A CASE OF DELAYED METASTATIC SARCOMA OF THE PLEURA, ILLUSTRATING THE DIAGNOSTIC VALUE OF ARTIFICIAL PNEUMOTHORAX

WILLIAM BROMME, A.B., M.D., H. P. NELSON, M.D., F.R.C.S., AND THOMAS FINDLEY, JR., M.D.

(From the Departments of Surgery and Internal Medicine, University Hospital, Ann Arbor, Michigan)

Diagnostic pneumothorax is not a new procedure, but most of the publications which have appeared since Brauer (1) first advocated its use in 1912 have dealt with the demonstration of mediastinal and pulmonary parenchymal lesions. The ease with which pleural adhesions could be visualized suggested the use of this technic in the diagnostic study of non-inflammatory diseases of the pleura, but only a few authors have presented cases wherein roentgenograms of the chest made after artificial lung collapse clearly revealed pleural neoplasm (2, 3). Artificial pneumothorax would seem particularly applicable to the study of pleural effusions of obscure origin because of the virtual worthlessness of ordinary roentgenograms in the face of massive opacity. In view of the common association between intrathoracic neoplasm and pleural effusion (4) it is probable that diagnostic opportunities are lost through failure to use a procedure which is simple and safe in competent hands.

The following case report is a pertinent illustration of the worth of this method. Moreover, it presents a death from metastatic sarcoma thirteen years after the appearance of the primary bone lesion, an event which seems worthy of emphasis in view of Crowell's recent analysis of the Bone Sarcoma Registry data (5).

Case Report

R. H., a white male aged twenty-eight, was admitted to the University Hospital in July 1931, stating that for approximately one year he had been troubled by a constant sense of pressure in the right chest, associated with attacks of severe pain in the same location, non-productive cough, and excessive fatigue. He believed that he had occasional fever but denied hemoptysis and loss of weight.

In 1919, following mild trauma to the left wrist, a tender swelling appeared at the distal end of the left radius, but x-ray examination at the time was understood by the patient to have been negative. The swelling disappeared within approximately one month. In 1921, however, a fracture was sustained at this site following a trivial blow, and x-ray studies then revealed definite osteoporosis in the region of the fracture site. Open reduction was performed, and upon a portion of the removed tissue Dr. Alfred Scott Warthin returned the following opinion: "Alveolar spindle-cell sarcoma" (Fig. 1). Radium implantation and deep x-ray therapy in unknown quantity were employed, but by 1923 so large a mass had developed that amputation through the mid-shaft of the humerus was carried out. The operative site healed primarily, and the patient was without

1 We are indebted to Dr. John B. Barnwell for permission to report this case.
symptoms until 1930, eleven years after the initial injury, and nine years after the exact diagnosis had been established.

In July 1930, following a severe throat infection, the chief complaints recorded above were observed, and the patient presented himself at the University Health Service for advice. X-ray films of the chest revealed a small circumscribed increase in density in

*Fig. 1. Biopsy Tissue from Left Radius in 1921; Diagnosis, Alveolar Spindle-cell Sarcoma. $\times 70$ (above) and $\times 305$ (below)*

the second right intercostal space, the nature of which was uncertain. Extension of this lesion into a well marked fluid collection took place rapidly, and in August 1930, 20 c.c. of clear yellow, sterile fluid were aspirated. The fluid contained a few lymphocytes and produced no specific lesions upon injection into guinea-pigs. Subsequent x-ray studies failed to clarify the diagnosis, and the patient was not seen again until he entered the University Hospital one year later, with marked exaggeration of symptoms.
Physical Examination: On admission in July 1931, two findings of distinct importance were observed. One was a massive right hydrothorax which must have been present for at least one year. The other was a smooth, flat, firm, non-tender mass approximately

![Image of biopsy tissue from chest wall in 1931]

These metastases are strikingly similar to the primary bone tumor removed ten years earlier. $\times 70$ (above) and $\times 305$ (below).

The size of a man's palm, overlying the 5th, 6th and 7th costochondral articulations and attached to the bony thoracic cage but not to the skin. The patient stated that this mass had been observed by his father at the time of the amputation in 1923, but that he himself had not noticed it until the spring of 1930, since which time it had grown slightly larger. It seemed unlikely to us that the patient would have overlooked its presence for seven years, and we are therefore inclined to doubt the parent's observation.

Laboratory Procedures: Routine blood counts, urinalyses, and blood Kahn tests were negative.
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X-ray Examination: Stereoscopic films of the chest (Fig. 3) revealed simply a massive fluid collection in the right side dense enough to obliterate all bony and soft tissue markings. The left lung appeared normal.

![Figs. 3 and 4. Roentgenograms on Admission and After Replacement of Fluid by Air](image)

Fig. 3 (left) shows massive pleural effusion, all diagnostic details being obscured by the fluid. Fig. 4 (right), taken after replacement of the fluid by air, shows clearly the nodules of pleural neoplasm indicated by arrows.

In brief, this was a patient who presented intrathoracic disease which might readily have been attributed to the antecedent sarcoma had he not been symptom-free for such an extraordinarily long period—eleven years after the probable onset and seven years after eradication of the primary lesion. The external growth on the thoracic wall was looked upon as a possible metastasis, but the confusing history of its development seemed to demand direct confirmation. Such information we believed could be obtained only by x-ray visualization of the obscured lung field and pleural cavity and by biopsy.

Clinical Course: Repeated thoracentesis was performed and more than 4 liters of fluid replaced with air before satisfactory visualization of the chest cavity was accomplished. The specific gravity of the fluid was 1.022 and it contained 130 white blood cells per c.mm., chiefly small lymphocytes, although occasional eosinophiles and large mononuclear cells suspected of being "endothelial leukocytes" were observed. X-ray films now showed nearly complete collapse of the right lung (Fig. 4). The roentgenologist reported as follows: "There is a soft-tissue polypoid tumefaction beneath the 2nd right interspace in the anterior axillary line, and a second soft-tissue tumefaction in the 5th right interspace anteriorly which apparently is producing a pressure deformity of the terminal end of the 5th rib at its costo-chondral junction. The appearance is suggestive of neoplasm. The left lung is well aerated."

Inasmuch as the identity of the external tumor with those visualized by x-ray was not entirely established, it was decided to inspect the right pleural cavity directly before proceeding with biopsy. Accordingly a thoracoscope was introduced under local anesthesia through the right midaxillary chest wall. It was then apparent that the external mass was contiguous with the subpleural growths indicated in the x-ray films, and that the parietal pleura was studded with firm, flat, blue-white nodules, varying widely in size.
and shape, but all presenting broad bases and abrupt margins. Elsewhere the parietal pleura was thickened and glazed in an irregular manner, presenting a "white lace" appearance. The visceral pleura was also somewhat thickened but seemed entirely free from tumor and nearly so from adhesions.

The continuity of the internal and external lesions having been established, extrapleural biopsy of the external growth was done under local anesthesia. Dr. Carl Vernon Weller reported on the biopsy material as follows: "Alveolar small spindle-cell sarcoma of precisely the same type as when examined by us in May, 1921, at which time material was removed from the left radius. Hematogenous metastasis must have taken place, since the primary and secondary lesions are on opposite sides of the body." (Fig. 2.)

It was believed that radiation therapy offered very little at this time, and the patient was discharged. A letter from him, in November 1931, stated that since the biopsy the external lesion had rapidly increased in size, and that he had become very weak. He died in February 1932. No autopsy was obtained.

**Comment**

Although permanent cures of sarcoma, particularly those involving the lower extremities, are no surgical curiosities today, the prognosis is still so gloomy that the "five-year cure" criterion is widely accepted. We point to the accompanying biopsy specimens taken more than a decade apart to show that in occasional instances the human organism may harbor sarcoma for a time considerably beyond the usual experience. Unawareness of this fact may, as in the present instance, lead to diagnostic uncertainty. Under such circumstances direct confirmation becomes essential, and diagnostic pneumothorax may meet the requirements. It may be that its use is more widespread than the current literature would indicate. At all events we have felt it worth while to emphasize its special applicability to lesions of the lung and pleura masked by effusion. If the films are taken while the patient lies on his sound side, residual fluid will not obscure the costophrenic angle and diaphragm in question.

It may be hazarded in this instance, from the appearance of the visceral pleura and the readiness with which the lung collapsed, that these metastases had not extended from the lung outward, as is usually the case in hematogenous spread.

The termination of this case forced us to regret the haste with which biopsy was undertaken. How frequently malignant spread is accelerated by surgical trauma we do not know (6), but a review of this history leads us to support the contention of others that biopsy should be done only after a preliminary period of irradiation (7, 8). While it is not likely that the biopsy greatly advanced this patient's death, yet it is certain that extension of the process, at least locally, occurred after each of his three surgical experiences.

**Summary**

(1) A death from osteosarcoma metastatic to the pleura thirteen years after the appearance of the primary lesion is reported, and microscopic sections from both sites are presented.

(2) Artificial pneumothorax is a valuable adjunct in the study of
pleural effusions of doubtful etiology. It should be carried to the point where complete visualization of the pleural surfaces is possible.

References