A rare case report of rib hemangioma mimicking a malignant bone tumor or metastatic tumor

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**A B S T R A C T**

Hemangioma of the rib is a rare benign vascular tumor. This benign disease induces osteolytic changes, and must be distinguished from a malignant bone tumor or metastatic tumor. Definitive diagnosis is achieved by excision biopsy or histological examination after surgical resection in many cases. We here present a rare case of hemangioma of the rib.

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1. Introduction

Hemangioma of the rib is a rare benign bone tumor that accounts for 1% of all bone tumors. This tumor may arise in all ribs, and it is asymptomatic or mildly symptomatic. About half of costal tumors are malignant, and it is difficult to distinguish an osteolytic hemangioma of the rib from a malignant tumor.

2. Presentation of case

A 73-year-old woman was referred to our hospital for surgical resection of sigmoid colon carcinoma. Physical examination in our hospital revealed an asymptomatic palpable tumor at the left chest wall. She had no remarkable family history or other physical examination findings. She had no history of chest trauma or rib fractures. Preoperative chest computed tomography (CT) showed an osteolytic, expanding left chest tumor extending from the left sixth rib to the adjacent soft tissues and measuring 2.9 × 2.5 × 1.9 cm. The tumor was enhanced by contrast medium (Fig. 1). Magnetic resonance imaging (MRI) of the tumor revealed low signal intensity on T1-weighted images, high signal intensity on T2-weighted images, and early-phase enhancement by contrast medium (Fig. 2). The muscles adjacent to the tumor in the left chest wall were edematous. Positron emission tomography with 2-fluoro-2-deoxy-D-glucose (\(^{18}\)F-FDG PET) revealed a maximum standardized uptake value (SUV\textsubscript{max}) of 2.66 (Fig. 3). An aspiration needle biopsy of the tumor showed mainly blood cells and benign mesenchymal cells, with no malignant cells. We could not exclude the possibility of a primary malignant bone tumor or metastatic chest tumor; therefore, the left sixth rib was excised en bloc with the adjacent soft tissues. The chest wall was reconstructed using a polytetrafluoroethylene patch. The postoperative course was uneventful, and the patient was discharged on postoperative day 15.

The postoperative histological diagnosis was hemangioma of the rib. Macroscopic examination revealed a polycystic tumor containing hemorrhagic tissue (Fig. 4). Microscopically, the tumor was composed of dilated vessels and capillary-sized vessels in the bone and adjacent soft tissues (Fig. 5). These vessels comprised a single layer of endothelial cells.

The patient had no evidence of recurrence 1 year after surgical resection.

3. Discussion

Hemangioma of the rib is a rare bone tumor that accounts for 1% of all bone tumors. Most bone hemangiomas arise in the skull or vertebral bodies. Two major histological types of hemangiomas have been defined: the cavernous or capillary type and other subtypes including the arteriovenous or mixed type [1,2].

To the best of our knowledge, 25 reports of hemangioma of the rib have been published to date (Additional file; Table 1). These
Table 1
Case reports of hemangioma of the rib.

| Case | Age (y) | Gender | Location          | Osteolytic changes in CT | Size                      | Symptom                         | Year | Author                  | Journal                     |
|------|---------|--------|-------------------|--------------------------|---------------------------|-------------------------------|------|-------------------------|-----------------------------|
| 1    | 53      | F      | Rt. 7th rib       | Negative                 | Not mentioned             | Epigastric discomfort         | 1979 | Feldman                 | Skeletal Radiol.            |
| 2    | 11      | F      | Rt. 6th rib       | Negative                 | Not mentioned             | Back pain                     | 1986 | ortega                  | J. Comput. Assit. Tomorg.   |
| 3    | 14      | M      | The sacrum and Rt. 7th rib | Positive                | Not mentioned             | Pain in leg and low back      |      |                         |                             |
| 4    | 65      | F      | Lt. 8th rib       | Negative                 | Not mentioned             | Back pain                     |      |                         |                             |
| 5    | 56      | F      | Lt. 7th rib       | Positive                 | 16 x 7 x 6 cm             | None                          | 1994 | Kuo                     | Kaohsiung J. Med. Sci.      |
| 6    | 76      | M      | Lt. 3rd rib       | Negative                 | 7.5 x 3.5 cm              | None                          | 1998 | Clements                | Am. Surg.                   |
| 7    | 61      | M      | Lt. 12th rib      | Negative                 | 5 cm                      | Contralateral chest pain      | 1995 | Filosso                 | J. Cardiovasc. Surg.        |
| 8    | 59      | F      | Lt. 7th rib       | Positive                 | 3.5 x 2.5 x 1.5 cm        | None                          | 2000 | Okumura                 | Jpn. J. Clin. Oncol.        |
| 9    | 50      | F      | Rt. 1st rib       | Negative                 | Not mentioned             | Arm discomfort                | 2001 | Yeow                    | J. Vasc. surg.              |
| 10   | 59      | M      | Lt. 4th rib       | Positive                 | 7.5 cm                    | None                          | 2002 | Shimizu                 | Ann. Thorac. Surg.          |
| 11   | 45      | M      | Rt. 7th rib       | Positive                 | Not mentioned             | Costal Pain                   | 2005 | Roy                     | CMA Med. Icn.               |
| 12   | 74      | M      | Lt. 5th rib       | Positive                 | 9.5 x 6.5 x 3.0 cm        | Left back pain                | 2007 | Nakamura                | Gen. Thorac. Cardiovasc. Xurg |
| 13   | 28      | F      | Rt. 5th rib       | Positive                 | 7 x 9 x 5 cm              | Back pain, dyspnea and cough  | 2007 | Sirmali                 | Acta. Chir. Belg.           |
| 14   | 62      | M      | Rt. 5th rib       | Positive                 | 6 x 5 x 3.5 cm            | None                          | 2008 | Ceberut                 | Asian Cardiovasc. Thorac. Ann. |
| 15   | 54      | F      | Lt. 8th rib       | Positive                 | 9 x 6 x 5 cm              | None                          | 2008 | Cakir                   | Acta. Chir. Belg.           |
| 16   | 29      | F      | Lt. 7th rib       | Negative                 | 4.5 cm                    | None                          | 2010 | Gourgiotis              | Case Rep. Med.              |
| 17   | 48      | F      | Lt. 7th rib       | Positive                 | 5 x 3.8 cm                | None                          | 2011 | Abrao                   | Ann. Thorac. Surg.          |
| 18   | 64      | F      | Lt. 5th rib       | Positive                 | 8 x 4 x 2.5 cm            | None                          | 2011 | Hashimoto               | Ann. Thorac. Surg.          |
| 19   | 54      | F      | Rt. 9th rib       | Positive                 | 7.5 x 5.2 x 4.5 cm        | None                          | 2011 | Jia                     | Clin. Nucl. Med.            |
| 20   | 20      | M      | Rt. 5th rib       | Positive                 | 5 cm                      | None                          | 2011 | Khuismeng               | Diag. Inter. Radiol.        |
| 21   | 26      | F      | Rt. 8th rib       | Positive                 | 3 x 3 cm                  | Back pain                     | 2011 | Sanjay                  | Indian J. Surg.             |
| 22   | 17      | M      | Rt. 6th rib       | Positive                 | 10 cm                     | None                          | 2012 | Bouchik                | Pan Afr. Med. J.            |
| 23   | 16      | M      | Lt. 8th rib       | Negative                 | 7.2 x 3.7 x 4.0 cm        | Back pain                     | 2012 | Burke                   | Ir. Med. J.                 |
| 24   | 73      | F      | Lt. 8th rib       | Positive                 | 12 x 11 x 11 cm           | None                          | 2013 | Yamamoto                | Kyobu Geka.                 |
| 25   | 30      | M      | Rt. 8th rib       | Negative                 | 6 x 4 cm                  | Cough, chest pain             | 2013 | Memdahun               | Intern. Med.                |
| 26   | 79      | F      | Lt. 6th rib       | Positive                 | 2.9 x 2.5 x 1.9 cm        | None                          | 2015 | Haro                    | This case                   |

* Tumor expansion and bony speculations within the mass.
Fig. 1. Chest CT examination revealed (A) an expanding osteolytic tumor and (B) enhancement by contrast medium.

Fig. 2. Magnetic resonance imaging showed (A) low signal intensity on T1-weighted images, (B) high signal intensity on T2-weighted images and (C) enhancement by contrast medium.

Fig. 3. FDG-PET examination revealed accumulation with an SUV$_{max}$ of 2.66 at the left sixth rib.

reports involved 15 males and 11 females patients, and the average age was 47.0 ± 21.2 years old (range, 11–79 years). In one report, osseous hemangioma was identified in middle-aged patients [1], but middle-aged patients with hemangioma of the rib constituted 38.5% (9/26 cases) of all patients. The proportions of younger and older patients among all affected patients were 38.5% (9/26 cases) and 30.8% (8/26 cases), respectively. A total of 42.6% (12/26 cases) of all patients had one or more symptoms, and all the symptoms were mild. Hemangiomas occurred in the right and left rib at the same frequency (13/26 cases each). The tumors arose in all ribs from the 1st to 12th. In one report, they most frequently arose in the 7th rib [3]. Overall most of the hemangiomas (21/26 cases) occurred in the 5th and 8th ribs.
About half of costal tumors are malignant, and it is difficult to distinguish an osteolytic hemangioma of the rib from a malignant tumor such as a metastatic tumor, multiple myeloma, chondrosarcoma, or osteosarcoma [1]. Previous reports described osteolytic changes in 65.4% of cases (17/26 cases) on chest CT examination. On the other hand, osteosclerosis with calcification was described in a few reports [3,4].

The SUV\textsubscript{max} values of the rib in 18\textsuperscript{F}-FDG PET examinations in previous reports were 2.5 [5] and 2.2 [3], showing weak positivity. The SUV\textsubscript{max} of 2.66 in the this case indicated that the tumor was a low-grade malignant tumor or metastatic tumor. Bone scintigraphy using technetium 99m labeling reportedly revealed accumulation in a rib tumor [6], and 18\textsuperscript{F}-FDG PET examination is considered to be more useful than bone scintigraphy because it can indicate low or high accumulation [3]. The usefulness of 18\textsuperscript{F}-FDG PET examination should be estimated in additional cases.

The necessity of preoperative biopsy is controversial [7]. Some reports have stated that preoperative needle biopsy should be avoided due to the risk of seeding or bleeding [4,8], while others have found this technique to be safe and useful for obtaining a definitive diagnosis [3]. No complications of needle biopsy occurred in the present case. Aspiration biopsy of the tumor showed mainly blood cells, suggesting a hematoma or hemangioma.

The standard treatment of rib hemangiomas is complete excision with clean surgical margins [9]. Surgical resection was selected in 96.1% of previous cases (25/26 cases); radiotherapy and steroids were used in one case [10].

Hemangioma of the rib should be considered as a differential diagnosis of a weakly 18\textsuperscript{F}-FDG PET-positive osteolytic tumor of the rib, especially in asymptomatic or mildly symptomatic patients.

4. Conclusion

We experienced a rare case of hemangioma of the rib. This tumor can be misdiagnosed as a malignant tumor. Hemangioma of the rib should be considered as a differential diagnosis of rib tumors, principally in asymptomatic or mildly symptomatic patients. 18\textsuperscript{F}-FDG PET examination can be helpful for distinguishing hemangiomas of the rib from malignant tumors.

Conflicts of interest

None.

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Ethical approval

Not requested.

Consent

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

Authors contribution

AH acquired the data and wrote the article. AN coordinated and critically revised the study. Both read and approved the final manuscript.

Guarantor

Akira Haro.

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