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Limb-Sparing En Bloc Resection of a Multiply Recurrent Pleomorphic Liposarcoma of the Thigh: A Case Report

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Abstract | **Case Report**

Pleomorphic liposarcoma is the rarest and most aggressive subtype of liposarcoma, with high rates of local recurrence and distant metastasis. We report the case of a 40-year-old man who presented with a painless mass of the lateral left thigh. Excision revealed a 7.5 cm pleomorphic liposarcoma (Fédération Nationale des Centres de Lutte Contre le Cancer [FNCLCC] grade 3) with close margins (0.5 cm), followed by adjuvant brachytherapy. A bifocal recurrence in the vastus lateralis was excised 16 months later with a 1 mm margin. Over the following four years, the patient received 32 sessions of radiotherapy and 20 sessions of chemotherapy. Five years after diagnosis, a second recurrence developed in the medial compartment, abutting the femoral shaft and deep femoral vessels, and grew from 45 to 67 mm despite treatment. Staging showed no distant metastasis. At 46 years of age, the patient underwent a third resection: en bloc removal of the adductor compartment through a medial approach, preserving the femoral vessels, sciatic nerve and periosteum. Histopathology confirmed a 9 cm pleomorphic liposarcoma with tumour necrosis and clear margins (closest 0.5 cm). There was no postoperative deficit, and adjuvant radiotherapy was planned. Close margins and high grade were the main drivers of recurrence. When the disease remains confined to the limb and the neurovascular structures are displaced rather than encased, limb-sparing en bloc resection remains possible after failure of multimodal treatment. The first excision should aim for wide margins.

Keywords: Pleomorphic liposarcoma, soft tissue sarcoma, local recurrence, limb salvage, en bloc resection, adductor compartment, thigh, case report.

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INTRODUCTION

Pleomorphic liposarcoma (PLS) accounts for about 5–10% of liposarcomas and is, by definition, a high-grade sarcoma [1,2]. It mainly affects adults in their sixth and seventh decades, and the extremities, especially the thigh, are the most common site [2–4].

The prognosis is poor. In the two largest clinicopathological series, local recurrence occurred in 34–45% of patients and distant metastasis, mostly pulmonary, in 32–42.5% [3,4]. Five-year overall survival ranges from 54% to 63% [2,4].

Wide excision with negative margins, combined with radiotherapy for deep high-grade tumours, is the standard treatment of extremity sarcomas [5]. Managing repeated local recurrences is more difficult: tissues have already been irradiated, and the tumour often lies closer to the neurovascular bundle and bone. Amputation is then frequently discussed, although it improves local control without improving overall survival [5,6].

We report a multiply recurrent thigh PLS after surgery, brachytherapy, external beam radiotherapy and chemotherapy, treated by limb-sparing en bloc resection.

CASE REPORT

Patient information.

A 40-year-old shopkeeper with no relevant medical history presented (Month 0) with a painless swelling of the lateral aspect of the left thigh.

Initial treatment (Month 1).

After histological diagnosis, the mass was excised. The specimen (20 × 16 × 8 cm, 190 g) contained a well-limited, myxoid tumour of 7.5 cm in contact with

muscle. Histology showed a mucoid matrix with lipoblasts and multinucleated anaplastic cells, 14 mitoses per 10 high-power fields, no necrosis, muscle infiltration and no vascular emboli: pleomorphic liposarcoma, FNCLCC grade 3 (differentiation 3, mitoses 2, necrosis 0) [7]. Margins were clear but close, 0.5 cm medially and laterally. Adjuvant brachytherapy was delivered shortly after surgery: 7 Gy to the left thigh in two fractions, with good tolerance.

First recurrence (Months 17–19)

At Month 17, MRI showed four nodules in the vastus lateralis. Core biopsy found a high-grade pleomorphic spindle-cell sarcoma with more than 20 mitoses per 10 high-power fields and necrosis. At Month 18, re-excision removed two foci of 2.5 and 2 cm, confirming a bifocal recurrence of pleomorphic liposarcoma. Margins were clear, but the anterior margin was 1 mm. From Month 19 to Month 68, the patient received a total of 32 radiotherapy sessions and 20 chemotherapy sessions, with six-monthly surveillance.

Second recurrence (Months 59–73)

Surveillance MRI showed a new lesion against the medial cortex of the proximal femoral shaft, growing from 14 × 11 × 21 mm at Month 59 to 36 × 41 × 48 mm at Month 62. Thoraco-abdomino-pelvic CT showed no distant disease. Angio-CT (Month 63) located the lesion in the medial compartment, infiltrating the adductor magnus and vastus medialis, against the deep femoral artery and vein with loss of the fat plane; the superficial femoral vessels and sciatic nerve were spared. A third resection was planned at Month 63, but radiotherapy and chemotherapy were continued first.

Despite treatment, the mass grew from 36 × 45 × 44 mm at Month 68 to 59 × 66 × 67 mm at Month 73 (Figure 1), and surgery was then indicated. It was heterogeneously hyperintense on T2 with internal septa and enhanced intensely around central necrosis. It abutted the femoral cortex without lysis or marrow signal change, displaced the patent deep femoral vessels medially, and infiltrated the vastus intermedius and the adductor magnus and brevis. Staging CT (Month 72) showed no metastasis; bilateral subcentimetric axillary lymph nodes, a calcified pulmonary micronodule, calcified hepatic granulomas and simple renal cysts were noted.

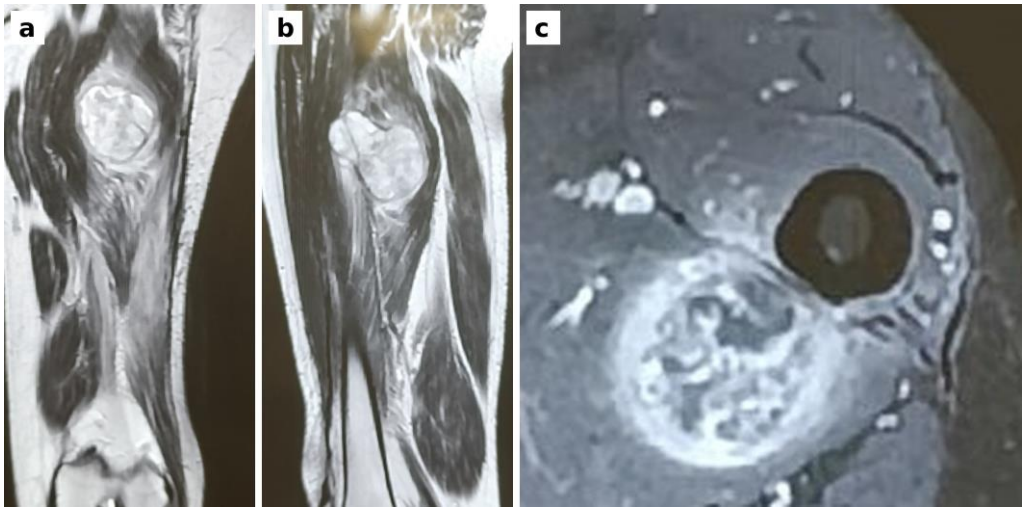


Figure 1: MRI of the left thigh at Month 73, before the third resection. (a) Coronal and (b) sagittal T2-weighted images showing a lobulated, heterogeneous mass with internal septa in the medial compartment of the proximal thigh. (c) Axial fat-suppressed T1-weighted image after gadolinium showing peripheral enhancement with central necrosis, in contact with the femoral cortex without bone lysis

Clinical findings (Month 75)

Examination found a mass of the medial aspect of the proximal left thigh and healed lateral scars from previous surgery. Neurovascular examination was normal, and the axillary nodes were painless and mobile.

Surgical procedure

At Month 75 (Day 0), under general anaesthesia in the supine position, the patient was operated on by the senior author (R.C.). Through a medial approach centred on the lesion, the adductor muscles were resected en bloc with the tumour (Figure 2). One branch of the deep femoral artery feeding the tumour was ligated; the femoral vessels and sciatic nerve were preserved. The periosteum was not resected, as imaging showed no bone invasion. The wound was closed in layers without a drain. The oriented specimen weighed 439 g and measured 11.5 × 8.5 × 10 cm; the tumour was entirely covered by a cuff of muscle without macroscopic breach (Figure 3).

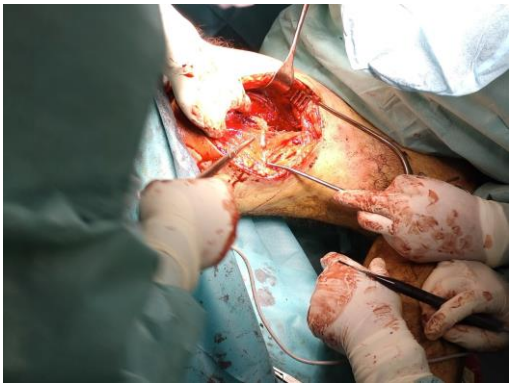


Figure 2: Intraoperative view of the medial approach to the left thigh during en bloc resection of the adductor compartment

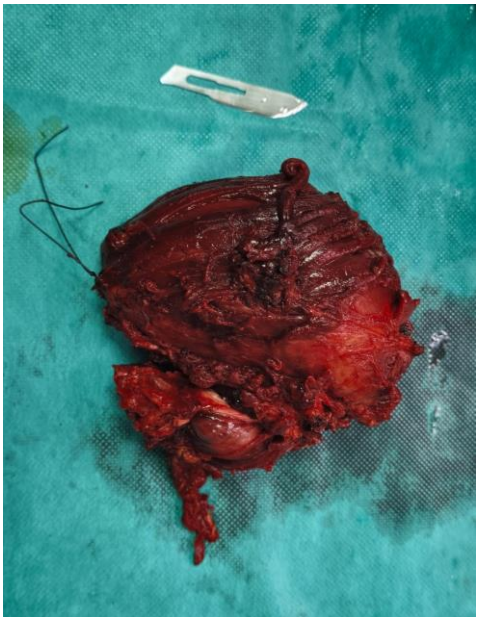


Figure 3: En bloc resection specimen (11.5 × 8.5 × 10 cm, 439 g), with the tumour covered by a cuff of muscle. The suture marks the distal end

Outcome and follow-up

The postoperative course was uneventful, with no neurological or vascular deficit. Final histology (Day 4) showed a myxoid tumour of 9 × 7.5 cm with pleomorphic spindle cells, abundant multinucleated cells, lipoblasts, numerous mitoses and tumour necrosis: pleomorphic liposarcoma, FNCLCC grade 3. All margins were clear: 0.5 cm medially, 1 cm at the superficial and deep margins, 2 cm proximally and distally, and 4 cm laterally. On Day 10, the oncology team decided on adjuvant radiotherapy; the wound had healed and the patient was in good general condition. The timeline is summarised in Table 1.

Table 1: Timeline of the case

Time point	Event
Months 0–1	Painless lateral thigh mass; excision of a 7.5 cm PLS, margins 0.5 cm
Month 1	Adjuvant brachytherapy, 7 Gy in two fractions
Months 17–18	First recurrence (four nodules, vastus lateralis); re-excision, anterior margin 1 mm
Months 19–68	32 radiotherapy sessions and 20 chemotherapy sessions; six-monthly surveillance
Months 59–62	Second recurrence, medial compartment: 21 → 48 mm
Month 63	Staging CT negative; angio-CT; planned resection deferred
Months 68–73	Growth from 45 to 67 mm under treatment
Month 75 (Day 0)	Third resection, en bloc adductor compartment, limb preserved
Day 4	Histology: 9 cm PLS, necrosis, clear margins (closest 0.5 cm)
Day 10	Oncology consultation: adjuvant radiotherapy decided

Ethics. Written informed consent for publication was obtained from the patient. Ethics committee approval was not required for a single case report at our institution.

DISCUSSION

Diagnosis

PLS must be distinguished from dedifferentiated liposarcoma with lipoblastic differentiation and from undifferentiated pleomorphic sarcoma. Pleomorphic lipoblasts support PLS, whereas MDM2 amplification points to dedifferentiated liposarcoma [4,8]. In our patient, the diagnosis rested on morphology, with pleomorphic lipoblasts on each specimen; MDM2 status was not reported.

Why did the tumour recur?

Our patient combined the main risk factors reported for PLS: high grade, deep location, size over 5 cm and multifocal recurrence [3,4,9]. Presentation with recurrent disease is itself a predictor of poor outcome [9]. Above all, both excisions achieved negative but close margins, 0.5 cm then 1 mm. A negative margin of a few millimetres corresponds to a marginal rather than a wide resection in Enneking's terms [10]. Guidelines recommend wide excision with a rim of normal tissue, or an intact anatomical barrier such as fascia, for high-grade extremity sarcomas [5]. Adjuvant brachytherapy and radiotherapy did not compensate for these close margins. The second recurrence arose in the medial compartment, away from the lateral operative bed; contamination along a previous biopsy tract or drain, a skip lesion or a new focus are all possible.

Response to systemic treatment

The lesion grew by about 50% during chemotherapy and radiotherapy. Systemic treatment, based on anthracyclines with or without ifosfamide, is mainly used for advanced sarcoma, and its benefit in localised recurrence is uncertain [5]. Here, progression left surgery as the only option for local control.

Limb salvage or amputation?

The randomised trial of Rosenberg *et al.,* showed that limb-sparing surgery with radiotherapy gave survival equivalent to amputation, despite more local recurrences [6]. Survival in PLS depends mainly on distant metastasis [2–4], and our patient had none after six years. The deep femoral vessels were displaced but patent, the superficial femoral vessels and sciatic nerve were free, and the femur was not invaded. These conditions allowed an en bloc compartmental resection with a functional limb, and final histology confirmed clear margins. Periosteal stripping or vascular resection would have added morbidity in irradiated tissue without clear benefit.

Postoperative management

Closure without a drain carries a risk of seroma in the dead space of irradiated tissue and requires close monitoring. Because the medial margin was only 0.5 cm, adjuvant radiotherapy was chosen despite the doses already received. Follow-up includes clinical examination, local MRI and chest CT every 3–4 months for 2–3 years, then every 6 months until 5 years [5].

Limitations

This is a single case with short follow-up after the latest resection. The chemotherapy regimen, the total external radiation dose and MDM2 status were not available.

CONCLUSION

Pleomorphic liposarcoma is a high-grade sarcoma whose local behaviour depends largely on the quality of the first excision. In this patient, close margins were followed by two recurrences over six years despite brachytherapy, radiotherapy and chemotherapy. Any suspected soft tissue sarcoma should be referred, before biopsy or excision, to a team able to plan a wide resection. At recurrence, imaging of the relationship between the tumour, the neurovascular bundle and the bone guides the choice between limb salvage and amputation. When the vessels are displaced rather than encased and the bone is not invaded, en bloc compartmental resection can still achieve clear margins and preserve a functional limb. Long-term surveillance remains essential.

Declarations

Conflict of interest: The authors declare no conflict of interest.

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