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Sclerosing variant of well-differentiated testicular liposarcoma: a case report

Lipossarcoma testicular bem diferenciado com variante esclerosante: um relato de caso

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ABSTRACT

Testicular cancers are classified in germ cell and non-germ cell tumors, as well as, liposarcomas. We report the case of a patient with a large testicular liposarcoma, submitted to surgical treatment with excision of scrotal pouch and segment of the spermatic cord, and the left testicle, showing a good evolution. This report presents one of the first cases of a sclerosing variant of well-differentiated testicular liposarcoma, large in size and with no association with another cancer. Due to their location, the diagnosis is difficult and unusual. Complete tumor resection and regular medical follow-up show a good prognosis, less recurrence, and little cellular differentiation.

Key words: testicular cancer; liposarcomas; sarcomas; testicular liposarcoma.

RESUMO

Os tumores testiculares são classificados em células germinativas e não germinativas, assim como os lipossarcomas. Relatamos o caso de um paciente com lipossarcoma testicular de grande dimensão submetido a tratamentos cirúrgicos com ressecção de bolsa escrotal e segmento do cordão espermático e do testículo esquerdo, apresentando boa evolução do quadro. O relato traz um dos primeiros casos de lipossarcoma testicular bem diferenciado com variante esclerosante, de grande dimensão e sem associação a outra neoplasia. Devido à localização, apresenta diagnóstico difícil e pouco habitual. Com a ressecção total do tumor e o acompanhamento médico regular, o paciente apresenta bom prognóstico, menor recidiva e pouca diferenciação celular.

Unitermos: *tumor testicular; lipossarcomas; sarcomas; lipossarcoma testicular.*

RESUMEN

Los cánceres de testículo se clasifican en tumores de células germinales y células no germinales, así como en liposarcomas. Presentamos el caso de un paciente con un gran liposarcoma testicular, sometido a tratamiento quirúrgico con exéresis de la bolsa escrotal y segmento de cordón espermático y testículo izquierdo, con buena evolución. Este informe presenta uno de los primeros casos de una variante esclerosante de liposarcoma testicular bien diferenciado, de gran tamaño y sin asociación con otro cáncer. Debido a su ubicación, el diagnóstico es difícil e inusual. La resección completa del tumor y el seguimiento médico regular muestran un buen pronóstico, menor recidiva, y poca diferenciación celular.

Palabras clave: *cáncer de testículo; liposarcomas; sarcomas; liposarcoma testicular.*

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INTRODUCTION

Affecting 1% of the world's male population, testicular cancer is the most common solid malignancy among men aged 15 to 35 years[1,2]. The incidence peak covers young adults in the second and third decades of life, while elderly people, over 65 years, represent a percentage of approximately 4%[3]. It has a lower incidence in Asia and Africa populations, and is more prevalent in developed countries, such as Norway, Denmark, and Switzerland[4].

As for races, in the United States, for example, whites are more affected than blacks[1]. Cryptorchidism, hypospadias, and first-degree family history are some of the risk factors for the development of testicular tumors[1]. Among these, germ cell tumors are the most common, representing 95% of cases[5]. They are classified into seminoma and non-seminoma[5] and mainly affect young people, while lymphomas prevail in the elderly[3,5]. The remainder comprises non-germ cell tumors, such as stroma/sexual cord tumors, paratesticular tumor[2], and liposarcoma – the latter with scarce reports in the literature.

Liposarcomas are malignant tumors of adipose tissue, with slow and painless expansion; they correspond between 15% and 20% of all soft tissue sarcomas[5]. They appear mainly in the retroperitoneum[6], and are uncommon in the genital region[5,7-10]. They are classified as well differentiated/undifferentiated, myxoid/round and pleomorphic cells[11].

The well-differentiated liposarcoma is classified into subtypes: lipomatous, sclerosing, and inflammatory[6] and is associated with the amplification of the chromosome segment 12q13-15, which contains the oncogenes MDM2 and CDK4[6,11]. It appears as a slow-growing mass[5,12], often between the fifth and seventh decades of life[12]. Despite having little or no metastatic potential, it has a tendency to recurrence[5,6,13] and can progress to dedifferentiated liposarcoma[11,13].

CASE REPORT

Male patient, white, 74 years old, smoker (192 packs/year), alcoholic in abstinence. In 2012, he sought medical attention referring to the appearance of a "hardened mass in the left testicle". He reported that the growth was progressive, slow, painless, and with no previous history of trauma.

After an imaging exam suggestive of malignancy, the patient underwent surgery with removal of the tumor, which measured 14 × 10 cm in its widest portions and weighed 542 grams.

When cutting, we observed a yellowish, shiny tissue, with pinkish-red areas of elastic consistency. We also observed four fragments similar to the main tumor, making it impossible to assess the surgical margin. The histological feature was suggestive of a well-differentiated liposarcoma with sclerosing areas, corresponding to grade 1 sarcoma.

After a surgical procedure, follow-up appointments remained irregular for seven years. After this period, the patient returned to medical care complaining of a new "hardened mass" growth in the left testicle.

After the reassessment, we opted again for surgical treatment, with surgical excision of the scrotum, containing the segment of the spermatic cord and the left testicle (**Figure 1**). The testicle measured 25 × 17 × 15 cm, and the surgical specimen weighed 3.572 grams. The tumor lesion presented a yellowish-gray color, shiny, with hemorrhagic points, occupying the organ almost completely (**Figure 2**).

In light microscopy, we observed a neoplasm composed of spindle cells, with myxoid areas, cell atypia, low mitotic activity, foci of necrosis, and some areas with intermingling adipocytes (**Figure 3**). In the immunohistochemistry panel, the tumor markers were positive for S100, desmin, and CD34; Ki-67 showed low cell proliferation. Immunohistochemistry associated with histology was again compatible with sclerosing variant of well-differentiated liposarcoma.

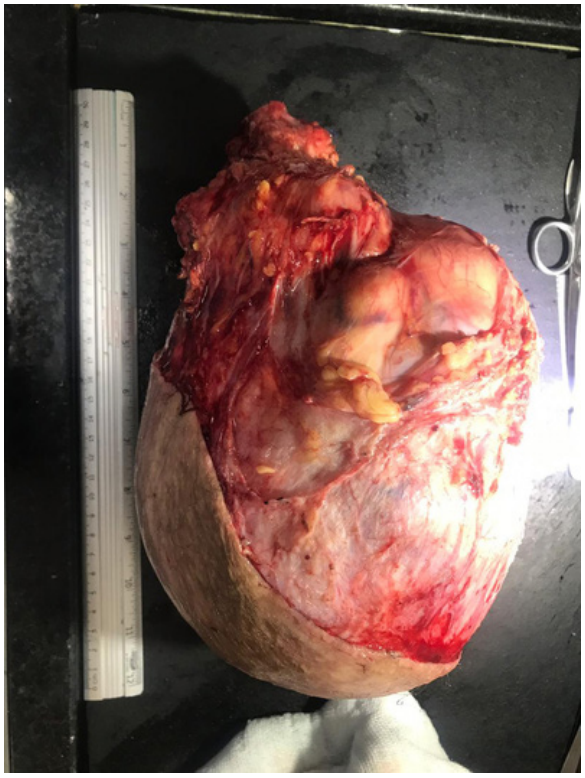

FIGURE 1 – *Surgical specimen containing scrotum and left testicle*

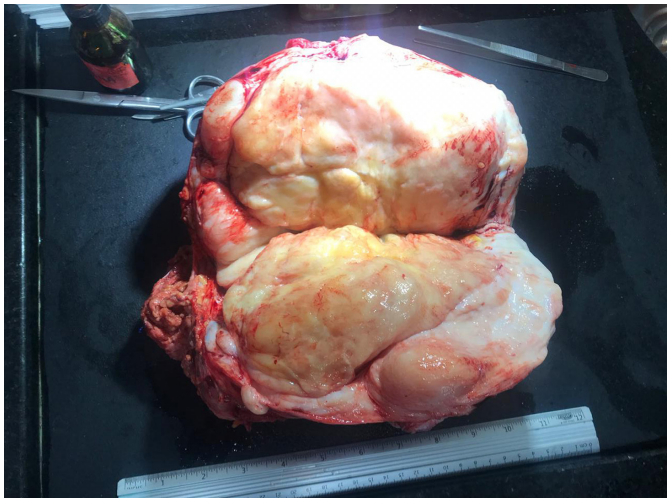

FIGURE 2 – *Surgical resection of the tumoral lesion*

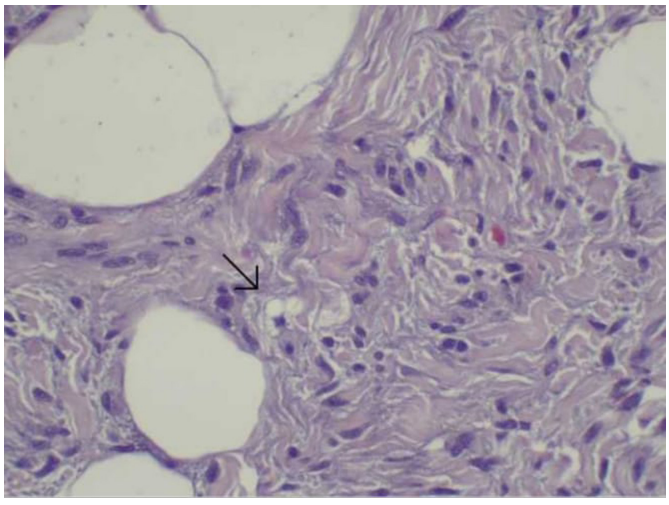

FIGURE 3 – *Optical microscopy of the tumoral lesion*
The arrow indicates an atypical adipocyte/adipoblast.

Currently, the patient remains with no clinical complaint and under regular medical monitoring.

DISCUSSION

Well-differentiated liposarcoma is a disease that is difficult to diagnose, since it has characteristics similar to lipoma, inguinal hernia, cysts, and hydrocele[7-9]. The presence of two factors corroborates a greater risk of recurrence: positive microscopic margins after resection, and sclerosing variant[14]. This is ratified in the case presented; the recurrence probably occurred due to possible incomplete excision of the lesion, suggested by the absence of free margins in the description performed, and to the histological subtype.

Regarding treatment, well-differentiated liposarcoma presents low sensitivity to chemotherapy[11]. For neoplasms with sclerosing areas, extensive resection of the lesion with preservation of function and free margins of 1 cm are the management of choice[14]. If this procedure is possible to be performed, radiotherapy will be unnecessary[14]. As in the case described, well-differentiated liposarcoma has a good prognosis[5], however, there is a high probability of recurrence and the possibility of progressing to the dedifferentiated subtype[5, 15], which requires regular monitoring.

According to extensive research, this is one of the first case reports on liposarcoma testicular, which still outstands because it was not associated with another neoplasia[16], and because of its large size. Bearing in mind that liposarcoma of the spermatic cord and paratesticular are atypical, with few case reports as most publications on the subject[7-10, 17], testicular liposarcoma is even more exceptional given the scarcity of literature.

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