Sclerosing liposarcoma with rib involvement detected by CT and ^{99m}Tc-MDP bone scintigraphy: A case report and review of the literature

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Keywords: Liposarcoma - Sclerosing liposarcoma - Dedifferentiated liposarcoma - Myxoid liposarcoma - SPECT

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Received:
16 May 2024
Accepted:
16 February 2025

Abstract
A 70-year-old male patient who presented with persistent upper abdominal pain was found to have a rare case of sclerosing liposarcoma with rib involvement. Despite the absence of significant weight loss or a history of infectious disease, elevated inflammatory markers suggested an underlying inflammatory or neoplastic process. Diagnostic imaging by computed tomography (CT) and single photon emission computed tomography (SPECT) played a critical role in identifying a soft tissue mass causing osteolytic damage to the left 9th rib. The rapid enlargement of the lesion and the specific imaging characteristics challenged conventional detection methods. Biopsy confirmed the diagnosis of low-grade sclerosing liposarcoma. This case highlights the range of abilities of current imaging modalities in detecting certain forms of liposarcoma and emphasizes the need for a comprehensive diagnostic approach that integrates clinical findings, radiology, and pathology to accurately diagnose and manage such complex cases.

Hell J Nucl Med 2025; 28(1): 75-78 *Epub ahead of print: 7 April 2025* *Published online: 30 April 2025*

Introduction

Liposarcomas represent the most prevalent form of soft tissue sarcomas, manifesting predominantly in the extremities or the retroperitoneum [1]. Their heterogeneity in pathological subtypes -ranging from well-differentiated to myxoid/round cell, dedifferentiated, and pleomorphic variants- poses significant challenges in diagnosis and treatment. Among these, sclerosing liposarcoma, a subtype of well-differentiated liposarcoma, presents with relatively lower malignancy but with a potential for complex diagnostic and therapeutic challenges, especially when involving bone. This paper presents a comprehensive case study of a patient with soft tissue tumor associated with osseous destruction, highlighting the diagnostic hurdles faced due to the limitations of current imaging technologies in detecting vertebral metastasis, and reviews the literature to explore the nuances of managing this rare condition. Through this discussion, we aim to emphasize the critical need for a nuanced approach in diagnosing and treating liposarcoma, leveraging a multidisciplinary strategy to enhance patient outcomes and advance the understanding of this multifaceted disease.

Case report

A 77-year-old male patient presented with persistent upper abdominal pain for over two months. He reported no significant recent weight changes. His medical history includes poorly controlled diabetes for more than two years due to irregular medication adherence. He denies any history of infectious diseases such as hepatitis or tuberculosis, as well as any major trauma or surgical history. Laboratory tests showed elevated ferritin levels at 1565μg/L (normal range: 30-400μg/L), IL-6 levels at 272.09pg/mL (normal range: 0-5.30pg/mL), and IL-10 levels at 6.54pg/mL (normal range: 0-4.91pg/mL), suggesting an inflammatory or neoplastic process.

To objectively determine the cause of the patient's symptoms, a contrast-enhanced chest CT scan was performed. Imaging showed a soft tissue mass with osteolytic dama-

ge to the left 9th rib, measuring 4.2×5.6cm. Axial CT images in the mediastinal window displayed arterial phase Hounsfield Units (HU) of 38 (A), venous phase HU of 37 (B), and delayed phase HU of 39 (C). A follow-up non-contrast CT scan two months later, indicated that the lesion had expanded to 5.3x 7.6cm in axial views through the mediastinal (D) and bone windows (E). A technetium-99m-methyl diphosphonate (^{99m}Tc-MDP) SPECT bone scan on February 16, 2023 (F), revealed markedly decreased tracer uptake in the area of the left 9th rib's soft tissue mass and increased tracer accumulation in the adjacent rib (Figure 1).

The lesion's rapid enlargement suggests the potential for malignant processes, such as metastatic cancer or a solitary plasmacytoma. Elevated tracer uptake was also noted on the left side of the T10 vertebra, yet the lack of definitive bone destruction on CT might not indicate a pathological state, possibly reflecting reactive or other benign conditions.

Following the procedure, a partial excisional biopsy of the mass on the left rib was conducted to determine its pathological type. Postoperative pathology, utilizing Hematoxylin and Eosin (H&E) staining at 200x magnification, revealed a sclerosing liposarcoma characterized by a dense fibrous matrix, mature adipose tissue, and scattered lipoblasts. Immunohistochemistry results were as follows: Bcl-2 (partially +), CD34 (-), CD99 (-), CK (pan) (+), Desmin (-), Ki67 (+), approximately 40%), S-100 (-), SMA (-), STAT6 (-), Vimentin (+), ALK (-), CDK4 (partially +), EMA (slightly +), and MDM2 (+) (Figure 2). These findings suggest a diagnosis of low-grade sclerosing liposarcoma.

Additionally, there was increased tracer uptake on the left side of the T10 vertebra; however, the absence of clear evidence of bone destruction on CT suggests that this finding may not necessarily indicate a pathological process, potentially representing reactive or other non-malignant changes.

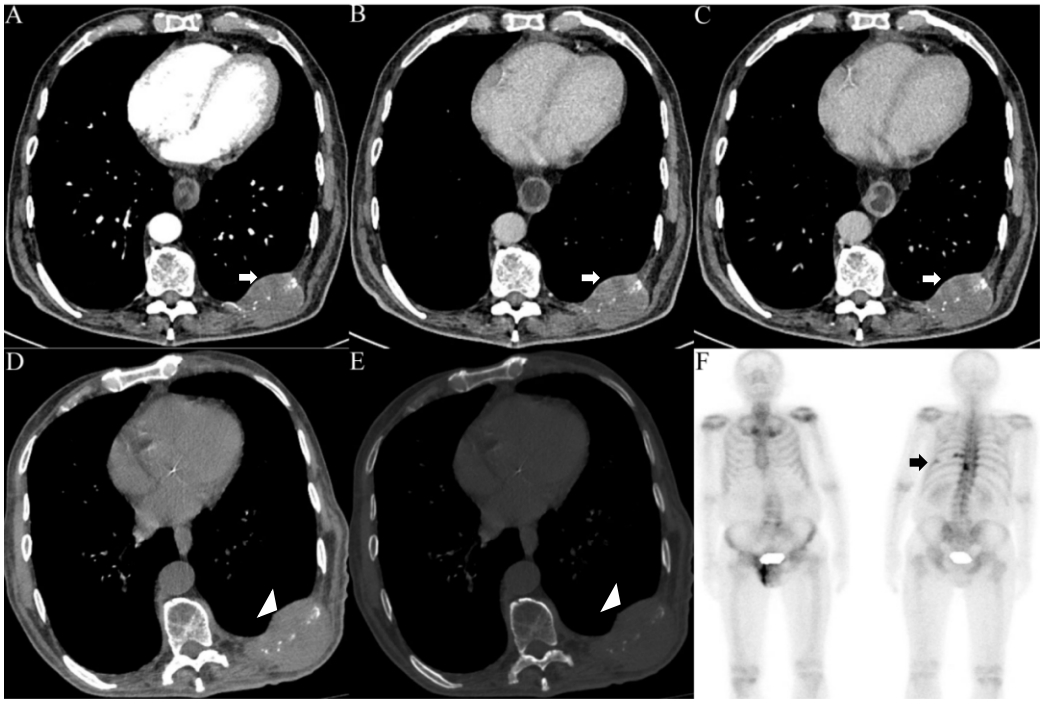

Figure 1. A 77-year-old male patient presented with two months of persistent upper abdominal pain and was subjected to an enhanced chest CT scan. The scan revealed a soft tissue mass with osteolytic destruction at the left 9th rib, measuring 4.2x5.6cm. Axial CT images in the mediastinal window (white arrows) demonstrated the lesion with arterial phase Hounsfield Units (HU) of 38 (A), venous phase of 37 HU (B), and delayed phase of 39 HU (C). Follow-up non-contrast CT two months later, showed an increase in size of the osteolytic lesion to 5.3x7.6cm on axial images (white arrowheads) through the mediastinal (D) and bone windows (E). Technetium-99m-MDP scintigraphy (F) depicted a significant decrease in tracer uptake in the left 9th rib soft tissue mass area with increased surrounding bone uptake (black arrow).

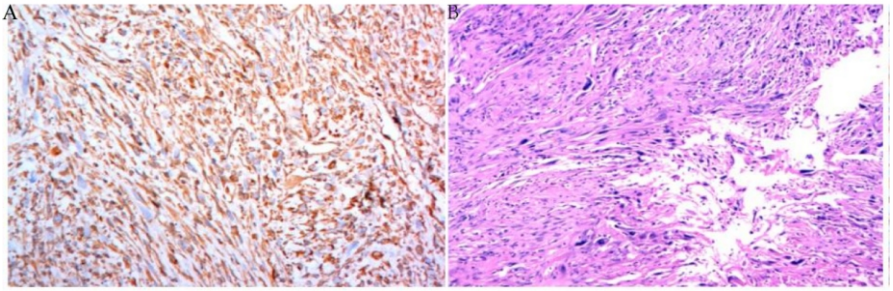

Figure 2. (A) Immunohistochemistry (200x) shows cells that are positive for the mesenchymal marker Vimentin. (B) Hematoxylin and eosin staining (200x) reveals a sclerosing liposarcoma, characterized by a dense fibrous matrix interspersed with mature adipose tissue and scattered lipoblasts.

Discussion

Liposarcoma is the most common form of soft tissue sarcoma and usually develops in the extremities or retroperitoneum [1]. The disease is classified into well-differentiated, myxoid or round cell, dedifferentiated, and pleomorphic subtypes, each with distinct pathological and clinical features. Among these subtypes, sclerosing liposarcoma is a subtype of well-differentiated liposarcoma. Therefore, as indicated by the pathology in this case, the malignancy is relatively low.

Primary intraosseous liposarcoma is a rare malignant neoplasm of the skeletal system, with the first case reported by Stewart in 1931 [2]. To better review the radiographic characteristics of bone liposarcoma, we conducted a literature review on bone liposarcoma in the Web of Science.

Taylor et al. (2018) [3] reported a case of a 54-year-old male with a recurrent thigh mass suggestive of an atypical mosaic-patterned lipomatous tumor intermixed with high-grade dedifferentiated liposarcoma, confirmed by PET/CT imaging showing slight metabolic activity, and histopathological examination revealing foci of high-grade spindle cell sarcoma co-mingling with the underlying atypical lipomatous tumor. In contrast to the low malignancy of well-differentiated liposarcomas, an 18-year-old male was diagnosed with a pleomorphic liposarcoma on the distal shaft of the left tibia, characterized by partial sclerotic lesions on plain radiographs, a heterogeneous tumor that was predominantly hyperintense on T1-weighted and T2-weighted MRI sequences with fat saturation suppression, and significant fluorine-18-fluorodeoxyglucose (^{18}F-FDG) uptake on positron emission tomography (PET)/CT scan (standardized uptake value (SUV)=17.1) showing metastases in the distal end of the right femur and multiple small lung metastases [4]. Ramirez et al. (2018) [5] reported on a 68-year-old woman with an atypical lipomatous tumor in the soft tissue of her left hand. The tumor was initially identified through magnetic resonance imaging (MRI) and X-ray as a large, calcified mass causing finger displacement. Despite biopsy and staged debulking, the tumor recurred locally and dedifferentiated. The patient has experienced metastasis to various parts of the body, including the lungs, pancreas, bone, and soft tissue. A whole-body PET scan conducted a year after debulking revealed intense ^{18}F-FDG uptake in the original mass, a nodule in the right lung base, left subpectoral adenopathy, and a mid-arm nodule, indicating extensive metabolic activity [5]. Zajicek et al. (2017) [6] reported on a 59-year-old male with a dedifferentiated liposarcoma in the lower extremity, notable for low-grade dedifferentiation and a rare low-grade osteosarcomatous component. This case highlights the complex nature of liposarcomas, emphasizing the unpredictable course and potential for aggressive behavior, including bone involvement and metastasis. Key findings included MDM2 and CDK4 amplification, underscoring the genetic aspects of dedifferentiation. The evolution of imaging features over time, from unrecognized myositis ossificans to pathologic fracture, illustrates the diagnostic challenges and the importance of thorough and ongoing imaging surveillance. Ultimately, this case, alongside others, underscores the diversity of liposarcoma presentations and the critical need for advanced diagnostic and therapeutic strategies to improve outcomes in such aggressive and complex cases.

Charest et al. (2008) [7] reported a case involving a 46-year-old male who was incidentally discovered to have a significant mass in the proximal right thigh during a routine examination. Further diagnostic evaluations, including a CT scan and MRI of the right thigh, revealed an hourglass-shaped soft tissue mass. The mass was subsequently classified as a myxoid liposarcoma, noted for the absence of round cell components through fine needle and trucut biopsies. An incidental osseous lesion was detected in the distal right tibia through a ^{99m}Tc-MDP whole-body bone scan. This presented a diagnostic challenge due to its localization and characteristics. The tibial lesion exhibited a notably higher ^{18}F-FDG uptake on PET/CT compared to the primary sarcoma, with a maximum SUV of 7.2. This was in stark contrast to the mild hypermetabolism (SUV 3.2) of the diagnosed liposarcoma. Upon further investigation, the lesion was identified as benign fibrous dysplasia.

The case reported by Charest et al. (2008) [7] serves as a pivotal example of the complexity involved in utilizing advanced imaging modalities like ^{18}F-FDG PET/CT and ^{99m}Tc-MDP whole-body bone scans in the oncological domain. This case contrasts the metabolic activities of a myxoid liposarcoma and incidental fibrous dysplasia in the same patient, underscoring the critical need for a nuanced understanding of imaging outcomes. Although ^{18}F-FDG PET/CT is highly sensitive in detecting metabolic differences across tissues, it has limitations in distinguishing between benign and malignant lesions based solely on metabolic activity. Therefore, cautious interpretation is necessary. An example of this is the incidental detection of fibrous dysplasia, which has higher ^{18}F-FDG uptake than the known malignancy, illustrating the potential for diagnostic challenges. This highlights the importance of comprehensive clinical evaluation. These scenarios highlight the significance of integrating metabolic and anatomical imaging results with clinical and histopathological insights to effectively navigate the complexities of diagnosis and staging, ensuring precise and personalized patient management.

In Nuclear Medicine, the ^{99m}Tc-MDP whole-body bone scan is commonly used to assess bone lesions. In this case, there was decreased radiotracer distribution in the left 9th rib with increased surrounding bone uptake. Additionally, an abnormal increase in radiotracer distribution was noted on the left side of the T10 vertebra. This finding is inconsistent with the metabolic pattern of the primary lesion, which requires careful interpretation of the results. Upon further review of the CT imaging, it was discovered that there was a decrease in bone density and coarsening of trabeculae in the T10 vertebra. However, there was no clear evidence of osteolytic bone destruction. Due to the patient's advanced age, metastasis from liposarcoma was not initially considered. Instead, reactive changes or other benign conditions were considered. Close follow-up and re-evaluation are recommended.

Unfortunately, further evaluation with PET/CT was not conducted to determine whether it was a primary bone liposar-

coma or metastatic liposarcoma. However, based on the pathology of low-grade malignant sclerosing liposarcoma and the lesion's localized nature, which is distinct from the biological characteristics of dedifferentiated and pleomorphic liposarcomas that often metastasize distantly, the initial inclination was towards a primary bone liposarcoma. The patient is currently receiving standardized chemotherapy and will undergo further follow-up. If necessary, additional prognosis assessment will be conducted using PET/CT.

Additionally, it is imperative to note that a retrospective study underscores the high incidence of extrapulmonary metastases in myxoid liposarcoma, primarily involving bones and soft tissues. Computed tomography imaging unveiled a spectrum of lytic and sclerotic changes, progressing to extensive bone destruction in later stages. Magnetic resonance imaging revealed significant heterogeneous enhancement in bone metastases. On the contrary, ^{18}F-FDG PET/CT scans failed to detect considerable ^{18}F-FDG uptake in any of the metastases, highlighting the unique imaging profiles of myxoid liposarcoma as opposed to its dedifferentiated counterpart. Thus, MRI is distinguished as the premier imaging technique for the evaluation of bone and soft tissue metastases in myxoid liposarcoma scenarios [8]. A 53-year-old male with myxoid liposarcoma experienced back pain due to vertebral metastasis in eight vertebrae, underscoring that bone scans and ^{18}F-FDG PET can fail to detect such metastases, even with extraskeletal extensions or in the presence of a pathological fracture [9].

In conclusion, bone liposarcoma presents a complex and diverse pathological landscape, with both primary and metastatic occurrences. This malignancy includes various pathological types, each with unique biological behaviors and imaging signatures. Therefore, a comprehensive and integrated approach is necessary for its assessment, which involves clinical evaluation, detailed histopathological analysis, and genetic testing. A multidisciplinary approach is essential for accurately diagnosing the specific subtype of liposarcoma, understanding its potential for malignancy, and creating a tailored treatment plan. This approach aids in the precise characterization of the disease and paves the way for more effective and personalized therapeutic interventions, ultimately aiming to improve the prognosis and quality of life for patients with this challenging condition [10-12].

The authors declare that they have no conflicts of interest

Acknowledgments
We express our gratitude to Professor Zhang, a renowned expert in soft tissue pathology, for his precise diagnosis of the liposarcoma subtype.

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