Scholars Journal of Medical Case Reports
Abbreviated Key Title: Sch J Med Case Rep
ISSN 2347-9507 (Print) | ISSN 2347-6559 (Online)
Journal homepage: https://saspublishers.com

OPEN ACCESS

Pathology

Bilateral Pleural Metastasis from Secondary Osteosarcoma Arising in a Previously Treated Giant Cell Tumor of Bone in a Young Woman: A Rare Case Report

Dr. Kharidehal Durga[1]*, Dr. Galeti Ayesha[2], Dr. B. Syam Sundara Rao[3], Dr. Rudresh Hiremath[4], Dr. Ramakrishna Rao[5] Baru

[1]Professor and HOD, Department of pathology, Narayana medical college Hospital, Nellore, Andhrapradesh, India
[2]Assistant Professor Department of Pathology, Narayana Medical College Hospital, Nellore, Andhra Pradesh, India.
[3]Professor, Department of Pathology, Narayana Medical College Hospital, Nellore, Andhra Pradesh, India.
[4]Professor, Department of Radiology, Narayana Medical College Hospital, Nellore, Andhra Pradesh, India.
[5]Professor and HOD Department of Radiology, Narayana Medical College Hospital, Nellore, Andhra Pradesh, India.

DOI: https://doi.org/10.36347/sjmcr.2026.v14i11.016 | **Received:** 24.08.2026 | **Accepted:** 29.10.2026 | **Published:** 05.11.2026

***Corresponding author:** Dr. Kharidehal Durga
Professor and HOD, Department of pathology, Narayana medical college Hospital, Nellore, Andhrapradesh, India

Abstract **Case Report**

Giant cell tumor of bone (GCTB) is an intermediate, locally aggressive primary bone neoplasm that rarely undergoes malignant transformation. Secondary osteosarcoma arising in GCTB is exceptionally uncommon, particularly in the absence of prior radiotherapy. Although the lungs are the most common site of distant metastasis in osteosarcoma, pleural involvement is rare. We report a rare case of a 27-year-old woman with a history of surgically treated GCTB who subsequently developed bilateral pleural lesions diagnosed as metastatic osteosarcoma. The case highlights the diagnostic challenges associated with identifying secondary malignant transformation in a patient with a previous history of conventional GCTB. Correlation of the clinical history, radiological findings, and histopathological features is essential to establish the diagnosis and distinguish malignant transformation from conventional metastatic GCTB and other primary pleural malignancies.

Keywords: Giant cell tumor of bone, secondary malignant transformation, osteosarcoma, pleural metastasis, histopathology.

Copyright © 2026 The Author(s): This is an open-access article distributed under the terms of the Creative Commons Attribution **4.0 International License (CC BY-NC 4.0)** which permits unrestricted use, distribution, and reproduction in any medium for non-commercial use provided the original author and source are credited.

INTRODUCTION

Giant cell tumor of bone (GCTB) is a locally aggressive primary bone neoplasm that predominantly affects skeletally mature young adults, with a predilection for the epiphyseal or epimetaphyseal regions of long bones. Although traditionally regarded as a benign tumor, GCTB is currently classified as an intermediate, locally aggressive neoplasm because of its capacity for local recurrence and, less frequently pulmonary metastasis. Pulmonary metastases occur in a small proportion of patients, generally consisting morphologically of conventional GCTB [1–4].

Malignant transformation of GCTB is an uncommon but well-recognized phenomenon. It may occur as a primary malignant GCTB, in which a high-grade sarcoma is identified synchronously with conventional GCTB, or as a secondary malignant transformation arising after a previously documented benign GCTB. Secondary malignant tumors include osteosarcoma and undifferentiated pleomorphic sarcoma. Osteosarcomatous transformation is particularly rare, and spontaneous transformation in the absence of previous radiotherapy has been documented in only a small number of cases [5–9].

The diagnosis of secondary osteosarcoma is particularly challenging when the original tumor and the recurrent/metastatic tumor are separated by a substantial interval. Demonstration of malignant osteoid produced by malignant mesenchymal cells remains the defining histological feature of osteosarcoma. Molecular studies have further supported the concept of clonal progression in selected cases, with persistence of H3F3A alterations reported in both the original GCTB and subsequent osteosarcoma [8,10].

The lung is the most common site of distant metastasis for osteosarcoma. In contrast, pleural involvement is distinctly uncommon and is usually associated with pulmonary metastatic disease. Radiologically, pleural metastases may manifest as focal or multiple pleural nodules, nodular pleural thickening, pleural masses, pleural effusion and in osteogenic tumors, areas of pleural mineralization or ossification [11,12].

We report a rare case of a 27-year-old woman with a previous history of surgically treated GCTB who subsequently developed bilateral pleural osteosarcoma metastatic disease. The case highlights the importance of recognizing malignant transformation in patients with a history of GCTB and emphasizes the diagnostic significance of correlating current pleural lesions with the patient's previous bone tumor.

CASE REPORT

A 27-year-old woman presented with respiratory symptoms/pleuritic chest pain, dyspnea with wheeze and cough with expectoration since 5 months. She had a significant past history of a ?? Giant cell tumor of bone involving the right knee for which she had undergone above knee amputation performed 2 years back.

The original histopathological material/slides were not available with the patient. Chest X ray revealed bilateral loculated pleural effusion (Fig 1).

CECT chest shows multiple bilateral heterogeneously enhancing pleural masse lesions with areas of calcification involving both hemithoraces with rib erosions. The radiological findings raised the possibility of pleural synovial sarcoma, pleural metastases and tuberculosis (Fig 2).



Fig.1: Chest X ray revealed bilateral loculated pleural effusion.

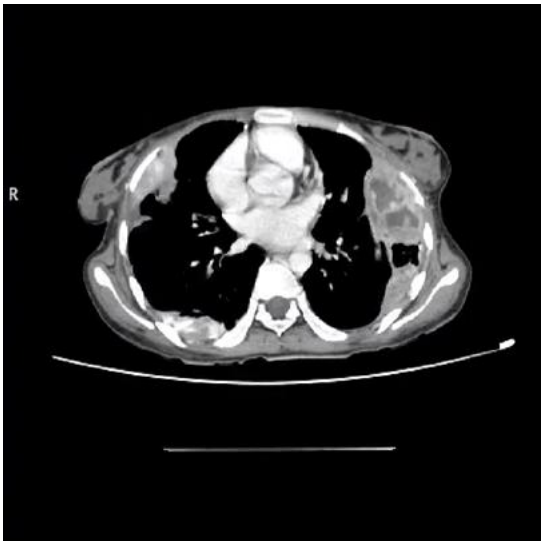


Fig.2: CECT chest shows multiple bilateral heterogeneously enhancing pleural mass lesions with areas of calcification involving both hemithoraces with rib erosions

Ultrasound guided biopsies were performed on bilateral pleural lesions. Histologically, the lesion was composed of highly atypical malignant spindle to polygonal cells arranged in loose fascicles, showing marked nuclear pleomorphism, hyperchromasia, prominent nucleoli and frequent mitotic figures, including atypical mitoses. In most areas irregular lace-like osteoid directly associated with malignant tumor cells was noted. Occational osteoclastic giant cells, fragments of mature bone were also seen. many dilated vascular channels lined by plump endothelial cells were seen within the tumour tissue (Fig 3,4).

There was no presence of conventional GCT foci noted in the deposits Immunohistochemical for Vimentin – Strongly positive (Fig .5), osteoid not stained, SAT B2 nuclear positivity of mesenchymal cells.

Correlation with the previous histopathological diagnosis of GCTB supported the interpretation of the current lesion as a secondary osteosarcoma arising in the setting of previously treated GCTB.

The patient was subsequently evaluated by the multidisciplinary sarcoma team and received chemotherapy regimen.

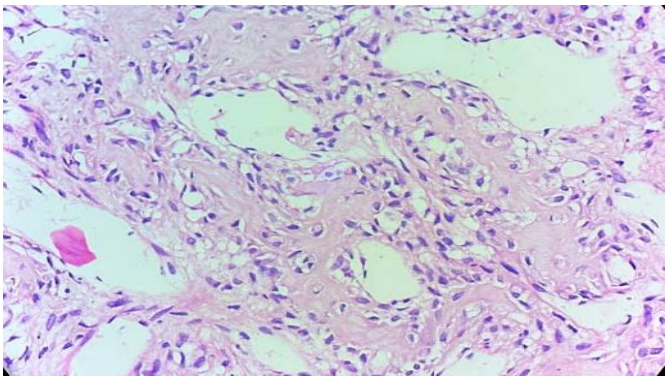


Fig.3: Irregular lace-like osteoid directly associated with malignant tumor cells

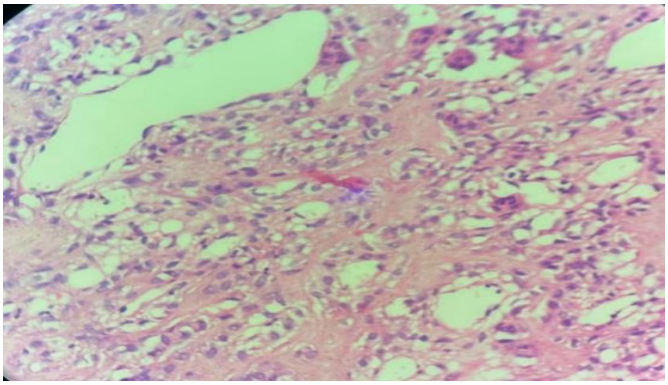


Fig.4: osteoclastic giant cells, fragments of mature bone and dilated vascular channels lined by plump endothelial cells were seen within the tumour tissue

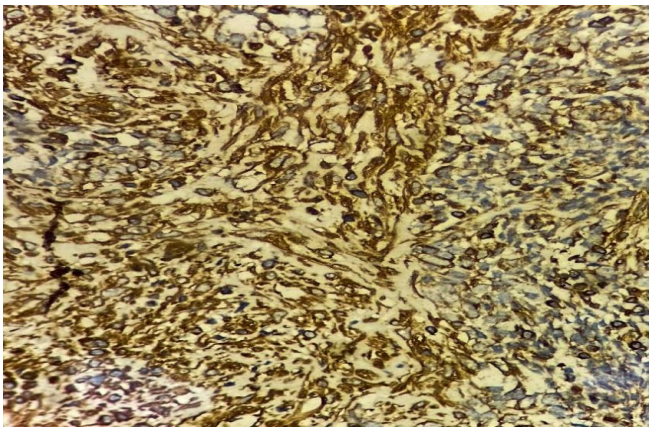


Fig.5: Immunohistochemistry shows strong positivity for vimentin

DISCUSSION

The present case is unusual because it demonstrates the intersection of two rare events: malignant transformation of a previously treated GCTB into osteosarcoma and subsequent bilateral pleural dissemination. GCTB generally behaves as a locally aggressive tumor, although a small proportion of patients develop pulmonary metastases. These pulmonary deposits usually retain the morphology of conventional GCTB and should not automatically be interpreted as malignant transformation [1–4].

Malignant transformation represents a distinctly different biological event. The reported frequency varies among series, reflecting differences in diagnostic criteria, treatment history and duration of follow-up, but the phenomenon remains uncommon. Secondary malignant transformation may occur years after the original diagnosis, emphasizing that a remote history of GCTB remains highly relevant when evaluating a new mass or metastatic lesion [5–7].

Secondary osteosarcoma is one of the recognized patterns of malignant transformation of GCTB. Histologically, the diagnosis rests on identifying a malignant mesenchymal neoplasm producing malignant osteoid. This distinction is important because GCTB itself may contain osteoclast-like giant cells and may show reactive bone formation, particularly after treatment. Reactive bone formation should not be confused with malignant osteoid. The latter is characterized by irregular, lace-like or "wickerwork" osteoid intimately associated with atypical malignant cells.

The distinction between conventional GCTB, malignant GCTB and secondary osteosarcoma can therefore be challenging, particularly in small biopsies. In the appropriate clinical setting, review of the original tumor is extremely valuable. Demonstration of a previously documented benign GCTB followed by development of a high-grade osteogenic sarcoma at a later time strongly supports secondary osteosarcoma. Recent studies have demonstrated that some secondary osteosarcomas arising from GCTB retain molecular alterations characteristic of GCTB, including H3F3A mutations, supporting the concept of biological continuity between the two lesions. [8,10] However, its interpretation in a malignant lesion requires caution, because H3F3A alterations can occasionally be encountered in other bone sarcomas. Molecular findings should therefore be interpreted in conjunction with morphology and clinical history [8,13].

An additional important issue in the present case is the unusual metastatic distribution. Osteosarcoma most commonly metastasize hematogenously to the lungs and less frequently to bone and other sites. Pleural metastases are uncommon. A recent radiological review of thoracic osteosarcoma metastases described pleural disease as typically occurring in association with pulmonary metastases, with imaging manifestations ranging from focal or multiple pleural nodules to diffuse pleural thickening and pleural masses. Pleural mineralization or ossification may be particularly suggestive of metastatic osteogenic disease [11].

The rarity of pleural metastasis is further illustrated by published case reports. Bilateral pleural metastatic disease with calcified pleural deposits has been described in young patients with osteosarcoma, and the presence of osteoid-producing tumor can result in conspicuous mineralization on imaging [12]. Thus, in a patient with a history of osteosarcoma or secondary osteosarcoma, bilateral pleural nodularity should prompt consideration of metastatic disease, even when the dominant radiological abnormality is pleural rather than pulmonary.

The present case also raises an important diagnostic consideration: whether the pleural lesion represents metastasis from a secondary osteosarcoma arising at the original GCTB site or represents metastatic conventional GCTB with subsequent malignant transformation. The distinction depends on morphology and, where available, comparison with the primary/recurrent lesion. If the pleural lesion demonstrates unequivocal high-grade malignant morphology with malignant osteoid production, the most appropriate diagnosis is metastatic osteosarcoma as in our case.

Secondary osteosarcomatous transformation of a previously treated giant cell tumor of bone is a rare but serious complication. Bilateral pleural metastases represent an unusual pattern of dissemination and may pose a significant diagnostic challenge specifically in the absence of ipsilateral pulmonary deposits.

In patients with a history of GCTB who develop new pulmonary or pleural lesions, the possibility of malignant transformation should be actively considered.

Management and prognosis in case of metastatic Osteosarcomas is determined by the number, site and size of metastasis [15].

CONCLUSION

Secondary osteosarcomatous transformation of GCTB with bilateral pleural metastases represents an exceptionally rare clinical presentation. This case emphasizes the importance of considering malignant transformation in patients with a history of GCTB who develop new thoracic lesions. Careful histopathological evaluation and correlation with the original bone tumor are crucial for accurate diagnosis and appropriate clinical management.

REFERENCES

1. WHO Classification of Tumours Editorial Board. Soft tissue and bone tumours. 5th ed. Lyon: International Agency for Research on Cancer; 2020.
2. Palmerini E, Picci P, Reichardt P, Downey G. Malignant giant cell tumor of bone: a review of the literature. Eur J Cancer. 2019;113:23-35.
3. Balke M, Schremper L, Gebert C, Ahrens H, Streitbuerger A, Koehler G, *et al.,* Giant cell tumor of bone: treatment and outcome of 214 cases. J Cancer Res Clin Oncol. 2008;134(9):969-78.
4. Viswanathan S, Jambhekar NA. Metastatic giant cell tumor of bone: are there any factors affecting the development of pulmonary metastases? Clin Orthop Relat Res. 2010;468(6):1641-7.
5. Bertoni F, Bacchini P, Capanna R, Ruggieri P, Biagini R, Picci P, *et al.,*Malignancy in giant cell tumor of bone. Cancer. 1997;79(9):1722-30.
6. Miller IJ, Blank A, Yin SM, McNickle A, Gray R, Gitelis S. A case of recurrent giant cell tumor of bone with malignant transformation and benign pulmonary metastases. Diagn Pathol. 2010;5:62.
7. Palmerini E, Saeter G, Gabella F, Picci P, Gambarotti M, Donati DM, *et al.,* Malignant transformation of giant cell tumour of bone: a review of literature and the experience of a referral centre. Cancer Treat Rev. 2022;107:102408.
8. Matsunobu T, Maekawa A, Jotatsu M, Makihara K, Hisaoka M, Iwamoto Y. Malignant transformation of giant cell tumor of bone 7 years after initial surgery: a case report and literature review. JBJS Case Connect. 2021;11(2).00417.
9. Brien EW, Mirra JM, Kessler S, Suen M, Ho JK, Yang WT. Benign giant cell tumor of bone with osteosarcomatous transformation ("dedifferentiated" primary malignant GCT): report of two cases. Skeletal Radiol. 1997;26(4):246-55.
10. Nielsen GP, Rosenberg AE. Diagnostic pathology: bone. 2nd ed. Philadelphia: Elsevier; 2018.
11. Choi Y, *et al.,* Osteosarcoma metastasis to the thorax: a pictorial review of chest computed tomography findings. Diagnostics (Basel). 2024;14(18):2085.
12. Yadav P, *et al.,* Diffuse pleural metastases of osteosarcoma detected by bone scan. Clin Case Rep. 2020;8: [page numbers to be verified].
13. Righi A, Paioli A, Cevolani L, *et al.,*H3F3A mutation as a marker of malignant giant cell tumor of the bone: a case report and review of literature. J Cancer Res Ther. 2023;19: [page numbers to be verified].
14. Ahuja S, Khan AA, Kabra A, Zaheer S. Giant cell tumor of bone with late-onset lung metastasis and secondary osteosarcomatous transformat
15. L. Pochanugool, T. Subhadharaphandou, M. Dhanachai, P. Hathirat, D. Sangthawan, R. Pirabul, et al, Prognostic factors among 130 patients with Osteosarcoma , Clin Orthop.Relat Res 345 (1997) 206- 214.