopathological subtypes may be distinguished: classical SCT, SCT of calcified giant cells, and sclerosing SCT.

The classical variant is the most common, and has a highly variable range of pathological characteristics. It has a tubular pattern, with cells of pale or eosinophilic wide cytoplasm, vesicular nucleus with nucleus and hyalinized stroma. In complicated cases, histochemistry is performed, which should be positive for cytokeratin and inhibit and negative for PLAP.

The SCT of calcified giant cells is another variant occurring at an earlier age. It mainly affects children and young adults aged approximately 20 years, and shows a solid growth pattern, polygonal cells, prominent nucleolus, and calcifications.

The sclerosing variant is considered extremely uncommon, and only 14 cases have been reported in medical literature to date. This tumor shows small tubes in a sclerous stroma, and is usually negative or weakly positive for cytokeratin.

The vast majority of SCTs are usually benign in nature. However, 10%-22% have a certain ability to metastatize (less than 50 cases reported) which is somewhat related to tumor subtype. Metastatic disease may be found in 15%-20% of SCTs of calcified giant cells and in 5%-10% of those of the classical type. No clear data are available on the dissemination capacity of sclerous SCTs.

No unified or standardized malignancy criteria exist currently, but some histopathological changes may be of great guidance, such as moderate or severe cell atypia, a diameter larger than 4-5 cm, the presence of more than 5 mitoses per every 10 fields, or necrosis and vascular-lymphatic invasion. The only unequivocal criterion continues to be the presence of metastasis (mean 2-year survival in these cases).

There are cases reported in medical literature of SCTs with positive neuroendocrine markers, like the one found in our patient. This makes pathological study difficult and requires that existence of a testicular carcinoid tumor, among others, is ruled out. The clinical implications of this incidental finding are currently unknown.

Standard treatment of these lesions is inguinal orchiectomy, associated to retroperitoneal lymphadenectomy.
if node enlargement is shown in malignant cases, as well as systemic polychemotherapy and radiation therapy (usually with an ominous prognosis). There are cases reported in medical literature where conservative surgery has been performed (tumor resection) in bilateral cases, prepubertal males, or clearly benign cases such as our patient. Long-term follow-up is required because metastases have been reported even 15 years after treatment. Because of the unusual nature of this condition, there is no established pattern to be followed, but regular tests of tumor markers (AFP, β-hCG, LDH) and hormones (estrogens, progesterone, and testosterone), physical examination, and CT scans of the chest and abdomen are recommended.

The low incidence and relative lack of understanding of the long-term course of this type of tumor, as well as the exceptional finding of neurosecretion characteristics in our tumor and the few references in medical literature in this regard, raise the doubt as to whether this could be a clinically significant finding or a mere chance finding.

REFERENCES


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