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CASE REPORT

Primitive Testicular Leiomyosarcoma

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Abstract Primary testicular leiomyosarcoma is an uncommon tumor with only few cases described in literature. In young people this rare tumor seems to be related to radiotherapy and anabolic steroids abuse. In older people there are apparently no risk factors. We describe one further case in a 77-years old man with full histological and ultrastructural evaluation. A short term follow-up of one year signals no recurrence of the disease.

Keywords Testis · Leiomyosarcoma

Introduction

Primary leiomyosarcoma of the testis is a rare tumor with only few cases described in literature. Some have been related to radiotherapy [1], and anabolic steroids abuse [2], and they all occurred in young patients (<50 years old). Only four cases have been described in older people (>65 years old), without apparently any correlation with known risk factors [3–6]. We herein present an immunohistochemical and ultrastructural analysis of a case in an elderly man and a review of the literature.

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Case Report

A 77-years-old man was referred to urologic consultation for a painless left testicular swelling lasting for around 5 years that has increased during the last year. The patient did not report any other significant symptom. The physical examination revealed a hard, firm tumor by palpation. The ultrasound evaluation showed an egg-shaped solid intratesticular hypoechogenic 40×28 mm mass with well-defined borders (Fig. 1) and with an unremarkable spermatic cord. Chest radiography was normal. A CT scan resulted negative. Serological markers (alpha-fetoprotein, AFP, B-human chorionic gonadotropin, B-HCG) were within normal range. The clinical history was negative for malignant diseases. The man underwent left inguinal orchiectomy. Within the testis, a hard and white tumor was observed. The lesion was confined within the tunica albuginea, with no invasion of the paratesticular structures. Light microscope examination revealed a monomorphic pattern with elongated cells arranged in interweaving fascicles (Fig. 2). The cells contained abundant cytoplasm and cigar shaped hyper-chromatic nuclei with mitotic activity and nuclear pleiomorphism (Fig. 3). The surrounding vascular space was without tumor invasion. Strong positive staining was observed for smooth muscle actin (1A4), and desmin. Tumor cells were negative for alfa-inhibin, vimentin, cytokeratins, myogenin, CD117, PLAP, S100, B-HCG, AFP, and CD30.

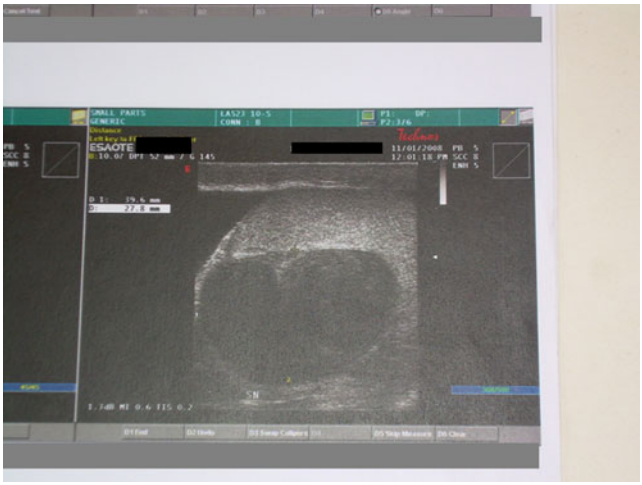

Fig. 1 Ultrasound of the testis showing a egg-shaped solid, hypoecogenic 40×28 mm mass with well-defined borders

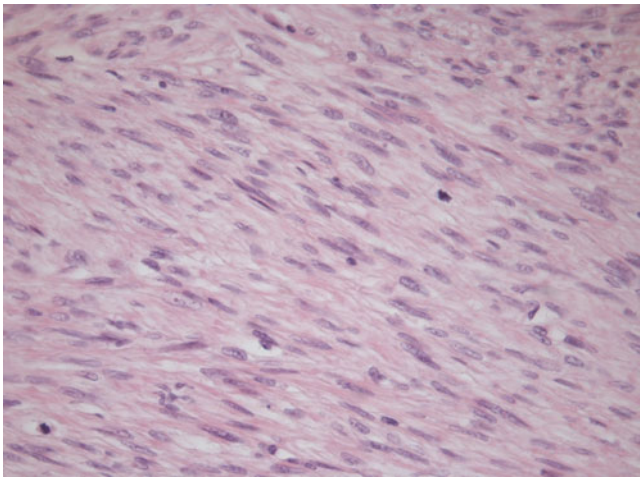

Fig. 3 Testicular leyomiosarcoma at an higher magnification showing atypic cells and mitotic figures (E & E, original magnification × 40)

Electron microscopic examination showed neoplastic cells with a spindle shape and with oval nucleus containing clumps of chromatin. The cytoplasm was almost entirely occupied by bundles of microfilaments. Basal lamina like material was occasionally present at the cell surface (Fig. 4).

The tumor was classified as a high-grade testicular leiomyosarcoma.

There was no evidence of recurrence or metastasis 12 months post-operation.

Discussion

Sex cord-stromal tumors comprise less than 5% of all testicular tumors, and they are mostly differentiated towards Leydig and Sertoli cell components [7]. In the testis there is the rare possibility of tumors with exclusive fibrous and spindle cell components which do not involve the paratesticular tissue. Few cases have been reported of benign testicular gonadal stromal tumor with only spindle cells [8]. Few cases have been observed with atypic spindle tumor cells with nuclear pleiomorphism, numerous mitoses and blunt edged nuclei suggesting malignant tumors of smooth muscle origin. These cells may be derived from the blood vessels or they are supposed to be contractile cells of the seminiferous tubules. The diagnosis of these rare malignant tumors should only be done after having excluded the more commonly seen paratesticular smooth muscle tumors or a possible metastasis from a primary leiomyosarcoma from another origin. In this case, scrotal and abdominal ultrasound evaluation, chest radiography were suggestive for a primitive testicular tumor; moreover previous clinical history was negative for malignant diseases. The histological evaluation confirmed

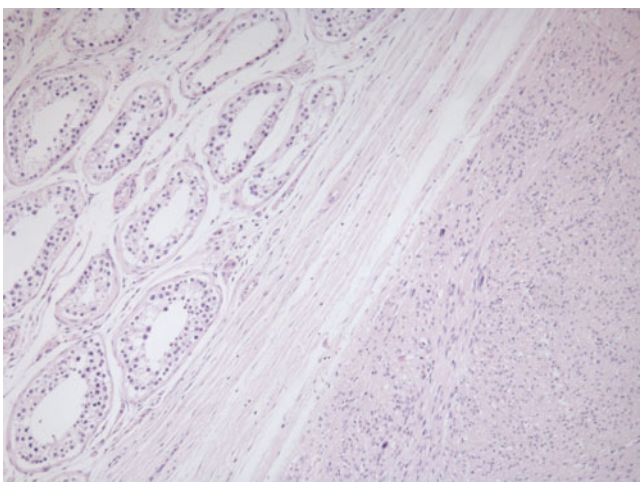

Fig. 2 Histologic features are characterized by monomorphic pattern of interweaving spindle elements in close proximity to seminous tubules. (E & E, original magnification × 10)

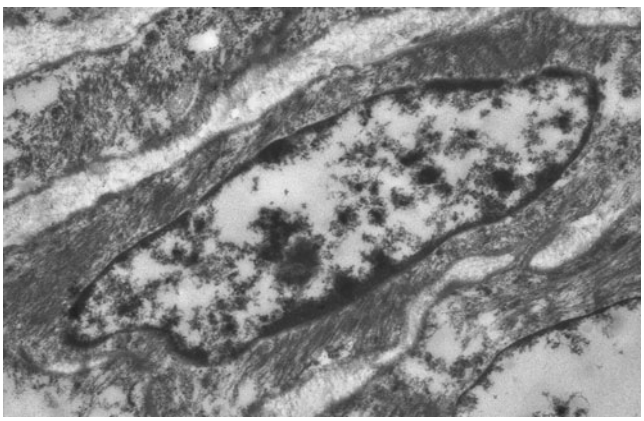

Fig. 4 Tumor cells with smooth muscle differentiation showing several intracytoplasmatic actin microfilaments (Original magnification × 6000)

the involvement of the testis by malignant tumor cells with smooth muscle differentiation among seminiferous tubules, and ruled out both the presence of germ cell tumor cells and the involvement of paratesticular structures. In the literature, in the elderly people (>65 years), only four testicular leiomyosarcoma cases have been reported and with very similar histological features. The longest follow-up was 12 months apparently with no recurrence of disease except for a 70-years old man whose death is reported 14 months after orchiectomy due to "pulmonary metastasis". Of course the prognosis of these patients is impaired by the advanced age which eventually does not allow a long term follow-up. Therefore radical orchiectomy followed by surveillance appears to be the treatment of choice.

This is the fifth case reported of a primary testicular leiomyosarcoma in an old patient with a full histological and ultrastructural evaluation. A short term follow-up signals no recurrence of disease.

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