Spontaneous Hemothorax Secondary to Immature Teratoma of the Mediastinum

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Spontaneous hemothorax in a 20-year-old boy was caused by rupture of an immature teratoma of the mediastinum. The tumor bled spontaneously into the right pleural space. This life-threatening complication necessitated emergency surgery. The unusual cause and the interesting clinical course of spontaneous hemothorax are described.

Key words: germ cell tumor, intrapleural rupture, hypovolemic shock

Introduction

Primary germ cell tumors of the mediastinum are rare and account for approximately 10% of all neoplasms of this structure (1). Immature teratoma of the mediastinum is particularly rare, with only 28 reported cases according to a review compiled by Carter et al in 1982 (2). Virtually all teratomas have been detected as an incidental finding (3). Some reports have described massive hemothorax secondary to rupture of mediastinal teratomas (4-7). Benign mediastinal teratomas appear to rupture more frequently than immature and other types of teratomas (8).

Rupture of a teratoma is associated with spontaneous rupture into the pleural space or with vague chest pain or symptoms due to vessel compression. We report a patient with immature teratoma of the mediastinum in whom spontaneous rupture into the right pleural cavity was the presenting manifestation; emergency surgery was necessary for hemostasis. This rare case of hemothorax secondary to an immature teratoma of the mediastinum took an interesting clinical course.

Case Report

A 20-year-old man with an 8-day history of dyspnea was admitted to our emergency room. A chest radiograph obtained elsewhere showed a massive right pleural effusion. An exploratory puncture of the effusion revealed the presence of blood; the hemoglobin concentration was 9.9 g/dl. The patient was referred to our hospital for further evaluation. There was no history of recent trauma. Six months previously, chest radiograph performed at a routine health checkup showed no abnormal shadow. On physical examination, the patient was alert, sweating, and pale. The blood pressure was 113/75 mmHg, the pulse was 113/min, and the body temperature was 35.0°C. Decreased breath sounds and dullness to percussion were heard over the right lung. Hematologic study disclosed normocytic-normochromic anemia with a hemoglobin concentration of 11.1 g/dl and an elevated leukocyte count. Serum chemical studies showed slightly elevated values of lactate dehydrogenase and creatine phosphokinase. The results of serologic studies were positive for C-reactive protein, and the CA-125 level was abnormally high. Other laboratory variables, including platelets and bleeding time, were within the normal range. Arterial blood gas analyses showed a pH of 7.36, an arterial carbon tension of 39.8 mmHg, and an arterial oxygen tension of 98.6 mmHg.

A chest radiograph obtained on admission (Fig. 1) showed a massive right pleural effusion with evidence of tracheal displacement and deviation of the cardiac shadow to the left. Because of his severe respiratory distress due to the large pleural effusion, an intercostal tube for continuous drainage was inserted. A large intercostal tube (28F) was inserted through the 5th intercostal space in the direction of the lower field. Non coagulated bloody fluid (1,900 ml) was removed during the first 3 hours. Continuous drainage was performed with volume resuscitation.

A computed tomographic scan of the chest (Fig. 2) obtained after drainage showed a tumor in the mediastinum. The tumor was difficult to distinguish from hematoma and had an indistinct border with the mediastinum.

After 6 hours of drainage, the hemoglobin concentration fell from 11.1 g/dl to 2.9 g/dl. Although we administered volume...
resuscitation with 2,700 ml of crystalloid solution, 1,750 ml of colloid solution, and 560 ml of blood transfusion, his condition worsened to nearly hypovolemic shock.

As the hemoglobin concentration obtained elsewhere the previous day was 12.2 g/dl, it was suspected that continuous drainage for relief of his respiratory distress induced re-hemorrhage into the pleural cavity. Angiography was not done because his condition was too unstable.

As progressive intra-thoracic hemorrhage was suspected, emergency surgery was performed. When the posterolateral incision was performed through 5th intercostal space, active hemorrhage was not detected. There was about 850 g of blood coagulation around the mass and 1,100 ml of blood in the pleural cavity. After the coagulation was removed, a mass was detected about the size of a child’s head, white, and elastic hard. The precise hemorrhagic point was unclear. The mass broadly invaded the major blood vessels, the right phrenic nerve, the right hilar area, and the right upper lobe. We thought these findings were suggestive of malignancy. We could only partially resect the mass for pathological diagnosis, because his condition was very unstable and preoperative systemic survey was not fully done.

The pathological specimen of the tumor showed diffuse proliferation of spindle or polyhedral cells. The nucleus was irregular and round shape, the chromatin showed fine granular proliferation and the many mitotic cells were shown. These cells had no remarkable differentiation. The immunohistological staining showed that the tumor was positive only for vimentin, a marker of non-epithelial tumors, and negative for all epithelial, myogenic, neurogenic, and histiocytic markers. The pathological diagnosis was immature teratoma (Fig. 3). The differential diagnosis was small cell round tumor such as lymphoma, pulmonary small cell carcinoma, and primitive neuro-ectodermal tumor (PNET). By the use of immunohistochemical study, we denied these diseases.

There was no metastasis by the computed tomographic scan of the brain, chest, and the abdomen. Bone scintigram was also intact. The final diagnosis was mediastinal immature teratoma. His family refused chemotherapy, and the patient was transferred to another hospital. He received no active chemotherapy there. About two months later, he died from respiratory failure. Autopsy was not done.

**Discussion**

Primary tumors of extragonadal origin are rare, with fewer than 1,000 cases described in the literature. The mediastinum is the most common extragonadal primary site of origin (9). Moran and Suster analyzed the histologic features of a series of 322 primary germ cell tumors of the mediastinum over a period of 34 years. Although teratoma was the most common
Hemothorax due to Immature Teratoma

type, accounting for 138 cases (43%) there were only 6 cases (4%) of immature teratoma (10). In Japan, only 15 cases were reported in an autopsy database covering 1974 to 1995.

Etiologically, primordial germ cells migrating from the allantois hindgut during the first few weeks of embryologic development may give rise to immature teratoma by straying into midline extragonadal sites, such as the mediastinum (3).

Immature teratomas are always potentially malignant, but their prognosis is influenced by the anatomic site of the tumor, patient age, and the fraction of immature elements within the tumor (11). Carter et al (2) noted that all patients younger than 15 years were all alive and well, except for one patient who died postoperatively, whereas those 15 years of age or older all died from their tumors within 1 year of diagnosis (2).

The tumor of the present case broadly invaded the surrounding organs, and his age was much older than 15 years, So our case therefore, the tumor was fully suspected to be of malignant. To our knowledge, there is no report of a long-standing case without complete resection.

Epidemiological studies have estimated that 50% to 60% of teratomas of the mediastinum are detected as an incidental finding, which may be associated with vague chest pain, a symptom due to vessel compression (3). However, only a few reports have previously described spontaneous hemothorax secondary to immature teratoma of the mediastinum (4–7).

A review of mechanisms of teratoma rupture reported that autolysis, chemical inflammation (non-infected enzymatic inflammation), ischemia, pressure necrosis, and infection are potential causes (12). Because there was no remarkable findings of infection, chemical inflammation, ischemia, and pressure necrosis were considered as the main causes in the present case.

References