American Journal of Case Reports

e-ISSN 1941-5923
© Am J Case Rep, 2021; 22: e930902
DOI: 10.12659/AJCR.930902

Received: 2021.01.05
Accepted: 2021.03.04
Available online: 2021.03.12
Published: 2021.04.17

Costal Osteoma: Report of a Case in an Unusual Site

Authors' Contribution:
Study Design A
Data Collection B
Statistical Analysis C
Data Interpretation D
Manuscript Preparation E
Literature Search F
Funds Collection G

BD Kyung Han Nam
AEF Bomi Kim

Department of Pathology, Inje University Haeundae Paik Hospital, Inje University College of Medicine, Busan, South Korea

Corresponding Author: Bomi Kim, e-mail: domabem96@paik.ac.kr
Conflict of interest: None declared

Patient: Male, 53-year-old
Final Diagnosis: Osteoma
Symptoms: Rib mass
Medication: —
Clinical Procedure: Excision biopsy
Specialty: Pathology • Surgery

Objective: Rare disease

Background: Osteoma is a benign bone tumor that typically arises in facial bones and less frequently in the long bones. It rarely occurs in the appendiceal skeleton. Pathologic findings are similar to those for parosteal osteosarcoma, periostitis ossificans, and osteoid osteoma. Diagnosing osteoma at an unusual site is always problematic. Here, we present a case of costal osteoma that was found incidentally on screening and produced mild symptoms in the patient.

Case Report: A 53-year-old man was examined because of a rib mass in the eighth rib on his left side, which had been present for 2 years. A computed tomography scan revealed that the intensely dense mass arose from the external surface of the eighth rib. Microscopic examination showed that the lesion consisted of mature lamellar bone with several Haversian systems, typical of an osteoma. No atypical spindle cells or necrosis were identified. The diagnosis was osteoma.

Conclusions: Because the anatomy of the ribs poses a challenge when performing needle biopsy, diagnosing bony lesions solely based on that technique is difficult. The diagnosis of costal osteoma should be made carefully, based on clinical, radiological, and pathological findings. To the best of our knowledge, ours is the first case report about a costal osteoma. It is useful for chest surgeons, pathologists, and radiologists as an example of a rare presentation of this tumor.

Keywords: Bone Neoplasms • Osteoma • Ribs

Full-text PDF: https://www.amjcaserep.com/abstract/index/idArt/930902

Indexed in: [PMC] [PubMed] [Emerging Sources Citation Index (ESCI)] [Web of Science by Clarivate]

Background

Primary rib tumors are extremely rare, comprising 5% to 7% of all primary bone tumors [1]. Moreover, distribution of benign and primary malignant tumors of the thoracic cavity is almost equivalent [2]. Among them, chondroma, osteochondroma, fibrous dysplasia, eosinophilic granuloma, and Ewing sarcoma are relatively common [2]. Because biopsying a rib is difficult and biopsies of bone tumors are not very accurate, the only method for diagnosing a rib tumor is excisional biopsy; therefore, correlation of clinical, radiological, and pathologic results is very important [3]. Even with excisional biopsy, pathologic confirmation of osteoma is difficult and the lesion rarely is found in the ribs. Osteomas generally occur in the bones of the skull and face and are rarely found in extracranial sites [4]. The tumors are classified as medullary or parosteal according to the bone location [5]. Pathologically, osteomas are composed of mature lamellar trabeculae with no cytological atypia and few mitoses [5]. Considering their locations and pathologic features, osteomas should be distinguished from parosteal osteosarcomas [5]. Here, we present a very unusual case of a costal osteoma that occurred in the rib of an adult.

Case Report

A 53-year-old man was referred to the Surgery Department with a rib mass that had been detected 2 years previously on a routine chest X-ray, for which he was prescribed no treatment. He experienced symptoms of pressure while supine but denied pain. He was diagnosed with a degenerative joint disease and had fractured a rib on his right side and his right ankle 3 and 5 years ago, respectively. Palpation of the patient's back revealed a hard, non-tender mass in the posterior aspect of the left side of his back. His preoperative laboratory test results were unremarkable. A chest posterior anterior X-ray and a computed tomography scan showed an exophytic solid mass measuring 2.5 cm on the posterior arch of the eighth rib on the left side (**Figure 1**). The radiological diagnosis was a bone tumor, such as an osteochondroma or osteosarcoma. Our clinical impression was a benign bone tumor, such as an osteochondroma.

The patient underwent partial resection of the posterior arc side of his eighth rib under general anesthesia. Gross examination showed a lobulated, solid, osseous mass on the surface of the bone (**Figure 2A**) that was a homogenous yellow-tan color (**Figure 2B**). The entire specimen was processed for microscopic examination. Low-power magnification showed that the lesion was not connected to the spongy part of the ribs. It was contiguous with and covered by the periosteum, which was a thin connective tissue (**Figure 3A**). It was composed of dense, cortical-type compact bone with several Haversian

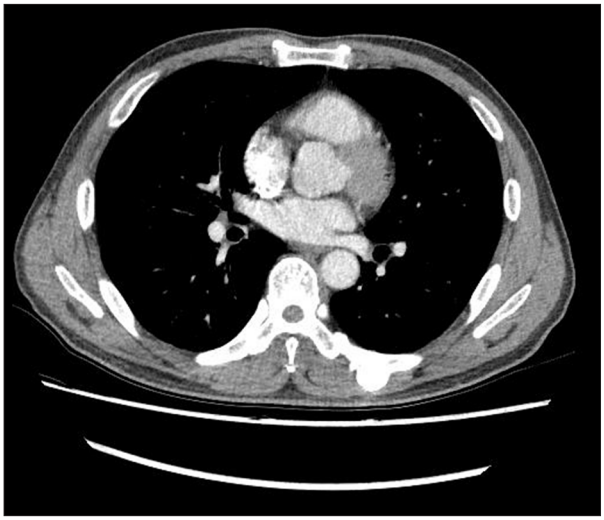

Figure 1. A computed tomography scan shows that the mass arises from the posterior costal arc of the eighth rib on the patient's left side and does not involve the medulla.

canals (**Figure 3B**). The cartilaginous cap was absent. High-power magnification showed that many of the lacunar spaces were empty or had myxoid stroma with vessels. Flattening of the osteoblastic rimming was inconspicuous. The woven bone was immature and there was no proliferation of spindle cells, granulation tissue, or necrotic bone. No cytological atypia, mitosis, or endochondral ossification was present. No inflammatory cells were found inside the lesion. Considering these pathological features, the diagnosis was osteoma. On follow-up after discharge, the patient had no back discomfort or any surgical complications.

Discussion

Osteoma is a benign osteogenic tumor that often occurs in craniofacial bones [5]. It is less frequently found in long bones such as the femur and tibia and rarely seen in vertebral columns, except in correlation with Gardner syndrome [5]. Its pathogenesis is unknown. There is a suggestion that osteoma is a form of exaggerated proliferation of intramembranous ossification that presents as hamartomatous lesions arising from the periosteum [6]. *APC* and *LEMD3* gene mutations are known to be related to osteoma [4].

The diagnosis of osteoma in an unusual site is very problematic and should be made with caution. Differential diagnoses of pathologic abnormalities include low-grade parosteal osteosarcoma, osteochondroma with an attenuated cartilaginous cap, reactive sclerosis, and myositis ossificans [5]. Owing to the unusual clinical manifestations in the present case, we considered all differential diagnoses. Osteosarcoma, in particular,

Indexed in: [PMC] [PubMed] [Emerging Sources Citation Index (ESCI)] [Web of Science by Clarivate]

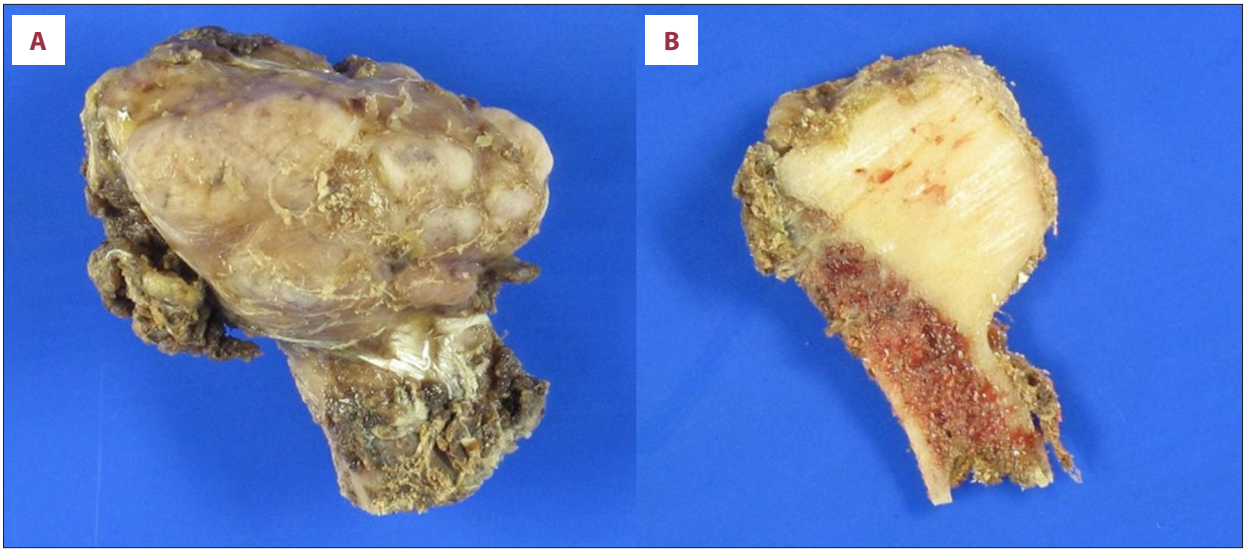

Figure 2. (**A**) Gross examination shows that the bony mass, which measured 2.5 cm, has a lobulated contour. The external surface is smooth and covered by a thin, fibrous, soft tissue membrane. (**B**) The mass is yellow or ivory in color and has a hard consistency, making it blend into the cortex of the rib bone.

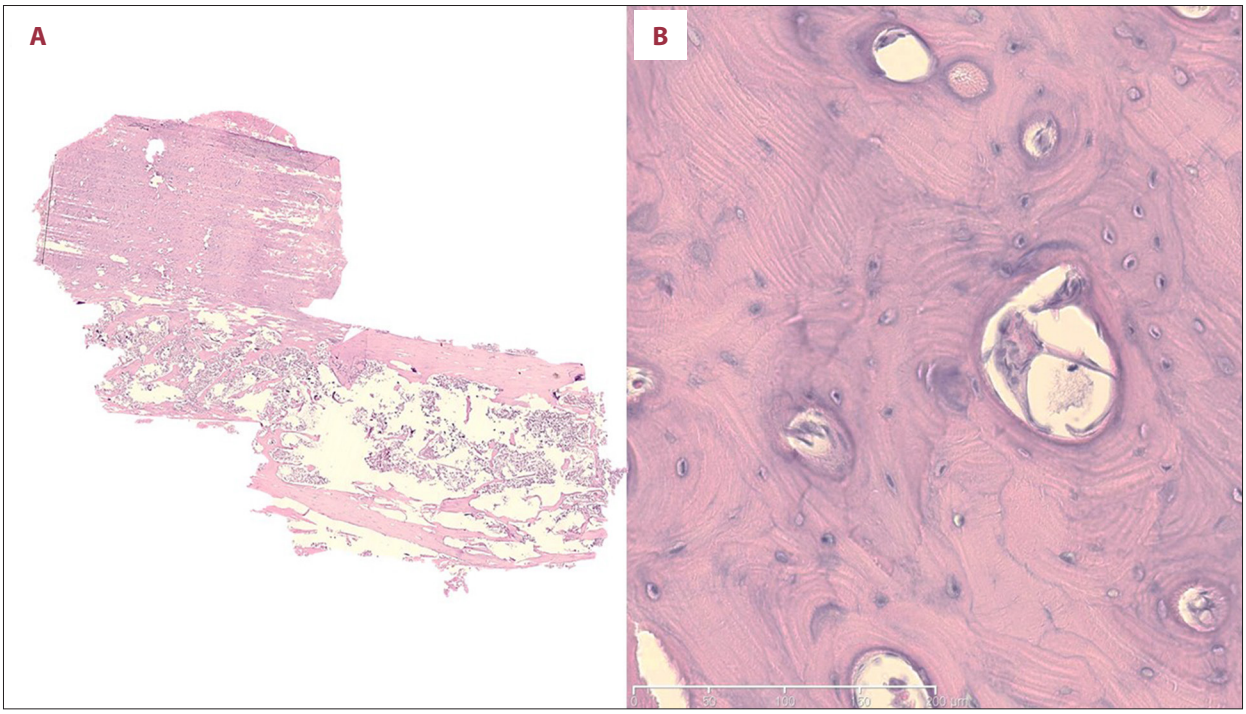

Figure 3. (**A**) A histologic section of the resected specimen shows that the solid mass was not connected to the marrow of the rib (hematoxylin and eosin [H&E], magnification×1). (**B**) The specimen is composed of compact, mature, lamellar bone with well-developed Haversian canals. (H&E, magnification×200).

is strikingly similar to osteoma on radiographic studies, although the clinical courses of the 2 tumors are different. We ruled out all of the previously mentioned diseases except osteoma because our patient's lesion was well demarcated with no destructive growth, lacked a cartilage cap, and did not have the zonal pattern typical of pathological findings for myositis ossificans. In adults, the cartilage cap of an osteochondroma is sometimes attenuated and replaced by cortical bone through endochondral ossification [7,8]. However, neither the continuous connection of its stalk and rib marrow cavities nor endochondral ossification were present in our patient. The histological diagnosis was consistent with an osteoma. Because

Indexed in: [PMC] [PubMed] [Emerging Sources Citation Index (ESCI)] [Web of Science by Clarivate]

we had never encountered a costal osteoma before, we consulted with 2 bone pathologists and they concluded that our patient had an osteoma.

Osteomas rarely occur in extracranial locations such as the femur, fibula, tibia, patella, pelvic bone, humerus, ulna, radius, metatarsal bone, scapula, or clavicle [6,8-20]. Bertoni et al reviewed 14 cases of parosteal osteoma of bones other than the skull and face and found that they arose more frequently in men than in women [11]. The patients with them ranged in age from 8 to 75 years [10,20]. Osteomas of long bones grew in the diaphysis and the metaphysis [11]. None of the patients who were followed up showed evidence of progressive disease or changes in size of their tumors [11]. We know of only 3 cases of costal osteoma discussed in the literature [21-23]. The first was a report by Flori in 1950, which described the tumor as an "osteoid osteoma" [21]. Humplik and Mayr diagnosed "osteoma" on 30 000 radiophotographs but did not conduct a pathological examination [22]. Steinberg described a stony-hard rib mass composed of pink, loose, fibrillary tissue with bony spicules, calcification, and fat [23]. We were not able to confirm if the lesions described in the previous literature were pathologically shown to be osteomas. In Korea, Joo et al reported on recurrence of an osteoma in grafts from frontal calvarian and rib bones [24]. Those authors suggested that the remaining periosteum triggered the recurrence, regardless of the location of the grafted bones. Osteoma arising from the clavicle has been shown to have pathologic features similar to the present case [6,11].

Excision of an osteoma is not necessary for treatment if the patient does not have any symptoms [4]. It is essential, however, for diagnosis. Pathological diagnosis of the tumor with a needle biopsy is difficult because the approach is challenging and it has low accuracy for bone lesions [3,25]. Osteoma often is difficult to identify in biopsy specimens because it closely resembles parosteal osteosarcoma, osteochondroma, and myositis ossificans. No reports exist of osteoma recurrence, metastasis, or malignant transformation [5,16]. Generally, patients with the tumor need no further treatment if they are asymptomatic. In our case, excision was performed for diagnosis and treatment, and it relieved the patient's symptoms.

Conclusions

We have described the case of a 53-year-old man who complained of pressure while supine. Radiological studies revealed an exophytic bony mass originating from the surface of the eighth rib on his left side. We completely excised the lesion because we were unable to completely rule out parosteal osteosarcoma during the preoperative work-up. Histological examination showed features compatible with an osteoma, not parosteal osteosarcoma or osteochondroma. Osteomas rarely occur in bones other than the skull and face. Ours is the first reported case of primary costal osteoma. Without excision, osteomas continue to grow slowly and the associated symptoms worsen; however, these tumors do not recur, metastasize, or progress to malignancy. To avoid overtreatment, accurate diagnosis of an osteoma is very important.

Ethics Statement

This study was approved by the Internal Review Board of the Inje University Haeundae Paik Hospital, Busan, Republic of Korea (no. 2020-11-015).

Conflict of Interest

None.

Indexed in: [PMC] [PubMed] [Emerging Sources Citation Index (ESCI)] [Web of Science by Clarivate]

References:

1. Hughes EK, James SL, Butt S, et al. Benign primary tumours of the ribs. Clin Radiol. 2006;61(4):314-22
2. Teitelbaum SL. Twenty years' experience with intrinsic tumors of the bony thorax at a large institution. J Thorac Cardiovasc Surg. 1972;63(5):776-82
3. Mankin HJ, Lange TA, Spanier SS. The hazards of biopsy in patients with malignant primary bone and soft-tissue tumors. J Bone Joint Surg Am. 1982;64(8):1121-27
4. WHO Classification of Tumours Editorial Board. Osteoma. Soft Tissue and Bone Tumours. In: WHO Classification of Tumours. 5th ed: World Health Organization; 2020
5. Santini-Araujo E, Kalil RK, Bertoni F, et al. Osteoma. In: Santini-Araujo E, Kalil RK, Bertoni F, Park YK, editors. Tumors and tumor-like lesions of bone: For Surgical Pathologists, Orthopedic Surgeons and Radiologists: Springer; 2015
6. Meltzer CC, Scott WW Jr., McCarthy EF. Case report 698: Osteoma of the clavicle. Skeletal Radiol. 1991;20(7):555-57
7. Khurana JS. The surgical pathology of bone tumors and tumor-like lesions. In: Khurana JS, editor. Bone patholgy Humana Press; 2009; 285-346
8. O'Connell JX, Rosenthal DI, Mankin HJ, et al. Solitary osteoma of a long bone. A case report. J Bone Joint Surg Am. 1993;75(12):1830-34
9. Ayas MS, Gul O, Dada ME. Extracranial location of the osteoma: Patella, case report and literature review. Eklem Hastalik Cerrahisi. 2018;29(3):189-92
10. Baum PA, Nelson MC, Lack EE, et al. Case report 560: Parosteal osteoma of tibia. Skeletal Radiol. 1989;18(5):406-9
11. Bertoni F, Unni KK, Beabout JW, et al. Parosteal osteoma of bones other than of the skull and face. Cancer. 1995;75(10):2466-73
12. Cervilla V, Haghighi P, Resnick D, et al. Case report 596: Parosteal osteoma of the acetabulum. Skeletal Radiol. 1990;19(2):135-37
13. Chikuda H, Goto T, Ishida T, et al. Juxtacortical osteoma of the ulna. J Orthop Sci. 2002;7(6):721-23
14. Dolan KD, Seibert J, Seibert RW. Gardner's syndrome. A model for correlative radiology. Am J Roentgenol Radium Ther Nucl Med. 1973;119(2):359-64
15. Houghton MJ, Heiner JP, De Smet AA. Osteoma of the innominate bone with intraosseous and parosteal involvement. Skeletal Radiol. 1995;24(6):455-57
16. Mirra JM, Gold RH, Pignatti G, et al. Case report 497: Compact osteoma of iliac bone. Skeletal Radiol. 1988;17(6):437-42
17. Soler Rich R, Martinez S, de Marcos JA, et al. Parosteal osteoma of the iliac bone. Skeletal Radiol. 1998;27(3):161-63
18. Stern PJ, Lim EV, Krieg JK. Giant metacarpal osteoma. A case report. J Bone Joint Surg Am. 1985;67(3):487-89
19. Sundaram M, Falbo S, McDonald D, et al. Surface osteomas of the appendicular skeleton. Am J Roentgenol. 1996;167(6):1529-33
20. Zhou BG, Liu MY, Lv LC, et al. Bone marrow osteoma of the tibia: A case report. Oncol Lett. 2014;8(6):2776-78
21. Flori D. [Case of costal osteoma]. J Radiol Electrol Arch Electr Medicale. 1950;31(9-10):549-50 [in French]
22. Humplik M, Mayr M. [Rib osteomas]. Acta Chir Orthop Traumatol Cech. 1970;37(6):374-75 [in Czech]
23. Steinberg I. Huge osteoma of the eleventh left rib. J Am Med Assoc. 1959;170(16):1921-23
24. Joo CS, Lee YH. Case report of recurrent osteoma at the grafted bone. J Korean Soc Plast Reconstr Surg. 2006;33(3):367-70
25. Andrianopoulos EG, Lautidis G, Kormas P, et al. Tumours of the ribs: Experience with 47 cases. Eur J Cardio Thorac Surg. 1999;15(5):615-20

Indexed in: [PMC] [PubMed] [Emerging Sources Citation Index (ESCI)] [Web of Science by Clarivate]