CASE REPORT

Spermatic cord mixed liposarcoma. Case report and review of the literature

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Abstract

A 75-years-old man presented with a clinical diagnosis of hydrocele. Grossly examination revealed a large (14×8×9 cm) tumor. Histologically, the tumor had a mixed pattern, with major pleomorphic and a minor sclerosing well-differentiated component. The anatomo-clinical features, the prognosis and the principles of treatment of this rare condition are shortly discussed.

***Keywords*:** spermatic cord, liposarcoma, mixed.

Introduction

Spermatic cord liposarcoma are very uncommon tumors [1, 2], only 161 cases having been described in the literature until 2005 [3]. Patients usually present with a painless, slowly growing bulge in the inguinal and scrotal region, which is clinically diagnosed as a hernia, the correct diagnosis being made postoperatively [2, 4–10].

In this paper, we report a case of spermatic cord liposarcoma with a peculiar clinical presentation and an unusual histological pattern – a mixed type liposarcoma with a major pleomorphic and a minor sclerosing well differentiated component. This histological type was not reported, to the best of our knowledge, in this localization.

Patient and methods

The patient, a 75-years-old man, presented with a massive enlargement of the scrotal sac, which was interpreted both clinically and echographically as a hydrocele.

Intraoperative examination confirmed the mass involving the spermatic cord and extending to the inguinal region.

Histopathology revealed a liposarcoma and management was extended to radical inguinal orchiectomy, funiculectomy and epididimectomy. Several pieces of the resected tumor were fixed in 10% formalin, embedded in paraffin, cut at 5 µm, and stained with Hematoxylin–Eosin and immunohisto-chemically for S–100 protein.

Results

Grossly examination revealed a large (14×8×9 cm), well-circumscribed, polilobated tumor. The cut surface was yellow with white and gelatinous speckles.

Microscopic examination revealed a liposarcoma with a mixed histologic pattern. The tumor was predominantly (>80%) a pleomorphic liposarcoma with a minor zone of sclerosing well-differentiated liposarcoma. The pleomorphic liposarcoma was composed of sheets of large lipoblastic cells with single or multiple bizarre scalloped nuclei (Figure 1). Nuclei, which were peripherally or haphazardly arranged either, were surrounded by a vacuolated or eosinophylic cytoplasm. Some cells contained "usual" vacuoles (Figure 2), while in other cells there were tiny vacuoles (Figure 3). Occasionally the two types of vacuoles were found in the same cell (Figure 4). A few cells contained small granules into the cytoplasm (Figure 5).

S–100 protein was focally positive. The mitotic activity was moderate (5 mitoses/10 HPF). Necrotic areas represented 15% of the tumor surface. The sclerosing well-differentiated component merged with the former component. It was characterized by the presence of fibrous septa containing atypical cells with large irregular hyperchromatic nuclei, which separated adipose lobules (Figures 6–8).

Discussions

Spermatic cord liposarcoma usually appear in old men [1, 11], although exceptional cases may involve young boys [12].

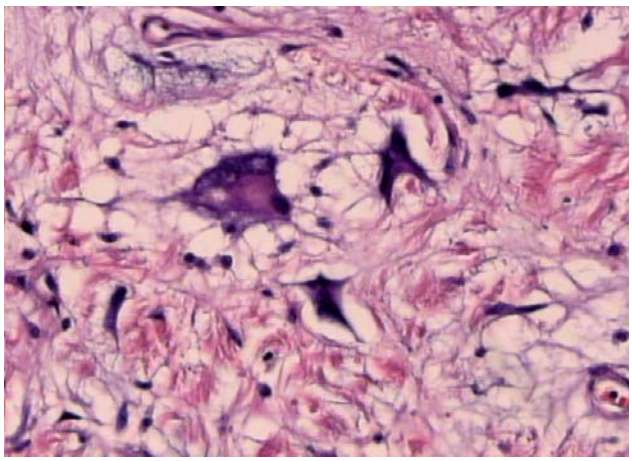

Figure 1 – ***Area of pleomorphic liposarcoma showing many atypical lipoblasts with large scalloped hyperchromatic nuclei (HE stain, ×50)***

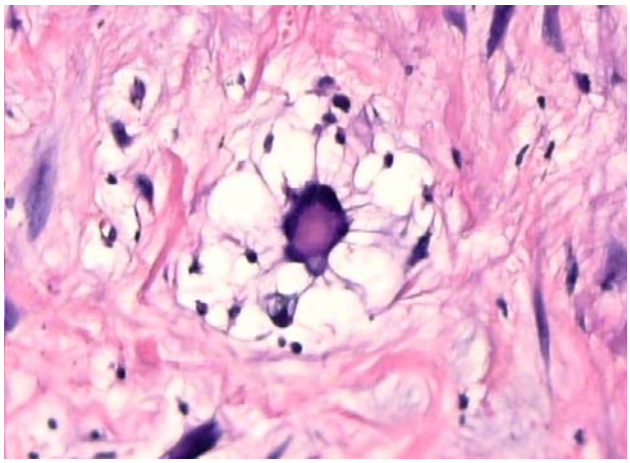

Figure 2 – ***View of a "classical" lipoblast with large cytoplasmatic vacuoles indenting the nucleus (HE stain, ×100)***

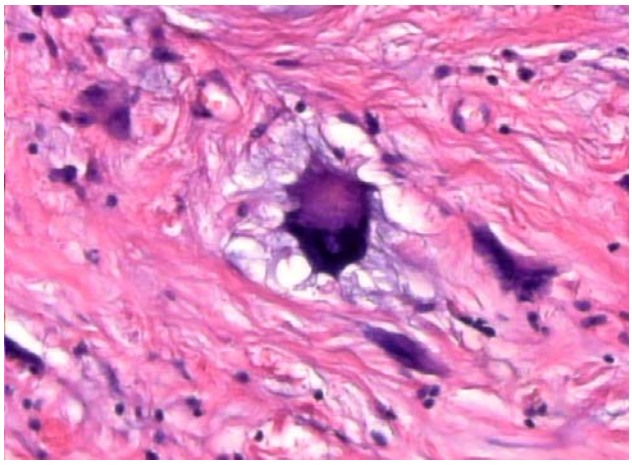

Figure 3 – ***Lipoblast – microvesicular aspect of the cytoplasm (HE stain, ×50)***

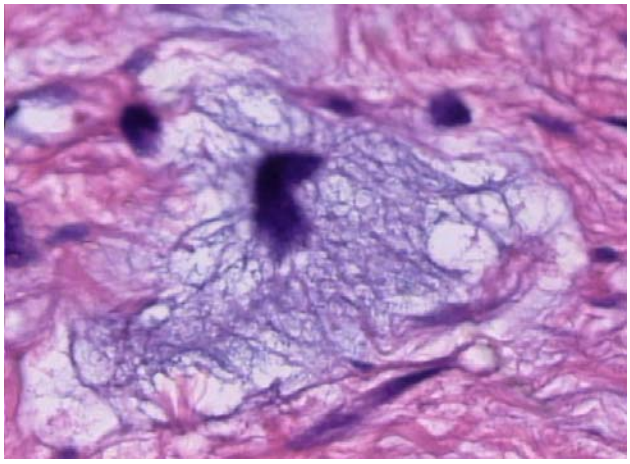

Figure 4 – ***High-power view of a large lipoblast showing a mixture of macro and microvacuoles filling the cytoplasm (HE stain, ×100)***

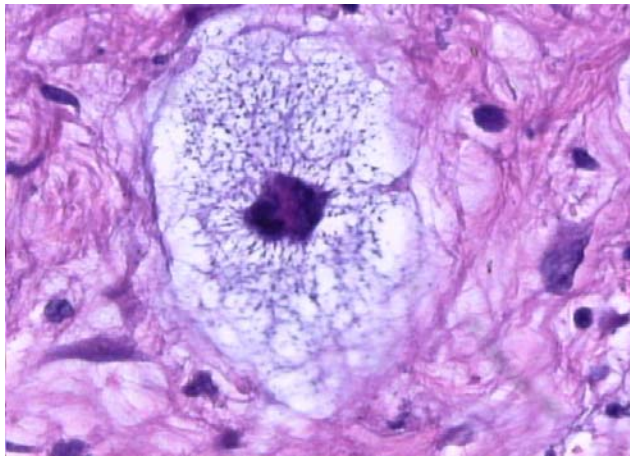

Figure 5 – ***Large lipoblast exhibiting numerous tiny cytoplasmatic granules (HE stain, ×100)***

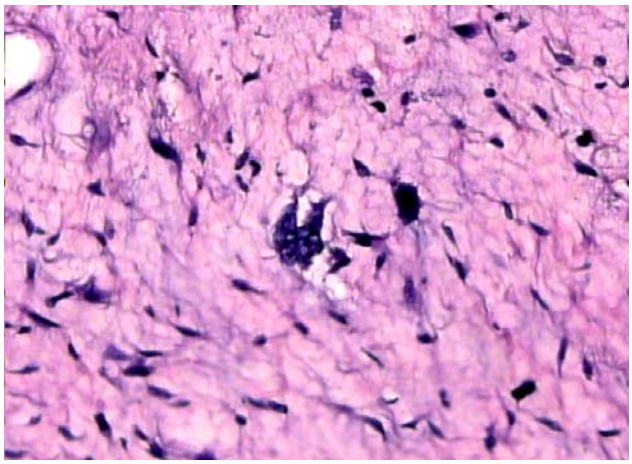

Figure 6 – ***A dense fibrous area containing lipoblastic cells (HE stain, ×50)***

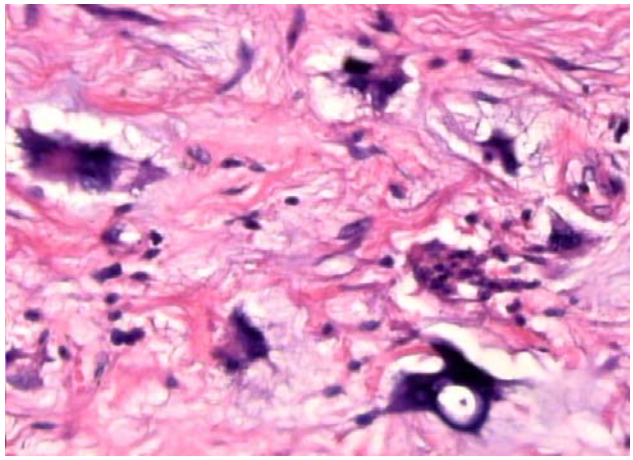

Figure 7 – ***View of the sclerosing component of the tumor. Atypical lipoblasts are embedded in a hyaline stroma. Some mitotic figures are seen (HE stain, ×50)***

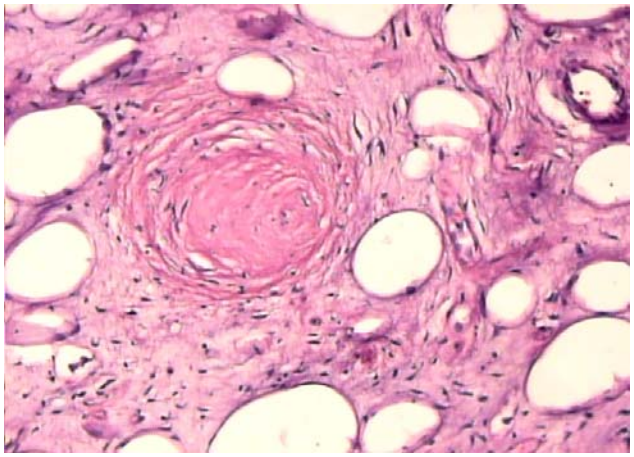

Figure 8 – ***Low-power view of the tumor showing adipose lobules surrounded by a fibrous stroma with glomeruloid-like features (HE stain, ×25)***

Grossly, the tumors are usually large (mean 12 cm), lobulated, yellow. These features were recapitulated by the tumor that we have studied. Several cases of giant tumors have also been reported [1, 13, 14]. Histologically, most cases are well differentiated, some cases belonging to the sclerosing or inflammatory type [11, 15–22].

Some well-differentiated liposarcoma comprised foci of myxoid liposarcoma [7, 23]. Myxoid and myxoid/round cell liposarcoma [1, 14, 24, 25] and dedifferentiated liposarcoma [11, 18] are rare types.

Peyrí Rey E *et al*. [26] reported a dedifferentiated liposarcoma which apparently developed by degeneration of an earlier resected lipoma.

Pleomorphic liposarcomas are an uncommon type of liposarcoma, which infrequently involve the spermatic cord [27–29].

The case reported by Hagiwara N *et al.* [30] recurred six years later showing areas of cartilaginous metaplasia. In the case, that we have studied – a mixed type liposarcoma – the pleomorphic areas represented approximately 80% of the tumor surface. Pleomorphic liposarcoma have a high local recurrence and metastatic rate [28] and an extensive pleomorphic component certainly influences the prognosis. This assumption is further supported by immunohistochemical studies and microarray approach, which demonstrate a differential expression of multiple genes in the well-differentiated and pleomorphic components of a mixed liposarcoma. It has been suggested that these genes are associated with the differences in the morphological characteristics and pathogenesis of mixed type liposarcoma [31].

The most difficult differential diagnosis of mixed liposarcoma is the dedifferentiated liposarcoma [32]. According to WHO [33], dedifferentiation is characterized by the emergence of a non-lipogenic component of variable histological grade within an atypical lipomatous tumor/well differentiated liposarcoma, a situation clearly different from our case. Most liposarcoma of the spermatic cord have a low level of malignancy, a prolonged course and an overall good prognosis. There is, however, a high rate of, sometimes late, recurrences so that a periodical follow-up is necessary [3, 18]. Relapses, when occur tend to be localized [34]. In some cases, the recurrence of a well-differentiated liposarcoma displayed foci of high-grade dedifferentiation [11]. Metastases, when present, occur through hematological route (especially lungs). Lymphatic spread is unusual [11, 34]. Spermatic cord liposarcoma are treated by radical orchiectomy [17, 34–38]. Radiation therapy is recommended in addition to surgery in high-grade tumors, lymphatic invasion, inadequate margins, or relapses [8, 20, 39]. The role of chemotherapy, still discussed, is reserved for high-grade tumors [1, 3, 26, 35, 38].

Conclusions

Spermatic cord liposarcoma are very uncommon tumors, mostly encountered in old men. The case we are exhibits an unusual histological pattern – a mixed type liposarcoma with a major pleomorphic and a minor sclerosing well-differentiated component. The diagnosis of this rare entity may represent a challenge even for an experimented pathologist.

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