

Synchronous Myxoid Liposarcoma of the Popliteal Fossa and Distal Femur Treated with Single-Session En Bloc Resection and Oncologic Endoprosthetic Reconstruction: A Case Report

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Abstract

Background: Myxoid liposarcoma (MLS) is a soft tissue sarcoma with a distinctive tendency to metastasize to extrapulmonary sites, including soft tissue and bone. Synchronous lesions in soft tissue and bone are rare, and distinguishing multicentric primary tumors from metastatic spread is difficult. We report a case treated with single-session en bloc resection and oncologic endoprosthetic reconstruction.

Case Description: A 38-year-old man presented with a four-month history of a posterior knee mass. A liposarcoma diagnosed at another center was confirmed as MLS on review of the paraffin block at our institution, and baseline chest CT was negative. A subsequent MRI showed an intramedullary lesion in the ipsilateral distal femur, and CT-guided biopsy again showed MLS. After tumor board discussion, both lesions were resected in a single session: en bloc resection of the popliteal mass in the prone position, followed by a 10-cm distal femoral resection and reconstruction with a cemented modular oncologic endoprosthesis in the supine position. Margins were negative in both specimens. Pulmonary lesions appeared two months after surgery, and the patient continues oncologic treatment. At 12 months he had no pain, and knee flexion was limited to 15°. Molecular studies in the literature indicate that multifocal MLS reflects metastatic spread rather than independent primary tumors. Clonality was not tested in our patient, so we regard the synchronous presentation as probable metastasis of one lesion to the other site.

Conclusions: Synchronous soft tissue and bone MLS should be approached as probable metastatic disease, with whole-body staging, molecular confirmation of clonality when possible, and management in a specialized sarcoma center. Single-session limb salvage was technically feasible and achieved negative margins, but at a functional cost, and early pulmonary progression underlines the aggressive biology of the disease.

Keywords: Myxoid Liposarcoma; Multifocal Liposarcoma; Bone Metastasis; Endoprosthetic Reconstruction; Popliteal Fossa; Limb Salvage

1. Introduction

Myxoid liposarcoma (MLS) is the second most common liposarcoma subtype [1] and accounts for approximately 5% of soft tissue sarcomas [2]. It is defined by the FUS::DDIT3 fusion resulting from t(12;16)(q13;q11) or, in fewer than 5% of cases, by the EWSR1::DDIT3 fusion from t(12;22)(q13;q12) [1,3].

Unlike most soft tissue sarcomas, MLS often metastasizes to extrapulmonary sites, mainly soft tissue, the abdominal cavity and bone. In a review of 14 series including 1,853 patients, the first metastasis involved soft tissue in 32%, the abdomen in 26%, the lung in 24% and bone in 17% [4]. In a series of 43 patients, four of the nine deaths occurred in patients with metastatic disease, and in all four the metastases were nonpulmonary [5]. For this reason, whole-body imaging has been proposed for staging and follow-up [2,4,5].

Multifocal MLS, defined as tumor at two or more anatomically separate sites before involvement of the lungs, has been debated as either an unusual pattern of metastasis or a set of independent primary tumors. Molecular analyses of multifocal cases have shown identical clone-specific rearrangements across sites, supporting a metastatic origin [3,6].

We report a patient with synchronous MLS of the popliteal fossa and the ipsilateral distal femur treated with single-session en bloc resection and oncologic endoprosthetic reconstruction. We discuss the metastasis-versus-multicentricity question and the oncologic and reconstructive implications of this presentation.

2. Case Presentation

2.1 Clinical presentation and diagnosis

A 38-year-old man with no relevant medical history presented with a palpable mass in the posterior aspect of the right knee that had been enlarging progressively for four months. He reported local discomfort and denied fever, weight loss or night sweats. Examination showed a firm, non-tender, well-defined mass in the popliteal fossa, with a normal neurovascular examination of the limb. MRI of the knee showed a heterogeneous soft tissue lesion in the popliteal fossa with areas of myxoid signal intensity and no other relevant findings.

A diagnosis of liposarcoma had been made at another center. The paraffin block was reviewed at our institution and confirmed myxoid liposarcoma. Chest CT at initial staging was negative for metastasis. A subsequent MRI revealed an intramedullary lesion in the distal femoral metaphysis of the same limb, with high signal intensity on T2-weighted images, suggestive of malignant involvement (Figure 1). CT-guided biopsy of the femoral lesion at our institution again showed myxoid liposarcoma. Immunohistochemistry was negative for S100, cytokeratin AE1/AE3, CD45 and CD20, which supported a mesenchymal origin and favored the diagnosis of myxoid liposarcoma.

Given the synchronous lesions, the differential diagnosis was bone metastasis from the popliteal tumor versus true multicentric MLS. After multidisciplinary tumor board discussion, surgery was indicated, with a two-stage, single-session plan under combined anesthesia with an epidural catheter.

2.2 Surgical procedure

In the first stage, with the patient prone, a posterior bayonet approach was made and dissected down to the subfascial plane. The biceps femoris and semimembranosus were identified proximally. The sciatic nerve and common peroneal nerve were released microsurgically along their course, and the popliteal vascular bundle was released without compromising oncologic margins. An en bloc resection was performed from proximal to distal and from superficial to deep, including the biceps femoris, semimembranosus, semitendinosus and the proximal portion of the medial gastrocnemius. The specimen was sent for pathology and the wound was closed in layers.

In the second stage, the patient was repositioned supine. An anterior midline approach extended proximally was used with a medial parapatellar arthrotomy. After posterior vascular release, a 10-cm distal femoral resection was performed according to preoperative planning, and the specimen was sent for pathology. The tibial cut was made with an intramedullary guide. Reconstruction used an OSS (Biomet/Zimmer Biomet) system with a 10-cm femoral module and a cemented 150 × 16.5 mm stem, and a size 71 tibial component with a 12-mm polyethylene insert. Implant stability was satisfactory. A muscle flap was used for coverage, a deep drain was placed and the wound was closed in layers. The procedure was uneventful, and postoperative radiographs showed satisfactory implant position (Figure 2).

2.3 Pathology

Macroscopically, the popliteal lesion measured $17 \times 12 \times 8$ cm (Figure 3). The distal femoral specimen measured 10.5 cm in length, with an intramedullary tumor in the metaphyseal region (Figures 4 and 5). Histopathology of both specimens confirmed MLS with similar morphology, with lipoblasts in a myxoid stroma and a prominent delicate capillary network. Areas of round cells with prominent nucleoli, consistent with a high-grade component, were identified (Figures 6 and 7). Resection margins were negative in both specimens.

2.4 Postoperative course and outcome

The postoperative course was uneventful, and the patient began rehabilitation with early mobilization and partial weight-bearing. After surgery he received systemic therapy and radiotherapy according to the protocol defined by the multidisciplinary tumor board. As this report focuses on the surgical management, the details of these treatments are not presented. Pulmonary lesions were detected in the second month after surgery, and the patient continues oncologic treatment at the time of writing. At 12 months he reported no pain, and knee flexion was limited to 15° .

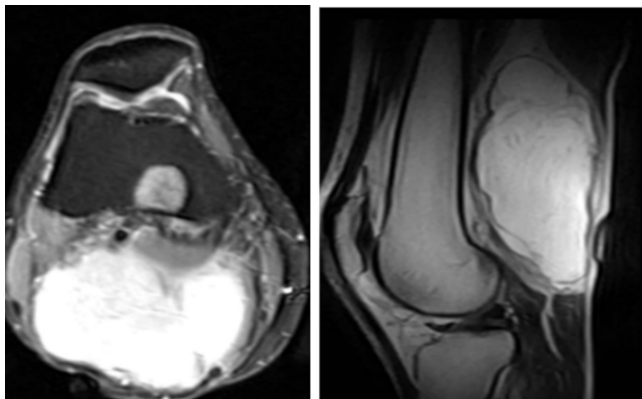


Figure 1. Preoperative MRI of the right knee. Axial image (left) showing the popliteal fossa mass and a rounded intramedullary lesion in the distal femur; sagittal image (right) showing the popliteal mass.



Figure 2. Postoperative radiographs of the knee showing the modular oncologic endoprosthesis (OSS) after distal femoral resection.

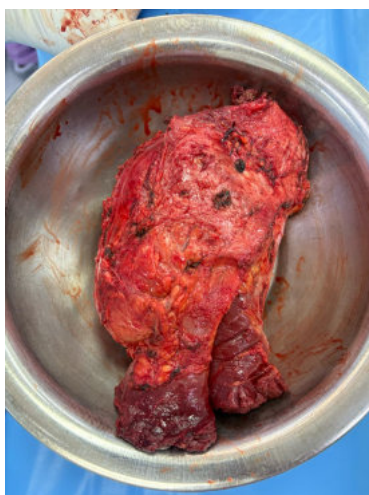


Figure 3. Resected popliteal fossa lesion ($17 \times 12 \times 8$ cm).

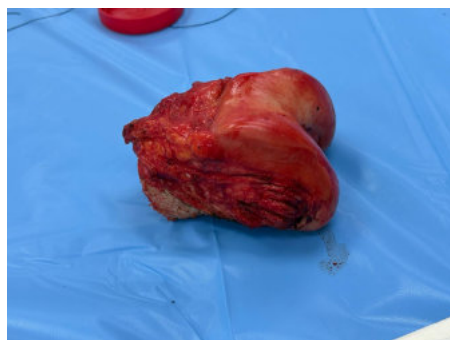


Figure 4. Distal femoral resection specimen (10.5 cm).



Figure 5. Distal femoral resection specimen, second view.

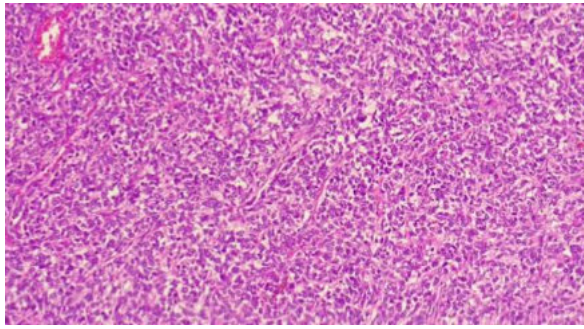


Figure 6. Hematoxylin and eosin, ×4. Proliferation of round cells in sheets with interspersed thin-walled vessels.

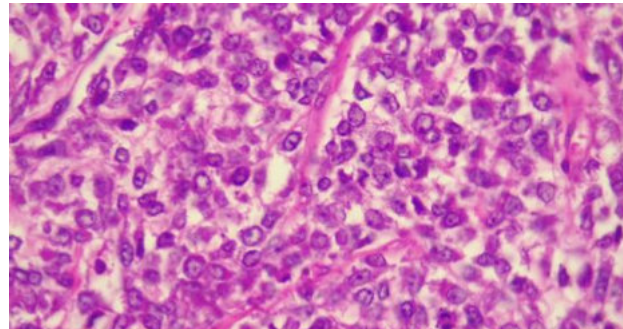


Figure 7. Hematoxylin and eosin, ×10. Cells with rounded nuclei and prominent nucleoli, corresponding to the high-grade component of myxoid liposarcoma.

3. Discussion

3.1 Metastasis or multicentricity

MLS spreads to extrapulmonary sites more often than other sarcomas [2,4,5], which complicates the interpretation of synchronous lesions. A round cell component above 5% identifies patients at higher risk of metastasis and death, and extrapulmonary metastases are a peculiar feature of the disease [7]. Antonescu et al. analyzed six patients with multifocal MLS and found identical clone-specific TLS-CHOP or EWS-CHOP rearrangements in every anatomically separate tumor sample of each patient, which established the metastatic nature of the distant soft tissue lesions; the prognosis in that small series was poor [3]. De Vreeze et al. studied 15 patients (three with synchronous lesions) and found identical FUS-CHOP fusion products at all sites, with loss of heterozygosity analysis suggesting clonal relationships and no evidence of second primary tumors [6].

In our patient both lesions showed the same histology. Clonality was not tested, but the molecular literature and the known bone tropism of MLS favor metastasis of one lesion to the other site over two independent primaries, and we cannot determine which lesion was the primary. The appearance of pulmonary lesions two months after surgery, despite a negative baseline chest CT, suggests occult disease at presentation and is consistent with the aggressive course described for multifocal MLS [3]. This contrasts with the favorable outcomes of localized MLS, with 5- and 10-year disease-specific survival of 93% and 87% in one large series [8].

3.2 Oncologic treatment

Wide resection with negative margins is the cornerstone of local treatment; in myxoid, dedifferentiated and pleomorphic liposarcoma, surgical margins significantly influence local recurrence-free, metastasis-free and overall survival [9]. MLS is highly radiosensitive. In the DOREMY trial, preoperative radiotherapy of 36 Gy achieved an extensive pathologic response in 70 of 77 patients (91%), with 100% local control and wound complications requiring intervention in 17% [10]. A retrospective series reported a mean tumor volume reduction of 52% with 40 Gy [11].

For systemic therapy, the ISG-STS 1001 randomized trial, which included high-grade MLS, did not show benefit of histotype-tailored chemotherapy (trabectedin for MLS) over standard epirubicin plus ifosfamide, and it was closed after a futility analysis [12]. A phase 2 trial of neoadjuvant trabectedin with 45 Gy radiotherapy in localized extremity or trunk MLS did not meet its primary endpoint, although it showed a complete pathologic response in 13% of evaluable patients and 10% or less viable tumor in 51% [13]. Trabectedin has also shown activity in advanced pretreated MLS [14]. The ESMO-EURACAN-GENTURIS guidelines recommend that soft tissue sarcomas be managed by a multidisciplinary team at a reference center [15]. In our patient, whose disease became metastatic early, treatment was defined by the multidisciplinary tumor board.

3.3 Surgical and reconstructive considerations

A single-session prone-then-supine sequence allowed resection of both lesions with oncologic margins while preserving the sciatic and common peroneal nerves and the popliteal vessels. Endoprosthetic reconstruction after resection of soft tissue sarcomas with bone involvement is uncommon and carries a high complication burden. In 29 patients with lower-limb soft tissue sarcoma, 48% underwent reoperation and 31% had infection, and overall and metastasis-free survival were lower than in a matched cohort of sarcomas not requiring endoprosthetic reconstruction [16]. In 12 patients treated with endoprosthetic reconstruction for soft tissue sarcoma attached to bone, 66% had no evidence of disease at a median follow-up of 62 months [17]. In a systematic review of 2,721 patients with primary bone tumors, 47% required reoperation, with aseptic loosening occurring at a mean of 75 months and deep infection at a mean of 24 months [18]. These figures justify long-term implant surveillance.

The functional result in our patient was limited: he had no pain, but knee flexion was limited to 15°. This probably reflects, at least in part, the resection of the knee flexors (biceps femoris, semimembranosus, semitendinosus) and the medial gastrocnemius required for oncologic clearance. This trade-off should be discussed with patients before surgery.

3.4 Surveillance

Because MLS metastasizes to extrapulmonary sites, follow-up should not be limited to chest imaging. Whole-body MRI, including the spine and pelvis, has been proposed for staging and surveillance [2,4,5].

3.5 Limitations

This is a single case with 12 months of follow-up and ongoing disease. The diagnosis rested on morphology and an immunohistochemical panel that was negative for S100, cytokeratin AE1/AE3, CD45 and CD20; FISH for DDIT3 rearrangement and DDIT3 immunohistochemistry [19] were not available. Molecular clonality testing was not performed either, so the metastatic interpretation rests on indirect evidence. Baseline staging in this report is limited to chest CT. Functional outcome was described without validated scores such as MSTS or TESS. The report focuses on surgical management, and the details and response of systemic and radiation therapy are not presented. These limitations restrict generalization of our observations.

4. Conclusions

Synchronous soft tissue and bone myxoid liposarcoma most likely reflects metastatic spread rather than independent primary tumors. Single-session en bloc resection with oncologic endoprosthetic reconstruction achieved negative margins and limb salvage, at the cost of markedly limited knee flexion, and pulmonary progression appeared early. Whole-body staging, molecular confirmation of clonality when possible, and management in a specialized sarcoma center are advisable for similar presentations.

Conflict of Interest

The authors declare no conflict of interest.

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None.

Informed Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images.

Ethics Approval

This report was authorized by the institutional ethics committee.

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Author Contributions

Agustín Davies and Marcos Galli Serra wrote the manuscript. Walter Parizzia, Bruno Terrarossa Retegui and Marcos Galli Serra were the operating surgeons.

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