ADAMANTINOMA OF THE MAXILLA METASTATIC TO THE LUNG*

CASE REPORT

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The adamantinoma (ameloblastoma) is a rare tumor most commonly found in the jaw. It grows slowly, usually over a period of many years. This epithelial tumor arises either from the remnant of the enamel organ or from cell rests scattered along the length of the tooth. It is closely related to the dentigerous cyst and might well be called an enamel cell tumor. Enlargement of the bone, with rarefaction and thinning of the cortex, occurs as a direct result of the growth of the tumor, which may be primarily either solid or cystic, although degeneration of the solid type to form pseudo-cysts is common.[1]

The literature reveals that 80 to 85 per cent of adamantinomas arise in the mandible and about 15 per cent in the maxilla. Isolated case reports indicate other primary sites such as the tibia, the lip, the cheek, or even the pituitary body. The tumor has been generally considered to be benign, but metastases to other organs have been reported.

Chont[2] briefly reviewed the literature in October 1943 and reported one additional case of distant metastasis from an adamantinoma of the mandible. There have been reported 18 cases in which distant metastases to various sites have occurred; in nine of these the metastatic lesion involved the pulmonary parenchyma (Table I). Schweitzer and Barnfield,[3] reporting also in October 1943, describe a case wherein metastasis to the lung occurred from an adamantinoma of the mandible. These authors mention the fact that in only one previous instance, that case reported by Vorzimer and Perla, was there histologic proof of the ameloblastomatous nature of the metastatic lung tumor. It is reasonable to assume that the clinical impressions of the nature of the metastatic deposits in the pulmonary parenchyma were adequate to substantiate these diagnoses in spite of the lack of histologic proof. Indeed, in that case reported by Chont, the nature of the metastatic lesion was not verified histologically, but the clinical history leaves little doubt as to its true nature. However, the fact remains that of the nine reported cases of adamantinomas metastasizing to the lung, only two have been verified histologically. We are reporting a third.

CASE REPORT

Mrs. M. C., a 56-year-old, white housewife, entered the University of California Hospital December 5, 1947. She was referred to us by her physician, Dr. Sidney Shipman. Sixteen months previously (September 1946) she had noted the onset of malaise and easy fatigability. Chest roentgenograms taken at that time demonstrated a soft shadow in the right lung field which was interpreted as bronchitis. The slight irritative

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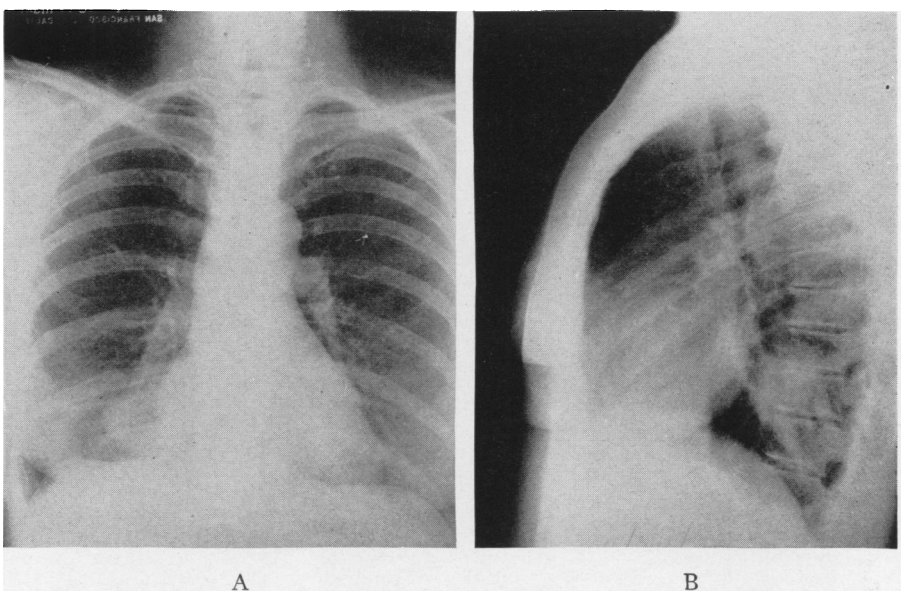
TABLE I.—*Summary of Reported Metastases from Adamantinoma*

Author of Case Report	*Site of Primary Tumor*	*Duration of Primary Tumor*	*Sex*	*Age*	*Race*	*Sites of Metastases*	*Remarks*
1. EVE	Left Mandible	13 Weeks	F	60	W	Lumbar lymph nodes	
2. EWING	Unknown	Unknown		Unknown		Cervical lymph nodes	
3. EWING	Unknown	Unknown		Unknown		Lung	
4. GENTSCH	Left Maxilla	Unknown	M	44	W	Lymph nodes, sphenoid sinus, and malar bone	
5. HAVENS	Unknown	Unknown		Unknown		Unknown	
6. HAVENS	Right Mandible	Unknown	F	49	W	Right submaxillary region	
7. HEATH	Both Mandibles	28 Years	M	67	W	Local recurrence, metastases to deltoid fossa and pelvis	
8. HORSLEY	Right Mandible	20 Years	M	39	W	Local recurrence, metastases to cervical lymph nodes and lung	
9. NEW	Mandible	Unknown		Unknown		Submaxillary gland	No recurrence after excision of primary tumor & involved submaxillary gland
10. SIMMONS	Left Mandible	15 Years	F	37	W	Recurred locally, 4 times; metastasized to cervical lymph nodes.	
11. SIMMONS	Right Mandible	15 Years	M	37	W	Local recurrence, metastases to cervical, submaxillary lymph nodes & lungs	
12. SPRING	Right Mandible	Unknown	M	5	W	Recurred 7 times, metastases to cranial bones, subcutaneous tissue of forehead	Metastases disappeared with X-radiation
13. VORZIMER AND PERLA	Right Maxilla	31 Years	M	38	W	Recurred 5 times, invasion of antrum & nasal septum & metastases to lung	Metastases to lung histologically proved.
14. WEISSENFELS	Left Mandible	14 Years	M	26	W	Recurred, metastases to cervical lymph nodes, skin, submaxillary lymph nodes, & lung	
15. CHONT	Left Mandible	8 Years	F	55	W	Local recurrence, metastases to lungs	Punch biopsy of lung. Tumor not adequate for diagnosis. No autopsy.
16. OKINOUYE	Mandible	Unknown	M	15	W	Metastases to lungs	Original lesion: ameloblastoma. Metastatic lesion: resembled carcinoma.
17. PHELPS	Maxilla	Unknown	M	42	W	Metastases to liver, ribs; tumor found within pulmonary vessels	
18. SCHWEITZER AND BARNFIELD	Mandible	Unknown	F	23	C	Recurred locally despite 24 operations; involved the maxilla & calvarium & metastasized to lungs	Histologically proved.
19. GRIMES AND STEPHENS	Left Maxilla	3 Months (Approximate)	F	46	W	No local recurrence, metastasis to lung	Histologically proved.

cough, without production of sputum or blood, persisted without change until her entry into this hospital. On only one occasion, while walking hurriedly, did the patient note right anterior chest pain. She received symptomatic medical therapy. There was no weight loss. Repeat roentgenograms in July 1947 and in September 1947 each showed an increase in the size of the lesion over that of previous roentgen examinations. Because of ankylosis of the temporomandibular joints, two attempts to perform bronchoscopy were unsuccessful.

In November 1937, ten years previously, an adamantinoma of the left maxilla was removed surgically. The operative procedure was preceded by an extraction of the left upper third molar tooth resulting in a chronically draining area which was finally removed with the subjacent tumor mass. She received postoperative radium therapy which resulted eventually in a partial ankylosis of the temporomandibular joints, some of which she had overcome.



A B

FIG. 1.—Roentgenograms taken in November 1947 illustrating the increase in size of the right lower lobe lesion over that of July 1947. The circumscribed nature of the mass is maintained. It is more apparent on the lateral view than in the previous films.

Major system inventory was non-contributory in respect to the pulmonary lesion. Progressive bilateral eighth nerve deafness had been present since the age of 18, requiring the constant use of a mechanical hearing aid.

Physical examination revealed a well nourished white female, not acutely ill, with normal temperature, pulse and respiration. The blood pressure was 126/72. The left upper alveolar ridge and the adjacent maxilla, as well as the floor of the left antrum of Highmore, had been surgically removed. The posterior aspect of the left half of the hard palate also had been removed, allowing one to view a rather large cavity in the roof of the mouth extending posteriorly along the base of the skull for a distance of approximately 6 to 7 cm. A well-fitting dental prosthesis effectively closed off these openings. The mucosa surrounding the previously involved area, although exhibiting considerable scarring, was completely smooth and without evidence of recurrence of the original tumor. There were no palpable lymph glands or other masses in the cervical region.

Expansion of the chest was equal bilaterally. There was an area of decreased breath

sounds, overlaid with an exactly similar area of dullness, in the right lower lung field posteriorly. There was no evidence of an intrapleural collection of fluid.

Examination of the abdomen, pelvis and rectum revealed only normal findings throughout. Urinalysis showed normal constituents, and the hematologic study showed no anemia.

Roentgenograms of the chest (November 1947) showed a circumscribed density of the lower lobe of the right lung occupying its posterior basal division. Comparison with the films taken three and five months previously (July and September 1947) showed that the lesion had again definitely increased in size. (See Figure 1.)

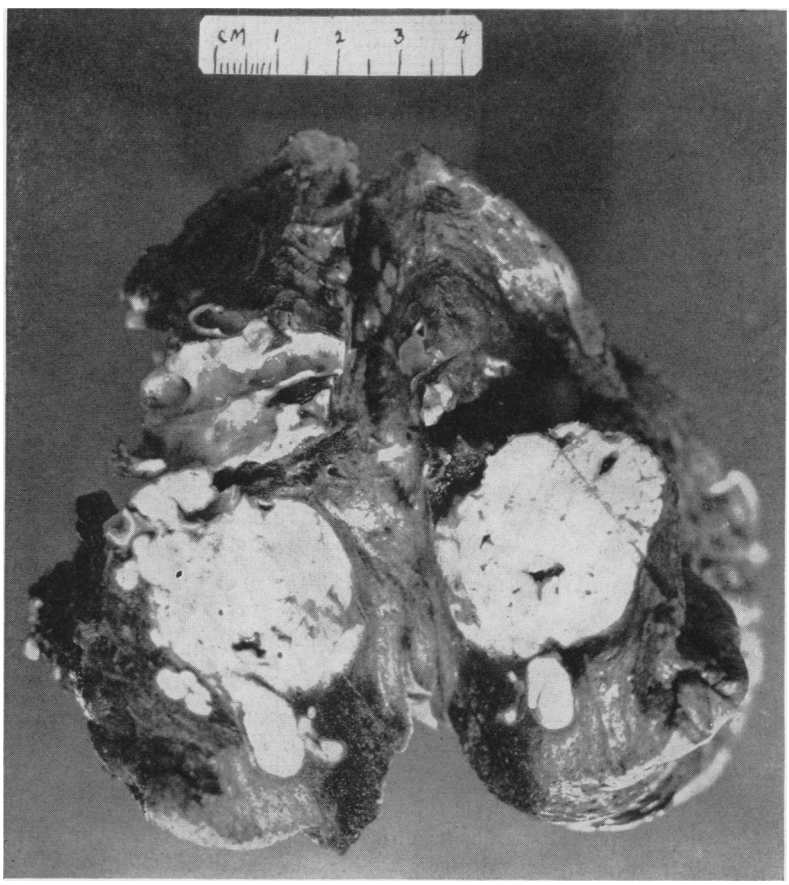


FIG. 2.—Photograph demonstrating the cut section of the tumor. The lower lobe bronchus (top left), though immediately adjacent to the mass in one area, is not involved by it. The surrounding pulmonary parenchyma is only slightly compressed and is not otherwise abnormal.

It was our opinion that we were dealing with (1) a slowly growing, low-grade primary malignancy of the right lower lobe, (2) a metastatic tumor from a primary lesion in one of the major systems not demonstrable by the usual clinical methods, or (3) a metastatic tumor from the original focus in the left maxilla.

Thoracotomy was carried out December 6, 1947. The hard, circumscribed rounded mass lay deep in the parenchyma of the right lower lobe and was entirely confined within it. The hilar and mediastinal lymph nodes in the area of dissection were soft,

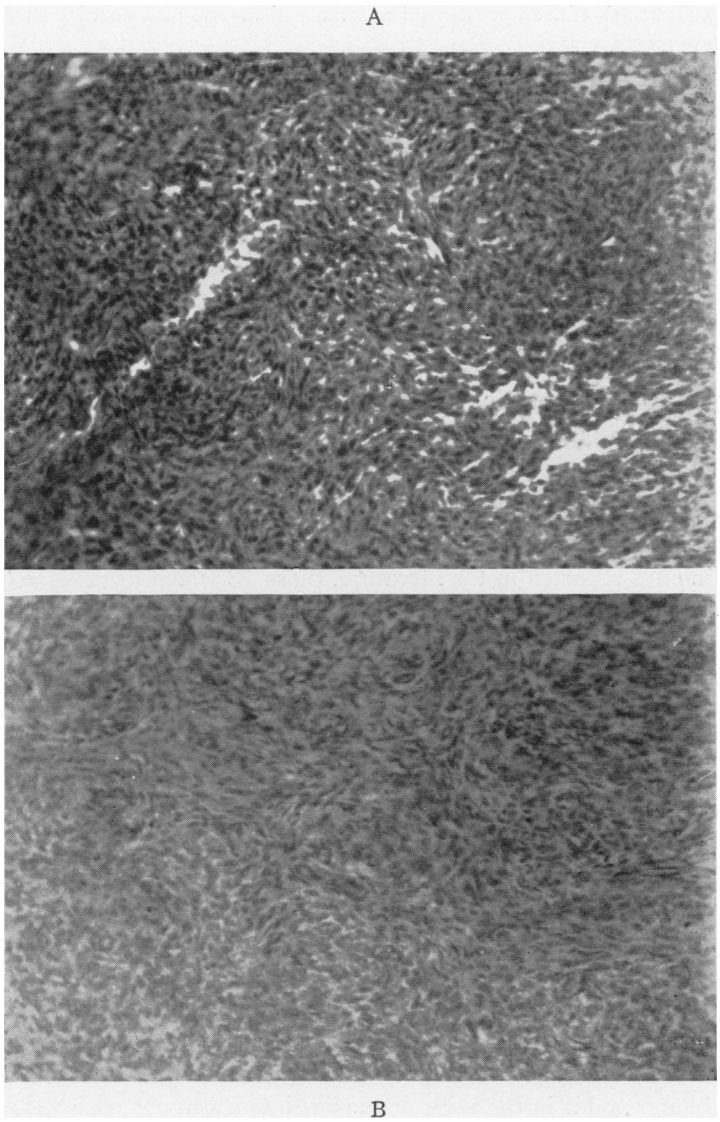


FIG. 3(A)—Photomicrograph of the original lesion of the left maxilla. The cells are closely packed together and exhibit ovoid or spindle-shaped nuclei but only scanty cytoplasm. In some areas there are whorls, in others suggestive glandular arrangements. FIG. 3(B)—Photomicrograph of the metastatic tumor of the lung. Note the dense cellular pattern corresponding to that of the original lesion. The similarity between the two lesions is marked. There is apparently no correlation between the histologic pattern of the adamantinoma and its ability to metastasize.

pigmented, and were not grossly involved by abnormal tissue. There were no notable adhesions. The nature and characteristics of the tumor made one think of a secondary metastatic deposit. A right lower lobectomy was therefore accomplished. The postoperative course was entirely uneventful and the patient was discharged to her home on the 11th postoperative day.

The excised lobe of the lung contained in its center a firm, round, well circumscribed mass 3.5 cm. in diameter which was homogeneous and pinkish-white on cut section. The surrounding lung tissue showed little evidence of compression, and in general was quite well aerated. No communication between the mass and a bronchus could be demonstrated. (See Fig. 2.)

Microscopically, the tumor was composed of closely packed masses of large cells with ovoid or spindle-shaped nuclei and scanty eosinophilic cytoplasm. The cells resembled basal cells, and in some areas were palisaded at the margin. There was invasion of the surrounding connective tissue by the malignant cells. In some areas the cells were arranged in whorl-like patterns suggestive of rosettes. There was a tendency toward glandular pattern in a few areas, and occasionally the cells resembled columnar epithelial cells. In most areas, however, the masses of tumor cells were separated from the pulmonary alveoli by a heavy layer of connective tissue. Microscopic diagnosis: Adamantinocarcinoma. (See Figs. 3A and 3B.)

DISCUSSION

There is general agreement that local recurrence of an adamantinoma frequently occurs, though distant metastases are rare. Of the approximately 500 adamantinomas reported in the literature, those 18 which metastasized regionally or distally represent 3.6 per cent, while those metastasizing to the lung represent 1.8 per cent of the total.

Ewing[4] considered all adamantinomas to be malignant because of their tendency to recur and to invade bony structures in the immediate neighborhood. Many writers on the subject agree that local recurrence is the rule rather than the exception. However, some explain the high rate of local recurrence by the fact that the tumor may infiltrate the bone more than is apparent to the naked eye so that incomplete removal may be followed not by actual recurrence due to inherent malignancy, but rather by persistence of the tumor.

Distant metastases, however, can be explained only by the malignant nature of the primary ameloblastoma. The three possible routes of transfer to the lung are (1) by inhalation through the tracheo-bronchial tree during the course of an operative procedure, (2) via the lymphatic chains, and (3) by blood transport. It is reasonable to assume that in the case being reported the tumor tissue reached the lung by way of the blood stream, since involvement of lymph nodes in the neck or mediastinum was not observed, nor was there an intimate association of the tumor with a bronchus.

The fact that metastatic adamantinomas are likewise slow growing is well borne out in this case. It is probable that the metastasis occurred before or during the period of treatment of the original tumor in 1937, since no evidence of tumor tissue has subsequently been noted in the region of the left maxilla.

SUMMARY AND CONCLUSION

A case history is presented in which an adamantinoma of the left maxilla metastasized to the right lung. Histologic verification of the nature of the

primary and secondary lesions is presented. This is the third reported case of metastatic adamantinoma of the lung substantiated by microscopic examination. Although the adamantinoma is characterized mostly by local recurrence, in a small percentage of cases there are distant metastases.

REFERENCES

[1] Boyd, William: Surgical Pathology, 5th ed., Philadelphia and London, W. B. Saunders Co., 1942.

[2] Chont, L. K.: Adamantinoma; Report of Eight Cases. Am. J. Roentgenol., **50**: 480-490, 1943.

[3] Schweitzer, F. C., and W. F. Barnfield: Ameloblastoma of the Mandible with Metastasis to the Lungs; Report of a Case. J. Oral Surg., **1**: 287-295, 1943.

[4] Ewing, James: Neoplastic Diseases, 3rd ed., Philadelphia and London, W. B. Saunders Co., 1928.

ANNOUNCEMENT OF VAN METER PRIZE AWARD

The American Goiter Association again offers the Van Meter Prize Award of Three Hundred Dollars and two honorable mentions for the best essays submitted concerning original work on problems related to the thyroid gland. The Award will be made at the annual meeting of the Association which will be held in Madison, Wisconsin, May 26th, 27th and 28th, 1949, providing essays of sufficient merit are presented in competition.

The competing essays may cover either clinical or research investigations; should not exceed 3000 words in length; must be presented in English; and a typewritten double spaced copy sent to the Corresponding Secretary, Dr. T. C. Davison, 207 Doctors Building, Atlanta 3, Georgia, not later than March 15th, 1949. The committee, who will review the manuscripts, is composed of men well qualified to judge the merits of the competing essays.

A place will be reserved on the program of the annual meeting for presentation of the Prize Award Essay by the author if it is possible for him to attend. The essay will be published in the annual Proceedings of the Association. This will not prevent its further publication, however, in any Journal selected by the author.