

BRIEF REPORT

A case report of rib osteosarcoma and literature review

Xiaofeng Hu¹ | Tianyi Bao²  | Chao Yan² | Yongliang Zhu² | Xiaofei Zheng¹

¹Department of Orthopaedics, Jinling Hospital of Nanjing Medical University, Nanjing, China

²Department of Orthopaedics, Nanjing Central Hospital, Nanjing, China

Correspondence

Xiaofei Zheng, Department of Orthopaedics, Jinling Hospital of Nanjing Medical University, 305 Zhongshan East Road, Nanjing 210002, Jiangsu Province, China.

Email: zxf163yxh@163.com

Yongliang Zhu, Department of Orthopaedics, Nanjing Central Hospital, No. 116 Chengxian Street, Nanjing 210001, Jiangsu Province, China.

Email: 18204314934@163.com

Abstract

About half of osteosarcomas occur near the knee joint, but other sites such as the humerus, upper femur, fibula, spine, and ilium can also occur. However, rib osteosarcoma is rarely reported. Here, we report the case of a 17-year-old female who was found to have a left dorsal mass on physical examination. Computed tomography (CT) revealed bone destruction in the seventh rib, leading to surgery for mass excision. Pathological results suggested chondroblastic osteosarcoma. After surgery, the patient was treated with chemotherapy and is doing well.

KEYWORDS

osteosarcoma, rib, surgical treatment

1 | INTRODUCTION

Osteosarcoma is a malignant mesenchymal tumor. It is mainly manifested as the formation of immature osteoid cells by tumor cells. Osteosarcoma is the most common primary bone tumor in children and adolescents, with a tendency to occur in the metaphysis of the long shaft. Osteosarcoma occurring at the rib end is very rare, and here, we report a case of primary rib osteosarcoma treated in an adolescent girl in order to provide some diagnostic aid to clinicians.

2 | CASE REPORT

We admitted a 17-year-old adolescent girl. She has a 1-month history of swelling and pain in her left chest and back. During a school-organized physical examination 4 days ago, a lump was found on her left back. The

patient has no symptoms such as fever, cough, dyspnea, and weight loss. We examined the patient and found that a lump could be felt at the level of approximately the seventh rib on the left chest and back, with an area of approximately 2.0×3.0 cm. The lump feels tough and lacks mobility, causing pain when pressed on this area. Complete blood cell count and serum biochemistry are within the normal range. Chest x-ray showed rib destruction and surrounding mass. The results of chest computed tomography (CT) showed that the seventh rib on the left side of the patient was partially damaged, with a soft tissue mass surrounding it, and no pleural effusion was found (Figures 1 and 2). Emission computed tomography (ECT) showed that the local strip radioactive uptake of the left seventh posterior rib increased, indicating that bone metabolism was active.

During the operation, the seventh rib mass protruded to the medial side. The mass is tough and has no obvious adhesion with the surrounding, and the capsule is intact. In order to completely remove the mass, we carefully

Xiaofeng Hu and Tianyi Bao contributed equally to this paper.

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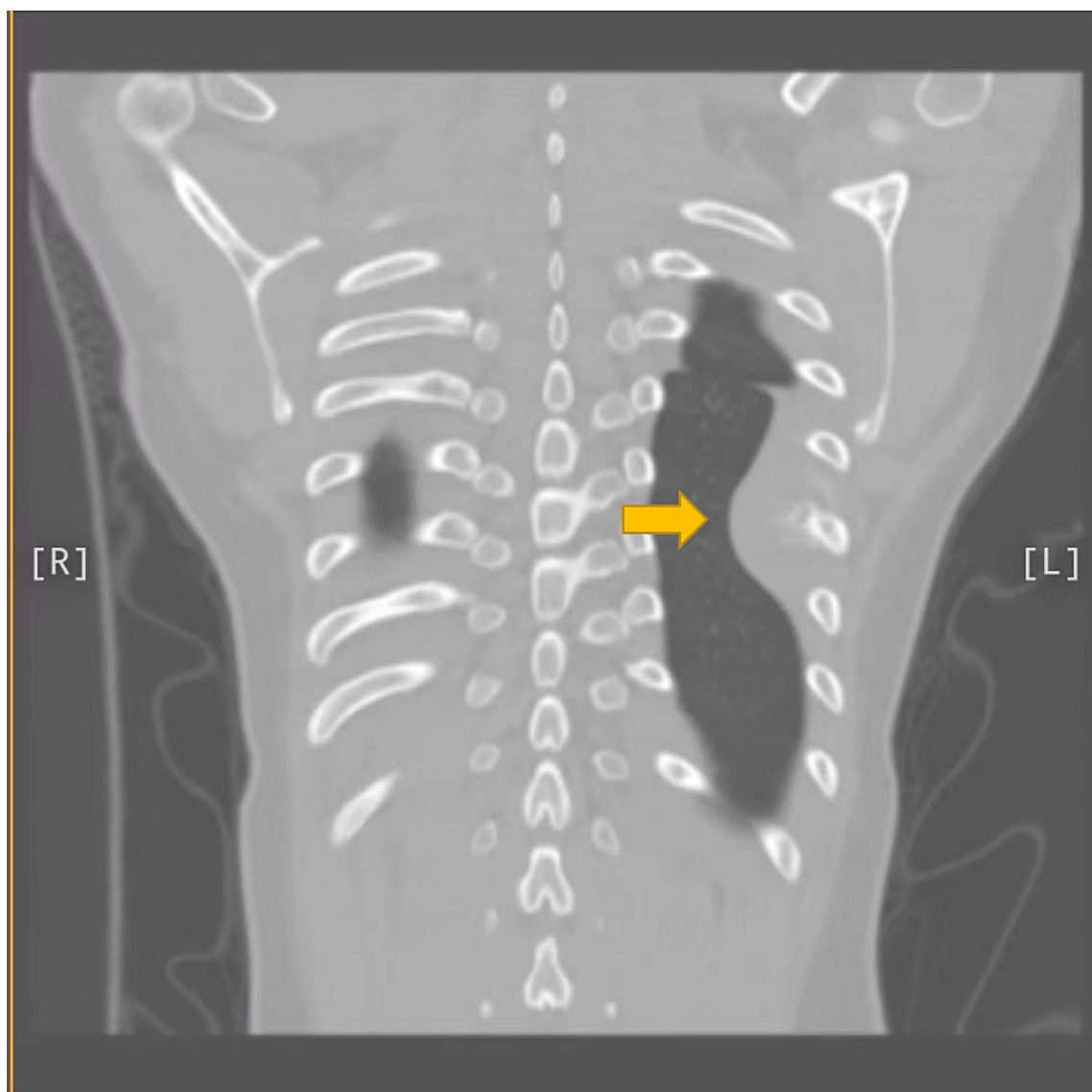


FIGURE 1 Preoperative coronal CT of patient. Notice a semicircle mass (pointed by yellow arrow) on left hemithorax and destruction of the seventh ribs.

separated the mass, removed part of the latissimus dorsi muscle, and cut the ribs at the normal tissue on both sides of the mass (Figure 3A). The mass was histopathologically diagnosed as chondroblastic osteosarcoma (Figure 3B,C). After surgery, the patient was transferred to the oncology department for chemotherapy. Chemotherapy was initiated with a protocol consisting of cyclophosphamide (2 g) in the first to third day, doxorubicin liposome (40 mg) in the first day and lobaplatin (40 mg) in the second day of the treatment. It is supplemented by symptomatic treatment such as stomach protection, liver protection, and antiemesis. After the treatment, the patient voluntarily asked for discharge. The patient is required to return to the hospital regularly for re-examination.

The patient has lived normally for one and a half years after treatment, and no special changes have been found in the ribs or other parts of the body.

3 | DISCUSSION

Osteosarcoma is one of the primary malignant tumors of the bone, which is more common in males. The incidence rate of osteosarcoma showed a bimodal pattern, reaching its peak at the age of 18 and 60 respectively.¹ Osteosarcoma mainly occurs around the knee (50%) and near the humerus (15%).² It is worth noting that osteosarcoma rarely occurs in flat bones, so there are few reported cases of rib osteosarcoma.

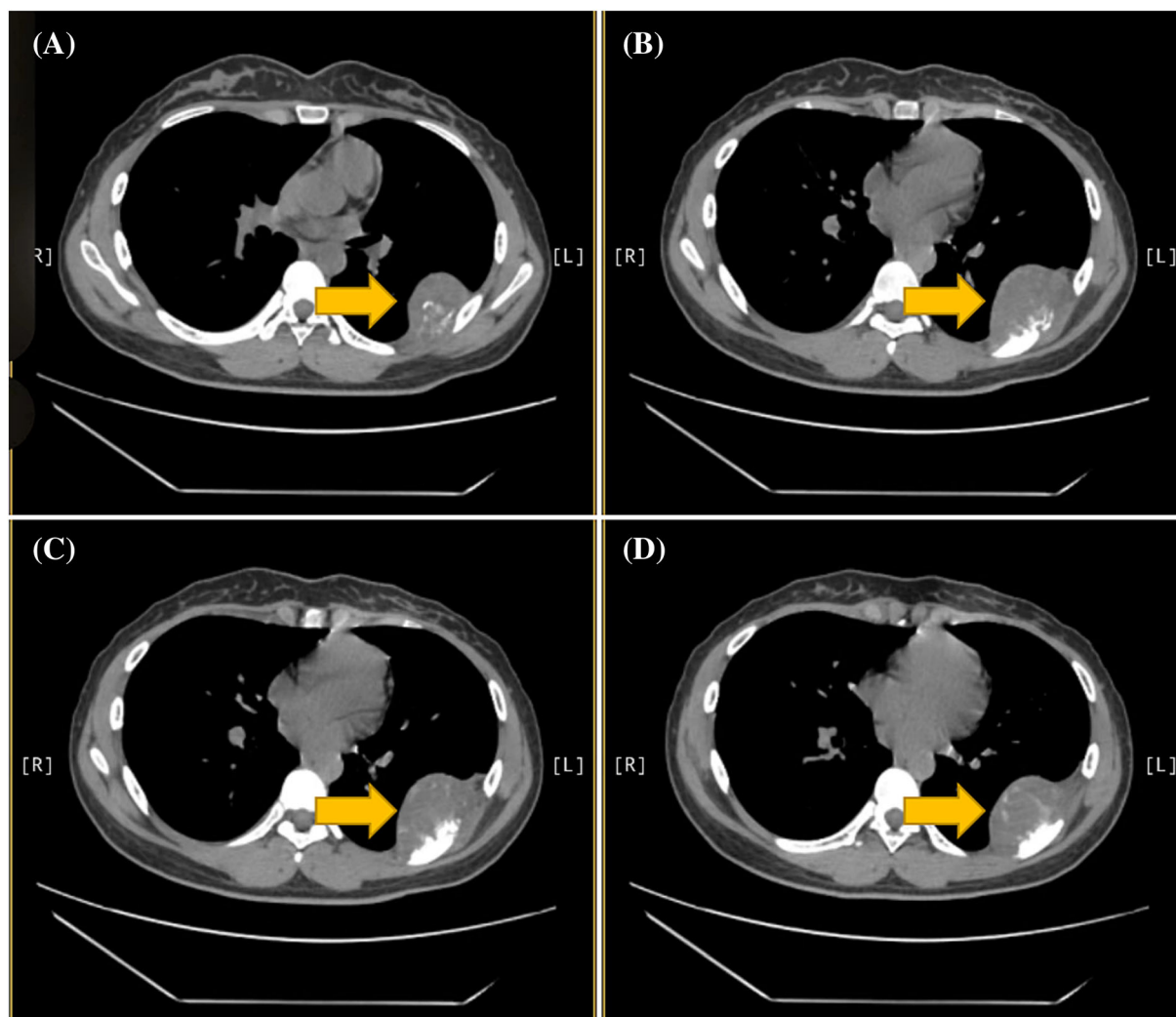


FIGURE 2 Preoperative CT showed destruction of the seventh rib and surrounded by mass (pointed by yellow arrow).

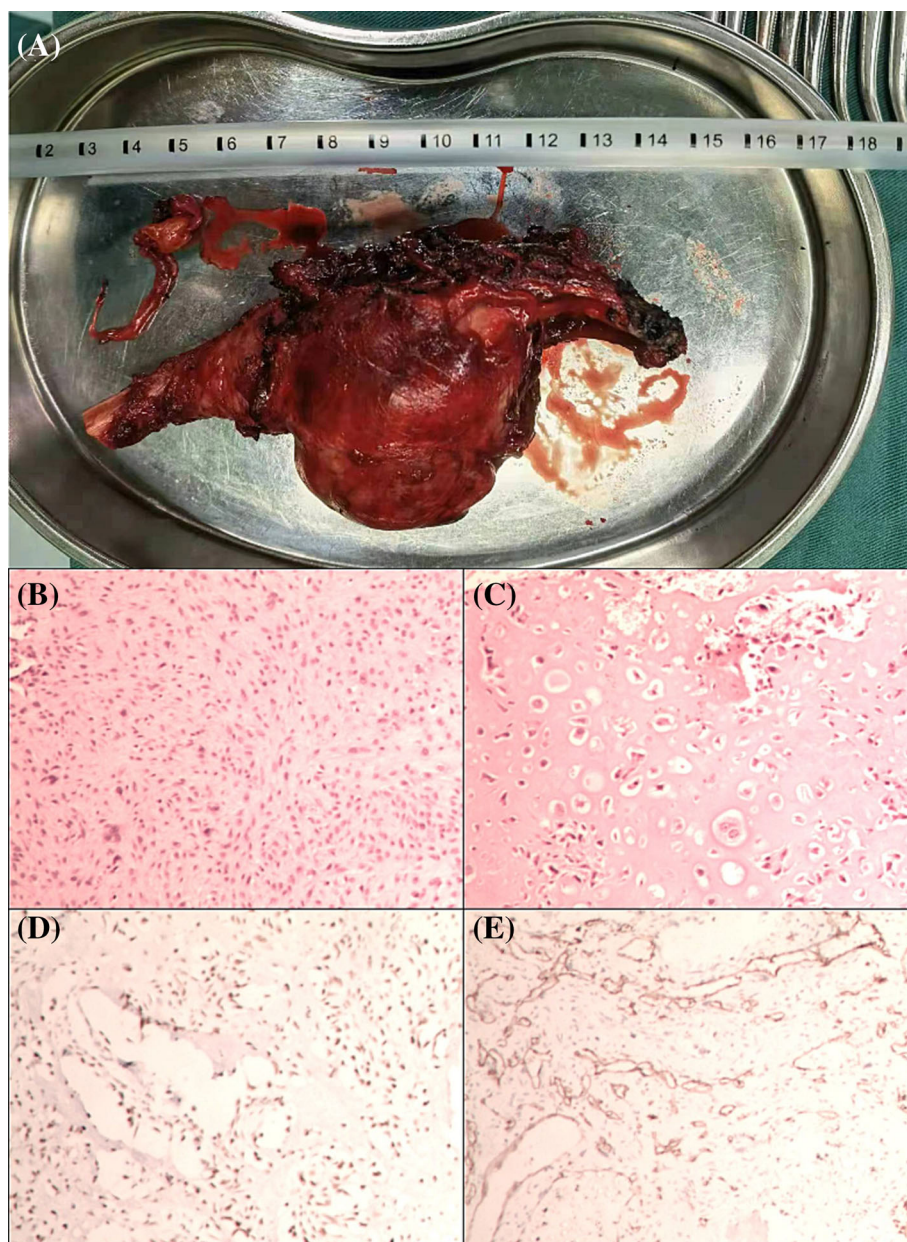
Wardoyo et al. found that rib osteosarcoma accounts for only 1.25% of all osteosarcoma cases.³ Burt et al. reported 1435 cases of osteosarcoma, with only 13 cases (0.9%) of primary rib osteosarcoma.⁴ Similarly, Bielack et al. found 14 cases (0.8%) of rib osteosarcoma out of 1702 cases.⁵ Among all types of osteosarcoma patients, the incidence rate of male patients is about 1.5 times that of female patients.⁶

Almost all patients had lumps or pain or both, and some had other symptoms such as cough, dyspnea, and hemoptysis.⁷ The overall 5-year survival rate for localized osteosarcoma is 60% to 80%,⁶ with a survival rate of approximately 60% for children and adolescents.⁸ Among metastatic osteosarcoma, patients with lung metastasis have the worst prognosis, with a 3-year survival rate of less than 30%.⁹ Fractures may also be a more invasive indicator of diseases. Patients with fractures have a higher incidence of lung metastasis at the onset and after treatment.¹⁰

At present, the key population for screening osteosarcoma is mainly patients with hereditary cancer susceptibility syndrome. The screening strategy advocates increasing awareness of the risk of osteosarcoma and conducting comprehensive physical examinations every year. For patients carrying the pathogenic TP53 germline variants, it is necessary to focus on monitoring blood indicators and imaging changes, which can help physicians detect solid tumors in patients early and improve their long-term survival rate.¹¹ Recommendations for patients and families with Li-Fraumeni syndrome include conducting annual whole-body MRI examinations to screen for various possible malignant tumors, including sarcomas, and maintaining a high suspicion index for rare cancers.¹²

X-ray examination is the standard examination for all patients with chest abnormalities. When examining patients with osteosarcoma, large lesions may be found, mainly manifested as destruction of normal bone trabeculae with unclear edges.¹³ Lesions usually stimulate the formation

FIGURE 3 (A) Excised tumor mass, (B and C) HE staining picture of tissue, and (C and D) immunohistochemical staining pictures.



of new bone in the periosteum, resulting in the characteristic Codman triangle. For a more detailed evaluation, CT or magnetic resonance imaging (MRI) can be performed.¹⁴ CT scans can provide sufficiently detailed images to evaluate almost all chest wall tumors. MRI can better represent related soft tissue masses, which can provide effective information for subsequent biopsy and final surgical resection. MRI often captures jumping metastases of local tumors. But if it metastasizes further away, a full body bone scan is required, which has an impact on treatment and prognosis. According to the UICC-TNM classification, 8 cm was defined as the prognostic cutoff for tumor size.¹⁵

The resection of primary osteosarcoma should be cautious to achieve complete resection while preserving

bone function. Otherwise, intra lesion or marginal resection will increase the local recurrence rate, which is related to a decrease in overall survival rate.^{5,16} The local recurrence rate of osteosarcoma in the limbs is relatively low, usually less than 5%, indicating that in most cases, complete resection can be achieved.¹⁷ Among patients with osteosarcoma who choose surgery, postoperative radiation therapy should be considered, especially for patients with closer surgical margins and lower degree of necrosis of the excised specimen.

In the English literature, we found about 17 cases of primary rib osteosarcomas. The characteristics of 18 patients with rib osteosarcoma, including this case, are shown in Table 1. As can be seen from the table, most

T A B L E 1 The characteristics of the patients with osteosarcoma of the rib.

References	Age (years)	Sex	Location	Primary/secondary	Size (cm)	Pathology	Treatment	Outcome
Wardoyo et al. ³	55	F	4th rib	Primary	5.5 × 5.3	Conventional osteosarcoma	Surgery	NA
Yaman Bajin et al. ⁷	14	M	5th rib	Primary	10.0	Conventional osteosarcoma	CT	Died with disease, 12 months
Moghadamfalahi and Alatassi ¹⁸	33	M	6th–8th ribs	Primary	4.0	Low-grade central osteosarcoma	Surgery	NA
Bay et al. ¹⁹	14	F	4th rib	Primary	12.0 × 13.0	Conventional osteosarcoma	NACT Surgery CT	Alive without disease, 8 months
Lim et al. ²⁰	15	M	4th rib	Primary	9.0 × 13.0	Conventional osteosarcoma	CT Surgery	Alive without disease, 10 months
Chattopadhyay et al. ²¹	11	M	9th and 10th ribs	Primary	3.5 × 1.5	Conventional osteosarcoma	Surgery CT	Alive without disease, 24 months
Ikeda et al. ²²	37	F	2nd–4th ribs	Primary	8.0 × 7.0	High-grade malignant osteosarcoma	Surgery CT	Alive without disease, 11 months
Botchu et al. ²³	7	F	7th rib	Primary	NA	Conventional osteosarcoma	Surgery ACT	Alive without disease, 12 months
Zheng et al. ²⁴	29	F	9th and 11th ribs	Primary	NA	Conventional osteosarcoma	CT Surgery Long-term immunotherapy	Alive without disease, 60 months
Das et al. ²⁵	12	M	6th rib	Primary	7.0 × 3.5 × 2.5	Chondroblastic osteosarcoma	Surgery	NA
Sinn et al. ²⁶	29	F	3rd–5th ribs	Primary	NA	Conventional osteosarcoma	CT Surgery	Recurrence, 4.5 years
Xie and Huang ²⁷	23	M	8th–10th ribs	Primary	8.8 × 8.3	Conventional osteosarcoma	NA	NA
Krishnamurthy and Arulmolichelvan ²⁸	24	M	5th–7th ribs	Primary	20.0 × 15.0	High-grade osteosarcoma	ACT Surgery CT	Alive without disease, 13 months
Hong et al. ²⁹	17	M	7th rib	Primary	5.0 × 6.0	Periosteal Osteosarcoma	Surgery	Alive without disease, 5 years
Xu and Zheng ³⁰	59	M		Primary	NA		Surgery	Recurrence, 1 month

TABLE 1 (Continued)

References	Age (years)	Sex	Location	Primary/secondary	Size (cm)	Pathology	Treatment	Outcome
			4th and 5th ribs			Conventional osteosarcoma		
Kuwabara et al. ³¹	67	M	3rd and 4th ribs	Primary	12	Conventional osteosarcoma	Surgery	Died with disease, 4 months
Anoop et al. ³²	16	M	2nd rib	Primary	12.3 × 9.5 × 9.0	Conventional osteosarcoma	CT	Died with disease, 1 week
Our patient	17	F	7th rib	Primary	2.0 × 3.0	Chondroblastic osteosarcoma	Surgery CT	Alive without disease, 18 months

Abbreviations: ACT, adjuvant chemotherapy; CT, chemotherapy; F, female; M, male; NA, not available; NACT, neoadjuvant chemotherapy.

patients can obtain a good therapeutic effect by surgery and adjuvant chemotherapy. The patient in this case was evaluated by experts in orthopedics and thoracic surgery immediately after the discovery of the chest mass. During the surgical resection of the mass, we removed at least 3 cm of normal rib tissue at both ends of the mass for complete removal of the mass. At the same time, the patient underwent chemotherapy after surgery. Our treatment strategy was broadly the same as the guidance given in the literature. The patient's postoperative feedback was also quite good, and no significant recurrence was found for one and a half years after the operation.

Several points should be noted in this case. This patient was found during physical examination, which is relatively early, and the surgical stage belongs to stage IIA (G2T1M0). According to our domestic treatment plan, surgery can be performed before chemotherapy. For stage IIB and III patients, neoadjuvant chemotherapy followed by surgery, and then chemotherapy. Preoperative neoadjuvant chemotherapy is very important for patients with limb salvage, but this patient's osteosarcoma is located in the ribs, and there is no need to reconstruct the bone structure. With or without preoperative chemotherapy, complete resection can be achieved without affecting the function. For patients with stage IIA rib osteosarcoma, early resection of the tumor also has certain advantages. For example, patients have good immunity and quick recovery after surgery. In addition, it can avoid some adverse consequences caused by puncture and obtain reliable pathological results at an early stage.

AUTHOR CONTRIBUTIONS

Xiaofeng Hu: Study concept or design; data collection. **Tianyi Bao:** Data analysis or interpretation; writing the paper. **Chao Yan:** data collection. **Yongliang Zhu:** Data analysis or interpretation. **Xiaofei Zheng:** Study concept or design; writing the paper.

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

CONFLICT OF INTEREST STATEMENT

The authors declare that they have no conflicts of interest.

DATA AVAILABILITY STATEMENT

Data sharing is not applicable to this article as no datasets were generated or analyzed during the current study.

ETHICS STATEMENT

Appropriate written informed consent was obtained from the patient for the publication of this case report and

accompanying images. It was approved by the Clinical Research Ethics Committee of Jinling Hospital of Nanjing Medical University and was implemented.

ORCID

Tianyi Bao  <https://orcid.org/0000-0002-0308-7039>

REFERENCES

- Beird HC, Bielack SS, Flanagan AM, et al. Osteosarcoma. *Nat Rev Dis Primers*. 2022;8(1):77-19. doi:10.1038/s41572-022-00409-y
- Meyers PA, Gorlick R. Osteosarcoma. *Pediatr Clin North Am*. 1997;44(4):973-989. doi:10.1016/s0031-3955(05)70540-x
- Wardoyo S, Kamal AF, Furqon MA, Grantomo J, Hutami WD. Osteosarcoma of the rib: a challenge of diagnosis and surgical intervention: a case report. *Int J Surg Case Rep*. 2021;81:105777. doi:10.1016/j.ijscr.2021.105777
- Burt M. Primary malignant tumors of the chest wall. The memorial Sloan-Kettering cancer center experience. *Chest Surg Clin N Am*. 1994;4(1):137-154.
- Bielack SS, Kempf-Bielack B, Delling G, et al. Prognostic factors in high-grade osteosarcoma of the extremities or trunk: an analysis of 1,702 patients treated on neoadjuvant cooperative osteosarcoma study group protocols. *J Clin Oncol*. 2002;20(3):776-790. doi:10.1200/JCO.2002.20.3.776
- Ritter J, Bielack SS. Osteosarcoma. *Ann Oncol*. 2010;21(Suppl 7):vii320-vii325. doi:10.1093/annonc/mdq276
- Yaman Bajin I, Kurucu N, Oguz B, et al. Primary osteosarcoma of the rib: a case report and review of the literature. *J Pediatr Hematol Oncol*. 2018;40(1):48-50. doi:10.1097/MPH.0000000000000896
- Gorlick R, Janeway K, Lessnick S, Randall RL, Marina N, Committee COGBT. Children's oncology group's 2013 blueprint for research: bone tumors. *Pediatr Blood Cancer*. 2013;60(6):1009-1015. doi:10.1002/pbc.24429
- Aljubran AH, Griffin A, Pintilie M, Blackstein M. Corrigendum to 'osteosarcoma in adolescents and adults: survival analysis with and without lung metastases': annals of oncology 2009; 20: 1136-1141. *Ann Oncol*. 2021;32(3):424. doi:10.1016/j.annonc.2020.12.011
- Zhou Y, Lu Q, Xu J, et al. The effect of pathological fractures on the prognosis of patients with osteosarcoma: a meta-analysis of 14 studies. *Oncotarget*. 2017;8(42):73037-73049. doi:10.18632/oncotarget.20375
- Villani A, Shore A, Wasserman JD, et al. Biochemical and imaging surveillance in germline tp53 mutation carriers with Li-Fraumeni syndrome: 11 year follow-up of a prospective observational study. *Lancet Oncol*. 2016;17(9):1295-1305. doi:10.1016/S1470-2045(16)30249-2
- Kratz CP, Achatz MI, Brugieres L, et al. Cancer screening recommendations for individuals with Li-Fraumeni syndrome. *Clin Cancer Res*. 2017;23(11):e38-e45. doi:10.1158/1078-0432.CCR-17-0408
- Papagelopoulos PJ, Galanis EC, Vlastou C, et al. Current concepts in the evaluation and treatment of osteosarcoma. *Orthopedics*. 2000;23(8):858-867; quiz 868-859. doi:10.3928/0147-7447-20000801-11
- Strauss SJ, Frezza AM, Abecassis N, et al. Bone sarcomas: ESMO-EURACAN-GENTURIS-ERN PaedCan clinical practice guideline for diagnosis, treatment and follow-up. *Ann Oncol*. 2021;32(12):1520-1536. doi:10.1016/j.annonc.2021.08.1995
- Bertero L, Massa F, Metovic J, et al. Eighth edition of the UICC classification of malignant tumours: an overview of the changes in the pathological TNM classification criteria-what has changed and why? *Virchows Arch*. 2018;472(4):519-531. doi:10.1007/s00428-017-2276-y
- Picci P, Sangiorgi L, Rougraff BT, Neff JR, Casadei R, Campanacci M. Relationship of chemotherapy-induced necrosis and surgical margins to local recurrence in osteosarcoma. *J Clin Oncol*. 1994;12(12):2699-2705. doi:10.1200/JCO.1994.12.12.2699
- David EA, Marshall MB. Review of chest wall tumors: a diagnostic, therapeutic, and reconstructive challenge. *Semin Plast Surg*. 2011;25(1):16-24. doi:10.1055/s-0031-1275167
- Moghadamfalahi M, Alatassi H. Low-grade central osteosarcoma of the rib: a case report and brief review of the literature. *Case Rep Pathol*. 2013;2013:798435. doi:10.1155/2013/798435
- Bay SB, Kebudi R, Iribas A, et al. Osteosarcoma of the rib: a rare presentation. *Turk Pediatri Ars*. 2018;53(1):57-60. doi:10.5152/TurkPediatriArs.2018.4689
- Lim W, Ahmad Sarji S, Yik Y, Ramanujam T. Osteosarcoma of the rib. *Biomed Imaging Interv J*. 2008;4(1):e7. doi:10.2349/biij.4.1.e7
- Chattopadhyay A, Nagendhar Y, Kumar V. Osteosarcoma of the rib. *Indian J Pediatr*. 2004;71(6):543-544. doi:10.1007/BF02724299
- Ikeda H, Takeo M, Kayata H, Mikami R, Nakamoto Y, Yamamoto M. A case of rapidly growing osteosarcoma of the rib. *Ann Thorac Cardiovasc Surg*. 2014;20(Suppl):521-524. doi:10.5761/atcs.cr.12.02069
- Botchu R, Ravikumar KJ, Sudhakar G, Meruva S, Anwar R. Osteosarcoma of rib in a seven-year-old child: a case report. *Eur J Orthop Surg Traumatol*. 2006;16(2):156-157. doi:10.1007/s00590-005-0032-5
- Zheng S, Wang F, Huang J, et al. Case report: sequential chemotherapy and immunotherapy produce sustained response in osteosarcoma with high tumor mutational burden. *Front Endocrinol (Lausanne)*. 2021;12:625226. doi:10.3389/fendo.2021.625226
- Das C, Mukhopadhyay M, Parvin T, Saha AK. Primary osteosarcoma of rib in a child-an uncommon case report. *Indian J Surg Oncol*. 2019;10(4):716-718. doi:10.1007/s13193-019-00980-z
- Sinn K, Hritcu R, Mosleh B, et al. Long-term survival after salvage surgery for a giant primary rib osteosarcoma. *Ann Thorac Surg*. 2021;111(1):e45-e47. doi:10.1016/j.athoracsur.2020.04.116
- Xie P, Huang J. Primary osteosarcoma of the rib identified on bone scintigraphy. *Clin Nucl Med*. 2016;41(5):390-391. doi:10.1097/RLU.0000000000001106
- Krishnamurthy A, Arulmolichelvan A. The management challenges in an unusual case of primary osteosarcoma of the rib in an adult patient. *Indian J Surg*. 2017;79(4):363-366. doi:10.1007/s12262-017-1591-5
- Hong JB, Cho KH, Choi JH. Periosteal osteosarcoma arising from the rib and scapula: imaging features in two cases. *Korean J Radiol*. 2014;15(3):370-375. doi:10.3348/kjr.2014.15.3.370

30. Xu G, Zheng K. Successful management of early recurrence after surgery for primary rib osteosarcoma in an adult. *Interact Cardiovasc Thorac Surg*. 2013;17(2):431-432. doi:[10.1093/icvts/ivt173](https://doi.org/10.1093/icvts/ivt173)
31. Kuwabara H, Fujita K, Yuki M, Goto I, Hanafusa T, Shibayama Y. Cytokeratin-positive rib osteosarcoma metastasizing to the small intestine. *Indian J Pathol Microbiol*. 2014;57(1):109-112. doi:[10.4103/0377-4929.130915](https://doi.org/10.4103/0377-4929.130915)
32. Anoop TM, Geetha N, Babanrao SA, Jayasree K. Primary osteosarcoma of rib mimicking lung mass with secondary aneurysmal bone cyst formation. *J Thorac Oncol*. 2014;9(5):738-739. doi:[10.1097/JTO.0000000000000155](https://doi.org/10.1097/JTO.0000000000000155)

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