

Oncology

A case of subcutaneous leiomyosarcoma of the scrotum presenting as a slowly growing mass in a 36-year-old male: A case report and literature review

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ABSTRACT

Leiomyosarcoma of the scrotum, not involving the testis, epididymis, or spermatic cord, is a very rare type of tumor. A 36-year-old male presented with a slowly growing right scrotal mass (size: 3 × 2 cm). An ultrasound examination revealed a hypoechoic, subcutaneous, solid mass. Laboratory investigations detected normal levels of serum alpha-fetoprotein, beta-human chorionic gonadotropin, and lactate dehydrogenase. The mass was slowly growing and surgically resection was performed diagnosing leiomyosarcoma with histological examination. Wide local excision with a 2-cm margin around the operative scar and right orchiectomy were performed as second surgery. The patient has not suffered recurrence for 28 months.

Introduction

Over 95% of paratesticular leiomyosarcomas originate from the spermatic cord or the epididymis, dartos muscle, or subcutaneous scrotal muscle. Superficial leiomyosarcomas are rare malignant smooth muscle tumors. They account for 4–6.5% of all soft tissue sarcomas, less than 2–3% of cutaneous soft tissue neoplasms, and 0.04% of all cancer.¹ Leiomyosarcoma of the scrotum, which arises in the subcutaneous scrotal layer, is a rare type of tumor. Subcutaneous scrotal tumors have been reported to be associated with an increased risk of local recurrence and distant metastasis.² To the best of our knowledge, less than 38 cases of leiomyosarcoma of the scrotum have been reported in the literature.

Herein, we report the case of a 36-year-old patient, who was initially diagnosed with an epidermal cyst, but was subsequently diagnosed with paratesticular leiomyosarcoma.

Case report

A 36-year-old male presented with a small scrotal mass, which had slowly enlarged over the last 8 months. It was a non-tender, firm, irregular, and mobile mass. It measured 2 cm in diameter and had an irregular elliptical shape. It did not exhibit continuity with the testis. The irregular form of the mass was confirmed by ultrasonography, and the interior of the mass was heterogeneous. No abnormal findings were

seen in either testis. Laboratory tests did not reveal any significantly abnormal values, including in the levels of tumor markers, such as cancer antigen 19–9, squamous cell carcinoma antigen, interleukin-2 receptor, human chorionic gonadotropin, and alpha-fetoprotein. The patient was initially diagnosed with an epidermal cyst, however, there was still a possibility that it has a malignant potential because of the increase in size. Therefore, the mass was surgically removed. No adhesion between the tumor and the neighboring structures was observed during the excision, and we did not see any continuity between the mass and the testis either (Fig. 1). A pathological examination involving hematoxylin and eosin staining revealed that the mass consisted of spindle-shaped cells, multiform cells, and mitotic cells. There were 15–17 mitoses per 10 high power fields (Fig. 2). An immunohistochemical examination demonstrated that the tumor cells were strongly positive for smooth muscle actin and 30% positive for Ki-67. Staining for S-100 was negative. Chest and abdominal computed tomography (CT) examinations did not show any abnormal findings. The patient was pathologically diagnosed with a primary subcutaneous leiomyosarcoma. Based on a discussion with the dermatologists and pathologists at our hospital, we decided to perform a secondary surgical procedure, with the intention of widely removing the surrounding structures, including the right testis. Japanese guidelines for the treatment of soft tissue tumor, recommend that soft tissue tumors should be resected together with a 20-mm safety margin. We followed this

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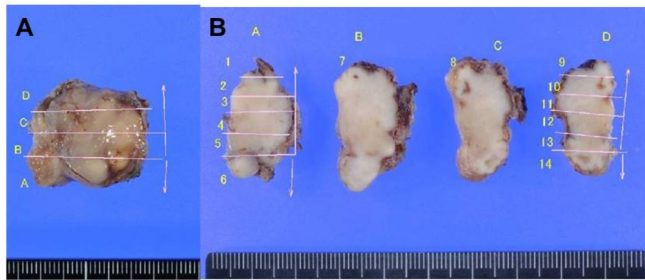


Fig. 1. (A) The gross specimen of the tumor measured 3 cm × 2 cm × 1 cm and had a grey-white cut surface. (B) The cross-sectional surface was yellow and elastic.

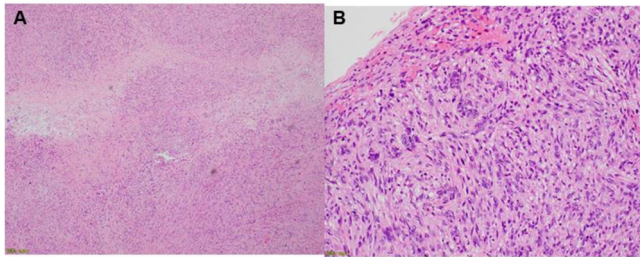


Fig. 2. (A) The histological features of subcutaneous leiomyosarcoma are shown (hematoxylin and eosin staining [H&E] staining, original magnification: ×40). (B) The spindle-shaped cells exhibited nuclear pleomorphism. Prominent nucleoli and 3–5 mitotic figures per high power field along with bizarre pleomorphic giant tumor cells were seen (H&E staining, original magnification: ×200).

approach so that scrotal skin around the prior operative scar with testis was excised. No leiomyosarcoma cells were found in the excised tissue. Follow-up CT was performed every 6 months, and no signs of local recurrence or distant metastasis have been seen for 28 months.

Discussion

Soft tissue sarcomas of the genitourinary tract are uncommon and only comprise 1% of all malignancies. They can originate from all types of soft tissue, including muscles, tendons, fat, fibrous tissue, synovial tissue, blood vessels, and nerves. Leiomyosarcomas constitute 10%–20% of soft tissue sarcomas and most often develop in the uterus, gastrointestinal tract, or retroperitoneum. Leiomyosarcomas are malignant mesenchymal neoplasms that arise from smooth muscle, and their etiology is unknown. However, the risk factors for these tumors include high-dose anabolic steroids, chronic inflammation of the testis, and testicular field irradiation for the treatment of leukemia.³ None of these factors were present in our patient.

Scrotal leiomyosarcomas are very rare. They arise between the fourth and eighth decades of life as painless, slowly growing skin lesions of 2–9 cm in diameter.⁴ They are described as smooth muscle tumors of the skin that are found in the muscle linings of arterioles and

veins in the subcutaneous layer. It is extremely rare for them to arise in the scrotal skin or subcutaneous layer, and only 38 cases of leiomyosarcoma of the scrotum have been reported.

The diagnosis of leiomyosarcoma is based on histological examinations. Leiomyosarcomas are composed of spindle-shaped cells with cigar-shaped nuclei arranged in interweaving fascicles. As for their pathological findings, leiomyosarcomas contain 2 to 3 mitoses per 10 high power fields⁴; however, in the present case 15 to 17 mitoses per 10 high power fields were detected during hematoxylin and eosin staining. Hence, we considered that this case exhibited higher malignancy. Immunohistochemical examinations of leiomyosarcoma show positive staining for actin, desmin, and CD34.

Leiomyosarcomas can metastasize to the lungs, liver, and bone hematogenously, and the prognosis of leiomyosarcoma depends on the size, depth, and grade of the tumor and the presence/absence of distant metastasis at diagnosis. Wide excision is required for the treatment of localized leiomyosarcoma of the scrotum because of the risk of recurrence/distance metastasis.⁵

Chemotherapy is an option for patients who refuse surgery. It can include gemcitabine, paclitaxel, and vincristine. For example, a 59-year-old male with lung metastasis from scrotal leiomyosarcoma was treated with gemcitabine and paclitaxel.

Radiotherapy has recently been recommended as an adjuvant therapy for testicular leiomyosarcoma after surgery regardless of the grade or histology of the tumor.

Long-term follow-up is required because of the reported risk of late local recurrence.⁵

Conclusion

This report describes how various common testicular tumors present. Leiomyosarcoma can mimic epidermal cyst. After a histological diagnosis has been obtained, appropriate medical management of scrotal tumors is essential, even for small scrotal masses.

Declaration of interest

None.

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