

Clinicopathological characteristics of costal hemangiomas: a case report and literature review

Journal of International Medical Research

2019, Vol. 47(2) 982–985

© The Author(s) 2018

Article reuse guidelines:

sagepub.com/journals-permissions

DOI: 10.1177/0300060518809589

journals.sagepub.com/home/imr



Chang Gok Woo¹, Ho-Chang Lee^{1,2},
Seung-Myoung Son¹, Jong-Myeon Hong^{2,3}
and Ok-Jun Lee^{1,2}

Abstract

Bone hemangioma accounts for approximately 1% of all bone neoplasms and commonly occurs in the vertebral body and skull. However, costal hemangiomas are extremely rare. We herein present a case involving a 52-year-old woman with a hemangioma in the third rib and review 29 cases of rib hemangiomas available in the literature. Rib hemangioma mainly affects women in their 50s and has expansile osteolytic features in radiographs and a weak maximum standardized uptake value in ¹⁸F-fluorodeoxyglucose positron emission tomography images. When these findings are displayed, clinicians should include rib hemangioma as a differential diagnosis and consider avoidance of preoperative biopsy because of the risk of life-threatening bleeding.

Keywords

Rib, bone, hemangioma, benign, neoplasm, osteolysis, maximum standardized uptake value

Date received: 14 June 2018; accepted: 5 October 2018

Introduction

Bone hemangioma accounts for approximately 1% of all bone neoplasms and commonly occurs in the vertebral body and skull.¹ Hemangioma of the rib is extremely rare, but this tumor may occur in all ribs. Distinguishing a hemangioma from other malignancies is difficult when the costal

¹Department of Pathology, Chungbuk National University Hospital, Cheongju, Korea

²Chungbuk National University College of Medicine, Cheongju, Korea

³Department of Thoracic and Cardiovascular Surgery, Chungbuk National University Hospital, Cheongju, Korea

Corresponding author:

Ok-Jun Lee, Department of Pathology, Chungbuk National University Hospital, Chungbuk National University College of Medicine, Chungdae-ro 1, Seowon-gu, Cheongju 28644, Korea.

Email: ojlee@chungbuk.ac.kr



lesion shows osteolytic findings in radiographs because approximately half of all costal tumors are malignant. To our knowledge, few studies have demonstrated the clinicopathological characteristics of costal hemangiomas. We herein present a case of a costal hemangioma and describe its clinical features.

Case report

A costal lesion was incidentally detected during a regular checkup of a 52-year-old woman. She had no history of trauma and no complaints. Her medical history and family history were unremarkable. Chest computed tomography demonstrated an expansile osteolytic tumor in the midportion of the left third rib, extending toward the thoracic cavity (Supplementary Figure 1(a)). ^{18}F -Fluorodeoxyglucose positron emission tomography revealed a maximum standardized uptake value (SUVmax) of 2.3 (Supplementary Figure 1(b)). Based on these findings, the differential diagnoses were a primary malignant bone tumor and other malignancies. The patient underwent en bloc excision of the tumor along with the adjacent soft tissues by axillary thoracotomy. Grossly, the lesion displayed a spongiform cut surface with hemorrhage and measured $5.0 \times 3.5 \times 2.5$ cm (Supplementary Figure 1(c)). Microscopically, the tumor comprised numerous irregular, dilated, and thin-walled vessels lined with a single layer of flattened endothelial cells without cytologic atypia, consistent with the findings of a cavernous hemangioma (Figure 1). The bony cortex was eroded by this tumor. Both lateral margins were free from the tumor in permanent sections. The patient's recovery was uneventful, and she demonstrated no evidence of local recurrence 1 year after surgery. This study was approved by the institutional review board (CBNUH IRB, Cheongju, No. 2018-08-015), and the patient provided informed consent.

Discussion

Rib hemangioma is a very rare tumor. Most bone hemangiomas occur in the skull and vertebral column, and they constitute approximately 1% of all bone tumors.^{1,2} To the best of our knowledge, approximately 30 reports of rib hemangiomas have been published to date. Previous articles were identified through electronic searches of the Google Scholar and PubMed databases. The search terms were "rib hemangioma," "costal hemangioma," "rib vascular tumor," "costal vascular tumor," and "bone vascular tumor." We excluded articles that were written in a non-English language and in which the clinicopathological features were not available. In the present report, we review 29 cases of rib hemangiomas available in the literature and describe 1 case of a rib hemangioma (Table 1 and Supplementary Table 1). Rib hemangiomas may manifest at any age, but they reportedly show a peak incidence at the age of 50 to 60 years (median, 50 years; range, 11–76 years). Additionally, they predominantly affect females; the cases reported herein comprise 19 females and 11 males. The tumor was incidentally detected in more than half of the patients, but approximately 43% (13/30) of the patients had one or more symptoms, such as localized back and chest pain. Some patients (11%, 3/28) had a history of trauma (a fracture at the location of the hemangioma). The tumors ranged in size from 2.9 to 16 cm (median, 6 cm). In one report, the tumors were found to occur in the right and left ribs at the same frequency,³ but more than half of them (59%) occurred in the left rib. They frequently developed between the fifth and eighth ribs (70%), and only three tumors (12%) involved the anterior arc of the rib. Many rib hemangiomas (73%, 22/30) showed osteolytic changes in radiographs, causing them to appear as malignancies. A preoperative biopsy was performed in 6 of 30 patients, and the need

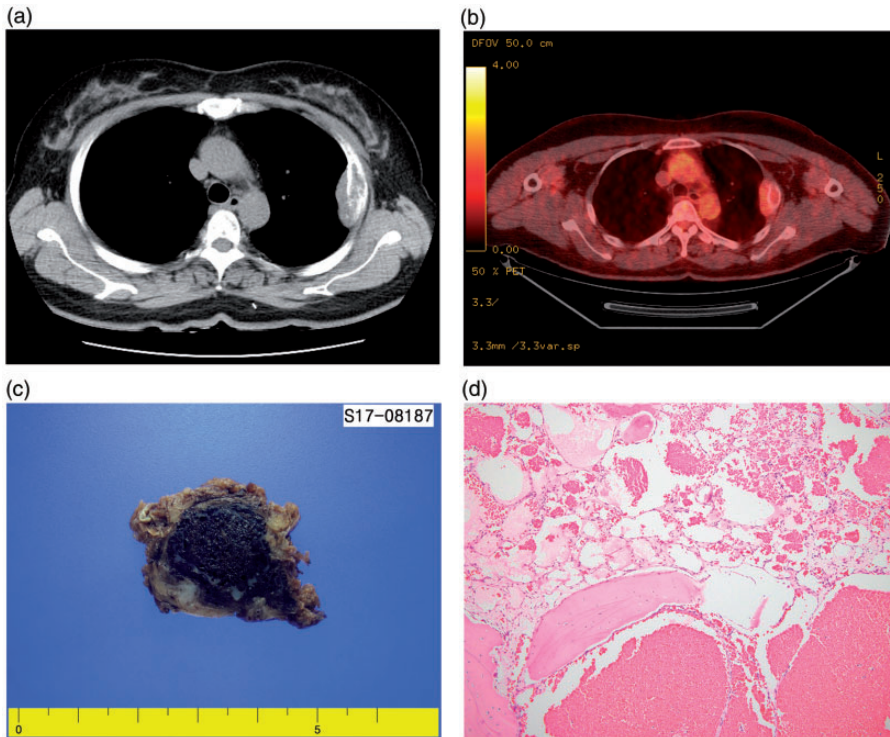


Figure 1. (a) Chest computed tomography demonstrates an expansile osteolytic lesion in the mid arc of the left third rib. (b) Positron emission tomography reveals ^{18}F -fluorodeoxyglucose uptake with a maximum standardized uptake value of 2.3 in the mid arc of the left third rib. (c) Grossly, the cut surface of the formalin-fixed tumor specimen is spongiform. (d) Microscopically, the tumor comprises irregular thin-walled vessels containing red blood cells.

for this procedure is controversial. Some authors have insisted that biopsy should be avoided because of a risk of seeding through the biopsy tract and bleeding,^{4,5} whereas others have emphasized the need for a needle biopsy to obtain definitive diagnosis.⁶ However, critical bleeding and hematoma formation were reported in some patients who underwent a biopsy.⁷ The pathological diagnosis of a hemangioma in biopsy specimens is challenging to the pathologist because of disrupted blood vessels and empty spaces caused by loss of red blood cells during the procedure. Therefore, biopsy should be avoided unless multiple myeloma or metastasis is suspected. The treatment of choice for rib hemangioma is

complete surgical excision; embolization, radiotherapy, and steroid therapy have been applied in a few patients, but these may increase the risk of local recurrence.⁸ The SUVmax of rib hemangiomas in previous reports and the current case were 2.2, 2.3 (present case), 2.4, 2.5, and 2.7. ^{18}F -Fluorodeoxyglucose positron emission tomography demonstrates slightly elevated glucose uptake.

The present case demonstrated characteristics that are typical of other costal hemangiomas, as noted above. When women in their 50s show an expansile osteolytic lesion with a low SUVmax in the mid-level rib, clinicians should consider costal hemangioma as a differential diagnosis and

Table 1. Clinicopathological characteristics of patients with rib hemangioma.

Parameters	N = 30
Age in years	50 (11–76)
Sex	
Male	11 (37)
Female	19 (63)
Symptoms	
None	17 (57)
Back pain + cough	7 (23)
Chest pain + cough	4 (13)
Other symptoms	2 (7)
History of trauma	
None	25 (89)
Rib fracture	2 (7)
Previous operation	1 (4)
NA	2 (7)
Size in cm	6 (2.9–16)
Side of the lesion	
Right	12 (41)
Left	17 (59)
NA	1 (4)
Location of affected rib	
1st–4th	6 (20)
5th–8th	21 (70)
9th–12th	3 (10)
Arc of the rib	
Anterior	3 (12)
Mid	12 (46)
Posterior	11 (42)
NA	4 (13)
Bone destruction in radiographs	22 (73)
Preoperative biopsy	6 (20)

NA, data not available.

Data are presented as median (range) or n (%).

avoidance of preoperative biopsy because of the risk of life-threatening bleeding. However, a biopsy is necessary if malignancy is clinically suspected. In addition, complete excision of the costal lesion is required regardless of the preoperative impression.

Declaration of conflicting interest

The authors declare that there is no conflict of interest.

Funding

This research received no specific grant from any funding agency in the public, commercial, or not-for-profit sectors.

References

1. Dorfman HD, Steiner GC and Jaffe HL. Vascular tumors of bone. *Hum Pathol* 1971; 2: 349–376. 1971/09/01.
2. Kuo YT, Lin MB, Sheu RS, et al. Imaging diagnosis of cavernous hemangioma of the rib—one case report and review of the literature. *Gaoxiong Yi Xue Ke Xue Za Zhi* 1994; 10: 469–473. 1994/08/01.
3. Haro A and Nagashima A. A rare case report of rib hemangioma mimicking a malignant bone tumor or metastatic tumor. *Int J Surg Case Rep* 2015; 16: 141–145. 2015/10/12. DOI: 10.1016/j.ijscr.2015.09.036.
4. Abrao FC, Tamagno M, Canzian M, et al. Hemangioma of the rib. *Ann Thorac Surg* 2011; 91: 595–596. 2011/01/25. DOI: 10.1016/j.athoracsur.2010.07.091.
5. Gourgiotis S, Piyis A, Panagiotopoulos N, et al. Cavernous hemangioma of the rib: a rare diagnosis. *Case Rep Med* 2010; 2010: 254098. 2010/06/30. DOI: 10.1155/2010/254098.
6. Nakamura H, Kawasaki N, Taguchi M, et al. Cavernous hemangioma of the rib diagnosed preoperatively by percutaneous needle biopsy. *Gen Thorac Cardiovasc Surg* 2007; 55: 134–137. 2007/04/24. DOI: 10.1007/s11748-006-0086-2.
7. Deshmukh H, Rathod KK, Hira P, et al. Hemangioma of rib: a different perspective. *Pol J Radiol* 2015; 80: 172–175. 2015/04/14. DOI: 10.12659/pjr.893011.
8. Ortega W, Mahboubi S, Dalinka MK, et al. Computed tomography of rib hemangiomas. *J Comput Assist Tomogr* 1986; 10: 945–947. 1986/11/01.