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【 CASE REPORT 】

Pulmonary Malignant Ameloblastoma without Local Recurrence 31 Years after Primary Resection: A Case Report and Literature Review

Haruyasu Sakuranaka[1], Akimasa Sekine[2], Ippei Miyamoto[1,3], Yuji Yamakawa[1], Akifumi Hirata[1,3], Eri Hagiwara[1,3], Koumei Igei[1,3], Naoki Okamoto[1,3] and Masahiko Ichioka[1]

Abstract:
A 78-year-old man with a history of surgical resection for ameloblastoma 31 years earlier visited our hospital for prolonged cough. Chest computed tomography showed multiple nodules in both lungs. Although there was no local recurrence in the mandible, the specimen taken from a transbronchoscopic bronchial biopsy showed recurrent ameloblastoma. Despite receiving no treatment, the disease in our patient remained clinically stable for 8.4 years. Chest physicians should be aware that pulmonary malignant ameloblastoma can first relapse several decades after curative surgery. In addition, pulmonary malignant ameloblastoma without local recurrence may be associated with a good prognosis.

Key words: ameloblastoma, lung, malignant ameloblastoma, metastatic ameloblastoma, recurrence, pulmonary metastasis

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Introduction

Ameloblastoma is a benign tumor of odontogenic origin. Although local recurrence after surgical resection is common, distant metastasis is rare. We herein report a case of pulmonary malignant ameloblastoma without local recurrence 31 years after primary resection.

Case Report

A 78-year-old man with a history of ameloblastoma resected by mandibulectomy 31 years earlier visited our hospital due to a 3-month history of dry cough. Chest radiography had not been routinely performed since his surgery because he had been asymptomatic for a long time.

A physical examination revealed neither abnormality nor local recurrence in the mandible. Chest radiography showed a mass in the right lower lung field (Fig. 1), and chest computed tomography (CT) revealed a 50-mm mass in the right B^6 and multiple nodules in both lungs (Fig. 2). On bronchoscopy, a raised lesion with an irregular margin was located in the right B^6 (Fig. 3). The pathological results of the specimen taken from the right B^6 showed an outer arrangement of columnar or palisaded ameloblast-like cells and an inner zone of stellate-like cells forming a follicle (Fig. 4A). Cytologic and nuclear atypia were absent. These features closely resembled those of the primary lesion (Fig. 4B), so pulmonary malignant ameloblastoma was diagnosed. Despite receiving no treatment beyond antitussive agents, the disease of the patient remained clinically stable for 8.4 years.

Discussion

Ameloblastoma is a benign tumor derived from odontogenic epithelial cells and accounts for 1% of all tumors and cysts of the jaws (1). Previous studies have reported ameloblastoma with metastasis as "metastatic ameloblastoma," "metastasizing ameloblastoma," "malignant ameloblastoma," "ameloblastic carcinoma," etc. and have

[1]Respiratory Medicine, Tokyo Metropolitan Toshima Hospital, Japan, [2]Respiratory Medicine, Kanagawa Cardiovascular and Respiratory Center, Japan and [3]Department of Internal Medicine, Division of Respiratory Medicine, Nihon University School of Medicine, Japan
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Correspondence to Dr. Haruyasu Sakuranaka, hal_amazon@yahoo.co.jp

lacked uniform diagnostic criteria. Under the current WHO classification system of 2005, malignant ameloblastoma is defined an ameloblastoma that metastasizes despite a benign histologic appearance. It does not show any features that can be distinguished from ameloblastoma that does not metastasize and is reclassified as such only in retrospect when metastasis occurs. Ameloblastic carcinoma has histologic features of cytologic atypia with or without metastasis. Our case was one of malignant ameloblastoma. Although local recurrence after surgical resection is common, distant metastasis is rare (<2%), and the most frequent metastatic site of ameloblastoma is reported to be the lungs (>70-80%) (2). Almost all malignant ameloblastoma and ameloblastic carcinoma repeat local recurrence. There have been only 10 reported cases of pulmonary malignant ameloblastoma or ameloblastic carcinoma without local recurrence, as shown in Table (3-11).

The present findings provide two important clinical implications.

First, chest physicians should be aware that pulmonary malignant ameloblastoma can first relapse several decades after curative surgery. Indeed, in the present case, the recurrence of ameloblastoma was confirmed 31 years after primary resection. Dissanayake et al. reported that the disease-free interval (time from primary to first metastasis) in malignant ameloblastoma ranges from 2 months to 42 years (pulmonary malignant ameloblastoma: from 2 months to 35 years, cervical LN malignant ameloblastoma: from 1.5 months to 42 years) (2). Each case showed repeated local recurrence before metastasis (2, 12). The previous studies showed that the interval from curative primary surgery to pulmonary metastasis without local recurrence ranged from 5 to 29 years, with a median of 13.4 years (Table) (3-11), so the present case was the longest case. Although routine long follow-up is not easy, chest physicians should be aware that pulmonary malignant ameloblastoma can first relapse several decades after curative surgery.

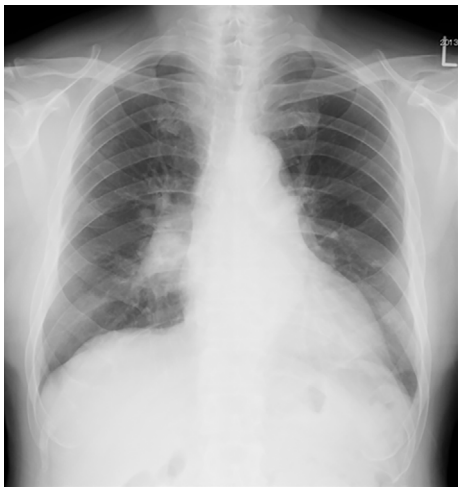

Figure 1. Chest radiograph shows a mass in the right lower lung field.

Second, pulmonary malignant ameloblastoma without local recurrence may be associated with a good prognosis. Indeed, the disease of our patient remained clinically stable for 8.4 years despite no treatment. Pulmonary malignant ameloblastoma has been reported to have a poor prognosis, with a median survival time of 2.6-3 years (2, 13). However, regarding pulmonary malignant ameloblastoma patients without local recurrence, previous studies have shown that the disease remained stable in all patients, and they were all still alive in the follow-up period (average 3.8 years: range 0-8.4 years) (Table) (3-11). The median survival time for pulmonary malignant ameloblastoma without local recurrence should be longer due to their excellent clinical stability. Given the present and previous findings, pulmonary malignant ameloblastoma without local recurrence seems to be associated with a good prognosis. Although why these patients have a good prognosis remains unclear, we believe that the presence of local recurrence can greatly affect the prognosis. In ameloblastoma, local recurrence and surgical procedures have been reported to be associated with hematogenous and lymphatic metastasis (5). In addition, repeated local recurrence may lead to malignant transformation (14), these mechanisms lead to systemic dissemination and a poor prognosis. However, pulmonary malignant ameloblastoma without local recurrence may be caused by aspiration of tumor cells from a resected primary oral lesion (1, 15), grow-

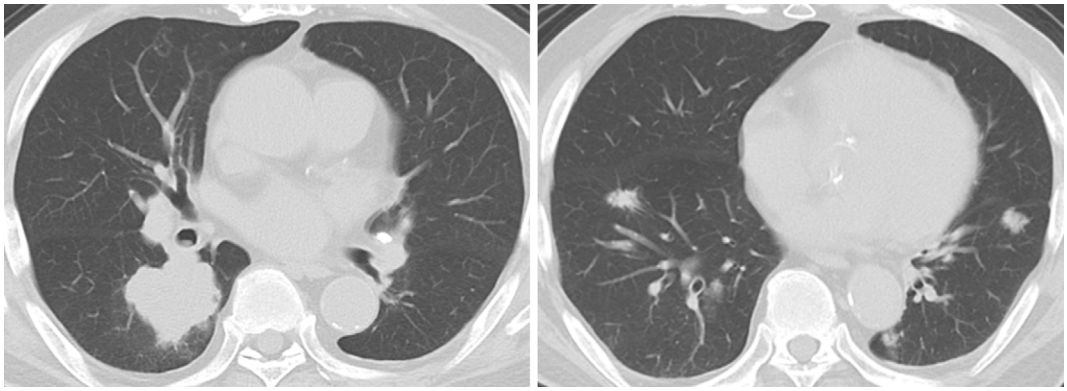

Figure 2. Chest CT shows a 50-mm mass in the right lower lobe and multiple nodules in both lungs.

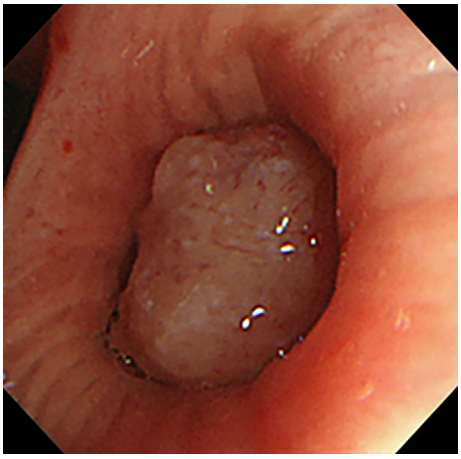

Figure 3. **A raised lesion with an irregular-margin mass is located in the right B^6 on bronchoscopy.**

ing very slowly and thus resulting in a better prognosis.

In conclusion, we experienced a case of pulmonary malignant ameloblastoma without local recurrence 31 years after primary resection. Chest physicians should be aware that pulmonary malignant ameloblastoma can first relapse several decades after curative surgery. In addition, pulmonary malignant ameloblastoma without local recurrence may be associated with a good prognosis. Because pulmonary malignant ameloblastoma is rare, the accumulation of case reports is necessary in order to confirm our results.

The authors state that they have no Conflict of Interest (COI).

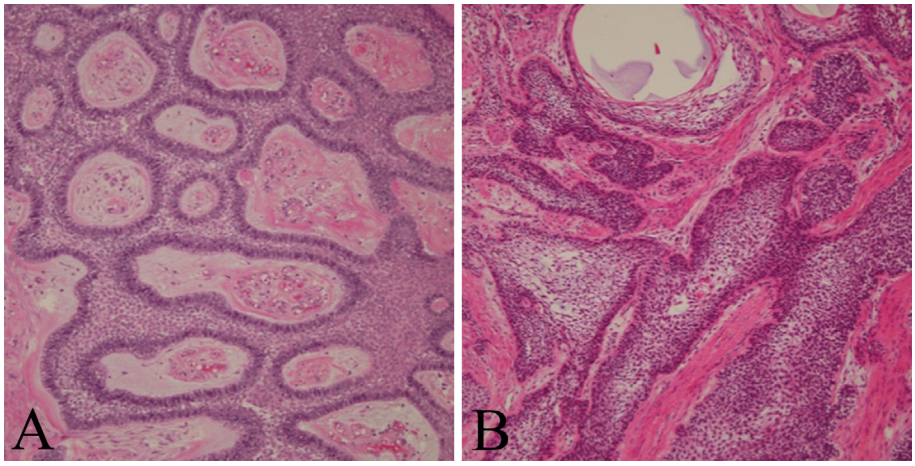

Figure 4. **Histological findings taken from right B^6 (A) and primary lesion (B) using Hematoxylin and Eosin staining. Both findings show an outer arrangement of columnar or palisaded ameloblast-like cells and an inner zone of stellate-like cells forming a follicle.**

Table. **Clinical Characteristics of All Reported Cases of Pulmonary Malignant Ameloblastoma or Ameloblastic Carcinoma without Local Recurrence.**

Case No.	Age/ Sex	Clinical symptom	Number of PM	Location of PM	Duration until PM diagnosis (years)	Cellular atypia of Primary lesions/ Metastatic lesions	Follow-up period after PM diagnosis (years)	Alive or Dead	Treatment for PM	Ref.
1	47M	None	Multiple	Bilateral	13	-/-	2	Alive	None	3
2	33M	None	Multiple	Bilateral	5	-/-	8	Alive	Operation Chemotherapy	4
3	55N/A	None	Multiple	Bilateral	29	-/-	1.5	Alive	None	5
4	37F	Cough Breathless	Multiple	Bilateral	20	-/-	1.6	Alive	Chemotherapy	6
5	56F	Malaise Fatigue	Single	Right lung	10	-/+	N/A	Alive	None	7
6	52N/A	Chest pain	Multiple	Bilateral	14	-/-	4.8	Alive	None	8
7	44N/A	Chest pain	Multiple	Bilateral	10	-/-	0.8	Alive	None	8
8	27F	None	Single	Right	7	-/-	2	Alive	None	9
9	47M	Hemoptysis	Multiple	Bilateral	9	-/+	5	Alive	Operation Chemotherapy	10
10	50F	Back pain	Single	Right lung	17	-/-	N/A	Alive	Operation	11
Our case	78M	Cough	Multiple	Bilateral	31	-/-	8.4	Alive	None	

PM: pulmonary metastases

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