

Hemangioma of the rib showing a relatively high 18F-FDG uptake: a case report with a literature review

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Abstract

Hemangioma of the rib is a rare benign tumor that is often difficult to distinguish from malignant bone tumors. Rib hemangioma often shows bony disruption with a slight cortical disruption, extraosseous lesion, and expanded bone on computed tomography (CT). We report the case of a 68-year-old man with atypical rib hemangioma with a slight cortical disruption and no expanded bone. The tumor showed relatively high 18FDG-uptake on positron emission tomography (PET)/CT. Rib hemangioma often shows higher 18FDG-uptake. PET/CT may not provide useful information for distinguishing rib hemangioma from a malignant tumor. Close observation without surgical resection may be feasible if the tumor is diagnosed as a rib hemangioma by biopsy.

Keywords

18F-fluorodeoxyglucose (FDG), computed tomography (CT), hemangioma, rib, positron emission tomography (PET)

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Introduction

Bone hemangiomas are benign hamartomatous vascular tumors. They account for less than 1% of all bone tumors (1). Approximately 50–80% of bone hemangiomas occur in the vertebral body or the skull (2). Hemangiomas of the rib are relatively rare; hence, they are often misdiagnosed as malignant tumors, such as metastatic tumors or primary malignant bone tumors. Given that 60–80% of the primary rib tumors are malignant (1), it is important to have knowledge of the features of rib hemangioma images.

We experienced a case with hemangioma of a rib showing relatively high 2-fluoro [fluorine-18]-2-deoxy-D-glucose (18F-FDG) uptake (maximum standard uptake value [SUV_{max}] = 2.9) on positron emission tomography (PET)/computed tomography (CT), which was difficult to distinguish from a malignant tumor. In this manuscript, we review the features of the rib hemangioma images and discuss the usefulness of PET/CT for distinguishing rib hemangiomas from malignant tumors. We discuss the surgical indications of rib hemangioma.

Case report

A bone tumor, 2 cm in diameter, of the fourth left rib was incidentally identified in a 68-year-old man upon a CT scan performed as part of his periodic examination for chronic pancreatitis. He did not want further investigation at the time, but the tumor had enlarged over half a year, and he then presented to our institution for a closer inspection.

He had no physical or visible symptoms. He had a history of myocardial infarction, hypertension, and

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chronic pancreatitis, but no history of malignancy. CT revealed a tumor in the neck of the fourth rib, $1.8 \times 1.6 \times 2.1$ cm in size, presenting as a multicystic lesion with soap bubble appearance, with a slight cortical disruption, extraosseous lesion, but no expanded bone (Fig. 1).

On magnetic resonance imaging (MRI), the lesion showed low to intermediate signal on T1-weighted (T1W) images, intermediate to high signal on the T2-weighted (T2W) images, and enhancement with gadolinium (Fig. 2a–c).

On PET/CT, the lesion showed relatively high ^{18}F FDG uptake ($\text{SUV}_{\text{max}} = 2.9$) (Fig. 3). Blood analysis demonstrated no remarkable findings and the tumor markers were negative. The tumor grew larger over the subsequent six months. In order to distinguish whether this was a malignant tumor, we performed open biopsy, with 20 mL bleeding. The specimen

showed hyperplasia of the small vessels in the bone marrow (Fig. 4). No malignant findings of endothelial cells of the vessel were made and he was diagnosed as having a hemangioma of bone.

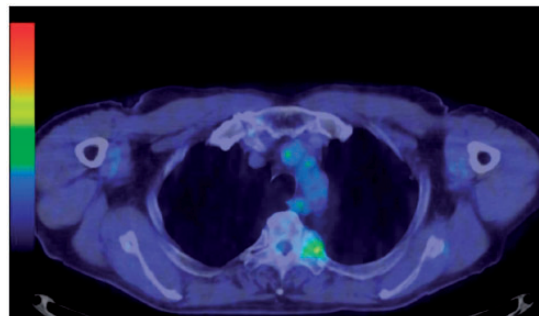


Fig. 3. Slightly increased FDG accumulation in the tumor on PET/CT image. The SUV_{max} value of the tumor was 2.9.

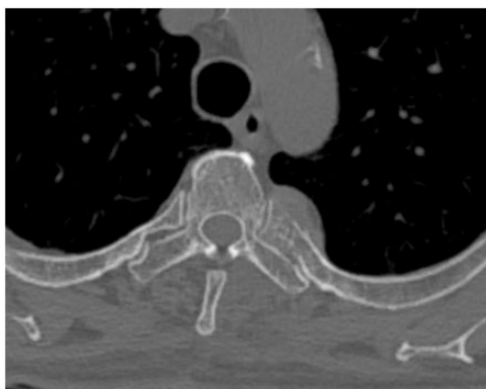


Fig. 1. CT image of the fourth left rib. The tumor occurred in the neck of the rib and showed bony disruption with a slight cortical disruption.

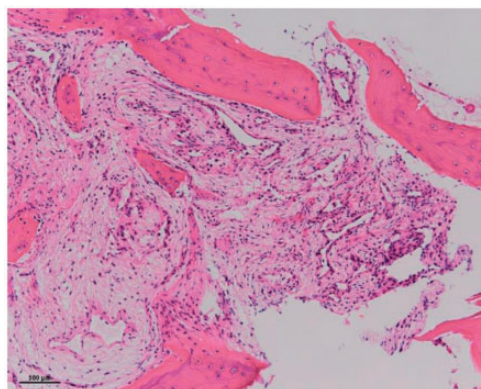


Fig. 4. Hematoxylin and eosin stain (original magnification $\times 100$). The tumor showed hyperplasia of the small vessels in the bone marrow, and a diagnosis of hemangioma was made.

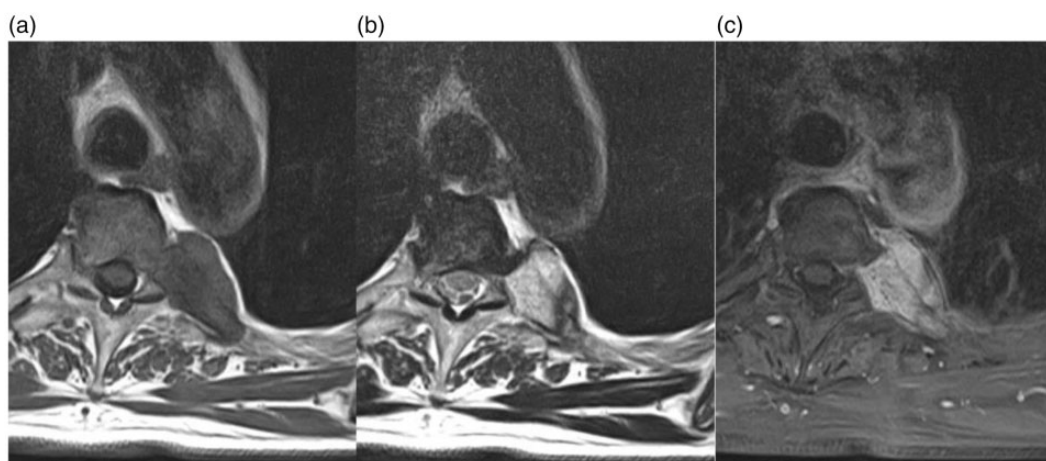


Fig. 2. MR images of the affected rib. (a) T1W sequence, (b) T2W sequence, and (c) T1W fat-saturation sequence with gadolinium enhancement.

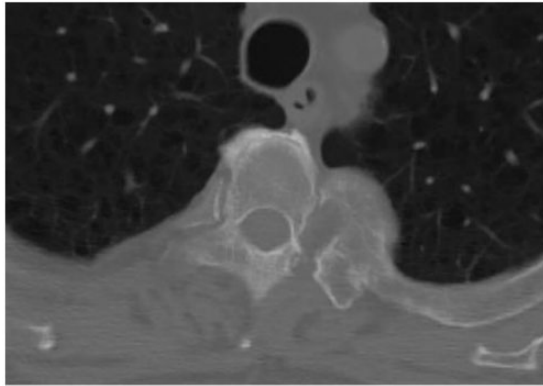


Fig. 5. CT image of the fourth left rib one year after the biopsy. The tumor showed calcification, but had not enlarged since the biopsy.

Surgical resection of the rib was not conducted as he was asymptomatic and hemangioma of the bone being a benign bone tumor. The patient was discharged on the eighth postoperative day without any complications. The tumor showed calcification formation but without any further growth, as assessed by CT one year after the biopsy (Fig. 5).

Discussion

Hemangioma represents 1% of primary bone tumors; the tumor occurs in the vertebral body or the skull in 60–80% of patients. Hemangioma of the rib is a relatively rare tumor. Hemangioma of the bone is usually asymptomatic, but one or more symptoms occur in 42.6% of cases if the lesion occurs in a rib (3).

Of primary rib tumors, 89% are malignant (2). The differential diagnosis of rib tumor is usually primary or metastatic. Malignant tumors include malignant lymphoma, myeloma, chondrosarcoma, osteosarcoma, and Ewing sarcoma (2). Thus, it is important to distinguish hemangioma from malignant tumors in the rib and this requires knowledge of the imaging features of rib hemangioma.

Hemangiomas in flat bones, including the rib, show cortical destruction with extraosseous mass and osteolytic lesions (4). We reviewed 22 previously published reports, 23 cases with 24 hemangiomas, obtained by using the key words “hemangioma AND rib,” and investigated clinical and radiological findings. Patients in these reports were aged 11–76 years.

Rib hemangioma is typically asymptomatic. Ten of the 23 described cases reported symptoms such as pain as the most common symptom. On CT, rib hemangioma often shows three features, i.e. bone disruption, extraosseous lesion, and expanded bone (2–23) (Table 1). Nineteen of the 24 hemangiomas showed bone disruption and 18 of the 24 hemangiomas

showed extraosseous lesions. In particular, all rib hemangiomas demonstrated expanded bone. However, in our case, the tumor showed bony disruption, with a slight cortical disruption, but the extraosseous lesion showed no expanded bone. A tumor may show expanded bone if the tumor grows larger; however, the tumor in our case was somewhat smaller than in the other reported cases. The literature review indicated that hemangiomas occur in the body of the rib more frequently than in neck of the rib.

Hemangioma of the rib demonstrates low to intermediate signals on T1W images, intermediate to high signals on T2W images, and enhancement with gadolinium on MRI (15), similar to Ewing sarcoma or malignant lymphoma on MRI. Hemangioma of bone shows various signals, according to the proportion of vessel and lipomatous soft tissue elements. Thus, MRI images of hemangioma may not be specific for diagnosis.

¹⁸F-FDG PET/CT is increasingly being used to obtain more information about benign and malignant tumors, including metastatic tumors. On PET/CT, hemangioma of the rib presents with an SUVmax of 2.2–6.7 (3,12,23). Therefore, PET/CT may not provide useful information for distinguishing rib hemangiomas from malignant tumors.

A mainstay treatment for hemangioma is reported to be surgical resection (20). In our review of the literature, resection was conducted in all cases. In some cases, embolization was conducted preliminary to reduce intraoperative blood loss or bleeding after biopsy (20,21). Resection of the tumors was often performed without biopsy. Complications, such as massive bleeding and hematoma, may occur during a preoperative biopsy, complicating the process of distinguishing malignant tumors from benign entities (15). Preoperative needle biopsy before resection was found to be valuable because the most suitable treatment for the rib tumor could then be determined (12). However, in some cases, resection was performed after the diagnosis of rib hemangioma by means of needle biopsy, because it was not possible to exclude the possibility of malignant tumor (3). We chose to use open biopsy, because the tumor occurred in the neck of the rib and obtained enough specimen with little bleeding.

Hemangioma is a benign tumor; hence, we did not perform surgical resection of the tumor. The tumor in our case showed calcifications and did not enlarge over the duration of one year after the biopsy (Fig. 5). Close observation without surgical resection may be feasible if the tumor is diagnosed as hemangioma by means of needle biopsy.

Preoperative embolization of large rib hemangioma can reduce the size (20), and cement or alcohol injection have been conducted as treatment of vertebral hemangiomas (24).

Table 1. Summary of the previous reported cases.

Age (years)	Gender	Location		Size (cm)	Expanded bone	Bony disruption	Extraosseous lesion	Symptom	Author	Year	References	
11	F	Right	6th	Neck	Not mentioned	+	-	+	Pain	Ortega	1986	(5)
65	F	Left	8th	Body	Not mentioned	+	-	+	Pain	Ortega	1986	(5)
56	F	Left	7th	Body	16 × 7 × 6	+	+	+	-	Kuo	1994	(6)
76	M	Right	3rd	Body	5 × 2.5	+	+	+	-	Clements	1998	(7)
59	F	Left	7th	Body	3.8 × 2.5	+	+	+	-	Okumura	2000	(8)
50	F	Right	1st	Body	Not mentioned	+	-	-	Thoracic outlet syndrome	Yeow	2001	(9)
45	M	Right	7th	Body	Not mentioned	+	+	+	Pain	Roy	2005	(10)
25	F	Left	8th	Neck	4 × 4 × 7	+	-	+	-	Ovali	2006	(11)
74	M	Left	5th	Body	9.5 × 6.5 × 3	+	+	-	Pain	Nakamura	2007	(12)
28	F	Right	5th	Neck	7 × 9 × 5	+	+	+	Pain	Sirmali	2007	(13)
54	F	Left	8th	Body	7 × 6 × 5	+	+	+	-	Cakir	2008	(14)
20	M	Right	5th	Neck	5	+	+	+	-	Tew	2011	(15)
48	F	Left	7th	Body	5 × 3.8	+	+	+	-	Abrao	2011	(2)
26	F	Right	8th	Neck	6.5 × 3 × 4.5	+	+	-	Swelling	Jain	2011	(16)
64	F	Left	5th	Body	8 × 4 × 2.5	+	+	-	-	Hashimoto	2011	(17)
54	F	Right	9th	Body	8 × 5 × 5	+	+	+	-	Jia	2011	(18)
57	F	Right	1st	Body	5.8 × 5.6 × 3.8	+	+	+	Not mentioned	Imai	2013	(19)
18	F	Right	3rd	Body	Not mentioned	+	+	+	Swelling	Deshmukh	2015	(20)
21	M	Left	5th	Neck	Not mentioned	+	+	+	Weakness in both lower limbs and loss of bowel bladder control	Shaik	2015	(21)
73	F	Left	6th	Body	2.9 × 2.5 × 1.9	+	+	+	-	Haro	2015	(3)
76	F	Left	4th to 6th	Body	10	+	+	+	Pain, intercostal paraesthesia, shortness of breath	Weinandt	2016	(22)
		Right	4th	Body	3	+	-	-				
63	M	Left	6th	Body	5.5 × 2.5 × 1.5	+	+	-	-	Park	2016	(4)
55	M	Right	3rd	Body	3.5 × 2.5 × 2	+	+	+	-	Saito	2016	(23)
68	M	Left	4th	Neck	1.8 × 1.6 × 2.1	-	+	+	-	This case	2017	-

If observation for rib hemangioma is decided, such percutaneous treatment might be one of the optional treatments.

In conclusion, the most common feature on CT was expanded bone. Observation without resection might be one of the treatments for rib hemangioma.

Declaration of conflicting interests

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