Adamantinoma of the Rib Metastasizing to the Liver

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Adamantinoma is a rare bone tumor, most of which originates in the long bone. Here, we present a case of adamantinoma of the rib with liver metastasis.

The patient, a 69-year-old man, complained of dull chest pain for over 6 months. Chest X-ray and CT film revealed osteolytic mass of the right 7th rib. Under the clinical diagnosis of hepatic tumor with rib metastasis, resection of the rib and partial hepatectomy were performed. Pathologically, the bone tumor was diagnosed as primary adamantinoma and the liver tumor was its metastasis. The patient has been well without adjuvant chemotherapy for 5 years after the operation.

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Introduction

Adamantinoma is a rare low-grade malignancy of the bone, occurring mostly in the long bones, and histologically resembles ameloblastoma and craniopharyngioma (1). Its histological origin still remains unclear. Maier (2) first reported an ulnar tumor with epithelial characteristics. Fischer (3) subsequently presented a similar long bone tumor and termed it adamantinoma. In Japan, 20 cases have been reported since 1973 (6–15, 16). Here, we report the first case of adamantinoma of the rib in Japan as far as we know. Furthermore, interestingly the tumor metastasized to the liver.

Case Report

A 69-year-old male was admitted to the Hiratsuka Kyosai Hospital on March 27, 1987 with complaints of rightside dull chest pain for 6 months. Physical examination failed to reveal any abnormality. Laboratory data on admission, including liver function tests and parameters relating bone metabolism were within normal limits (Table 1). The levels of serum tumor markers were also within normal limits. A tumor shadow at the right 7th rib was noted on the chest roentgenograph (Fig. 1a). The shadow consisted of multilocular, sharply-circumscribed lucent zones with irregular margins (Fig. 1b). The lytic area was predominantly situated in the medulla. The chest CT scan (Fig. 2) revealed bulging of the posterior surface of the right 7th rib with a mass, measuring 5×4×3 cm. The mass presented a soap-bubble appearance and was surrounded by a sclerotic margin.

The abdominal CT scan (Fig. 3) revealed a well-circumscribed, low-density area in the inferior medial region of the left hepatic lobe (S₄), measuring 5×5×2 cm. The bone scintigram demonstrated an abnormal uptake only in the right 7th rib (Fig. 4).

Although a hepatic malignancy metastasizing to the rib was suspected, the biopsied rib tumor did not show the representative features of any common hepatic malignancy. Under the

Table 1. Laboratory Data on Admission

WBC	7,100/mm ³	Na	137 mEq/l	Arterial blood gas	
RBC	511×10 ⁴ /mm ³	K	4.1 mEq/l	pH	7.32
Hb	17.2 g/dl	Cl	104 mEq/l	PCO ₂	42 Torr
Ht	48.8%	Ca	8.6 mg/dl	PO ₂	68.4 Torr
Plt	20.7×10 ⁴ /mm ³	BUN	17.8 mg/dl	HCO ₃	23.6 mmol/l
		Cr	1.0 mg/dl	BE	-1.1 mmol/l
		AMY	174 mg/dl		(room air)
TP	7.4 g/dl			CRP	(-)
A/G	1.51				
T.Bil	0.59 mg/dl	AFP	1.6 ng/dl		
GOP	21 IU/l	PIVKA-II	<0.1 AU/ml	PTH-intact	44 pg/ml
GPT	29 IU/l	CEA	1.7 ng/dl	Calcitonin	40 pg/ml
ALP	169 IU/l	CA19-9	6 U/ml		
γ-GTP	19 IU/l	TPA	110 U/ml		
CHE	320 IU/l				
LDH	319 IU/l	HBs Ag	(-)		
T-cho	224 mg/l	HCV Ab	(-)		
T.G.	287 mg/l	Urinalysis	n.p.		

Underlined data indicate abnormal findings. T-cho: total cholesterol, T.G.: triglyceride, AMY: amylase, TPA: tissue polypeptide antigen and n.p.: not particular.

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Fig. 1. Chest roentgenogram on admission (note the abnormal shadow in the right 7th rib; Arrow, DV (Fig. 1a) & right lateral oblique (Fig. 1b), and expanded figure in the inset picture). Note the osteolytic shadow of the right 7th rib.

clinical diagnosis of hepatic tumor with rib metastasis, the patient underwent resection of the right rib and partial hepatectomy, and has done well for 5 years after the operation without adjuvant therapy.

Histopathological findings

Gross findings

The resected specimen consisted of the right 7th rib and the

Fig. 2. Chest CT on admission. An osteolytic tumor was noted in right 7th rib; arrow and expanded figure in the inset.

Fig. 3. Abdominal CT scan on admission. Liver tumor (arrow) was noted as a low density area in the right lobe.

S₄ of the liver.

The middle portion of the rib contained a dark gray tumor measuring 5×4×3 cm. The tumor was well demarcated and soft in consistency. The liver contained a tumor, which was situated in the hepatic parenchyma, measuring 4×4×1 cm, and was well demarcated with a fibrous capsule. The tumor of the liver was homogeneously whitish-yellow in color, and elastic and firm in consistency. The surrounding hepatic parenchyma showed no cirrhotic change. There was no continuity between the rib and the hepatic tumor.

Microscopic findings

The histological characteristics of both tumors were similar. They were composed of solid cell nests surrounded by dense

Fig. 4. Bone scintigram. Note the abnormal galium uptake (arrow) in the right 7th rib.

fibrous stroma (Fig. 5a and 5b). The tumor cells were uniform in size and tightly connected to each other (Fig. 5c). In the margin of the cell sheets, the tumor cells were arranged in a palisading manner, showing a basaloid pattern. The cell sheets showed irregular branchings, and occasional gland-like spaces; Spaces filled with erythrocytes were also observed. The nucleocyto-plasmic ratio of the tumor cells was moderate. The nuclei were irregular in shape, and contained coarse chromatin and small nucleoli. The cytoplasm was weakly eosinophilic. Mitotic figures were rare. No bizarre giant cells were observed. The only difference between both tumors was the capsule formation. The tumor in the liver was well demarcated from the surrounding tissue with a dense fibrous capsule, while that in the rib was associated with osteolytic change without capsule formation.

PAS staining revealed basement membrane-like matrices around the cell sheets and intercellular portions, but no intracytoplasmic glycogen. Silver impregnation demonstrated reticulin fibers in the same portions. Alcian blue intensely stained the cytoplasm of the tumor cells and fibrous stroma.

Immunohistochemically, the tumor cells were positive for epithelial membrane antigen (DAKOPATTS, Glostrup, Denmark), keratin (DAKOPATTS), and carcinoembryonic antigen (DAKOPATTS), but negative for vimentin (DAKOPATTS), suggesting the tumor to be of epithelial origin. Based on the above findings, we concluded that the tumor originated in the rib and metastasized to the liver.

Fig. 5. Light micrographs of the tumor. a. Primary tumor of the rib (HE stain, ×40). The tumor was composed of solid cell sheets. Note the osteolytic change of the bone tissue. b. Liver metastasis (HE stain, ×100). c. Detailed features of the tumor cells (HE stain, ×200). The tumor cells form solid cell sheets with irregular-shaped nuclei and weakly eosinophilic cytoplasm; mitosis was rarely observed.

Discussion

Adamantinoma is a bone tumor which histologically resembles ameloblastoma and craniopharyngioma which are histologically characterized by solid cell nests. Adamantinoma

Table 2. Cases of Adamantinoma in Japan

No.	Author	(year)	Sex	Age	Laterality & anatomical site	History of traumatic injury	Local recurrence	Metastasis	Treatment	References reviewed in
1	T. Kushibiki et al	1973	M	29	R. humerus	—	—	—	S	6)
2	N. Takahashi et al	1976	M	29	tibia	—	+	—	S	6)
3	S. Sidahara et al	1977	M	53	L. tibia	+	—	—	S	6)
4	S. Masuda et al	1977	F	35	L. tibia	—	+	—	S	6)
5	(2 cases)		F	36	R. femur	—	—	—	S	6)
6	N. Mitsui et al	1977	F	67	L. tibia	—	—	—	S	6)
7	H. Miyaoka et al	1979	M	23	R. tibia	+	—	—	S	6)
8	H. Mori et al	1980	M	70	R. tibia	—	—	—	S	6)
9	H. Fukushima et al	1980	F	34	R. tibia	+	—	—	S	6)
10	R. Shima et al	1981	M	70	R. tibia	—	—	—	S	6)
11	K. Tachikawa et al	1983	F	58	R. femur	—	+	+ Lung	S	7)
12	K. Sakae et al	1984	M	31	L. tibia	+	—	—	S	8)
13	S. Akagi et al	1984	M	49	R. fibula	+	—	—	S	9)
14	Y. Miyauchi et al	1985	F	66	L. tibia	+	—	—	S	6)
15	Y. Ohsato et al	1988	F	1	L. tibia	+	—	—	S	10)
16	K. Asanuma et al	1989	M	17	R. tibia	—	—	—	S	11)
17	H. Ikeda et al	1990	F	51	R. tibia	+	—	—	S	12)
18	T. Tokita et al	1990	M	21	R. humerus	+	—	—	S & C	13)
19	R. Yokoyama et al	1990	M	51	R. radius	—	+	—	S	14)
20	G. Ishida et al	1990	M	23	R. humerus	—	—	—	S & C	15)
21	Present case	1994	M	69	R. rib	—	—	+ Liver (S ^d)	S	

Primary Adamantinoma of Rib										
No.	Author	(year)	Sex	Age	Laterality & anatomical site	History of traumatic injury	Local recurrence	Metastasis	Treatment	
1	David Plump	1986	F	49	R. 5th rib	—	+	+ Lung	S & R	17)
2	Present case	1994	M	69	R. 7th rib	—	—	+ Liver (S ^d)	S	

S: Surgical resection, C: chemotherapy, R: radiation.

generally occurs in the long bone, especially in the tibia. Among 21 Japanese cases of adamantinoma (6–15), 13 (61.9%) occurred in the tibia, 3 (14.3%) in the humerus, and 2 (9.0%) in the femur (Table 2). The present case was considered to be the first case of adamantinoma originating from the rib.

The patient ages ranged between 1 and 70 years old (mean; 42.0 years) and most were between 30 and 40 years old. A slight male preponderance was noted (male/female ratio, 13:8) (Table 2).

In general, the tumors grow slowly, and are frequently associated with dull pain or painless bone swelling. The present case showed typical dull pain for 6 months, no swelling was observed due to the internal growing of the adamantinoma. The low incidence of rib origin was the reason for the lack of consideration of adamantinoma in the initial diagnosis.

Roentgenologically, adamantinoma usually presents a well-circumscribed multilocular cystic and expansive shadow generally located in the diaphysis of the long bone, which resembles a soap-bubble appearance. The present case however, showed typical chest X-ray appearance of the adamantinoma. The rarity of adamantinoma of the rib mislead the diagnosis.

The histogenesis of adamantinoma has been controversial because of its various histological patterns. Many investigators

have concluded that the tumor should be considered to be of epithelial origin (5). The results of immunohistochemical examinations of the present case are consistent with epithelial origin; that is, positive for keratin and epithelial membrane, but not for vimentin.

Due to the high incidence of previous local traumatic injury (63%), Moon (16) proposed that adamantinoma is derived from posttraumatic inlets of epidermis. Among the Japanese cases, 43% (9 cases) also presented a past history of local traumatic injury. However, the present case had no previous chest trauma.

En-bloc resection is the operation recommended in various reports (4, 6, 16). However extensive treatment could be performed for recurrence and metastasis.

The prognosis of the adamantinoma reported by Moon (16) is generally fair; 82.5% survived also after the 4.4-year follow-up period (average). All of the Japanese patients except one (7) were alive at the time of the report. The good prognosis might be due to the relatively short follow-up period.

The apparent metastases in the 91 cases reported by Moon (16) were as follows; disseminated, 1; lungs, 9; ribs, 2; inguinal nodes, 3; abdomen, 2 and vertebrae, 2. Among the Japanese cases, only one patient except for the present case showed metastasis, which was found in the lung (7).

In summary, this is the first case of adamantinoma originating in the rib (17).

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