Case of spontaneous haemothorax associated with an endodermal sinus tumour

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Abstract

Spontaneous haemothorax is rare in infants. A case is reported of a nine month old infant who was found to have an endodermal sinus tumour.

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Spontaneous haemothorax is an occasional presentation of intrathoracic tumours. We describe its occurrence in an infant who was found to have an intrathoracic endodermal sinus tumour.

Case report

A nine month old boy presented with dyspnoea, cyanosis, and fever of sudden onset. The respiratory rate was 55/minute, heart rate 160/minute, and blood pressure 110/70 mm Hg. An opacification in the right hemithorax was visible on the chest radiograph but no abnormality of the mediastinum was detected. Thoracentesis yielded 140 ml of pure blood (haematocrit 29%). Analysis of

the pleural fluid did not reveal any malignant cells or bacteria. A thoracic computed tomographic scan showed a small pneumothorax on the right side and atelectasis in the basal segments of the right lower lobe. The patient improved, but on the 15th day of admission dyspnoea and cyanosis suddenly developed. Chest radiography showed complete opacification of the right hemithorax and 280 ml of bloody fluid was drained and a thoracotomy performed. At exploration a haematoma within the anterior wall of the thorax and a necrotic, haemorrhagic tumour (5 × 4 × 4 cm in size) was detected. This mass was adherent to the right lateral thoracic wall and a metastasis was found on the right lobe of the liver. No tumour was found in the mediastinum. The neoplasm was excised by right lower lobectomy. On macroscopic examination the right lower lobe was 6 × 6 × 4 cm in size and the visceral pleura thickened. Firm, gray-yellow irregular pieces of tumour tissue were seen. The biggest was 4 × 2 × 1.5 cm in size and some were haemorrhagic.

Microscopic examination showed atelectatic and interstitial fibrotic changes in the lung tissue. There was some compensatory emphysema within the lobe. Fibrous thickening and some haemorrhagic areas were seen in the pleural tissues. The tumour comprised a loose, vacuolated network with small cystic spaces forming a honeycomb pattern (fig 1). Numerous round hyaline globules were present inside and outside the cells (PAS positive) (fig 2) with perivascular formation (Shiller-Duvel bodies) and, in some areas, alveolar gland formation was seen. It was diagnosed as an "endodermal sinus tumour" and thought to be a metastasis originating from a primary tumour in the mediastinum. No other component was found to suggest that it was a teratoma. Computed tomographic scans of the brain, mediastinum, abdomen, and pelvis, and ultrasonography of the gonads showed no other foci. Laboratory test results showed increased plasma levels of α -fetoprotein (660 milliunits/ml) and carcinoembryonic antigen (18 ng/ml). The level of β -human chorionic gonadotrophin was normal (1 milliunit/ml). Chemotherapy was initiated and eight months later the patient was still in remission and α -fetoprotein levels had returned to normal.

Discussion

Spontaneous haemothorax is rarely seen in childhood. Neoplasms are among the rare causes of spontaneous haemothorax. In our patient the diagnosis of an endodermal sinus tumour was made at thoracotomy because of recurrent haemothorax. In 1988 Templeton *et al* first reported an extragonadal germ cell tumour in an adult who presented with a spontaneous haemothorax simulating an aortic dissection.¹ Klott and colleagues reported a child with malignant thoracic teratoma who had presented with a spontaneous haemothorax.²

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Figure 1 Loose vacuolated network, with small cystic spaces. Stain: haematoxylin and eosin. Original magnification × 125.

Figure 2 A loose vacuolated network of tumour tissue with hyaline globules both inside and outside the cells. Stain: haematoxylin and eosin. Original magnification $\times 500$.

Endodermal sinus tumours are the most common malignant germ cell neoplasm seen in childhood. They usually originate from the

gonads,³ but on rare occasions they may be seen in the retroperitoneum, anterior mediastinum, pleura, pericardium, sacrococcyx, central nervous system, liver, and vagina.^{3,4} The discovery of the metastasis to the peritoneum, pericardium, lymph nodes, and the lungs may be the first manifestation of the tumour.⁴ In our patient there was no evidence of any other lesion.

Despite the fact that malignant tumours are rare causes of spontaneous haemothorax they must not be overlooked in the differential diagnosis. In cases of haemothorax of unknown aetiology thoracotomy may be indicated.

- 1 Templeton PA, Vanright JR, Rodriquez A, Diaconis JN. Mediastinal tumours presenting as spontaneous haemothorax, simulating aortic dissection. *Chest* 1988;93:828-30.
- 2 Klott J, Walliser G, Sacher P. Nontrauma-induced haemothorax a certain leading symptom of a malignant tumour. *Z Kinderchir* 1989;44:115-8.
- 3 Kuzur ME, Cobleigh MA, Greco FA, Einhorn LH, Oldham RK. Endodermal sinus tumour of the mediastinum. *Cancer* 1982;50:766-74.
- 4 Truong LD, Harris L, Mattioli C, Hawkins E, Lee A. Endodermal sinus tumour of the mediastinum. *Cancer* 1986;58:730-9.

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Catamenial haemoptysis: a rare cause

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Abstract

Since the first report of pulmonary endometriosis as a cause of catamenial haemoptysis all cases have been assumed to be due to pulmonary endometriosis, even in the absence of histopathological proof. A case is presented where the histological findings were of a pulmonary arteriovenous malformation.

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Periodical haemoptysis occurring in association with the menses (catamenial haemoptysis) is a rare entity. Since the first published case, which was due to endometriosis of the lung,¹ there have been fewer than 20 reported cases. All the cases described have been attributed to pulmonary endometriosis although less than one third have had histological evidence.

Case report

A 32 year old woman presented with a 10 month history of recurrent haemoptysis occurring during every menstrual cycle. Symptoms started on the second day of her menses and lasted for three days. The haemoptysis was associated with mild right sided pleuritic type chest pain and never occurred without a period. No other symptoms were noted.

Two years previously she had been treated with methotrexate for nine months for an invasive hydatidiform mole. She had had normal menstruation and serum gonadotrophin levels ever since chemotherapy.

Clinical examination was normal. Laboratory investigations revealed normal full blood count, erythrocyte sedimentation rate, urea and electrolyte levels, liver function tests, bone profiles, coagulation studies, and serum gonadotrophin levels. Chest radiography during haemoptysis was normal but fiberoptic bronchoscopy revealed blood originating from the posterior segment of the right upper lobe but no endobronchial lesion was seen. Computed tomography during haemoptysis revealed a fairly well demarcated area of patchy consolidation posteriorly in the right upper lobe. Computed tomography was repeated in the middle of the cycle and revealed a small subpleural nodule at the same site with no evidence of surrounding consolidation or haemorrhage.

The patient was treated by posterior seg-

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