well circumscribed, its medial site invaded the vagal nerve and membranous portion of the trachea between the carina and the fifth ring from the carina. Since the examination of frozen section suggested malignant lymphoma, subtotal excision leaving some tumor on the membranous portion of the trachea was performed, and we planned on adjuvant chemotherapy in combination with radiotherapy.

Routine histologic sections, however, showed large epithelioid cells with abundant cytoplasm, suggestive of malignant melanoma or poorly differentiated carcinoma (Fig 2). Then, immunohistochemical staining was performed for keratin, S-100 protein, and melanoma-associated antigen as defined by HMB-45 (DakoPatts, Denmark). The sections were positive for S-100 protein but negative for both keratin and HMB-45. The findings were not compatible with either malignant melanoma or carcinoma. Examination of the entire lesion revealed ill-defined nodules composed of melanoma-like cells set in a myxoid background (Fig 3) and sarcomatous spindle components resembling conventional malignant schwannoma. A diagnosis of MES was therefore made.

To resect the residual tumor, the membranous portion of the trachea involved by the tumor was resected, and tracheoplasty was performed on the esophagus 2 weeks after the first operation. Radiotherapy of the mediastinum was performed after the second operation at a total dose of 50 Gy. A few months later, multiple metastases appeared in lung, cervical spine, and neck lymph nodes. The patient died on August 17, 1993, although no local recurrence was identified.

**Discussion**

Malignant epithelioid schwannoma is a rare malignancy that is classified as an unusual form of malignant schwannoma closely resembling a poorly differentiated carcinoma or melanoma. About 5% of all malignant schwannomas are of the epithelioid type; however, that figure may be too high because it does not account for many ordinary cases of malignant schwannomas. To our knowledge, there have been no reports of this type of tumor located in the mediastinum in the English-language medical literature.

The most constant histologic appearance of this tumor is that of ill-defined nodules composed of melanoma-like cells set in a variably myxoid background. However, if the area of epithelial differentiation is predominant, it is indistinguishable from a carcinoma or melanoma by hematoxylin and eosin-stained sections. Fortunately, since most of these tumors express S-100 protein but lack a melanoma-associated antigen and keratin, application of immunohistochemistry will provide adequate additional information to resolve this dilemma. The correct diagnosis has also largely depended on established an origin from a nerve. In our case, in situ characteristics of the tumor involving the vagal nerve were also helpful in making a correct diagnosis.

Clinically, this tumor does not have a particularly distinctive presentation. It may occur at any age and have a lower association with von Recklinghausen's neurofibromatosis than the ordinary form of malignant schwannoma. Although the number of reported cases is limited, at least 50% of the patients reported in the literature died of distant metastases, usually in the lung, while the incidence of local recurrence is low, which appears to be attributable to radical excision of the primary lesion. The efficacy of radiotherapy and chemotherapy appears to provide little additional benefit. However, no tumor-related death within the group of patients and superficial lesions (total 16 cases) was indicated in the most recent report. These tumors were reported to be often confined by a nerve sheath. The explanation of those results is that malignant epithelioid schwannoma may be eminently curable despite fully malignant characteristics if the radical local excision is accomplished in the early stage. This probably holds true for our case because he noticed the cough 4 months before the operation.

**References**


**An Unusual Case of Lung Torsion**

*John P. Fogarty, MD; and Gary Dudek, MD, FCCP*

We report a case of complete torsion of the right lung induced by transthoracic needle biopsy. While a delay in diagnosis occurred, viability of the torsed lung was maintained, thereby allowing for effective surgical repair.

(CHEST 1995; 105:575-78)

*From the Divisions of Pulmonary and Critical Care, Departments of Medicine, The Genesee Hospital and Strong Memorial Hospital, University of Rochester School of Medicine, Rochester, NY. Reprint requests: Dr. Dudek, 224 Alexander St, Rochester, NY 14607*
Pulmonary torsion is an infrequent event that most often occurs as a postthoracotomy complication or as a consequence of thoracic trauma. When it does occur, it is considered a surgical emergency, as impaired pulmonary blood supply can result in infarction and loss in viability of the torsed lobe or lung. We report a case in which torsion of the entire right lung developed as a delayed complication of transthoracic needle biopsy of a right upper lobe mass. This was successfully treated despite a clinical duration of torsion of greater than 1 week and a septic presentation.

**CASE REPORT**

An 84-year-old woman presented to the emergency department complaining of a 1-day history of fever, and a 1-week history of cough and right-sided chest pain. She had previously been followed up for an asymptomatic right upper lobe nodule that had been radiographically stable for 10 years. One year prior to hospital admission, the nodule was noted to increase in size; however, she declined further evaluation. Two months prior to hospital admission, she experienced weight loss, cough, and sputum production, and a chest radiograph demonstrated an 8-cm right upper lobe mass. A transthoracic needle aspiration revealed a well-differentiated adenocarcinoma. This was complicated by a small asymptomatic pneumothorax. She declined surgical evaluation and began a course of palliative radiation therapy. Two weeks after the initiation of radiation therapy and 1 week prior to hospital admission, she experienced cough with scant bloody sputum and right-sided chest pain. Twenty-four hours prior to hospital admission, the patient developed nausea, vomiting, diarrhea, and chills, and she presented to the emergency department.

Physical examination revealed a blood pressure of 80/40 mm Hg that improved to 110/60 mm Hg with intravenous hydration. Other vital signs included heart rate of 120, temperature of 38°C, and respiratory rate of 18. A maculopapular rash was present on the trunk and extremities. Chest examination revealed absent breath sounds in the lower right anterior chest with intermittent wheezing noted throughout the right side.

Significant laboratory data included a WBC count of 1,500 with 34 segmented neutrophils, 20 bands, 29 lymphocytes, 10 monocytes, and 2 eosinophils. The hematocrit was 34, and the platelet count was 127,000. An arterial blood gas value on 3 L/min O2 by nasal cannula was as follows: pH, 7.48; Pco2 33 mm Hg; Po2 65 mm Hg; and O2 saturation, 94%. Serum electrolytes, renal function, and urinalysis results were normal.

Figures 1 through 3 demonstrate three corresponding chest radiographs: the first (Fig 1) is after transthoracic needle biopsy, the second (Fig 2) on initiation of radiation therapy, and the third (Fig 3) on admission in the emergency department, which reveals opacification of the lower half of the right hemithorax with no visible right upper lobe mass as was previously apparent.

The patient was referred for chest CT scan. Multiple high-resolution CT images of the chest revealed the mass to be in an anterior, inferior position. The right main pulmonary artery was located posterior and inferior to the right mainstem bronchus. Alteration of the normal anatomy of vascular structures of the upper and lower lobes was noted, consistent with torsion of the entire right lung. A ventilation-perfusion scan was obtained that demonstrated preservation of perfusion to the right upper hemithorax, suggesting potential viability of what was presumably the right lower lobe.

The patient’s medical condition prior to surgery was optimized. Antibiotics for presumed postobstructive pneumonia were administered. Granulocyte colony-stimulating factor was administered to correct neutropenia. One week after hospital admission, all cultures were negative, the WBC count had increased to 17,000, fever had resolved, and the patient was taken to thoracotomy. Operative findings included a 180° torsion of the entire right lung without apparent infarction, a small right pleural effusion, a rudimentary right pulmonary ligament, a complete fissure between the right upper and middle lobes, and a right upper lobe mass. Detorsion and right upper lobectomy were performed. An air leak resolved after 15 days, and the patient was discharged home from the hospital on the 17th postoperative day. One year later, she claimed no respiratory symptoms and had been treated with palliative radiation ther-
apy for a pathologic lumbar compression fracture.

**Discussion**

Pulmonary torsion is a rare event with less than 60 reported cases in the medical literature. Torsion is most likely to occur when the normal stabilizing structures of each lung and lobe are altered or interrupted, enabling a bronchovascular pedicle to rotate on itself. This typically occurs in one of three settings: after trauma, postoperatively, or spontaneously. The incidence of pulmonary torsion is likely much greater than that reported. A recent survey of 117 thoracic surgeons in Great Britain revealed 30% having seen at least one case of lobar torsion, most of those complicating prior pulmonary resection.

Mechanical contributions to the development of lung torsion include the following: transection or congenital absence of an ipsilateral pulmonary ligament; absence of a parenchymal bridge between contiguous lobes (complete vs incomplete fissure); a long, free lobar pedicle; a heavy, compact lobe (due to the presence of a mass, consolidation, or atelectasis); or the presence of a pneumothorax or pleural effusion. While trauma and thoracic surgery are the most common inciting events, other contributing conditions include spontaneous pneumothorax, pneumothorax induced by percutaneous needle biopsy, diaphragmatic hernia, presence of an accessory lobe, pneumonia, neoplasm, and pleural effusion. Presenting signs and symptoms are nonspecific and include acute chest pain, diminished breath sounds over the involved area, copious secretions that may be bloody, hypoxemia, hypotension, and cardiovascular collapse.

Radiologic findings include a change in position of an opacified lobe, alteration or inversion of pulmonary vasculature, a collapsed or consolidated lobe that occupies an unusual position on plain radiographs, and “cutoff” or distortion of a bronchus. Other potentially helpful investigations include bronchography to reveal a bronchial “cutoff” or change in location of major bronchi, and pulmonary arteriography revealing distortion of the pulmonary vascular tree. Bronchoscopy (often with nonspecific findings of airway edema, purulent secretions, or obstruction of an affected lobe) and ventilation-perfusion scanning offer supportive but nonspecific findings. Chest CT has also been utilized and may be the best single diagnostic test, revealing an altered relationship between the trachea and pulmonary arteries, and change in position of a previously located mass.

On recognition of lung torsion, immediate thoracotomy and detorsion with resection of devitalized lung segments has been widely recommended, suggesting that a delay in surgery may result in increased mortality.

Our case was unusual for several reasons. The pneumothorax present after transthoracic needle biopsy in the presence of a large upper lobe mass suggests this as the most likely mechanical change to permit the occurrence of torsion. To our knowledge, this is the first report of torsion of an entire lung secondary to this common procedure, although lobar torsion due to needle biopsy has been reported previously. It would seem that the incompletely formed right inferior pulmonary ligament was the necessary prerequisite to permit torsion of the entire right lung.

The duration of lung torsion in our patient is decidedly unusual, likely occurring at least 1 week prior to presentation, with surgical correction performed 1 week after hospital admission. Survival of prolonged whole lung torsion has been described in children. In one case, the lung was left in situ as it was technically unresectable, resulting in a small, fibrotic lung 1 year after the torsion developed. In another case described by Daughtery, resection of all but a still viable left lower lobe superior segment was performed 6 weeks after traumatic left lung torsion in a 7-year-old. We are unaware of previously reported cases of adult survivors of prolonged lung torsion.

The intraoperative findings of viable, noninfarcted right middle and lower lobes despite a 180° torsion are equally unusual. Occlusion of the pulmonary veins is an anticipated complication of torsion. Experimentally, Swan and Mulligan have shown the development of fibrosionpleuritis and alveolar hemorrhage within 12 h of pulmonary vein ligation. This is seen clinically as the radiographic opacification of a torsed segment or lung within 24 h of the inciting event. This scenario has led to the recommendation by Oddi et al that immediate thoracotomy be performed on recognition of torsion in order to limit loss of lung tissue due to hemorrhagic infarction. The lack of alveolar hemorrhage and the maintenance of lower lobe perfusion despite 180° torsion in our patient is unique and has not been reported previously (to our knowledge).

Finally, the existence of neutropenia has not been described in the presence of pulmonary torsion. We cannot ascertain if this was due to the torsion, to an underlying infection, or less likely to prior chest irradiation.

In summary, we report the development of right lung torsion in an 84-year-old woman as a delayed complication of transthoracic needle biopsy. The patient subsequently underwent thoracotomy and detorsion of the right lung some 2 weeks after the clinical event with continued middle and
lower lobe viability. The upper lobe was resected due to its tumor burden. She continues to do well 1 year postoperatively. While we agree with a general approach of emergency surgery on diagnosis of pulmonary torsion, individual patients may demonstrate varied pathophysiologic responses to its occurrence.

REFERENCES
9 Felson B. Lung torsion: radiographic findings in nine cases. Thorac Rad 1987; 162:631-38
10 Berkman YM. Uncomplicated torsion of the right upper lobe secondary to spontaneous pneumothorax. Chest 1985; 87:695-97
13 Epplen F, Jacobson AL. Twisted pedicle of accessory lobe of the lung. JAMA 1930; 94:1135
14 Huang TY, Cho SR. Torsion of the lung without trauma. Radiology 1979; 132:25-6
15 Meissel R. Case of the spring season. Semin Roentgenol 1980; 15:115-16

Reflex Sympathetic Dystrophy Following Arterial Blood Gas Sampling in the Intensive Care Setting*

Chris Criscuolo, MD; Gaylon Nepper, DO; and Scott Buchalter, MD

A 54-year-old woman developed signs and symptoms consistent with reflex sympathetic dystrophy in her left upper extremity following arterial puncture. Diagnosis was confirmed by bone scan, and sympathetic blockade with intravenous regional bretulnium completely relieved her severe, intractable pain. (CHEST 1995; 108:578-80)

Key words: arterial puncture; causalgia; reflex sympathetic dystrophy; sympathetic blockade

Reflex sympathetic dystrophy (RSD) is a form of sympathetically maintained pain that is characterized by constant burning, swelling, hyperesthesia, allodynia, and vasomotor changes that usually affects the extremities. The mechanism responsible for these signs and symptoms is thought to be an abnormal reflex mediated by the sympathetic nervous system, usually in response to trauma. Other predisposing factors might include surgery, myocardial ischemia and infarction, primary neurologic disorders, or infection. In approximately 10% of cases, no cause can be identified.1 Without early recognition and treatment, RSD can progress through three stages, resulting in intense pain and irreversible trophic changes. In this case report, we present a patient who developed symptoms consistent with reflex sympathetic dystrophy following arterial puncture.

CASE REPORT

A 54-year-old woman was admitted to the intensive care unit (ICU) for respiratory failure that required endotracheal intubation and mechanical ventilation. Four days after admission to the ICU, the patient developed pain in her left wrist. Although she was intubated, she was able to communicate that her pain developed after a particularly painful and difficult arterial blood gas sampling. She described the pain as burning in nature. Her left arm, wrist, and hand were erythematous and edematous, as well as extremely tender to light touch, palpation, and motion. She was afebrile, and there was no elevation of her white blood cell count. Plain radiographs failed to show any bony abnormalities. A CT scan was also normal. A bone scan revealed increased activity and uptake in the phalangeal joints and metacarpal bones of the left hand (Fig 1). These clinical and radiographic findings prompted a diagnosis of RSD, and the pain service was consulted for management. Treatment of sympathetically mediated pain is based on sympathetic blockade. Inter-