coronary embolization of thrombotic and atheromatous material has been recognized as a complication of balloon angioplasty of saphenous vein grafts.\(^7\)

In this patient with unstable angina, forceful injection of contrast medium caused significant reflux resulting in retrograde propagation of a filling defect. This filling defect probably represented thrombus associated with an active coronary lesion. Migration of the filling defect toward the systemic circulation argues against injection of an air bubble. Since the procedure was performed using nonionic dye, one must consider the possibility of injection of a small thrombus formed in the syringe or catheter. However, this would not explain the direction of defect motion or the subsequent improvement in symptoms. The phenomenon of gradual resolution of perfusion defect following reperfusion, as seen in our patient, is well documented in the literature.\(^8\) The significant improvement and eventual normalization of thallium stress images after catheterization, despite reduction in medical therapy, support a causal relationship between the expulsion of suspected thrombus and the disappearance of unstable angina.

**REFERENCES**


---

**Pulmonary Metastatic Disease in Ameloblastoma**

**Brett C. Sheppard, M.D.; Barbara K. Temeck, M.D.; Jeffery K. Taubenberger, M.D., Ph.D.; and Harvey I. Pass, M.D., F.C.C.P.**

Ameloblastoma is a rare disease of odontogenic origin with indeterminate metastatic potential. The first site of metastatic disease is usually the lung. We report aggressive surgical treatment of a patient with bilateral disease with five subsequent recurrences. A review of the literature suggests that in the absence of effective chemotherapy or radiation, surgery should be considered the treatment of choice for metastatic ameloblastoma confined to the lung.

*(Chest 1993; 104:1933-35)*

A meloblastoma is a rare tumor of odontogenic origin that comprises 1 percent of all tumors and cysts of the jaw. The malignant potential of ameloblastoma cannot be predicted due to the lack of well-defined morphologic criteria. When metastases occur, the most frequent site is the lung. To date, effective chemotherapy or radiation therapy has not been developed and surgery remains the mainstay of any curative option. A case of aggressive management of pulmonary metastases for ameloblastoma and a review of the literature reveals that in the absence of randomized studies, repeated metastastectomy is justified for this difficult situation.

**CASE REPORT**

A 28-year-old man was first diagnosed as having ameloblastoma of the left mandible in 1980. He underwent wide excision, bone graft, and strut placement without incident and was free of disease for 5 years. In 1985, a routine chest radiograph demonstrated bilateral pulmonary metastases and the patient was referred to the National Cancer Institute.

The patient had a median sternotomy in April 1985 and had resection of a 4-cm right middle lobe lesion, 1-cm, 2-cm, and 4-cm right lower lobe lesions, a 0.5-cm left upper lobe lesion, and three 1.5-cm lesions in the left lower lobe. All lesions were consistent with metastatic ameloblastoma. The patient recovered uneventfully but had a recurrence in his lungs 6 months later. At that time, he refused surgery and received 150 mg/m² bolus methotrexate followed by 30 mg/m²/h for 24 h and 500 mg/m²/d of fluorouracil. Despite six cycles of treatment, his condition progressed and he then reconsidered the surgical option.

In May and June 1986, staged thoracotomies were performed. The right side of the chest was explored surgically first, and upper lobe and lower lobe lesions were removed. In June, a left lower lobectomy was performed due to the presence of disease near the hilum. Three nodules were removed with the specimen.

The patient had a recurrence in January 1987 (6 months later) and again refused surgery. This time he underwent five cycles of 80 µg/m²/d of cisplatin over 24 h followed by fluorouracil, 800 mg/m²/d for 5 days as a bolus. Again, his disease progressed and he agreed to surgery. In January 1988, he underwent right thoracotomy with resection of an upper lobe lesion and two lower lobe lesions. He remained disease free until a recurrence was noted in November 1988. The patient then had staged bilateral thoracotomies in

*From the Thoracic Oncology Section, Surgery Branch, National Cancer Institute, National Institutes of Health, Bethesda.*

Reprint requests: Dr. Harvey Pass, National Institutes of Health, Bldg 10, Rm 2842, Bethesda, MD 20892

---

CHEST / 104 / 6 / DECEMBER, 1993 1933
Table 1—Therapy and Patient Outcome

<table>
<thead>
<tr>
<th>Therapy</th>
<th>Total No. of Cases</th>
<th>No. of Cases With Follow-up</th>
<th>Mean Survival, yr</th>
<th>Reference No.</th>
</tr>
</thead>
<tbody>
<tr>
<td>Surgical resection</td>
<td>7</td>
<td>7</td>
<td>6.6</td>
<td>5, 19, 24, 26, 27, 29</td>
</tr>
<tr>
<td>Chemotherapy</td>
<td>6</td>
<td>5</td>
<td>1.1</td>
<td>10, 14, 15, 17, 18, 20, 30</td>
</tr>
<tr>
<td>Radiation</td>
<td>1</td>
<td>1</td>
<td>5</td>
<td>6</td>
</tr>
<tr>
<td>Radiation and chemotherapy</td>
<td>1</td>
<td>1</td>
<td>.25</td>
<td>12</td>
</tr>
<tr>
<td>None</td>
<td>14</td>
<td>10</td>
<td>1.1</td>
<td>3, 4, 7, 8, 9, 11, 13, 16, 21, 22, 23, 25, 29, 31</td>
</tr>
</tbody>
</table>

December 1988 with resection of a left upper nodule and a right lower lobe basal segmentectomy. A fifth surgical exploration in May 1992 was performed with resection of a right upper lobe nodule, followed in August 1992 with removal of a residual right lower lobe metastasis.

He is currently free of disease without activity limitation, despite eight thoracotomies. His original pulmonary function tests revealed an FVC of 2.69 (60 percent predicted) and FEV1 of 2.50 (69 percent predicted). Most recently (after seven thoracotomies) his FVC is 1.92 L (43 percent predicted) with a FEV1 of 1.66 L (47 percent predicted). His activities continue to include pick-up basketball games (“full court”).

Histologic examination of the primary mandibular lesion and all metastatic nodules confirmed ameloblastoma.

DISCUSSION

Unlike soft-tissue sarcomas, it is difficult to grade ameloblastoma with regard to malignant potential. Metastasis in an otherwise typical ameloblastoma is rare; malignant behavior is observed in 2 percent of these tumors.1 When metastatic disease occurs, however, the lung is the most common site, occurring in 88 percent of cases of disseminated ameloblastoma. Some investigators believe the pulmonary capillaries are an effective barrier to further dissemination only if the limited number of patients with extrapulmonary metastatic disease can be identified.2 This consideration, taken together with the lack of effective nonsurgical therapy for metastatic disease, suggests that an aggressive surgical approach to pulmonary metastatic disease may be warranted.

Review of the literature for reported cases of ameloblastoma with documented pulmonary metastases2–31 and the inclusion of the case presented suggest several trends that support this concept. Table 1 outlines these data for therapy and patient outcome. Most of the reports (21 of 31) describe a nonoperative approach via simple biopsy to document disease without resectional therapy. The time from initial diagnosis to pulmonary metastases ranges from 0.3 to 31 years with a mean of 12.1 years, suggesting a rather indolent course. In all but two of the cases reviewed, there was at least one local recurrence prior to the appearance of pulmonary disease. Thus, patients who have recurrences locally seem to have a propensity for dissemination, and warrant frequent (every 3 months) metastatic evaluation.

The time from appearance of metastatic disease until death ranged from 3 months to 5 years (mean = 16 months) for the 24 patients with sufficient follow-up. In one patient, metastatic disease was found at autopsy. Eleven patients did not receive therapy for their metastatic disease. In this group, the mean time from diagnosis of metastatic disease until death was 1.1 years and there were two survivors longer than 2 years. Likewise, the survival from diagnosis of metastatic disease to death in 5 of 6 patients receiving multiagent chemotherapy was 1.1 years. Survival with radiation therapy was 5 years for 1 patient and 3 months for another who also received chemotherapy. In the 7 cases in which surgical therapy for metastatic disease was undertaken, there were no operative morbidities and survival ranged from over 1 year to 19 years with a mean of 6.6 years.

In the case reported, the patient initially had bilateral pulmonary metastatic disease. Despite multiple recurrences, the patient has undergone complete resection of disease which conforms to our recommendations for management of multiple pulmonary recurrence from soft-tissue sarcoma.32 Therefore, the presence of multiple nodules or multiple recurrences should not preclude surgical exploration if by computed tomography all lesions can be removed and the patient will be left with sufficient pulmonary reserve.

The lack of a significant number of cases of ameloblastoma will probably preclude the development of effective treatment strategies based on randomized protocols. Moreover, since medical or radiation therapy at present does not impact on survival from metastatic ameloblastoma, surgical excision of pulmonary metastases offers the best chance for long-term palliation or cure.

Documentation of disease confined to the thorax should be performed in all cases with liberal use of tomogram of the jaw, computed tomogram of the abdomen, head, and bone scan. In cases of older patients or patients having repeated thoracotomies, pulmonary function testing should document eligibility for surgical exploration/resection.

CONCLUSION

In the absence of randomized study, patients with their first local recurrence from ameloblastoma should probably be considered as a high-risk subgroup and followed with serial (3 to 6 months) computed tomographic scans of the chest. If pulmonary metastases are identified, the patient should undergo metastatic workup. In the absence of disease other than in the lungs, patients should undergo median sternotomy, bilateral surgical exploration, and metastectomy. Recurrence of disease can be anticipated but does not contraindicate resection, and subsequent recurrences should be approached surgically, if there is sufficient functional reserve, since this is the only treatment that may offer long-term disease-free survival.
REFERENCES

7 Tsukada Y, de la Pava S, Pickren JW. Cranial-cell ameloblastoma with metastasis to the lung. Cancer 1965; 18:916-25
15 Gall JA, Santiano GP, Shreiner DP. Ameloblastoma of the mandible with pulmonary metastasis. Oncology 1975; 32:118-26