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Case Report

Recurrent Myxoid Liposarcoma of the Upper Thoracic Spine: A Case Report

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Keywords

Liposarcoma · Myxoid liposarcoma · Recurrent myxoid liposarcoma · Upper thoracic spine · Case report

Abstract

Introduction: Liposarcoma, one of the most common soft tissue sarcomas, originates from primitive mesenchymal cells. However, spinal involvement of liposarcoma, whether primary or metastatic, is rare. According to literature reports, myxoid/round cell liposarcomas are the most common types affecting the spine. Spinal liposarcomas, whether primary or metastatic, have only been sporadically reported. ***Case Presentation:*** The author presents an unusual case of recurrent myxoid liposarcoma originating from the erector spinae muscle, infiltrating the upper thoracic spine in a 65-year-old female. The patient presented with a 1-week history of progressively worsening right chest and back pain. The treatment approach involved surgical intervention with wide tumor resection, spinal cord decompression, posterior instrumentation, and thoracoplasty, complemented by radiotherapy. ***Conclusion:*** The preferred treatment is en bloc resection with wide margins. Postoperative radiation therapy can serve as complementary treatment for piecemeal resection. A follow-up is necessary because of the potential for recurrence.

Introduction

Liposarcoma, one of the most common soft tissue sarcomas in adults, originates from primitive mesenchymal cells and accounts for approximately 20% of all mesenchymal malignancies [1]. The most frequent sites for liposarcoma occurrence are the thigh and femur.

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Myxoid liposarcoma represents the second most common histological type, typically manifesting between the ages of 40 and 60 [1–3]. However, primary or secondary involvement of the spine is exceedingly rare, and only sporadic case reports have been published [4–6]. The objective of this study was to present an exceptionally rare case of recurrent myxoid liposarcoma involving the upper thoracic spine in an elderly patient.

Case Presentation

A 69-year-old female underwent excision surgery for a mass in the right scapular region at an external hospital 2 years ago. Postoperative pathology revealed myxoid liposarcoma, but no further treatment was administered. Six months later, the patient presented to our thoracic surgery department with anterior chest pain. Magnetic resonance imaging results showed a soft tissue mass within the erector spinae muscles at the level of thoracic vertebrae 2–4, invading the right appendage without encroachment on the spinal canal, suggesting tumor recurrence. Subsequently, the patient was started on oral treatment with "anlotinib 12 mg qd*14d," and regular follow-up indicated stable disease. However, 1 week before admission, the patient experienced worsening right chest and back pain described as stabbing and progressively intensifying, leading to hospital admission.

Imaging Studies

The chest thoracic computed tomography (CT) scan reveals a mixed-type extensive lytic lesion involving the right appendage and adjacent ribs from T1 to T5. The lesion exhibits radiolucent components and peripheral sclerosis, encompassing the right pedicle, vertebral body, and transverse process, with T4 being the most severely affected and extending into the spinal canal through the intervertebral foramen (Fig. 1). Subsequent magnetic resonance imaging assessment demonstrates invasive and uneven low signal intensity within the erector spinae muscles at the T1–5 level on T1-weighted images. T2-weighted images reveal an infiltrative mass with high signal intensity, displaying diffuse and nonuniform enhancement, spreading to the right paravertebral area and the extradural space (Fig. 2). Preoperative physical, laboratory, and imaging examinations, including chest CT scan, abdominal-pelvic CT scan, and bone scan, did not reveal evidence of tumors in other organs.

Surgical Procedure

The patient underwent a posterior approach tumor resection surgery with the design of a right-skewed oval incision, encompassing the original surgical scar (Fig. 3a). At positions 5 centimeters proximal and distal to the tumor, the right erector spinae muscles were severed, and dissection outward exposed the 2nd to 5th posterior ribs. At a distance of 5 centimeters from the tumor, the ribs were cut, followed by the excision of the right T2–5 lamina, costovertebral joints, and transverse processes. The tumor was excised in its entirety, revealing its infiltration through the right T4/5 intervertebral foramen into the spinal canal. Post-tumor resection, internal fixation was performed using the T2–5 pedicle screw-rod system. Shaping was achieved with a connecting rod inserted into the residual ends of the 2nd and 5th ribs, while the other end utilized a domino connector to connect with the pedicle screw-rod system. Repair for the deficient pleura was conducted using a biological mesh (Fig. 3b, c).

Pathological Result

Histopathological examination revealed a malignant mesenchymal origin tumor, microscopically characterized by extensive spindle-shaped cell proliferation with residual tumor observed. Invasion into striated muscle tissue is evident, accompanied by interstitial

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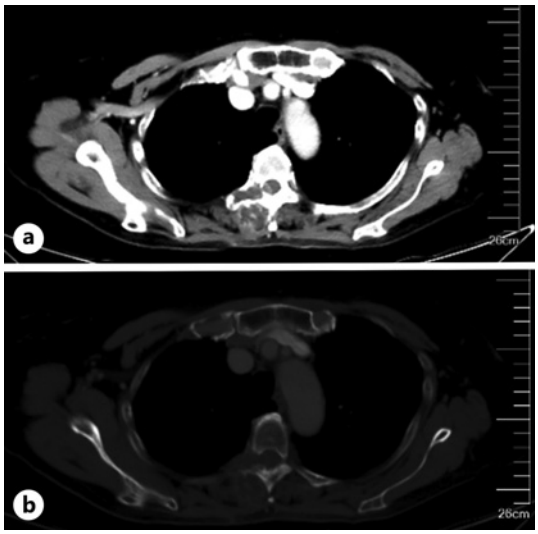

Fig. 1. a, **b** The chest thoracic computed tomography scan reveals a mixed-type extensive lytic lesion involving the right appendage and adjacent ribs from T1 to T5. The lesion exhibits radiolucent components and peripheral sclerosis, encompassing the right pedicle, vertebral body, and transverse process, with T4 being the most severely affected and extending into the spinal canal through the intervertebral foramen.

inflammatory fibrous tissue proliferation and mucoid degeneration. Considering the medical history, these findings are consistent with alterations following treatment for myxoid liposarcoma (Fig. 4a, b). Immunohistochemically, the tumor cells did not show MDM2 expression. FISH (DDIT3 (12q13) gene breakpoint probe) analysis showed DDIT3 rearrangement (Fig. 4c).

Postoperative Treatment

Postoperatively, the patient developed pleural effusion and encapsulated fluid in the surgical area, which resolved after treatment. The original chest and back pain significantly alleviated. One month later, the patient received adjuvant radiotherapy to the surgical site (50GY/25F) and declined chemotherapy. At the 12-month follow-up, the patient had a good recovery with no signs of recurrence or metastasis (Fig. 5).

Discussion

Liposarcoma is the most common soft tissue tumor and accounts for approximately 20% of all mesenchymal malignancies [2, 4, 5, 7]. Based on morphology and genetics, the World Health Organization (WHO) classified it into four distinct histologic subtypes: well-differentiated/dedifferentiated, myxoid/round cell, pleomorphic, and mixed. The myxoid/round cell liposarcomas (MRCLs) were the most common ones affecting the spine according to the literatures [3]. Myxoid liposarcoma is the second most common histological type, accounting for 30%–40% of cases, and occurring more frequently in younger patients between 40 and 60 years of age [2]. The most frequent sites of myxoid liposarcomas are buttocks, retroperitoneum, trunk, and proximal limb girdle, but primary or secondary spinal involvement is a rare entity.

Surgical resection is considered the most effective method for treating liposarcomas, as it can directly remove the tumor and achieve better local control [8]. For spinal liposarcomas, complete tumor removal without violating the capsule is not always a reliable ideal treatment strategy due to the large tumor size and complex anatomical structures. The goals of surgery for spinal tumors include not only removing the lesions but also protecting the spinal cord and/or nerve roots and reconstructing spinal stability. As mentioned earlier, spinal liposarcomas typically invade and erode bone structures, along with a large amount

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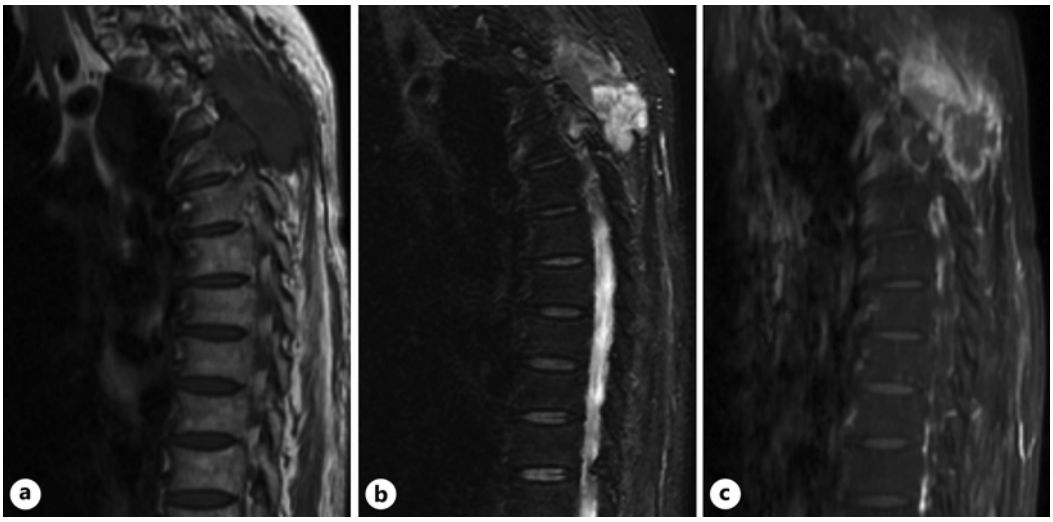

Fig. 2. a Magnetic resonance imaging demonstrates invasive and uneven low signal intensity within the erector spinae muscles at the T1–5 level on T1-weighted images. T2-weighted images reveal an infiltrative mass with high signal intensity (**b**), displaying diffuse and nonuniform enhancement, spreading to the right paravertebral area and the extradural space (**c**).

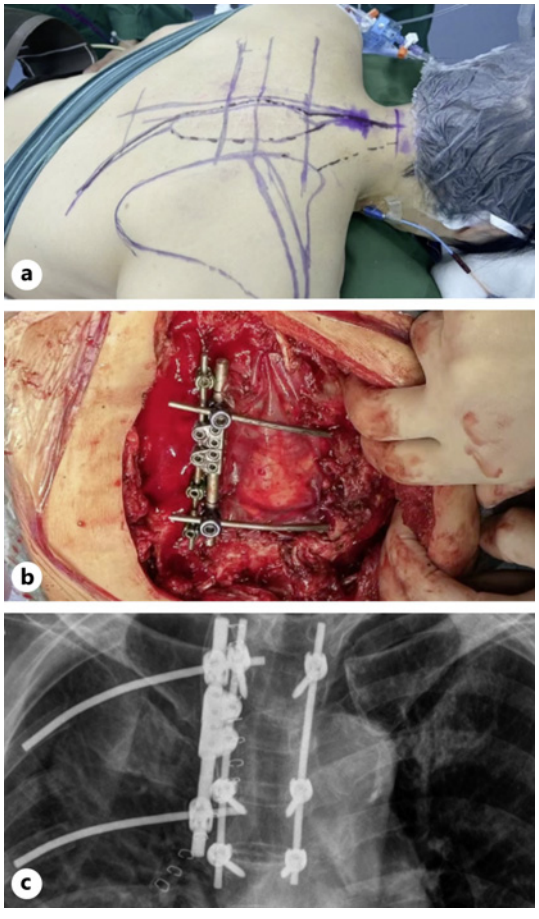

Fig. 3. Incision design (**a**): a right-skewed oval incision, encompassing the original surgical scar. Intraoperative tumor resection and reconstruction status (**b**): posterior fusion used T2–5 pedicle screw-rod system. Rib reconstruction: a connecting rod inserted into the residual ends of the 2nd and 5th ribs, while the other end utilized a domino connector to connect with the pedicle screw-rod system. The deficient pleura repair was conducted using a biological mesh. **c** Intraoperative fluoroscopy.

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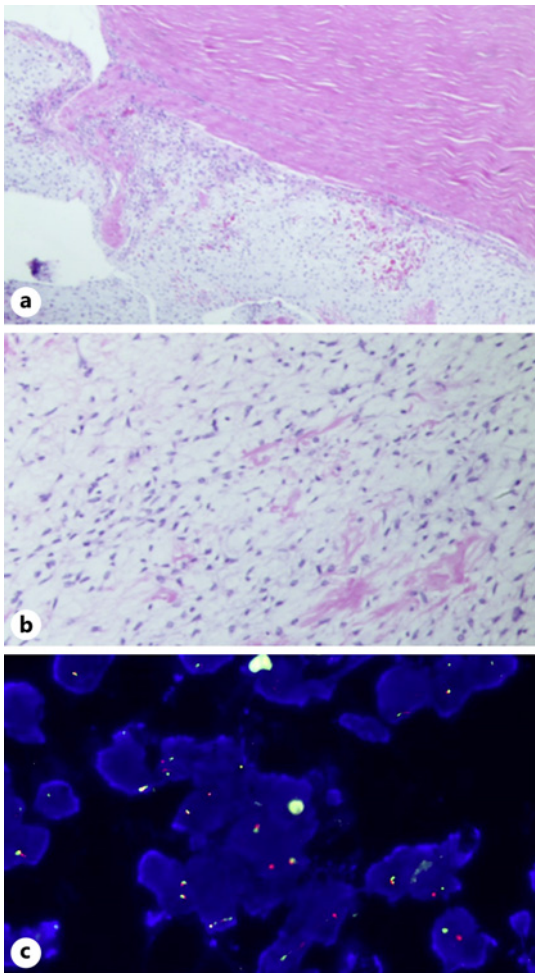

Fig. 4. Histopathological examination revealed a malignant mesenchymal origin tumor, microscopically characterized by extensive spindle-shaped cell proliferation with residual tumor observed. **a**, **b** Invasion into striated muscle tissue is evident, accompanied by interstitial inflammatory fibrous tissue proliferation and mucoid degeneration. Immunohistochemically, the tumor cells did not show MDM2 expression. **c** FISH (DDIT3 (12q13) gene breakpoint probe) analysis showed DDIT3 rearrangement.

of soft tissue mass, leading to the involvement of the spinal cord and/or nerve roots in the tumor. Therefore, whether primary or metastatic, spinal liposarcomas are usually removed piece by piece, although total en bloc spondylectomy has been proven effective with a lower incidence of local recurrence and is recommended for isolated spinal lesions [9, 10]. Our case also involved a piecemeal resection approach. Based on specific characteristics, only a small portion of appropriate patients can undergo total en bloc spondylectomy resection of spinal liposarcomas. Tumor contamination of the surgical field is difficult to exclude; therefore, intraoperative chemotherapy is used to reduce tumor residuals and/or local recurrence by rinsing and soaking the surgical field with cisplatin dissolved in desalted water [11–13].

Unfortunately, for patients with tumors that are too large to be resected and/or have multiple metastases, effective treatment options are currently limited. Radiotherapy can offer symptom palliation for some of these patients as a means of local control, but it does not address systemic disease [14, 15]. Cho et al. [2] emphasized the importance of radiotherapy for local control despite its limitations in treating patients with multicentric metastatic myxoid liposarcoma.

MRCL is the most chemosensitive subtype of liposarcoma. Anthracycline-based combination chemotherapy regimens remain the standard first-line treatment. The overall response rate (ORR) by RECIST with anthracycline-based chemotherapy is approximately 40% [16]. However, trabectedin is also effective and may be considered in the first-line setting

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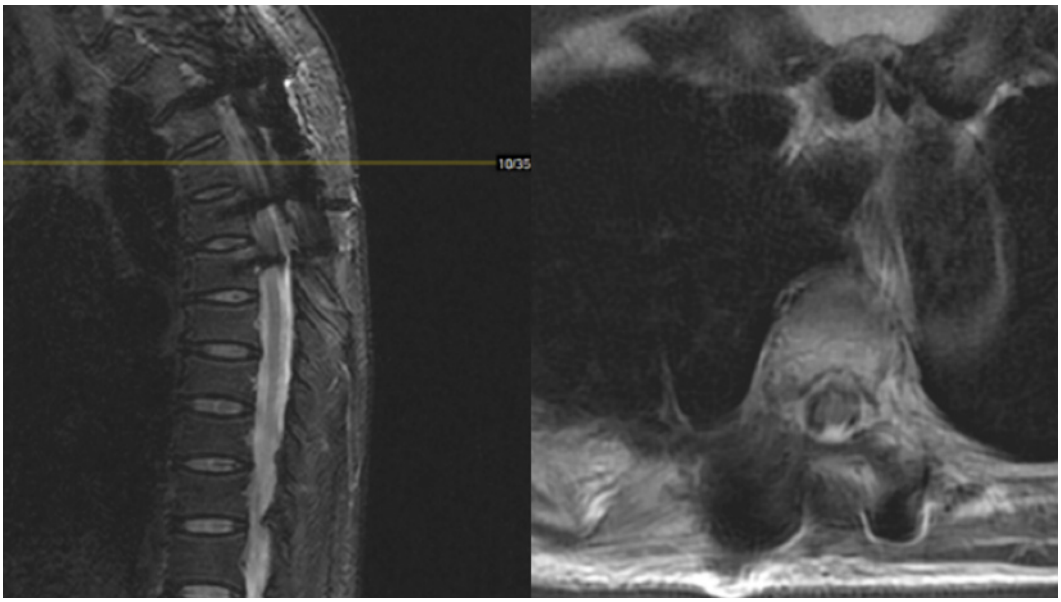

Fig. 5. After 12 months of postoperative follow-up, there is no local recurrence observed, as indicated by the magnetic resonance imaging.

when anthracyclines are contraindicated [17]. Beyond chemotherapy, new therapeutic classes are being developed, including autologous adoptive modified T-cell receptor therapies, which have shown promising results [18]. These therapies leverage the immunogenic potential of cancer-testis antigens, such as NY-ESO-1 and MAGE-A4, which are expressed in the majority of MRCL cases. Early-phase trials have demonstrated encouraging outcomes, with ORR rates up to 40% and a median progression-free survival of up to 8.7 months [19, 20]. Genomically, >90% of MRCLs harbor the chromosomal translocation t(12; 16) (q13; p11), resulting in the oncogenic fusion transcript FUS-DDIT3. Preclinical data have shown that the fusion oncoprotein FUS-DDIT3 affects JAK-STAT signaling, which is associated with a subpopulation of cells exhibiting cancer stem cell properties in MRCL [21]. Currently, a trial is underway testing itacitinib, a JAK1 inhibitor, in various sarcoma histologies, including MRCL (NCT03670069). Overall, the available treatment options for patients with MRCL are likely to evolve in the coming years, potentially improving patient prognosis.

Although complete surgical resection is the gold standard for all spinal tumors, our case, due to the extensive range of tumor invasion, could not achieve total excision and had to undergo piecemeal resection. Subsequently, radiation therapy was administered. Considering that this is a recurrence for the patient, and there may be residual tumor in the surgical area, we recommend the patient undergo adjuvant chemotherapy. However, the patient has declined chemotherapy due to concerns about its side effects. Fortunately, there has been no recurrence in the 12-month post-surgery. However, further observation during follow-up is needed to assess long-term outcomes.

Conclusion

Myxoid liposarcoma involving the spine (primary or metastatic) is rare. The preferred treatment is en bloc resection with wide margins. Postoperative radiation therapy can serve as complementary treatment for piecemeal resection. Chemotherapy remains the primary

treatment for recurrent or metastatic cases, but the ORR is suboptimal. However, biomarker-selected treatments are being developed. A follow-up is necessary because of the potential for recurrence.

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Statement of Ethics

Written informed consent was obtained from the patient for publication of the details of their medical case and any accompanying images. This study protocol was reviewed, and the need for approval was waived by the Ethics Committee of Shantou University Medical College Affiliated Cancer Hospital. The CARE Checklist has been completed by the authors for this case report, attached as online supplementary File (for all online suppl. material, see https://doi.org/10.1159/000542503).

Conflict of Interest Statement

The authors have no conflicts of interest to disclose.

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Author Contributions

Conceptualization, supervision, and writing – original draft preparation: Huaitai Lin; methodology and validation: Xinjia Wang and Chanping Yang; and writing – review and editing: Huaitai Lin, Chenyu Yang, Jinhao Zhu, Zijian Xu, and Weidong Wang.

Data Availability Statement

All data generated or analyzed during this study are included in this article and its online supplementary material files. Further inquiries can be directed to the corresponding author.

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