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Case Report

Recurrent, giant subcutaneous leiomyosarcoma of the thigh

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ABSTRACT

We present a case of recurrent, massive subcutaneous leiomyosarcoma involving the left thigh in a 29-year-old male from Madagascar. The patient had earlier undergone local resection of subcutaneous leiomyosarcoma a half year before. After surgical intervention, local recurrence developed at this site and was rapidly growing. The patient was surgically treated with a 2-cm-wide margin local excision in our hospital. The patient has remained recurrence free at 1-year follow-up.

Case report

A 29-year-old male from Madagascar presented to our hospital because of recurrence of subcutaneous leiomyosarcoma in the left thigh after surgery performed a half year before. The patient has not undergone chemotherapy and adjuvant postoperative radiotherapy. Physical examination revealed a mulberry appearance, tenderness, and poorly circumscribed mass, with easy superficial hemorrhage when touched (Fig. 1).

The patient was in apparently good health. Full blood count, biochemical test results, and tumor markers were within the normal range. There was no inguinal lymphadenopathy. In addition, chest x-ray and electrocardiography were within normal limits.

Magnetic resonance imaging (MRI) revealed a multilobulated, heterogeneous, subcutaneous lesion that measured 9.0 cm × 5.1 cm × 7.2 cm in the lateroposterior aspect of the left thigh (Fig. 2). In addition, multiple hemorrhages in the mass demonstrated high signal intensity in T1- and T2-weighted images (Figs. 3 and 4). After intravenous contrast-enhanced MRI, the mass appeared prominent, heterogeneous, and enhanced (Figs. 5 and 6).

The mass originated from a subcutaneous region at a lateroposterior aspect of the left thigh and broke out of the epidermis. The adjacent deep fascia was free of involvement.

The patient underwent a wide local excision with 2-cm lateral margins. All surgical margins were negative. Gross specimen appeared as a fleshy, gray-white, glistening, and predominantly cystic necrotic mass with hemorrhage (Fig. 7).

Histopathologic examination showed a poorly delineated tumor with intersecting, sharply marginated fascicles of spindle cells with eosinophilic cytoplasm and elongated, blunt-ended, and hyperchromatic nuclei (Fig. 8). Immunohistochemically, the cells were positive for actin and desmin (Fig. 9).

Competing Interests: The authors have declared that no competing interests exist.

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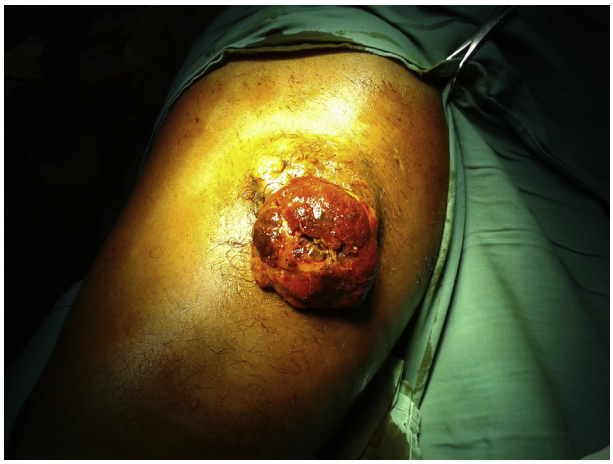

Fig. 1 – A recurrent subcutaneous leiomyosarcoma involving the patient's left thigh, with a loss of the continuity of focal skin.

Discussion

Superficial leiomyosarcomas are rare malignant tumors accounting for 4%-6.5% of all soft-tissue sarcomas [1]. They are divided into cutaneous or dermal and subcutaneous leiomyosarcomas depending on their location in the skin and presumed site of origin [1]. Subcutaneous leiomyosarcoma arises from the smooth muscle lining of arterioles or veins in subcutaneous tissue and accounts for 1%-2% of all soft-tissue sarcomas [2]. They may arise anywhere on the body, and the thigh has been reported as the most frequently affected site [3].

Subcutaneous leiomyosarcoma may occur at any age, most commonly between the ages of 40 and 60 years [2]. It usually presents as a hemispherical skin elevation ranging from 0.6 to 5 cm in diameter and larger than its dermal counterparts [3]. Subcutaneous leiomyosarcomas are typically fast growing and without surface changes [3]. However, our recurrent case presented with mulberry appearance and was associated with skin ulceration. A review of the literature suggests that the case reported here is the largest recurrent subcutaneous leiomyosarcoma arising in extremities reported so far.

The imaging features of subcutaneous leiomyosarcoma are typically nonspecific [1]. Lesions are typically isointense to

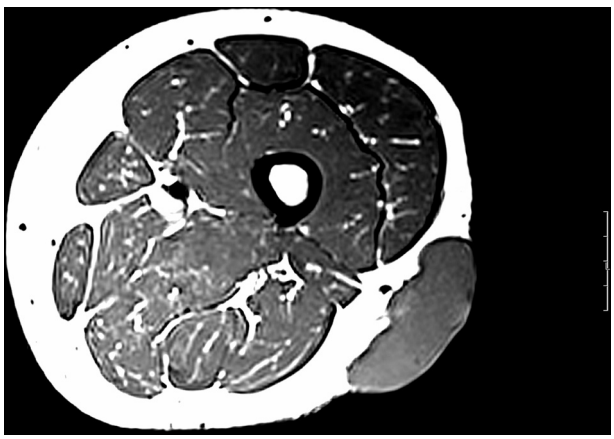

Fig. 2 – Axial T1-weighted magnetic resonance image demonstrates the lobulated mass isointense to muscle and lack of deep fascia involvement.

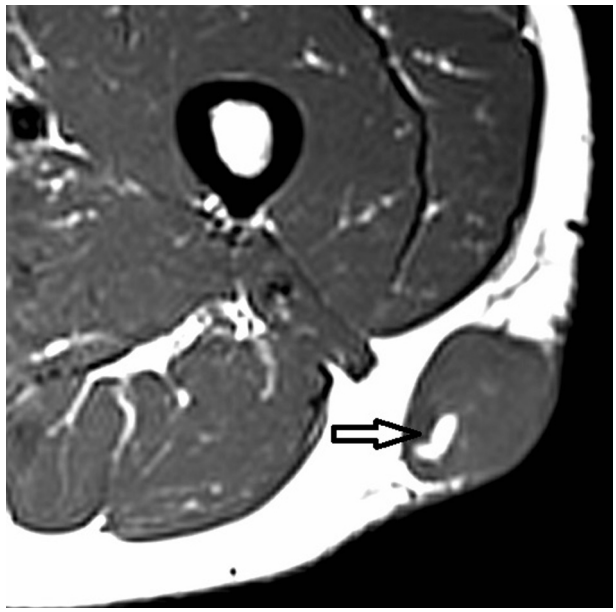

Fig. 3 – Axial T1-weighted magnetic resonance image demonstrates predominantly isointense mass and nodular focus of high signal of hemorrhage (arrow).

Fig. 4 – Coronal short time inversion recovery magnetic resonance image demonstrates lobulated, high-signal-intensity mass, and the hemorrhage shows high signal intensity (arrow).

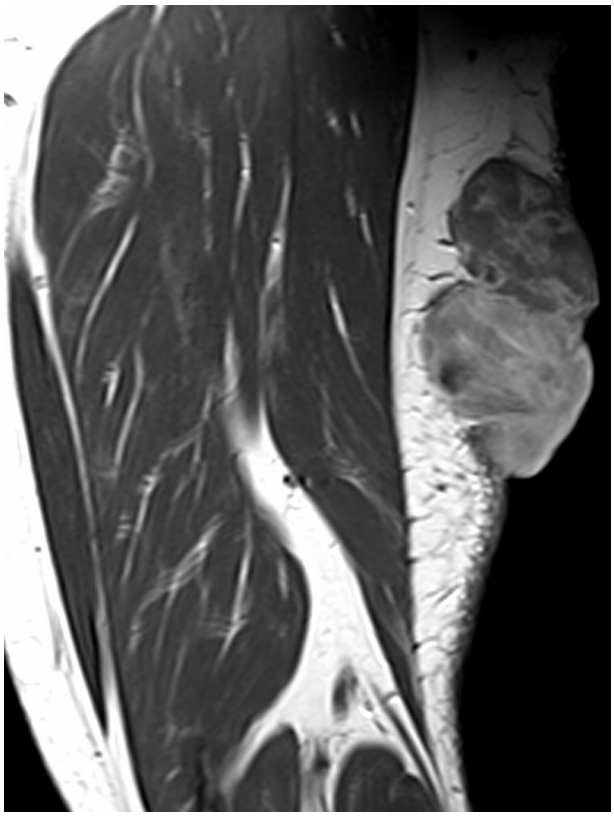

Fig. 5 – Coronal T1-weighted contrast-enhanced magnetic resonance image demonstrates prominent, patchy-enhancing subcutaneous mass.

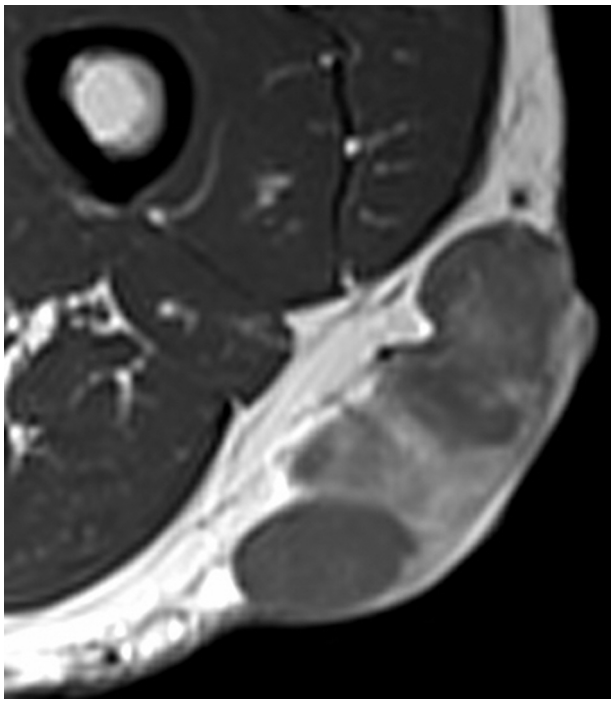

Fig. 6 – Axial T1-weighted contrast-enhanced magnetic resonance image demonstrates prominent, patchy-enhancing subcutaneous mass.

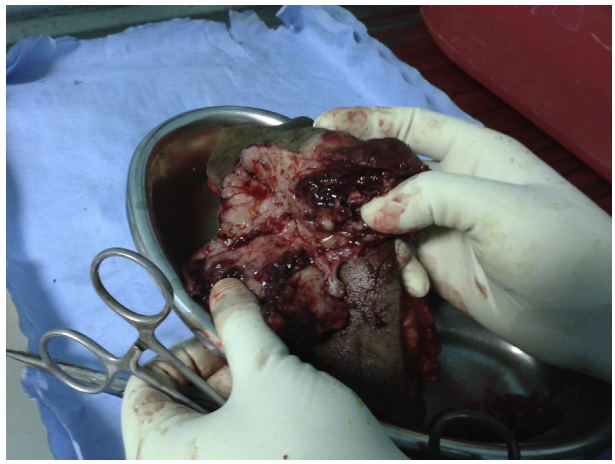

Fig. 7 – Gross specimen shows a multiple necrotic mass with typical fleshy appearance and hemorrhage on its cut surface.

muscle on T1-weighted images and variably hyperintense relative to muscle on T2-weighted images, with prominent contrast enhancement. Large lesions are usually more heterogeneous because of the presence of hemorrhage, necrosis, and cystic changes, whereas microcalcifications may be demonstrated in 10%-15% of the cases on radiographs or computed tomography scans [4]. MRI should be used for staging purposes in subcutaneous tumors larger than 3-5 cm [4].

As subcutaneous leiomyosarcoma is resistant to radiation and chemotherapy, early surgical excision with wide margins is the treatment recommendation [3,5]. Prognosis after excision is generally considered poor. Factors that have correlated with

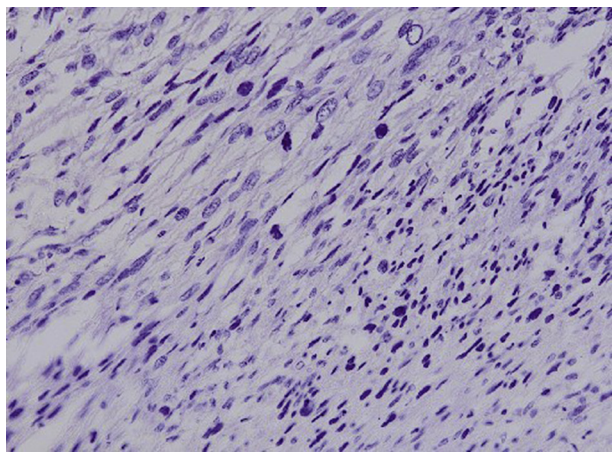

Fig. 8 – Microscopic examinations revealing the lesion to be of predominantly fascicular pattern, with tumor bundles intersecting each other at wide angles. The individual tumor cells have elongated blunt-ended nuclei and acidophilic fibrillary cytoplasm. Cytoplasmic vacuoles are located at both ends of the nuclei. Some of the tumor cells show large bizarre appearance.

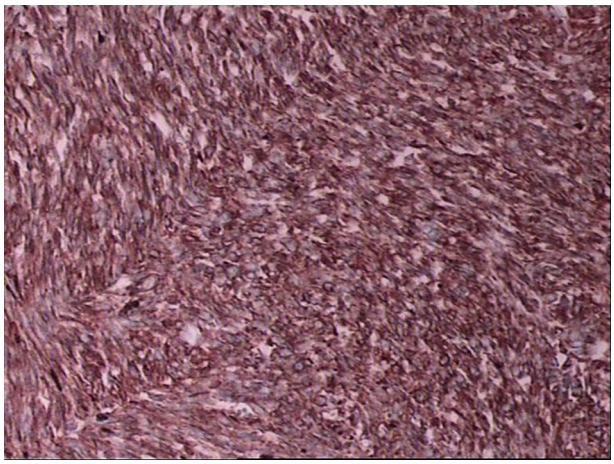

Fig. 9 – Immunohistochemistry: the tumor is positive for smooth muscle actin (original magnification ×100).

poor prognosis included tumor size, histologic grade, extensive necrosis, deep tumors with fascia involvement, and intratumoral vascular invasion [2,6]. The risks of local recurrence have ranged from 14% to 42% and have been reported even after a wide excision. Recurrences have been reported after the removal of very small primary tumors measuring 0.5 cm, whereas 50% of the tumors recur within 4-18 months after initial diagnosis [3,7].

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