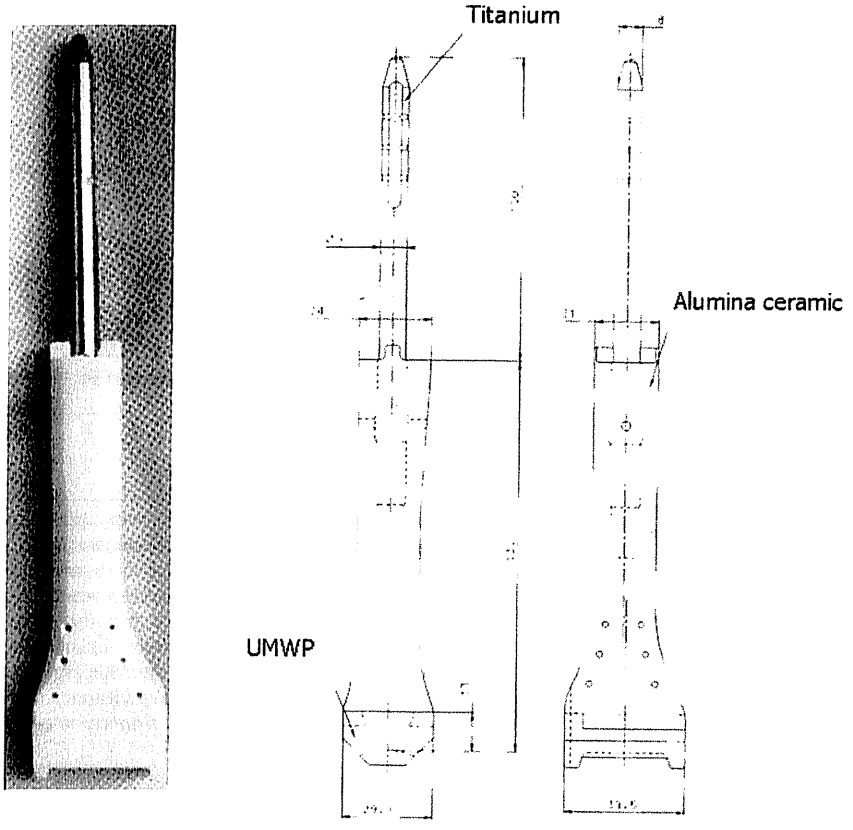

Fig. 1. Custom-made distal tibial prosthesis (Japan Medical Materials). The component is made from titanium (Ti–6Al–4V) intramedullary stem and alumina ceramic body. The articular surface of the joint element is covered with ultrahigh molecular weight polyethylene (UMWP) in order to achieve contact smoothly with the talar surface.

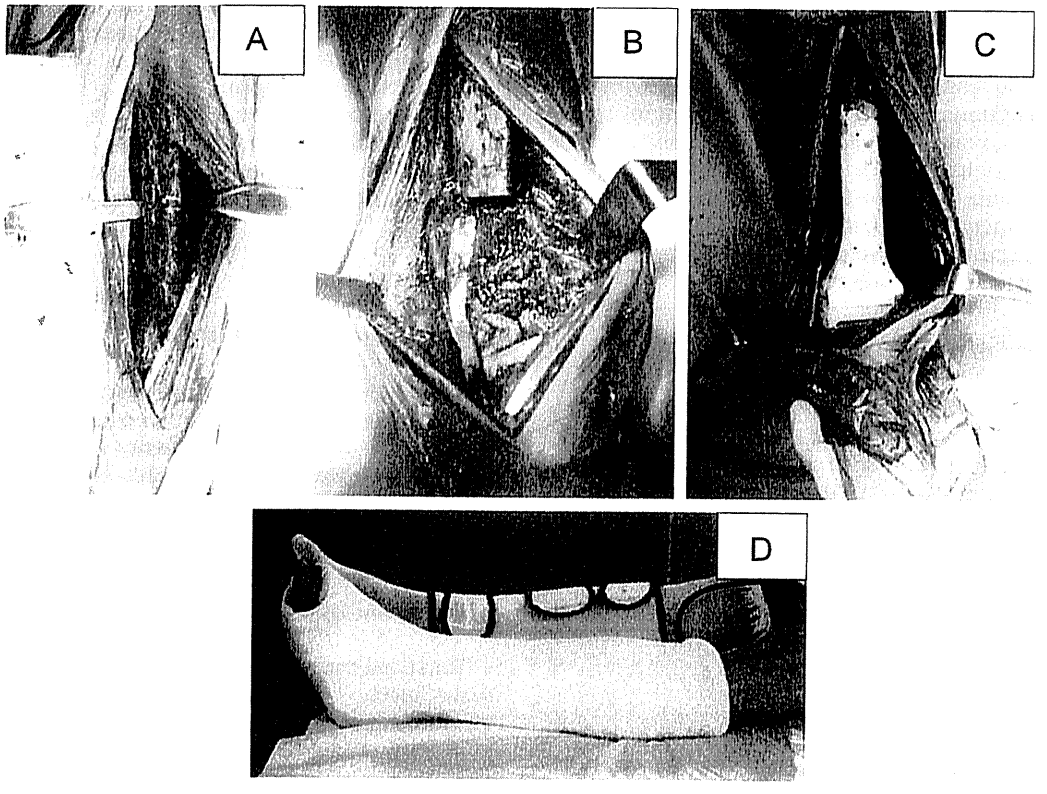

Fig. 2. (A, B) The bone was divided 5 cm above the upper margin of the tumor and resected en bloc. (C) The proximal intramedullary canal was reamed to the appropriate size. The stem of the tibial component was inserted into the medulla of the proximal portion of the tibia and fixed with bone cement, with the clinical outcome of appropriate rotational alignment in mind. (D) A short leg splint was applied for four weeks.

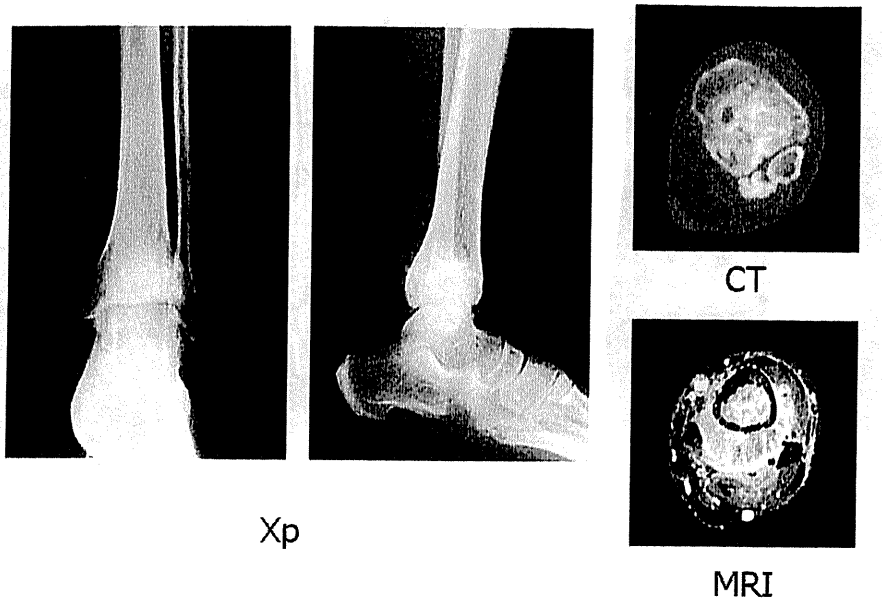

Fig. 3. A 52-year-old man with metastatic bone tumor from colon cancer. Anteroposterior, lateral radiograph, and CT showed mixed lytic and sclerotic lesions involving the distal tibia. Axial T-1 weighted image (Gd-enhanced) MRI showed extensive destruction of the distal tibia and extra-osseous soft tissue extension.

6. Case reports

6.1. Case 1

A 52-year-old male was treated at age 46 for adenocarcinoma of the colon with low anterior resection and chemotherapy. Four years after operation, the patient developed left-sided ankle pain. A needle biopsy led to the diagnosis of bone metastasis from colon cancer. Radiofrequency ablation (RFA) to this lesion was performed in a previous hospital; however, the treatment was not effective. Radiation therapy was additionally performed; however, after eight months, local recurrence occurred. Radiograph of the lower extremity showed mixed lytic and sclerotic lesions in the distal tibia (Fig. 3). The patient refused amputation for psychological and social reasons when informed of the recommended treatment. Instead, tumor resection with marginal resection and prosthetic reconstruction were carried out (Fig. 4). Although the fibular artery, flexor digitorum longus, and fibularis longus muscle were resected, the tibial nerve and posterior tibial artery were preserved. Intraoperative complication did not occur. Subsequently, the patient had local

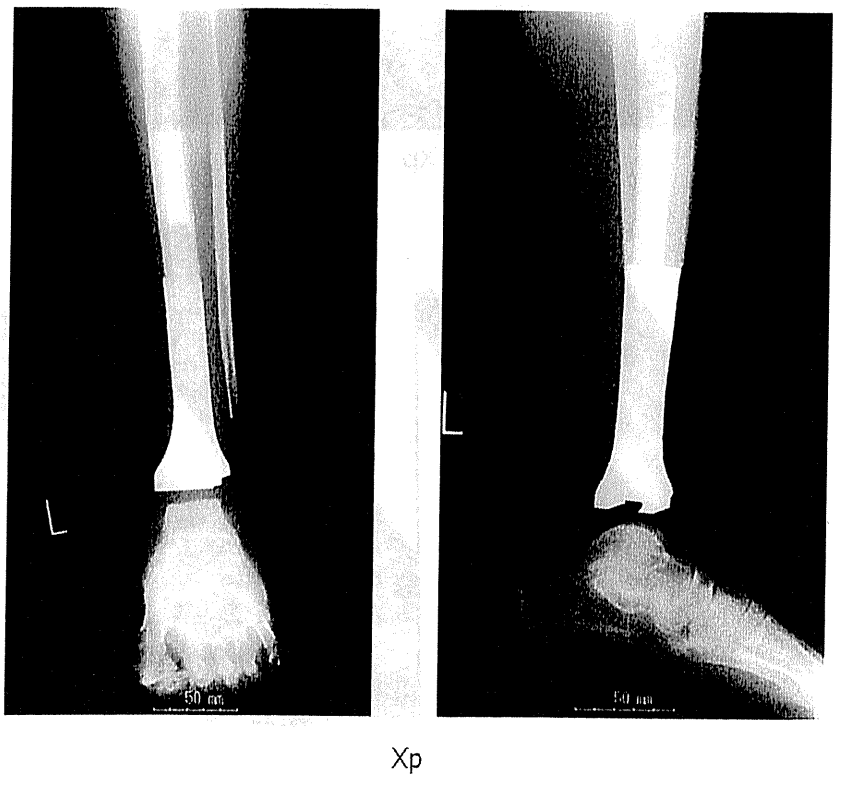

Fig. 4. Postoperative radiographs.

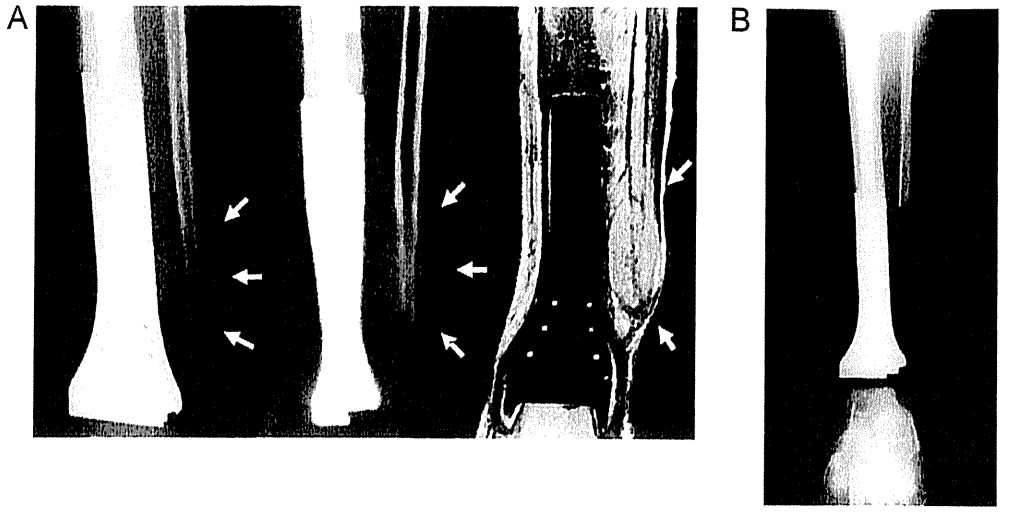

Fig. 5. (A) Local recurrence occurred in the distal end of the fibula eight months after primary surgery (arrow). (B) Second operation was performed and the recurrent lesion was resected.

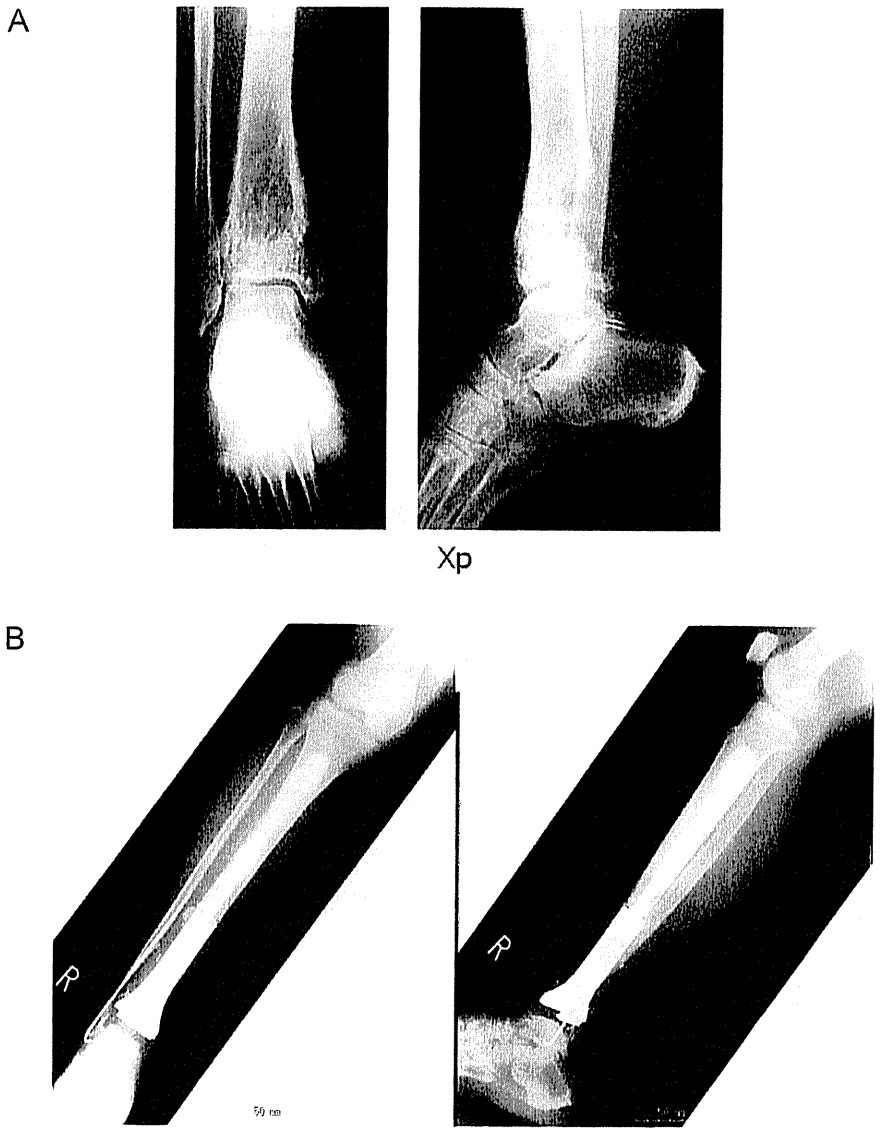

Fig. 6. (A) 52-Year-old woman with low grade central osteosarcoma. Anteroposterior and lateral radiographs showed destructive and osteolytic changes in the right distal tibia (A). Postoperative radiograph (B). Intraoperative fibular fracture occurred.

recurrence in the distal fibula (Fig. 5A). Resection was performed 8 months after the primary surgery (Fig. 5B). The patient remained free from disease at the latest follow up and showed neither radiological loosening nor subsidence of the talar component for three years. The range of ankle doriflexion was 10° and plantar flexion was 30°. The mean MSTS functional score was 25 (pain 5, function 3, emotional acceptance 5, walking ability 4, gait 4, and use of walking aids 4), which indicated 83.3% normal function.

6.2. Case 2

A 51-year-old female presented with a three-year history of swelling and slight tenderness on the right ankle. Pain slightly increased one month ago. A tumor in the distal tibia was pointed out in the previous hospital and the patient was referred to our hospital. Radiograph showed moth-eaten pattern of bone destruction in the right distal tibia (Fig. 6A).

Open biopsy was performed and histopathological diagnosis was low grade central osteosarcoma. The tumor with part of the tibial posterior muscle and flexor hallucis longus muscle were resected and custom-made prosthesis replaced the defect. The posterior tibial artery and nerve were not resected.

Fibular fracture occurred intraoperatively and the fracture was fixed intramedullary with *k*-wire (Fig. 6B). The range of ankle doriflexion after operation was −5° and plantar flexion was 30°. The patient remained free from disease at the latest follow-up and showed neither radiological loosening nor subsidence of the talar component for eight years. The mean MSTS functional score was 23 (pain 5, function 3, emotional acceptance 5, walking ability 3, gait 3, and use of walking aids 4).

7. Discussion

The distal tibia is an uncommon site for primary malignant bone tumors [1,2]. Abudu et al. [2] reported that of 190 patients with primary bone sarcoma of the tibia treated at one center for over 25 years, only 21 (11%) had the distal tibia as the location of the tumor. The most common of the primary bone tumors are the osteosarcomas and Ewing sarcoma [4].

Metastasis below the knee is also uncommon [5]. Lesson et al. [6] reported an approximately only 4% incidence below the knee.

There are limited indications for limb-preserving surgery in patients with malignant tumors of the distal tibia [2]. Most of the patients with malignant tumors of the distal tibia were treated by below knee amputation [1,2,7]. The principal concerns in the choice of limb-salvage surgery are the extent of soft-tissue invasion and the involvement of the distal blood vessels [1]. If neither the posterior tibial artery nor the dorsalis pedis artery proved to be salvageable, amputation should be carried out [1].

The many options of reconstruction, such as autograft, allograft, arthrodesis, implanted prostheses, and composites had been reported [1–3,8–11]. Casadei et al. [8] reported good functional and oncological results in patients with malignant bone tumors of the distal tibia, which were treated by resection and arthrodesis with autogenous bone graft. However, arthrodesis of the ankle has several limitations, including the loss of joint movement, long period of recovery, unpredictable outcome, possibility of non-union, need for multiple operations to achieve arthrodesis, and loss of ankle motion [1,2,12]. Although Bishop et al. [13] reported good results in a reconstruction using a vascular-free fibular graft in patients with malignant bone tumors of the distal tibia, allograft reconstruction is time-consuming, associated with significant risk of graft infection, prone to fractures, related to prolonged immobilization, and the outcome is unpredictable [2].

Ankle replacement arthroplasty has been occasionally carried out in patients with advanced arthritis [9–11]. Kofoed and Sørensen [10] observed a 73% rate of survival for their prosthesis at 14 years in arthritic patients; wherein, they emphasised accurate alignment and congruency of the joint. In this study, resection and prosthetic replacement for malignant bone tumor of the distal tibia were performed in two patients. Both patients were able to achieve rapid relief from pain and early return to maximum function of the limbs without major complications. Good early function, excellent stability of ankles, and long term local control of the tumor at the latest follow-up were observed. The patients still achieved 80% of the pre-morbid functional capacity at the time of last follow up and remained satisfied. Lee et al. [1] reported that talar collapse progressed until the talar component reached to the level of the subtalar joint; however, replacement of the talar surface in this study is not necessary to prevent talar collapse. Disadvantages are inevitable since the custom-made prosthesis takes 6 weeks to manufacture and there is risk for late failure and subsequent need for surgery.

Limb-salvage surgery using tumor prosthesis has an acceptable functional outcome in the medium-term analysis of two patients. This technique provides a safe and effective method of stabilization for properly selected malignant bone tumors of the distal tibia. To avoid amputation, it is important that patients with a suspected malignancy should be referred to a specialist center.

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Original Article

Eleven Cases of Cardiac Metastases from Soft-tissue Sarcomas

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Objective: Cardiac metastasis is a highly life-threatening condition because it leads to cardiac failure. However, it is difficult to diagnose because its precise clinical features are unknown. Here, we report 11 cases of cardiac metastasis from soft-tissue sarcoma, and discuss its diagnosis and treatment.

Methods: Of 641 patients with soft-tissue sarcoma treated in our institute between 1996 and 2009, we retrospectively reviewed the medical records of 11 patients whose cardiac metastases were diagnosed while they were alive.

Results: The most common primary tumor was leiomyosarcoma ($n = 5$), followed by clear cell sarcoma ($n = 2$). In all cases, metastases to other organs, including lungs ($n = 10$), soft tissues ($n = 5$) and bones ($n = 4$) were found along with cardiac metastases. Cardiac metastasis was diagnosed by echocardiography in six cases and by computed tomography in four cases. In four patients, cardiac metastasis was not detected by chest computed tomography as follow-up to lung metastases and echocardiography was required to make the diagnosis. Although five patients complained of exertional dyspnea, four were asymptomatic. Seven cases were treated with radiotherapy. No patient had surgery for their cardiac metastasis. The median survival of patients who received radiation therapy was 10.5 months; that of those who did not was 3.5 months.

Conclusions: Cardiac metastasis is often asymptomatic. Echocardiography is better than computed tomography for diagnosing cardiac metastasis, and should be considered in all patients presenting with soft-tissue metastases. Owing to the highly life-threatening nature of cardiac metastases and the possibility of soft-tissue dissemination, treatment with radiation therapy is recommended immediately on diagnosis.

Key words: cardiac metastasis – sarcoma – radiation

INTRODUCTION

Cardiac metastases are the immediate cause of death in as many as one-third of cases in which they occur (1). They may cause cardiac failure, arrhythmia and pericardial effusion, with cardiac failure being the main cause of death as a result of restrictive myocardial disease, ventricular inflow and outflow tract obstruction and pericardial constrictive disease. According to autopsy reports, cardiac metastases have been found in 25% of patients with soft-tissue sarcoma (1). The incidence of cardiac metastasis has increased during recent decades because better diagnostic tools and aggressive treatment of localized malignant tumors have led to longer

survival of patients (2). Despite their frequency, cardiac metastases are seldom diagnosed in living patients because other metastatic symptoms prevail. We reviewed 11 cases of cardiac metastasis from soft-tissue sarcoma, which had been diagnosed while the patients were alive, to discuss how to detect and treat them.

PATIENTS AND METHODS

We retrospectively reviewed the medical records of 641 patients with soft-tissue sarcoma treated at Osaka Medical Center for Cancer and Cardiovascular Diseases between 1996 and 2009. The 11 cases that had antemortem diagnoses of myocardial or pericardial metastases were further evaluated.

ILLUSTRATIVE CASES

PATIENT NO. 1

A 40-year-old woman with a history of mesenterium leiomyosarcoma treated with surgery three times, for the primary lesion and two local recurrences, showed lung metastases (treated with radiotherapy and chemotherapy), a right-buttock metastasis (treated with surgery), a left chest wall metastasis (treated with radiotherapy) and a left pelvic metastasis (treated with radiotherapy). Six years after the initial operation, she began to experience dyspnea and general fatigue. An echocardiogram revealed a 53 mm left atrial mass that blocked the left ventricular inflow tract (Fig. 1). Radiation therapy was impossible due to her inability to stay in position. She died on the day the cardiac metastasis was detected.

PATIENT NO. 2

A 55-year-old woman with a history of a right-buttock leiomyosarcoma treated with surgery and chemotherapy showed several metastases to her lung, left back, right forearm, left thigh and right buttock, all of which were surgically resected. Three years after the initial surgery, she presented with general fatigue. An echocardiogram revealed a 30 mm right atrial mass and cardiac tamponade resulting from a large pericardial effusion (Fig. 2). She received continuous drainage of pericardial effusion and radiation therapy for cardiac metastasis, receiving a total dose of 25 Gy in five fractions of 5 Gy. Twenty months after the radiation for cardiac metastasis, re-growth was observed, which was refractory to 25 Gy re-radiation. She died 3 months after the second radiation.

RESULTS

Metastatic disease to the heart was found in 11 cases of soft-tissue sarcoma, including the myocardium in 8 cases and the pericardium in 3 cases. The clinical features of these patients are shown in Table 1. The median age at initial diagnosis was 33 years. The median interval from initial diagnosis to cardiac metastasis was 36 months (range 8–108 months). The most common primary tumor was leiomyosarcoma ($n = 5$), followed by clear cell sarcoma, rhabdomyosarcoma, alveolar soft-part sarcoma, synovial sarcoma and malignant fibrous histiocytoma. Myocardial metastases favored the left over the right side of the heart, with the left ventricle predominating. In all cases, metastatic disease was found in other organs before or concurrently with the cardiac metastasis. The lungs were involved in 10 cases (91%), soft tissues in 5 cases (45%), bone in 4 cases (36%), pancreas in 2 cases (18%), kidneys in 2 cases (18%) and liver in 2 cases (18%).

Echocardiography provided the definitive diagnosis in six cases of this series, with four cases being diagnosed using computed tomography (CT). In one case, pericardium metastasis was found accidentally during surgery of lung metastasis. Clinical symptoms at diagnosis of cardiac metastasis included dyspnea in five cases, and general fatigue in two cases. Four patients were asymptomatic, which is more than might be expected.

Three patients required continuous drainage of pericardial fluid. In two of these three patients, pericardial effusion increased during chemotherapy. Seven cases of cardiac metastasis were treated with radiotherapy. No patient in this series had surgery for their cardiac metastasis. Survival from diagnosis of cardiac metastasis, excluding the one surviving case, was 8 months on average. Median survival of patients who received radiation therapy was 10.5 months, compared with 3.5 months for patients who did not receive radiation.

DISCUSSION

Hallahan et al. (1) reported that cardiac metastases were present in 25% of consecutive autopsies of patients with soft-tissue sarcoma, which is higher than recognized clinically, suggesting that most cases are probably missed. Cardiac metastasis often causes immediate death of a patient, so establishment of appropriate management is very important. However, little is known in terms of its precise clinical features.

The most common symptom of cardiac metastasis in this series was dyspnea, but, as many of the patients also had lung metastases, this is not clearly indicative of cardiac metastasis. It was not unusual for patients with cardiac metastasis to be asymptomatic. Furthermore, there were four cases whose cardiac metastases were not detected on chest CT as follow-up for lung metastases and were later found by echocardiography. It is supposed that chest CT failed to detect cardiac metastases even though tumors existed in hearts because chest CT showed mitral valve calcification and heart enlargement in case 1 and a small amount of pericardial effusion in case 2 but not cardiac metastases. In two cases (cases 6 and 8), patients received echocardiography despite being without symptom in order to investigate

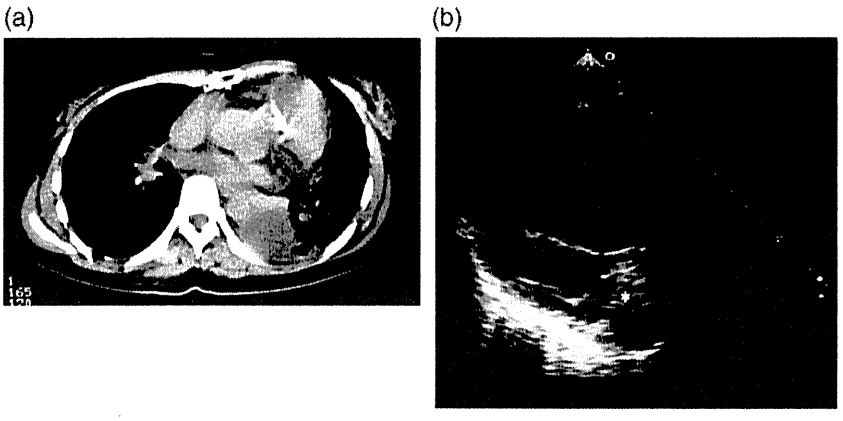

Figure 1. Chest computed tomography (CT) and echocardiogram of Patient 1. (a) and (b) Chest CT showed mitral valve calcification, heart enlargement and multiple lung metastases, but not cardiac metastasis. Two months later, her dyspnea and general fatigue had worsened. An echocardiogram revealed a 53 mm left atrial mass (*) that blocked the left ventricular inflow tract.

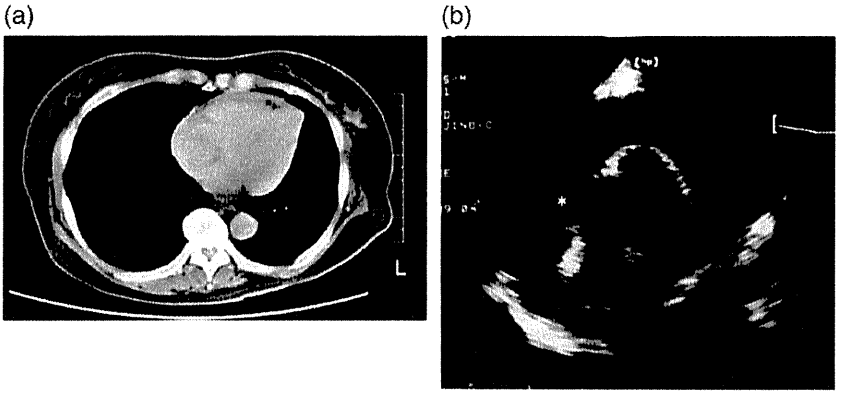

Figure 2. Chest CT and echocardiogram of Patient 2. (a) and (b) Chest CT showed a small amount of pericardial effusion and lung metastases, but not cardiac metastasis. Three months later, she presented with general fatigue. An echocardiogram revealed a 30 mm right atrial mass (*) and cardiac tamponade resulting from a large pericardial effusion.

whether she had myocardial damage by doxorubicin (case 6) and whether he had cardiac function to tolerate surgery for bone metastasis (case 8). Their cardiac metastases were found accidentally by those echocardiograms. In this case series, myocardium metastasis was often accompanied by soft-tissue metastases, possibly as a result of the intracardial metastasis sending malignant tumor cells into the circulation. We recommend that echocardiography should be performed on all patients with soft-tissue metastases.

While theory suggests that the filtration of systemic venous return by the pulmonary circulation would effectively shield the left ventricle compared with the right ventricle, previous autopsy studies have concluded that the left ventricular myocardium is most often the focus of metastatic disease, purportedly due to its level of vascularity (3). In this study, metastases of the heart were observed more often on the left than the right side. Left-side metastasis of the heart is considered to arise from coexistant lung metastases.

In this study, the most common histological type of cardiac metastasis was leiomyosarcoma. The histological types of cardiac metastases have not been included in autopsy reports (1). Leiomyosarcoma is the predominant sarcoma arising from large blood vessels, and it has been reported to be as common as angiosarcoma in primary cardiac sarcoma (4). These facts suggest that leiomyosarcoma can grow easily in the heart and vessels.

Hughes et al. (5) reported that cardiac metastasis correlated positively with pericardial effusion in pediatric sarcoma patients. Instillation of tetracyclines (6), radioactive phosphorus (7) or cisplatin (8) was reported to be a useful palliative procedure for malignant pericardial effusion. Radiation therapy prolongs life in patients with pericardial effusion, compared with repeated pericardiocentesis alone (9). In this series, continuous drainage of pericardial effusion in three cases and emergency pericardiotomy in one case were required for cardiac tamponade. Pericardial effusion increased during chemotherapy in two cases due to hydration of chemotherapy. After radiation therapy for their cardiac metastasis, the need for continuous drainage of cardiac effusion ceased. Therefore, radiation therapy for cardiac metastasis should be performed prior to chemotherapy.

Table 1. The clinical features of 11 cases of cardiac metastases from soft-tissue sarcomas

Pt. no.	Age	Sex	Diagnosis	Primary site	Primary size (cm)	Depth	Histological grade	AJCC staging	Time to initial met (month)	Time to heart met (month)	Heart met site	Other metastases	Chief symptom	Diag. modality	Therapy	Survival from heart met (month)
1	40	F	Leiomyosarcoma	Mesenterium	N/A	Deep	N/A	N/A	32	65	LA	Lung, soft tissue, bone	Dyspnea	Echo	–	0
2	55	F	Leiomyosarcoma	Rt buttock	8	Deep	2	II B	8	36	RV	Lung, soft tissue	Fitigue	Echo	RT (25 Gy/5Fr)/DCE	24
3	63	F	Leiomyosarcoma	Uterus	N/A	Deep	N/A	N/A	24	108	LV	Lung, soft tissue, liver, stomach, bladder, renal	Asymptom	CT	RT (45 Gy/15Fr)	13
4	47	M	Leiomyosarcoma	Chest wall	12	Deep	2	IV	0	30	LV	Lung, soft tissue, bone	Dyspnea	Echo	RT (50 Gy/25Fr)	7
5	26	F	Leiomyosarcoma	Rt groin	7	Deep	2	II B	33	51	LV	Lung	Asymptom	CT	RT (60 Gy/30Fr)	18 (AWD)
6	30	F	Clear cell sarcoma	Rt knee	7	Deep	3	IV	0	31	LV	Lung, soft tissue, bone, lymph	Asymptom	Echo	Immunotherapy	4
7	33	F	Clear cell sarcoma	Rt foot	3	Deep	3	IV	0	64	Pericardium	Lung	Asymptom	OP	–	5
8	23	M	ASPS	Lt shoulder	8	Deep	2	IV	0	27	LV	Lung, bone, pancreas	Asymptom	Echo	RT (60 Gy/ 30Fr)/Chemotherapy	8
9	69	F	Undifferentiated pleomorphic sarcoma	Rt thigh	10	Deep	3	III	11	13	RA	Pancreas	Dyspnea	CT	–	5
10	16	M	Rhabdomyo sarcoma	Lt buttock	8	Deep	3	III	8	8	Pericardium	Lung	Fatigue	Echo	RT (40 Gy/20Fr)/ Chemotherapy	6
11	13	F	Synovial sarcoma	Lt buttock	12	Deep	3	IV	0	57	Pericardium	Lung, liver, renal	Dyspnea	CT	RT (32 Gy/16Fr)/ DCE/Pericardiotomy/ Chemotherapy	5

Histological grade: according to Fédération Nationale des Centres de Lutte Contre le Cancer grading system for leiomyosarcoma, undifferentiated pleomorphic sarcoma and synovial sarcoma, but National Cancer Institute grading system for others.
AJCC staging: American Joint Committee on Cancer (AJCC) staging at the first presentation according to AJCC staging manual seventh edition.
OP: pericardium metastasis was found accidentally during operation of lung metastasis.
CT, computed tomography; RT, radiation therapy; DCE, drainage of cardiac effusion; N/A, not available; ASPS, alveolar soft part sarcoma; LA, left atrium; RV, right ventricle; RA, right atrium.

The seven patients who received radiation therapy tended to survive longer than those who did not. The tolerance of the heart to radiation therapy has been reported previously (10). Al-mamgani et al. (11) suggested that a total dose of 45 Gy, in 25 fractions of 1.8 Gy, would be appropriate for cardiac metastasis in a curative setting; an additional dose of 10–15 Gy may be delivered through small portals when indicated. In this study, cardiac metastasis treated with 25 Gy of radiation therapy showed re-growth only after 20 months, when re-irradiation was required.

There was no patient who underwent biopsy or surgery for tumor in the heart in this series. Although the possibility remains that tumors in the heart are primary tumors, we decide they are metastases from soft-tissue sarcoma from clinical course. In some cases, we performed resection of the tumors in the lung and/or soft tissue and confirmed they were metastases from soft-tissue sarcoma. While there have been some reports of long-term survival after resection of cardiac metastasis, the perioperative mortality rate remains high (40%) (12). Patients with diffuse cardiac metastases, massive intracavitary disease or uncontrolled metastatic disease have been reported to be poor-risk patients for surgical resection (1).

In conclusion, we suggest that echocardiography should be considered in cases of sarcoma presenting with soft-tissue metastases, and, because of the highly life-threatening nature of the condition and the possibility of soft-tissue dissemination, cardiac metastases should be treated by radiation therapy, a total dose of more than 45 Gy, immediately and prior to chemotherapy.

Funding

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Conflict of interest statement

None declared.

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Malignant Mixed Tumor of the Soft Tissue Occurring After Total Knee Arthroplasty

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abstract

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A 70-year-old woman developed a malignant mixed tumor of the soft tissue 2 years after total knee arthroplasty. A 5×3×3-cm elastic hard tumor at the lateral side of the surgical scar was resected. The tumor showed focal infiltration into surrounding adipose and fibrous tissues, focal necrosis, and vascular infiltration. It was diagnosed as malignant. Mixed tumor, or myoepithelioma, of the soft tissue is a relatively rare tumor that was recently recognized as a disease entity; the vast spectrum of myoepithelial cell differentiation and the resultant morphologic diversity might increase the difficulty of the histological diagnosis. Postoperatively, the patient did not receive adjuvant therapy and no recurrence of the tumor was observed for 6 years. Range of motion of her left knee is –5° extension and 90° flexion; however, her activities of daily living are restricted because of general fatigue, partly due to hepatoma and chemotherapy.

Despite the increase of artificial implant use worldwide, reports of peri-implant tumor formation are rare. Although we do not know the exact mechanism of tumor genesis, we consider the fibroblast formation in the routine healing process to be a possible mechanism. Further investigation is necessary to identify coexisting factors that increase the risk of tumor formation after implantation.

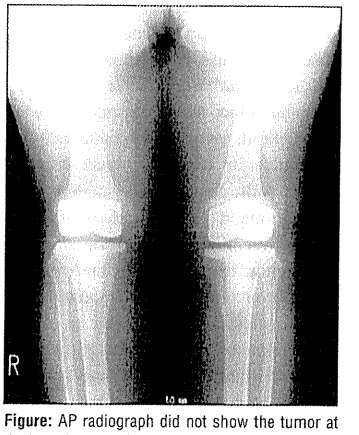

Figure: AP radiograph did not show the tumor at the lateral side of the implant of the left knee.

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The oncogenic potential of artificial implants has long been a concern, but the precise risk of tumor formation after implantation is unclear.[1] However, several case reports and animal model studies may provide information about the tumorigenic risk after implantation.[2-4] Formation and dissemination of metal particles originating from the implant and chronic inflammation have been discussed as causes of implant-inducing tumorigenesis.[3,4]

Myoepithelioma is a neoplastic proliferation of myoepithelial cells commonly observed in the salivary glands, also known as pleomorphic adenoma.[5] Myoepithelioma is also reported in soft tissue, skin, lungs, and breasts.[6-8] In soft tissue, myoepithelioma shows reticular growth of epithelioid, ovoid, and spindle cells in collagenous or chondromyxoid stroma. Tumors with ductal differentiation are known as mixed tumor, and those without ductal differentiation are known as myoepitheliomas; these 2 tumors are considered to lie on a morphologic continuum.[7]

The criteria of malignancy in myoepithelioma and mixed tumors of the soft tissue are slightly different from that in myoepithelioma and mixed tumors of the salivary glands. In the salivary glands, tumors showing cytologic atypia, increased mitotic activity, or infiltration into surrounding tissue are regarded as malignant.[9] Infiltration is not a criterion for malignant myoepithelioma or mixed tumors of the soft tissue.[7]

CASE REPORT

A woman underwent bilateral total knee arthroplasty (TKA) because of osteoarthritis. Two years after the TKA, at age 70, she noticed a lump in her left knee with associated pain and numbness. Tinel's sign was observed. A 3.5-cm mass that was elastic hard on palpation was found on the lateral side of the surgical scar (Figure 1). Magnetic resonance imaging and computed tomography scanning were not performed because TKA had been performed. 201Tl Scintigraphy (GE Healthcare, Chalfont St Giles, Buckinghamshire, United Kingdom) showed high radioactivity, suggesting the possibility of malignancy. Cytologic examination and biopsy were performed.

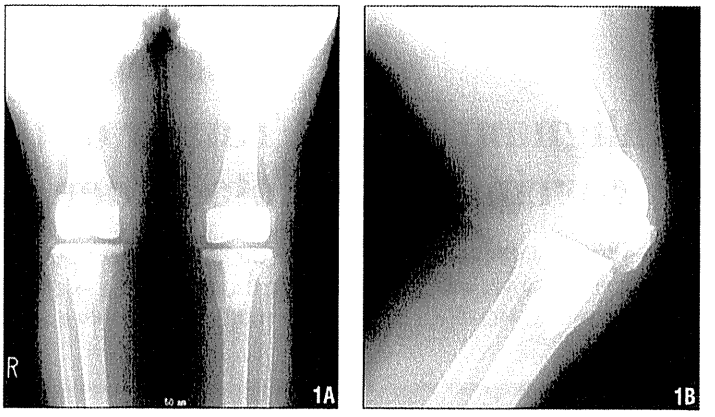

Figure 1: Anteroposterior (A) and lateral (B) radiographs did not show the tumor at the lateral side of the implant of the left knee.

Histological examination suggested a mesenchymal tumor because there was proliferation of stromal cells in the prominent fibrous tissue. Examination also did not suggest a diagnosis of malignancy. With these findings, surgical resection of the tumor with wide local margin was performed, including the fascia on the medial margin and a part of capsule on the distal margin. The tumor was located close to the implant; however, no direct contact of the tumor with the implant was observed. The implant was exposed during the operation, and irrigation was performed to avoid infection.

Postoperatively, the wound healed well with no infection, and pain was controlled. Three months postoperatively, range of motion (ROM) of the knee joint was –5° extension and 90° flexion, and slight weakness of quadriceps was observed. Because of the resection of the lateral collateral ligament, lateral instability of the knee joint was observed. With the use of knee brace and a T-cane, the patient's gait became stable.

During the 6-year follow-up period, no local recurrences or distant metastases were observed. However, hepatcellular carcinoma was diagnosed 5 years postoperatively and the patient was receiving chemoradiotherapy for it. At 6 years postoperatively, ROM of the left knee was –5° extension to 90° flexion; however, the patient's activities of daily living were restricted because of general fatigue, partly due to the hepatoma and chemotherapy.

Biopsy specimen showed mesenchmal cell proliferation surrounded by fibrous tissue (Figures 2A, B). The cells were immunohistochemically negative for cytokeratin AE1/3, CD31, CD34, desmin, S100 protein, and CD68. Although a mesenchymal tumor was surmised, histological subtype and malignant potential of the tumor could not be decided.

After the resection, the tumor was macroscopically and microscopically examined. The 5×3×3-cm tumor, which was located in the articular caosule, contained the myxomatous area and was circumscribed by a 4- to 5-mm thick white and amorphous layer (Figure 3). Microscopically, tumor cells grow in lobules, showing a predominantly reticular growth pattern with intersecting cords of epithelioid, ovoid, or spindle cells with partial

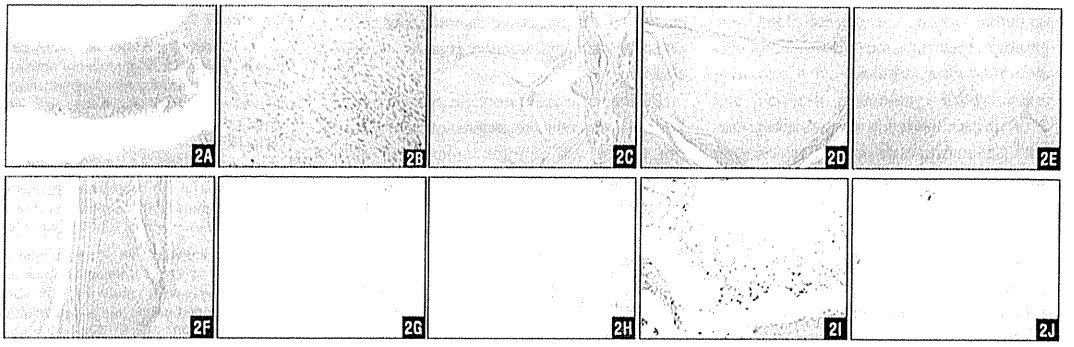

Figure 2: Biopsy specimen showed mesenchmal cell proliferation surrounded by fibrous tissue (hematoxylin-eosin stain; original magnification, ×10) (A). The proliferating cells did not show marked cytologic atypia (hematoxylin-eosin stain; original magnification, ×100) (B). Surgical specimen of the tumor. The tumor cells grow in lobules (hematoxylin-eosin stain; original magnification, ×20) (C). The tumor cells show in a reticular growth pattern (hematoxylin-eosin stain; original magnification, ×20) (D). Focal necrosis of the tumor was observed (hematoxylin-eosin stain; original magnification, ×20) (E). Vascular invasion of the tumor was seen (hematoxylin-eosin stain; original magnification, ×20) (F). The tumor cells show positive reactivity to cytokeratin AE1/3 (original magnification, ×20) (G), S100 protein (original magnification, ×20) (H), vimentin (original magnification, ×20) (I), and muscle actin (original magnification, ×20) (J).

hyalinization, collagenous stroma, and ductal differentiation (Figures 2C, D). The tumor showed focal infiltration into the surrounding adipose and fibrous tissues, focal necrosis, and vascular infiltration (Figures 2E, F).

Tumor cells were immunohistochemically positive for cytokeratin AE1/3, S100 protein, vimentin, epithelial membrane antigen, and muscle actin (HHF35) (Figures 2G-J). Mixed tumor of the soft tissue with malignant potential was diagnosed. Tumor cells were not observed in the resected margin. No metal particles or inflammatory features were observed in the tumor or surrounding tissue.

Discussion

Malignant tumor formation around artificial implants was first described in 1956 by McDougall,[2] who reported a malignant bone neoplasm resembling a Ewing sarcoma occurring at the site of bone plating. Since then, the incidence of surgical use of artificial implants has dramatically increased worldwide; however, reports of tumor formation concerning implants are rare. In this respect, risk of tumor formation after implantation seems to be negligible. In fact, several cohort studies on patients receiving hip or knee arthroplasty indicate no increases of cancer risk by implantation.[1]

However, several case reports, including the current report, clearly demonstrate the correlation between implants and tumor formation.[2-4] Furthermore, experimental studies in rats have shown that implantation of cobalt, chromium, nickel, polyethylene, and methylmethacrylate can induce the development of sarcomas.[3]

In the current study, the tumor was located close to the implant; however, no direct contact of the tumor with the implant was observed. This is distinct from the previous reports of implant-induced tumors arising close to the implants.[2,4] However, the close location of the tumor to the implant indicates the fibroblast formation in the routine healing process in the surgery area might have triggered tumorigenesis. Nevertheless, it should be recognized that this finding was entirely coincidental; thus, further studies are necessary.

Mixed tumor, or myoepithelioma of the soft tissue, is a relatively rare tumor that was recently recognized as a disease entity. Before the report by Hornick and Fletcher,[7] myoepithelioma of the soft tissue was a difficult tumor to diagnose, mainly due to the vast spectrum of myoepithelial cell differentiation and the resultant morphologic diversity in myo-

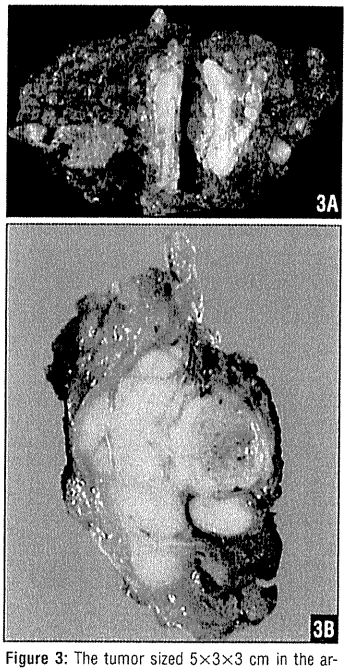

Figure 3: The tumor sized 5×3×3 cm in the articular capsule (A). The tumor contained myxomatous and collagenous areas (B). Scale bar: 1 cm.

epithelial tumors. Despite the broad morphologic spectrum, most cases of myoepithelioma of the soft tissue show a positive reactivity for cytokeratin, vimentin, and S100 protein, which is diagnostically useful.[7] The current case showed positivity in these immunohistochemistry.

Because of the rarity of the tumor, establishment of criteria for malignancy in myoepithelioma of the soft tissue is difficult. In the salivary gland, tumor infiltration into adjacent normal tissue is the most important criterion for malignancy, with or without the cytologic atypia.[9] However, this criterion was challenged in myoepithelial lesions of the soft tissue by Hornick and Fletcher,[7] who argued that infiltrative growth is insufficient for the diagnosis of malignancy but that cytologic atypia is essential in the diagnosis of malignancy. The current case did not show apparent cytologic atypia; however, the tumor was diagnosed as malignant because necrosis and vascular invasion were observed.

Further case collection and prospective studies are needed to determine whether this tumor was simply coincidental or whether a true association exists between myoepithelioma and TKA.

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Cardiac Synovial Sarcoma Swinging Through the Aortic Valve

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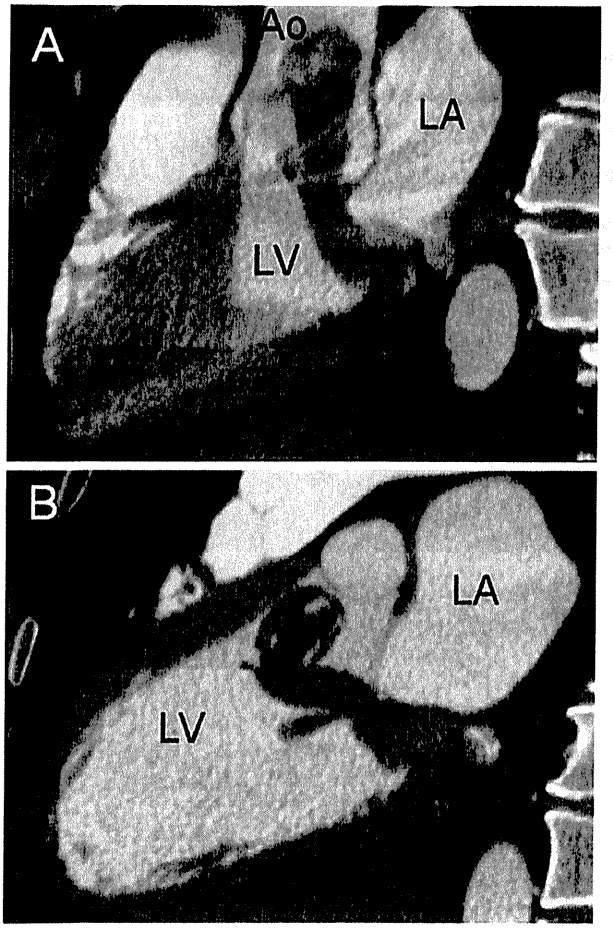

Fig 1.

A 34-year-old man presented with transient atrial fibrillation. Contrast-enhanced electrocardiographic-gated, 64-multidetector computed tomography revealed a left ventricular mass (Fig 1; Ao = aorta; LA = left atrium; LV = left ventricle) located on the posterior wall of the LV beneath the mitral annulus. The head of the tumor plunged through the aortic valve into the aorta in systole (Fig 1A) and retracted into the LV in diastole (Fig 1B) like a pendulum. Cardiac magnetic resonance images indicated the tumor tissue to infiltrate into the myocardium (Fig 2; arrow).

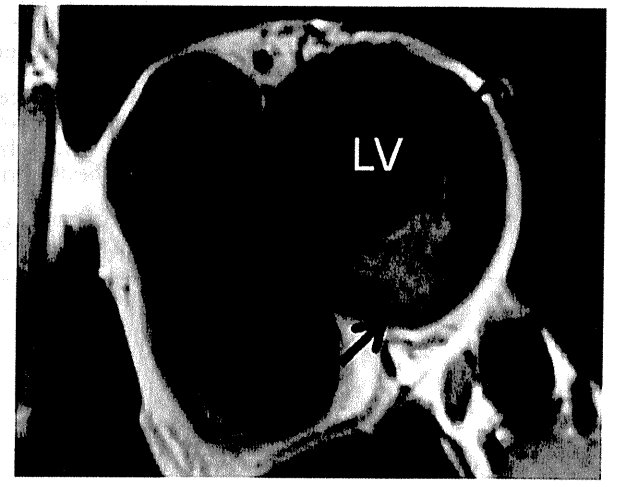

Fig 2.

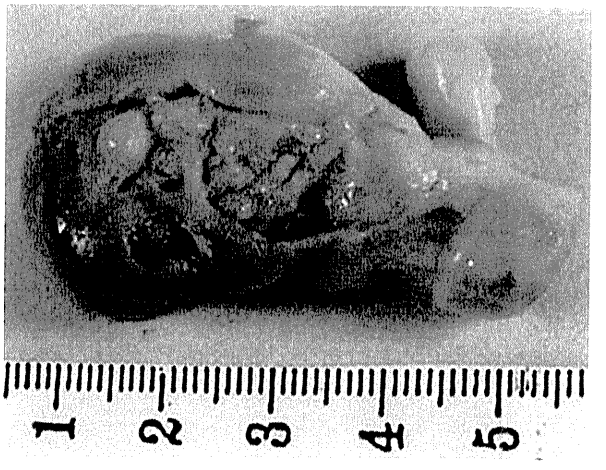

Fig 3.

During surgery, the tumor was severed from the left ventricular wall leaving a slight stump, preventing a potential wall rupture. The surgical specimen showed an elastic hard mass with a smooth surface (Fig 3). Histologic and molecular examination revealed that the tumor was a synovial sarcoma, with t(×18) chromosomal translocation. The patient underwent postoperative chemotherapy to treat any residual tumor cells.

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CASE REPORT Open Access

Case of an unusual clinical and radiological presentation of pulmonary metastasis from a costal chondrosarcoma after wide surgical resection: A transbronchial biopsy is recommended

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Abstract

Chondrosarcomas are the most frequently occurring primary malignant chest wall tumors. Furthermore, the lungs serve as the most frequent sites for metastases. Pulmonary metastases from sarcomas usually appear as round nodules of varying sizes on roentgenograms. Here, we report an unusual clinical and radiographic presentation of pulmonary metastasis from a costal chondrosarcoma. Bilateral pulmonary metastases developed soon after wide surgical resection. Thoracic computed tomography revealed unusual radiological findings: consolidation accompanied with ground-glass opacity. To confirm the metastasis, we recommend a transbronchial biopsy in cases where unusual pulmonary findings are detected.

Background

Chondrosarcomas are the second most frequent primary malignant bone tumors, after osteosarcomas [1,2]. They are also the most common primary malignant chest wall tumors: 5-15% of chondrosarcomas are located in the thoracic wall [3]. Since radiotherapy and chemotherapy are generally ineffective against chondrosarcomas, surgery is the only curative treatment, and the quality of the surgery is an essential prognostic factor [2]. Enneking et al. classified surgical margins into wide, marginal, and intralesional [4]. A wide resection is accomplished by a procedure in which the lesion, its pseudocapsule and/or reactive zone, and a surrounding cuff of normal tissue are taken as a single block. Therefore, resection for chest wall chondrosarcoma should be wide, taking intact pleura internally, intact muscle fascia externally, and transverse rib resection > 2 cm from the tumor on both directions [4,5]. Clinically, the involved rib en bloc should be resected along with the 2 intercostal spaces above and below the tumor.

On roentgenograms, pulmonary metastases usually appear as multiple peripheral, round nodules of varying sizes. Here, we describe an atypical presentation of pulmonary metastasis occurring soon after wide surgical resection of a costal chondrosarcoma. In this case, a thoracic computed tomography (CT) scan showed consolidation, predominantly in both the lower lobes, surrounded by ground-glass opacities and air bronchograms, mimicking serious pneumonia.

Case presentation

A 62-year-old woman was admitted to our hospital because of a mass that grew gradually in the right lateral chest wall for 1 year. Physical examination revealed a tumor (5 × 3.5 cm) in the right eighth rib. The mass was hard with an unclear border, no mobility, redness, or local heat, but it was tender. An X-ray revealed a mass with coarse calcification located on the right eighth rib, expanding beyond the irregular cortex. Thoracic CT revealed a 70 × 60 × 30 mm low-density mass

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(CT value, +18 HU) along the right eighth rib; it arose at the bone-cartilage border and destroyed these tissues (Figure 1a-c). No pulmonary metastasis was observed (Figure 2a). Other metastatic workup, including PET scan, was negative. The physical examination and imaging findings strongly indicated primary chondrosarcoma. Therefore, wide surgical resection was performed without performing a biopsy; the tumor was resected together with the right seventh, eighth, and ninth ribs. Transverse rib resection was performed >4 cm from the tumor in both directions. The chest wall was reconstructed using a Dexon mesh® (US Surgical, Connecticut, USA). Histological examination revealed a grade II chondrosarcoma with increased cellularity and myxoid stroma (Figure 3). All resected surgical margins were wide. The postoperative course was uneventful, and the patient was discharged 2 weeks after the operation.

However, 7 weeks after the definitive surgery, she presented with a slight fever, dyspnea, persistent dry cough, and purulent nasal discharge of 1-week duration. The white blood cell count (WBC)and C-reactive protein (CRP) level were 8.2 × 10^9 cells/L (neutrophils, 75%; lymphocytes, 16%; monocytes, 4.7%) and 3.7 mg/dL (normal: <0.30 mg/dL) respectively. Findings of other biochemical and serologic tests were normal. The chest roentgenogram showed air-space consolidation accompanied with an air bronchogram in the right upper and left lower lung fields (Figure 4) - a finding highly suggestive of bacterial pneumonia. Antibiotics (tazobactam/piperacillin [TAZ/PIPC]) administered for 7 days showed no results. Thoracic CT revealed pulmonary non-segmental consolidation, predominantly in the peripheral lung field, surrounded by ground-glass opacities; bronchovascular bundle thickness and interlobular septal thickness were absent (Figure 2b). Bronchoscopy and consequent transbronchial biopsy revealed blood vessel proliferation in the bronchial wall. Therefore, we considered this as a case of interstitial pneumonia such as cryptogenic organizing pneumonia, and initiated glucocorticoid therapy without waiting for the biopsy results. However, 3 days after the onset of the treatment, transbronchial biopsy sample through the left S8 bronchus confirmed the same histological features as the primary tumor in the peritumoral lumen structure, which was negative for CD34 and D2-40 (Figure 5a, b). The bronchoalveolar lavage fluid culture was negative. The patient died 12 weeks after the definitive surgery.

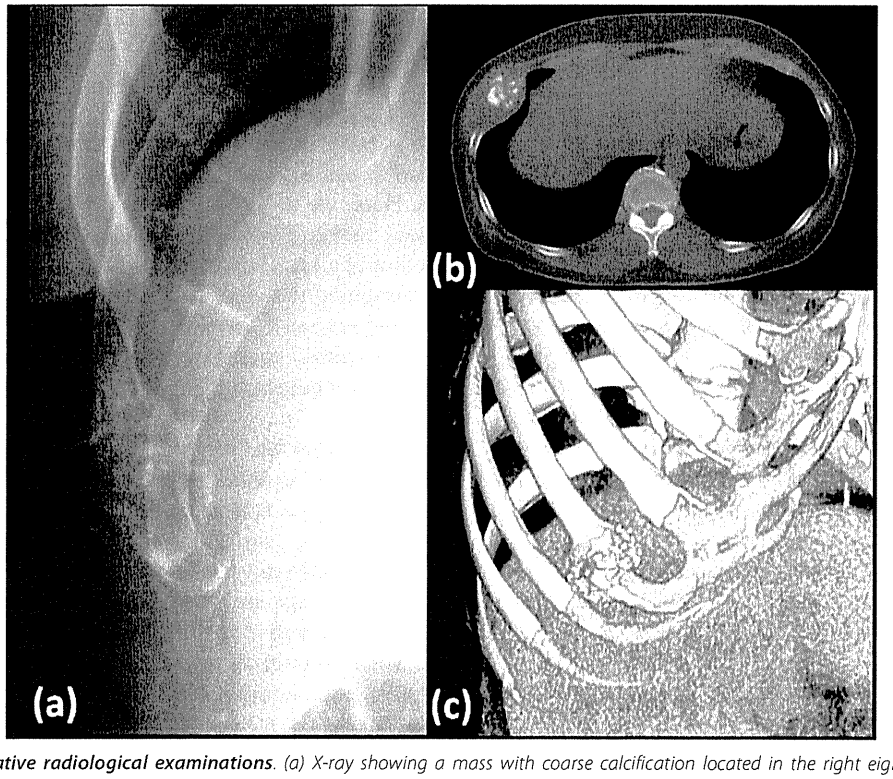

Figure 1 ***Preoperative radiological examinations***. *(a) X-ray showing a mass with coarse calcification located in the right eighth rib, expanding beyond the irregular cortex. (b) CT scan showing a low -density mass with coarse calcification along the right eighth rib; the mass arose at the bone-cartilage border. (c) 3D-CT scan showing destruction of bone and cartilage destruction, with expansive growth of the tumor at the right eighth rib.*

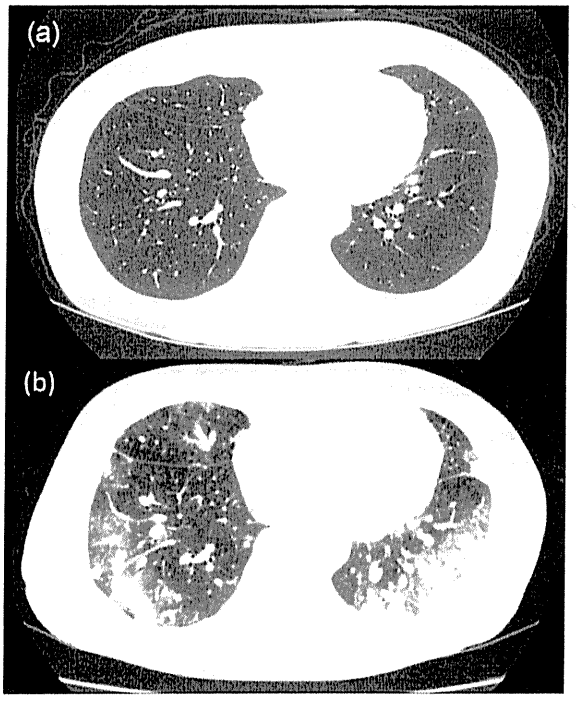

Figure 2 *Chest CT scan*. *(a) Preoperative CT scan showing no pulmonary metastasis. (b) Postoperative CT scan showing pulmonary non-segmental consolidation, predominantly in the peripheral lung field, with surrounding ground-glass opacities; no bronchovascular bundle thickness or interlobular septal thickness was observed.* The tumor was resected together with the right seventh, eighth, and ninth ribs.

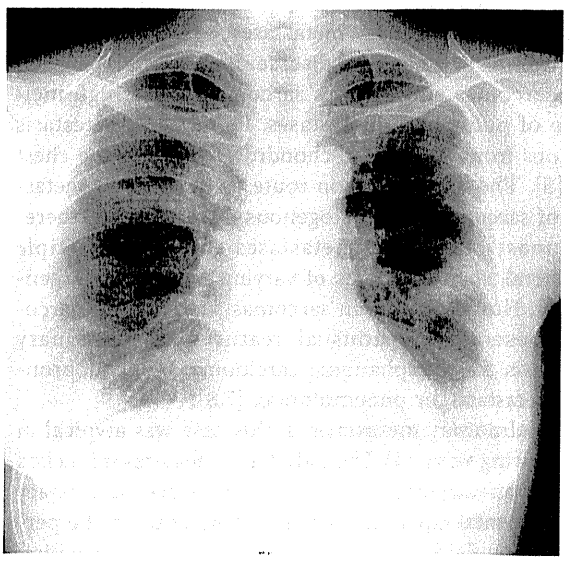

Figure 4 *Chest X-ray*. *Chest roentgenogram showed air-space consolidation with an air bronchogram, predominantly in the right upper and left lower lung fields.*

Discussion

Chondrosarcomas are classified on the basis of their aggressiveness into 3 grades according to their cellular density, degree of anisokaryosis, and nuclear hyperchromatism [6]. The histologic grades of chondrosarcoma correlate well with prognosis, especially for metastases [6]. The most frequent site of metastasis is the lungs; other sites include the bones, brain, regional lymph nodes, and liver [5]. The metastasis rates for grades I, II,

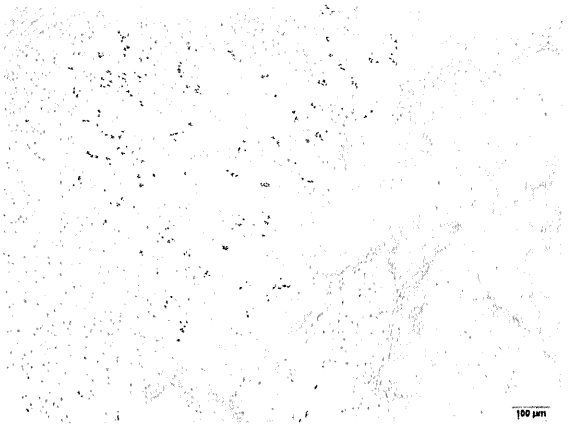

Figure 3 *Resected tumor specimen*. *Hematoxylin and eosin staining of the resected tumor showed a mild increase in cellularity and nuclear atypia. Doubly nucleated cells were seen in the field.*

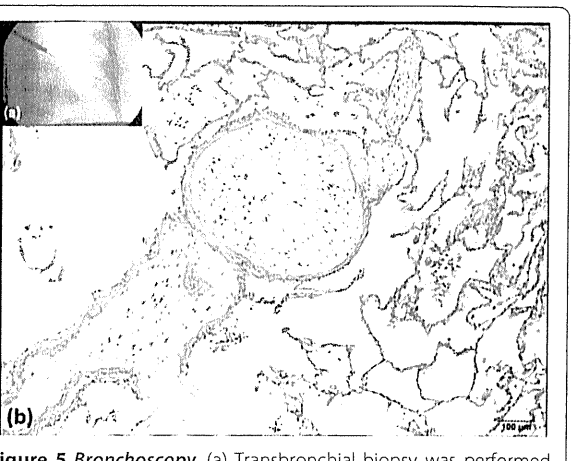

Figure 5 *Bronchoscopy*. (a) Transbronchial biopsy was performed through the left S8 bronchus. *(b) Hematoxylin and eosin staining of the biopsy sample showed a bone tumor in the lumen structure, with the same histological features as the primary bone tumor.*

and III tumors were 0, 13, and 23%, respectively [5]. The incidence of pulmonary metastases varies with the primary tumor and stage of disease. Bone tumors such as osteosarcomas and Ewing's sarcoma show a high incidence of pulmonary metastases. Pulmonary metastasis develops from 20% of the chondrosarcomas of the chest wall [5]. The most common route for pulmonary metastasis of sarcomas is hematogenous dissemination; therefore, most pulmonary metastases appear as multiple peripheral, round nodules of varying sizes on roentgenograms. However, certain sarcomas such as osteosarcomas present with unusual features of pulmonary metastasis, i.e., lymphangitic carcinomatosis, endobronchial metastasis, or pneumothorax [7,8].

The pulmonary metastasis in this case was atypical in the following ways: (1) The radiological features mimicked those of pneumonia. Thoracic CT revealed pulmonary non-segmental consolidation, predominantly in the peripheral lung field, surrounded by ground-glass opacities. This indicated interstitial pneumonia such as cryptogenic organizing pneumonia. (2) Although the operation involved only the right side, bilateral pulmonary metastases developed after the resection. Time taken for metastasis to develop has been reported to be an average of 20 months [2]. In this case, bilateral pulmonary lesions rapidly developed into metastases. Thus, histologic examination was needed in order to confirm the diagnosis.

Transbronchial biopsy, endobronchial biopsy, or surgical lung biopsy can be performed to obtain tissue specimens. Surgical lung biopsy includes video-assisted thoracic surgery (VATS) and open lung biopsy. The procedure chosen is based on clinical judgment, which entails weighing the yield versus the risk to the patient. In particular, transbronchial biopsy is usually the procedure of choice for the initial examination due to its high yield and relatively low risk [9], and therefore, we chose this approach. The transbronchial biopsy revealed pulmonary metastasis from costal chondrosarcoma although the mechanism underlying the pulmonary metastasis remains unknown. The possibility of lymphangitic carcinomatosis was eliminated because of the absence interlobular septal thickness.

Soon after the curative surgery is performed, to confirm the pulmonary metastasis, we recommend that transbronchial biopsy should be performed in cases where unusual clinical and radiological pulmonary findings are detected.

Informed consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-chief of this journal.

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Authors' contributions

ME: assisted in the writing of the manuscript and in the orthopedic workup of the patient; KH: assisted in the drafting of the manuscript and in the orthopedic workup of the patient; TK: assisted in the writing of the manuscript and performed the radiological evaluation; KN: performed the radiological evaluation; YT: performed the pathological evaluation; NN: assisted in the orthopedic workup of the patient; NA: evaluated critically the manuscript and gave final approval for the manuscript to be published. All authors read and approved the final manuscript.

Competing interests

The authors declare that they have no competing interests.

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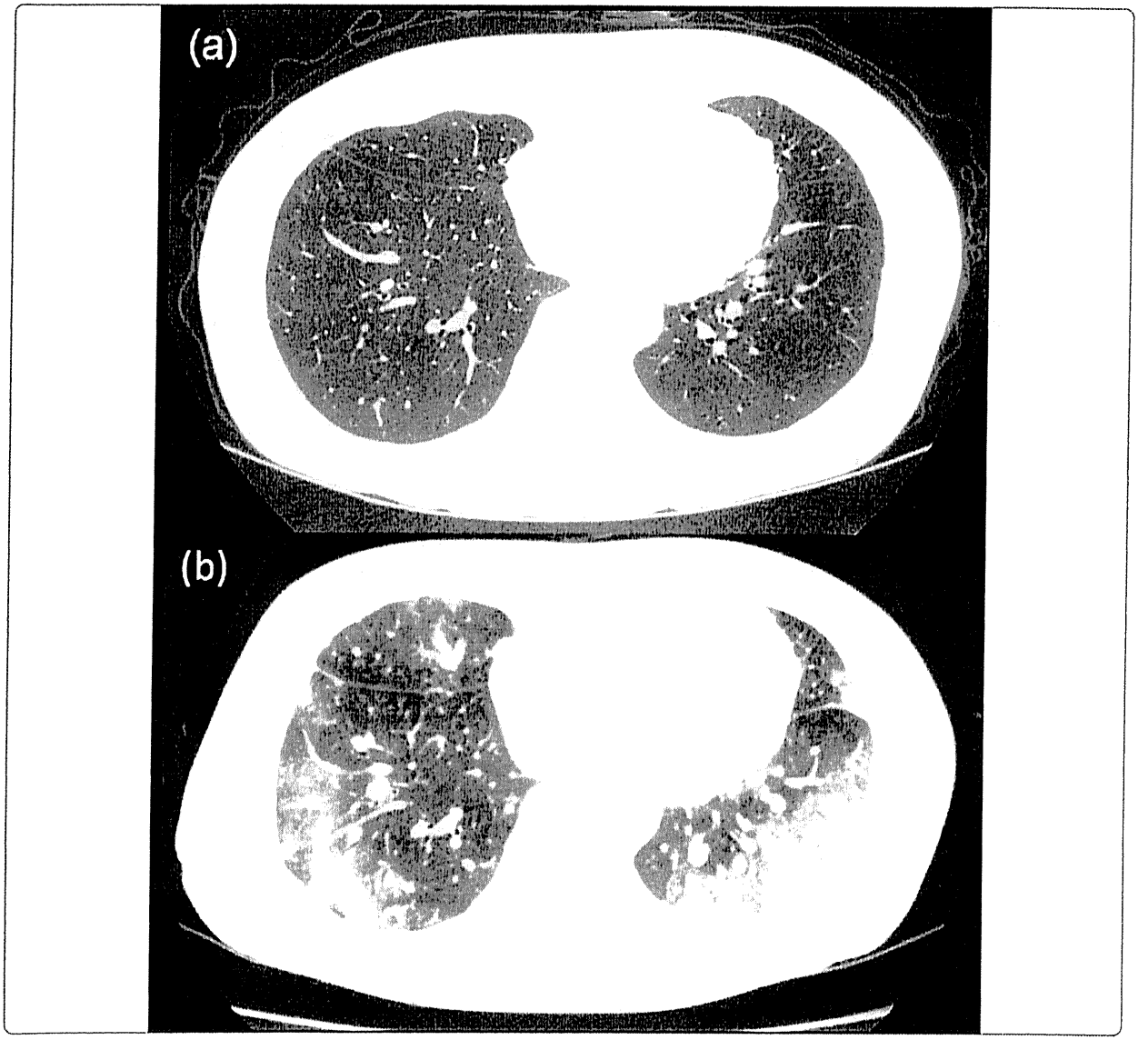

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