Carcinoma Arising in Chronic Empyema Cavity

Case Report with Review of the Literature

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Carcinoma is known to arise not infrequently in longstanding, chronically draining sinuses such as are seen with osteomyelitis, cutaneous ulcers, and various types of fistulae. In contradistinction, carcinoma arising within chronic empyema cavities apparently is quite rare, a search of the literature revealing only three such case reports. A detailed study of such a case is herein presented.

Case Report

This 63-year-old farmor was first seen on August 24, 1955 with a complaint of coughing up blood. Three weeks before, he had begun coughing up flecks of dark blood; the night of August 23, he coughed up about an ounce of bright blood. He had not had chills, fever, chest pain, sputum, weight loss, anorexia or weakness. Twenty-three years ago he had had a mass spontaneously arise just below the right nipple, and his physician had lanced it to release a large amount of pus. The area promptly healed never to bother him again. Physical examination showed a moderately obese man appearing his stated age. There was a four-inch, well-healed surgical scar just beneath the right nipple. Beneath this area the percussion note was flat and breath sounds were absent. No other abnormality of the thoracic organs was noted, although he did have hypertension (180/120).

X-ray examination showed a density in the right anterior lung field that was thought to be atelectasis of the middle lobe (Fig. 1).

Bronchoscopy was performed and blood was seen in the right middle lobe bronchus. No other abnormality was noted. Thoracotomy was performed and the density found to be an encapsulated chronic empyema with atelectasis of the middle lobe. Middle lobectomy and decortication were performed.

Pathologically, the findings were of interest in that the lining of the empyema cavity showed squamous epithelium. Scattered in the resected middle lobe were small foci composed of epithelioid cells, lymphocytes, plasma cells, and multi-

Figure 1: Initial (August 26, 1955) posteroanterior and lateral x-ray films of the chest, showing density in the area of the right middle lobe.
nucleated giant cells. No evidence of caseation was present. Acid-fast stains were negative. Complete pathologic diagnosis was: (1) organized empyema with squamous epithelization; (2) fibrosis and atelectasis of the middle lobe; (3) bronchiectasis and chronic bronchitis of the middle lobe; (4) granulomatus inflammation of the lung (as described above), etiology unknown.

Postoperatively he developed empyema at the site of the decortication. Open drainage was instituted through a large bore tube. Improvement was prompt and he was discharged to home in a few days (September 17, 1955). Three months later (December 14, 1955) a sinogram showed the cavity to be only a tiny tract and the tube was removed.

On June 8, 1956, he again coughed up a little blood. Examination and plain chest x-ray films showed no change since March 7, 1956 when the last film had been taken. However, he was readmitted to the hospital on June 10 for bronchoscopy and bronchograms. At bronchoscopy, a small amount of blood was seen coming from the right lower lobe bronchus; the middle lobe stump was well-healed. Bronchograms showed moderate tubular bronchiectasis of the right lower lobe. A broad spectrum antibiotic was given with prompt cessation of hemoptysis and he was discharged on June 16. In the next year, he was seen several times, for each time he contracted a cold he promptly had bronchitis. One of the broad spectrum antibiotics gave prompt relief each time.

On July 12, 1957, the anterior end of the thoracotomy incision broke down and started draining purulent material after having been painful for two or three days. He concurrently began to cough up purulent material. He continued to drain and cough purulent material and on August 8 was admitted to the hospital for further studies.

X-ray examination (Fig. 2) showed a density involving the right upper and lower lobes that was thought to represent pleural thickening, or fluid, or both. A sinogram showed a large cavity with a small but definite bronchial communication as had been suspected. At thoracotomy, the cavity was found to be about the size and shape of a hand with the fingers extended. Involved were all the surfaces of the upper and lower lobes, about one-half of the diaphragm, and all of the right side of the pericardium. All of the granulomatous material was removed insofar as possible. The overlying bone and thickened parietal pleura were also removed and a large bore tube introduced through a stab wound for drain-

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**Figure 2:** August 8, 1957: Chest film, with no significant change in the past 17 months, although breakdown of the incision, with drainage, had occurred.
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The fistula was not demonstrable. The immediate postoperative course was uneventful.

The specimen this time was described as consisting, in addition to the obvious bone and muscle, of many pieces of firm gray tissue and some fibrous yellow tissue. Microscopically there was a marked proliferation of squamous epithelium which in most instances appeared to form a surface or wall of the tract, but also penetrated the underlying connective tissue, bone and cartilage to appear as isolated nests. The squamous epithelium was well differentiated with only slight atypia. There were a few mitotic figures. In decalcified sections of bone there was sufficient atypia to justify a definite diagnosis of squamous-cell carcinoma (Fig. 3). In discussion, the pathologist, Herbert Z. Lund, M.D., stated that the sections showed more squamous-cell proliferation than was noted in the original surgical specimen. After discussing the case with other pathologists, Dr. Lund made a final diagnosis of low-grade squamous-cell carcinoma.

It was felt that further surgery had nothing to offer in view of the extensiveness of the tumor growth even though it was low-grade. Complete removal of the entire right half of the chest wall, right pneumonectomy and pericardiectomy, and removal of the right half of the diaphragm, seemed too formidable a procedure to undertake with questionable benefits to be obtained.

Roentgen-ray therapy was given to the entire right hemithorax through several portals over a two-month interval with considerable improvement. The bronchopleural fistula closed and coughing ceased.

Nine months later, in April, 1958, he was seen with anorexia, fever, malaise, and a cough, productive of purulent material and blood. The drainage site was closed off with friable granulomatous tissue. This friable tissue was curedtted out thoroughly to open up the bronchopleural fistula and he rapidly improved.

Pathologic report this time reported marked proliferation of squamous-cell epithelium with irregular pegs of this tissue penetrating into the underlying fibrous tissue as had been seen previously. The diagnosis was epidermalization of fistula and empyema cavity, probably low-grade squamous-cell carcinoma.

He was discharged to home in a few days and was seen at intervals for the next 20 months. He was admitted to the hospital for the fifth time on January 26, 1959. For the prior six to eight weeks, he had had an increase in weakness and for the preceding week had had diarrhea. Rectal examination showed a large fecal impaction which was subsequently relieved by digital extraction. There was no change in the chest x-ray findings; multiple draining sinuses were still present. The night before he was to be discharged from the hospital, he fell and broke the neck of the left femur. There was no radiologic evidence of metastases of the bone. Open reduction and pinning of the fracture was performed. He recovered from the direct results of the operation, but nevertheless weakened steadily and expired on the 17th postoperative day. This was three and one-half years after the first operation and 26 and one-half years after initial drainage of the empyema.

Necropsy was performed by H. Z. Lund, M.D. Positive findings related to the thoracic condition were as follows: “The chest is asymmetrical with the lower portion on the right side rather depressed. There is a 6 cm. in diameter cavity present in the right anterior axillary line; the margins of this cavity are thick and granular, and dirty white in color. There are multiple small ulcers with the same type of margins scattered around this large cavity. Exploration of the cavity showed a granular undermining friable mass extending laterally, invading the intercostal tissues and ribs as far posteriorly as the vertebral column and medially to the sternum, which is also involved. There is tumor present at each of the satellite ulcers. This tumor growth also extends down to and across the diaphragm involving the entire thickness thereof.” Dissection of the right lung showed that the middle lobe bronchus ended blindly about 1 cm. from its origin. There was no apparent tumor in the lung itself, although its surface was covered with tumor in most areas.

Microscopic sections of the tumor taken from many areas showed a well differentiated squamous-cell carcinoma.
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The final diagnosis was, "Bronchopleural cutaneous fistula with well differentiated squamous-cell carcinoma originating in a fistulous tract and spreading to involve the ribs, sternum, skin, intercostal bundles, parietal and visceral pleura."

DISCUSSION

Schmidt reported a case of carcinoma arising in an empyema that started with pneumonia and subsequent effusion. In 1916, thoracotomy was performed, to become complicated by persistent draining sinus. In 1935, or 19 years later, malignant changes were noted and both roentgen and radium irradiation were given. Nevertheless, the patient expired in a few months.

McAnally and Dockerty reviewed all cases seen at the Mayo Clinic in which malignant changes had taken place in chronic cutaneous sinuses and fistulae. Thirteen such cases were noted with only one of these being associated with empyema. This patient, a 55-year-old man, in 1918 had had a thoracotomy for pleural effusion. He subsequently developed a draining sinus that remained unchanged until 1945, 27 years later, when the discharge became bloody for the first time. The sinus was excised with portions of two ribs involved. Microscopically the lesion was diagnosed as Grade II epithelioma. Unfortunately, the patient was lost to followup.

Peabody reported a case of carcinoma arising in a bronchopleural cutaneous fistula following pneumonectomy for bronchiectasis. The pneumonectomy was performed in 1947. The bronchus opened to cause empyema, and in spite of a subsequent Schede thoracoplasty, the fistula persisted. Eleven years later carcinoma was identified in the sinus tract. Roentgen-ray therapy was instituted with no discernible aid, and the patient expired in 1959, 12 years after the initial resection.

In the present case there was an interval of 23 years between the onset of the empyema and the production of bloody sputum. The patient was symptom free in this interval in contradistinction to the other patients who drained constantly after the initial operation. In two of the cases, the appearance of a bloody discharge marked the onset of malignant changes.

The origin of the squamous epithelium that became malignant is not clear. Three explanations are available: (1) extension of metaplastic bronchial epithelium into the cavity; (2) metaplasia of the pleural endothelium; and (3) ingrowth of epithelium from the skin margins of the tract. In Schmidt's, McAnally's, and Peabody's cases, any one of these three explanations could be advanced, but the third was not available in the case being reported, as drainage did not occur until after the malignant change had occurred. McAnally subscribes to the dictum that epithelium comes from epithelium and believes that epithelization proceeded from outward within in his case. In the case being reported, I believe the bronchopleural fistula was a late development, as was the cutaneous sinus. Therefore, it seems that this squamous-cell carcinoma arose in an empyema cavity that was lined by epidermalized endothelium, said epidermalization being in response to the chronic infection present.

REFERENCES

1 Schmidt, R.: Quoted by McAnally and Dockerty. Ref. No. 2.