

Primary Pure Testicular Low-Grade Leiomyosarcoma

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Abstract

Herein we report a case of primary leiomyosarcoma of testis, which was believed to originate from normal testicular structures. It contained smooth muscle cells, blood vessels and contractile cells of the seminiferous tubules. No evidence of tumor spread was found. Treatment consisted of orchiectomy with high ligation of the spermatic cord. The patient received no adjuvant therapy and there was no evidence of tumor 30 months after the operation. Pertinent literature is reviewed and the differential diagnosis is discussed.

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Keywords • Testis • testicular neoplasmas • leiomyosarcoma

Case Report

A 27-year-old man, with a history of slowly enlarging left testicle over the past six months, presented to our center. No other constitutional or urologic symptoms were present. Sonogram showed an intratesticular mass with unremarkable spermatic cord. Chest radiogram, abdominal and chest computed tomography, alpha-fetoprotein (AFP) and beta-human chorionic gonadotropin (β -HCG) levels were normal. The patient underwent a left inguinal orchiectomy with high ligation of the spermatic cord. No other abnormalities were noted on physical examination and a staging procedure was negative. No additional treatment was done and the patient was well with no evidence of disease after 30 months.

Pathologic Findings

The specimen consisted of a testis measuring 6.5 cm in diameter and attached to a spermatic cord of 10 cm in length and 1.5 cm in diameter. A cut section of the testis revealed a well-circumscribed mass of 4.5 cm in diameter. The tumor was predominantly solid and white in color. The tunica albuginea was intact, and the rete testis as well as epididymis and the spermatic cord were unremarkable (Fig 1). Microscopic examination showed a cellular tumor composed of cigar-shaped elongated cells with central nuclei showing mild pleomorphism. A few mitosis were also seen, about 3-4 per 10 HPF. No germ cell component was present in the examined 25 sections of the tumor. (Fig 2) The surrounding rim of testis, epididymis and spermatic cord were unremarkable. Immunoperoxidase staining revealed the presence of smooth muscle actin in the tumoral cells but was negative for desmin and PLAP leading to a diagnosis of low-grade leiomyosarcoma.

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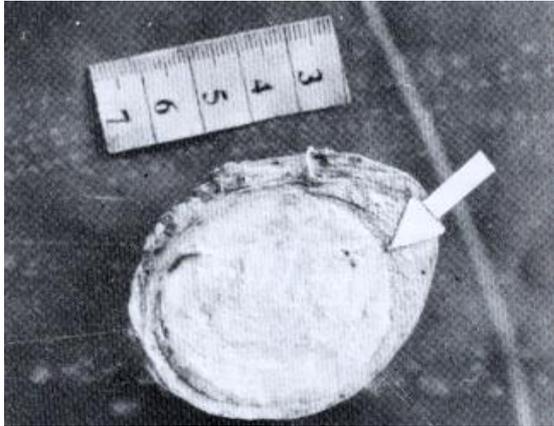


Figure 1: Macroscopic appearance of longitudinally opened testicle shows tumor displacing testicular tissue.

Discussion

Leiomyosarcomas are tumors that grossly resemble leiomyomas. They may arise anywhere in the body from tissues containing smooth muscle. Intrascrotal sarcoma and testicular leiomyosarcoma are rare¹, and are mainly rhabdomyosarcomas that occur in children². However, after treatment a few germ cell tumors may show sarcomatous transformation containing leiomyosarcomatous components.³⁻⁴ According to the anatomic site, leiomyosarcomas are divided into three groups: deep, superficial (cutaneous) tumors and those arising from

the walls of large blood vessels.⁵ Primary intratesticular sarcoma is very rare and must be grossly distinguished from paratesticular sarcoma. Since 1935 many cases of testicular sarcoma have been reported, most of them being rhabdomyosarcomas however, there are few reports on the primary testicular leiomyosarcoma in the literature.¹ Intrascrotal sarcomas are uncommon neoplasms and have been traditionally separated into paratesticular and the intratesticular tumors.² Paratesticular sarcomas are mostly rhabdomyosarcomas seen in children.¹⁻⁴ In contrast to paratesticular sarcomas which are almost always unaccompanied by other components, intratesticular sarcomas are usually associated with a germ cell tumor. Occasional sarcomatous transformation of gonadal tumor is well known²⁻³ and sarcomatous elements in testicular germ cell tumors have been recognized.⁴ Intratesticular primary leiomyosarcoma unassociated with a germ cell component is extremely rare. Only three cases of pure testicular leiomyosarcoma are reported.¹⁻⁴

Leiomyosarcoma arising from tunica vaginalis is also extremely rare,⁶ as are other testicular sarcomas like osteosarcoma and fibrosarcoma. Androgenic steroids have been suspected as carcinogens in the development of intratesticular leiomyosarcoma.⁷ Primary testicular leiomyosarcoma should be differentiated from paratesticular sarcoma, leiomyosarcoma of the scrotum,⁸ aggressive fibromatosis of the spermatic cord⁹ and smooth muscle hyperplasia of the testicular adnexa.¹⁰

In summary, we reported a rare case of primary low grade intratesticular leiomyosarcoma unaccompanied by germ cell component. Testicular

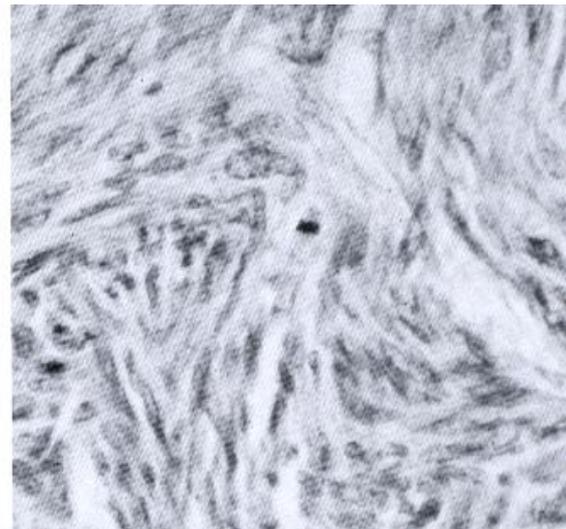
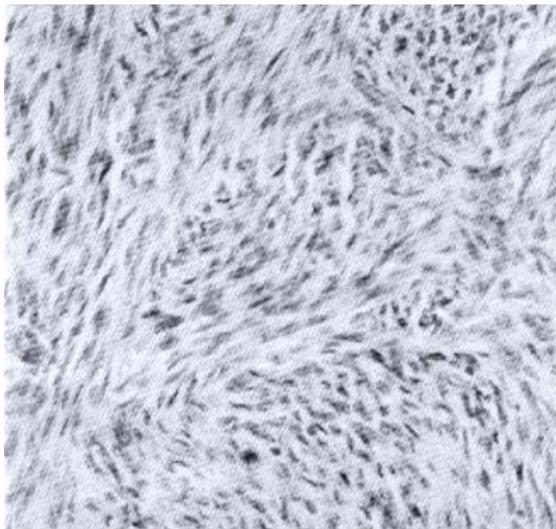


Figure 2: Microscopic section revealed interlacing bundles of spindle shaped cells with a mitotic figure. H&E: x120; x400

M. Sattary, B. Hazraty, M. Bagche Saraii

leiomyosarcoma appears to be a stromal tumor of unknown malignant potential. Germ cell tumor element must be carefully excluded before establishing the diagnosis. At present, inguinal orchiectomy with careful follow-up appears to be a sufficient treatment.

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