demonstrated high signal intensity on T1-weighted pulse sequences. One possible explanation for the high signal intensity is the presence of proteinaceous material within the cyst. Cyst fluid that has a low specific gravity and is mainly serous (a “spring water” cyst), will show very low signal intensity on T1-weighted images and have very high signal intensity on T2-weighted images. Most bronchogenic cysts contain large amounts of proteinaceous material and characteristically have high signal intensity on T1-weighted images.5,6

Enucleation is the procedure of choice, usually through a mediastinal laparotomy for subdiaphragmatic cysts. When the patient has a history of laparotomy, adhesions may make resection hazardous. Bronchogenic cysts located in the retroperitoneum can be excised using either a laparotomy incision or a flank incision.1 Resection of previously infected cysts is more difficult, giving further impetus to early removal of asymptomatic cysts.

The treatment of asymptomatic bronchogenic cysts remains a controversial topic. Most bronchogenic cysts are benign and remain asymptomatic. Therefore, the argument has been made that intervention is not warranted as long as the cyst is not causing problems. However, this philosophy is shortsighted because asymptomatic cysts do not always remain so.6 Infection is a well-known complication in these lesions, and the morbidity and mortality of surgery to remove an infected cyst is higher than that for a purely elective procedure. Furthermore, carcinomas and fibrosarcomas have been reported arising from benign-appearing bronchogenic cysts.5 Since it usually is not possible to establish an unequivocal diagnosis of bronchogenic cyst preoperatively, and it is impossible to anticipate infection, early surgical resection of bronchogenic cysts is warranted in all good surgical candidates.

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Malignant Ameloblastoma Metastatic to the Lungs 29 Years After Primary Resection
A Case Report

Lawrence M. Ciment, MD, FCCP; and Ari J. Ciment, MD

We describe a case of a 55-year-old man presenting with a metastatic malignant ameloblastoma 29 years after the primary tumor was resected. This represents the longest period between initial diagnosis and first subsequent metastasis recorded as a case report. This case illustrates distinctions between the terms metastatic and malignant; it also highlights the difficulties derived from the accumulation of data by new diagnostic modalities (electron beam CT and positron emission tomography) and their integration into assessment algorithms.

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Key words: ameloblastoma; electron beam CT; malignant; metastatic; metastatic ameloblastoma; positron emission tomography; recurrent ameloblastoma

Abbreviations: EBCT = electron beam CT; PET = positron emission tomography

We describe a case of a 55-year-old man presenting with a metastatic malignant ameloblastoma 29 years after the primary tumor was resected. This represents the longest period between initial diagnosis and first subsequent metastasis recorded as a case report. This case illustrates distinctions between the terms metastatic and malignant; it also highlights the difficulties derived from the accumulation of data by new diagnostic modalities (electron beam CT [EBCT] and positron emission tomography [PET]) and their integration into assessment algorithms.

Ameloblastomas represent 1% of all jaw tumors. They generally are regarded as benign tumors; however, since over half of resected tumors recur, several authorities consider ameloblastomas locally malignant but not metastasizing.1 Metastases, however, are known to occur in roughly 2 to 5% of cases. Over 80% of such metastases involve the lung.2,3

CASE REPORT

A 55-year-old practicing pulmonologist underwent a screening EBCT because of hypercholesterolemia and strong family history of coronary atherosclerosis. Coincidentally, the pulmonary win-
and benign.4 The lung is the most frequent site of metastases, high recurrence rate (50 to 72%), the fact that ameloblastoma have been reported. Though it is locally aggressive with a metastatic sites along with a pattern that otherwise resembled metastatic malignant ameloblastoma (Fig 1). No treatment was offered, and the patient remained in stable condition for an additional 18 months.

**DISCUSSION**

According to the World Health Organization, an ameloblastoma is a neoplasm in which ameloblastic features "are shown by the primary growth in jaws and by any metastatic growth."2 In the literature, "malignant ameloblastoma" and "ameloblastic carcinoma" have been differentiated. Whereas malignant ameloblastomas are tumors considered metastatic despite the appearance of well-differentiated or benign histology, ameloblastic carcinoma portrays histologically malignant features in both primary and metastatic sites along with a pattern that otherwise resembles an ameloblastoma.

Fewer than 45 cases of ameloblastoma with metastases have been reported. Though it is locally aggressive with a high recurrence rate (50 to 72%), the fact that ameloblastoma rarely metastasizes explains why it is considered benign.1 The lung is the most frequent site of metastases, occurring in up to 88% of disseminated cases. The next most frequent metastatic sites were regional lymph nodes, pleura, vertebra, skull, diaphragm, liver, and parotid gland.5

Many factors have been associated with the likelihood of developing metastases. Metastatic tumors occur usually in cases of long duration with multiple surgical procedures or radiation therapy. Extensive local disease and mandibular focus of the primary tumor also tend to be associated with development of metastases.6,8 Although tumor cells have been found invading blood vessels supporting the concept of hematogenous spread, Vorzimer and Perla6 have suggested that aspirated neoplastic cells were often the cause of pulmonary metastases. Finally, the incidence of lymph node metastatic disease argues for lymphatic dissemination as well.6,8

In 1989, Laughlin8 reviewed 43 patients with previously documented cases of metastatic ameloblastoma; the disease-free interval between diagnosis of tumor and appearance of metastases was 9 years, and the median survival time after metastases was 2 years. In 1993, Sheppard et al6 reviewed cases metastatic to the lungs and found that time from initial diagnosis to pulmonary metastases ranged from 0.3 to 31 years, with a mean of 12.1 years. The time from appearance of metastatic disease until death ranged from 3 months to 5 years, with a mean of 16 months for the 24 patients with sufficient follow-up. In the vast majority of cases, there was a recurrence of the primary tumor before the appearance of spread to the lungs.

Metastatic ameloblastoma can be diagnosed accurately by transbronchial biopsy. Whereas imaging that displays multiple nodules to the lung concurrent with a history of multiple recurrences of the primary ameloblastoma may be suggestive of a metastatic lesion, only a biopsy is truly diagnostic. A thoracotomy has developed into the diagnostic test of choice since it can be simultaneously therapeutic. The utility of the PET scan may lie in its negativity diagnostic test since it can be simultaneously therapeutic. This would suggest a more indolent process with a low mitotic rate and would thereby influence the differential diagnosis.

The treatment for metastatic ameloblastoma has been somewhat elusive. In 1987, Lanham6 reviewed chemotherapeutic options and noted that doxorubicin, 5-fluorouracil, 1-(2-chloroethyl)-3-cyclohexyl-1-nitrosourea, methotrexate, methotrexate with cyclophosphamide, cyclophosphamide, nitrogen mustard, vincristine, prednisone, bleomycin, 5-fluorouracil with dacarbazine, and 5-fluorouracil with doxorubicin did not produce any effective objective improvement. Whereas various studies16,11 have shown that ameloblastoma has responded unpredictably to radiation, Ellison et al11 point out that early radiotherapy failures occurred before development of megavoltage external irradiation. Significant resection with preservation of as much viable lung tissue as possible has been the treatment of choice, as this is the only way to offer a significant disease-free survival. In their 1993 case report, Sheppard et al6 reported a patient who underwent eight thoracotomies before achieving disease-free status with no limitation of activities.

Our case is unique for a number of reasons. The time interval between the diagnosis of primary tumor and subsequent evidence of metastatic pulmonary disease was 29 years, 16 to 19 years more than the mean of previously reported metastatic ameloblastoma cases. It

**Figure 1.** Thoracotomy biopsy specimen demonstrating characteristic organization of metastatic ameloblastoma (hematoxylineosin, original ×100).
is important to note that the metastatic lesions in our case were discovered only incidentally, the patient discovering them after undergoing a screening EBCT to assess his cardiac function. As EBCT is being used more often to evaluate cardiac function and disease, the conundrum of lung cancer screening is resurfacing.12 EBCT scans performed as screening procedures have the ability to provide high-resolution images and often present us with diagnostic dilemmas: to what extent is it cost-effective to pursue pulmonary irregularities incidentally discovered on cardiac screening? The findings of two previous Japanese nonrandomized studies13,14 on prevalence screening using low-dose CT with sputum sampling did not demonstrate clear decreases in mortality. A 1999 Mayo Clinic trial and the American College of Radiology Imaging Network have ongoing investigations, but no definitive evidence exists as to whether radiography for screening purposes has any impact on mortality.15 Our case highlights the efforts necessary to pursue the diagnosis entailed by the discoveries made available by these newer modalities; cost effectiveness remains to be elucidated.

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Venous Dilatation Seen on Routine Mammography*

A Clue to Superior Vena Cava Obstruction

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A diagnosis of superior vena cava obstruction (SVCO) generally is made on clinical grounds and can be confirmed by SVCO-specific diagnostic tests. When the obstruction is long-standing, clinical recognition may be compromised as venous drainage of the head, neck, chest, and upper extremity is diverted via collateral venous channels that bypass the obstructed superior vena cava. In such situations, only the visualization of this collateral flow will suggest the presence of SVCO. We describe a patient in whom the unanticipated diagnosis of SVCO was first suggested when routine mammography revealed grossly dilated superficial veins of both breasts, which were the result of collateral flow.

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Key words: mammogram; radionuclide venography; superior vena cava obstruction

Abbreviation: SVCO = superior vena cava obstruction

Clinical recognition of superior vena cava obstruction (SVCO) rests on the presence of a constellation of symptoms and signs that result from impaired venous drainage of the head, neck, chest, and upper extremities. SVCO-specific diagnostic tests such as radionuclide venography, contrast venography, and contrast-enhanced chest CT scan can be performed to confirm the diagnosis of SVCO. Obstruction of the superior vena cava results in collateral circulation diverted via the internal mammary, axillary, hemiazygos, lateral thoracic, thoracoepigastric, and vertebral veins.1 The improvement of venous drainage by collateral flow that bypasses the obstructed superior vena cava can allow SVCO to remain unrecognized. If these collateral venous channels are not visualized, SVCO is not suspected and SVCO-specific diagnostic tests are not performed. This was the case in our patient in whom chronic SVCO

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