BENIGN LOCALIZED FIBROUS PLEURAL MESOTHELIOMA IN A PATIENT WITH HYPOGLYCEMIC SYMPTOM

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ABSTRACT

A case of localized fibrous pleural mesothelioma in a 55 year-old male patient with hypoglycemic sympotoms was presented. The patient had also clubbed fingers. A large mass occupied 1/2 to 2/3 of right hemithorax was shown by plain film. An isoechoic mass to the liver was seen by ultrasonography with low echoic areas. A well defined border mass with isodensity to the liver with areas of low density was observed by CT scan. Ultrasonography helped separating the mass from the normal liver. Surgery was performed with easiness and the tumor was totally removed with the disappearance of the hypoglycemic symptoms.

INTRODUCTION

Pleural mesotheliomas was first described by Lieutaud in 1767 (1). It was classified into diffuse and localized mesotheliomas by Klemperer and Rabin (2,3), according to the gross, microscopic and prognostic viewpoints. The diffuse type is better known because of its dramatic gross appearance, its rapidly fatal course and its induction by asbestos (4-6). Localized mesotheliomas, in contrast, are usually purely fibrous and carry a good prognosis (7,8). It has received a variety of names, including fibrous mesothelioma, benign mesothelioma, localized mesothelioma, subpleural fibroma, and localized fibrous tumor of the pleura (4).

We present a case of benign fibrous pleural mesothelioma in a patient with hypoglycemic symptoms. Images are illustrated by plain film, ultrasonography and CT scan.

CASE REPORT

A 55 year-old male farmer from Bureeram Province, in the Northeastern part of Thailand, presented to us with seizure 2 days prior to the admission. His problem began 3 months ago; he was easily tired and had weight loss. One and a half months prior to the admission, he had a behavioural change; having been aggressive only in the morning every day. He, later, developed seizure and finally status epilepticus. He was found to have decreased blood sugar at a local hospital. He smoked 10 cigarettes per day for 30 years and was a regular alcoholic drinker. The positive physical examination findings were hepatomegaly, clubbing of fingers, chachetic, absent Babinskin's sign. His serum blood sugar was 20 mg/dl, calcium 8 mg/dl, and a normal serum insulin level.

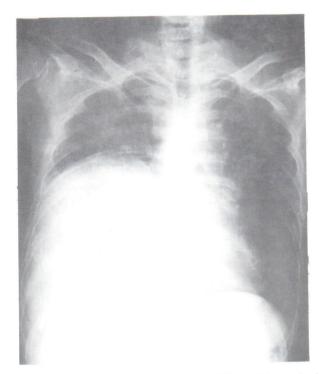
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Plain chest film in AP and lateral views showed a large intrathoracic mass at the lower half of the right hemithorax (Fig. 1). Ultrasonography of the right upper quadrant showed a solid mass at right lower hemithorax and a normal liver (Fig. 2). CT scan showed a large well defined border solid mass at right lower hemithorax without adjacent organs invasion; multiple small necrotic areas was noted within the mass (Fig. 3).



The tumor from the right lower hemithorax was removed surgically and totally. There was some adhesion to the right hemidiaphragm. The colour of the mass was white with necrotic areas. Histology revealed a benign fibrous mesothelioma.

The hypoglycemic symptoms and seizure disappeared after tumor removal.

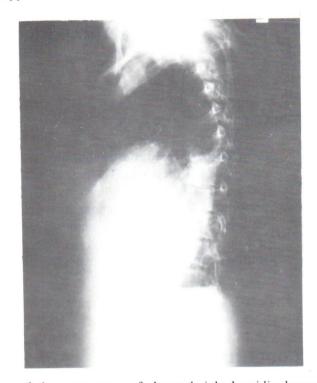


Fig. 1 Plain film of the chest in AP and lateral views showed the appearance of elevated right hemidiaphragm VS a large mass at the lower half to the hemithorax, obliterating right hemidiaphragm.

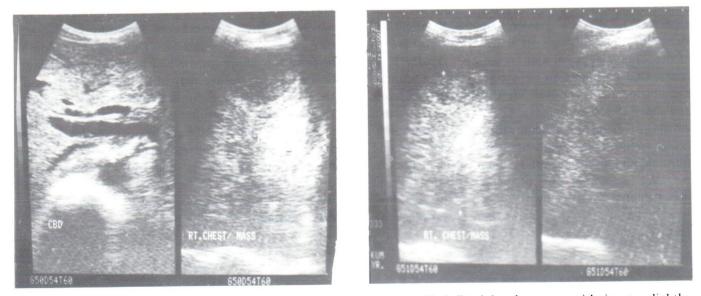


Fig. 2 Ultrasonography in the region of the mass revealed a well defined border mass with iso to slightly hyperechoic to the liver parenchyma and there were areas of low echo. The liver was normal.

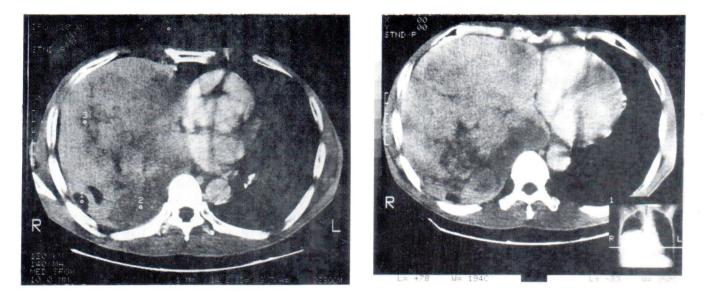


Fig. 3 I.V. contrast axial CT scan of the chest showed a well defined border mass with isodensity to mildly hypodensity to the adjacent heart and aorta. Multiple small low density areas were noted.

DISCUSSION

Solitary fibrous tumors of pleura achieved a classic clinical and pathologic description: a large pedunculated mass attached by a narrow pedicle to the visceral pleura, in an asymptomatic individual who occasionally had hypoglycemia or osteoarthropathy (7,9-13). A minority of localized fibrous tumors of pleura were intrapulmonary lesions (14-15).

The localized fibrous tumors of the pleura occured equally in both sexes, most commonly in the sixth to seventh decades of life. Presenting symptoms included chest pain, dyspnea, and cough; they were observed in three-fourths of patients with a malignant tumor. One in every four of these patients had hypoglycemia, clubbed digits or pleural effusion. Two-thirds of the tumors were attached to visceral pleura, often by a pedicle. The rest arose from the parietal pleura of the chest wall, diaphragm, or mediastinum. Neoplasms in these atypical sites, together with fissural lesions and tumors "inverted" into peripheral lung, were more often malignant. Most neoplasms measured 5-10 cm and weighed 100-400 g. Microscopically, the "patternless pattern" or hemangiopericytic type, was seen in the majority of cases, and mixed patterns were seen in nearly 40% of tumors. All of the benign and 45% of the malignant tumors were cured by simple excision. Patients surgically cured of a malignant neoplasm had pedunculated or well circumscribed lesions. However, 55% of patients with malignant tumors succumbed to their disease secondary to invasion, recurrence, or metastasis. Resectability is the single most important indication of clinical outcome. No tumor expressed epithelial differentiation, either immunohistochemically or ultrastructurally; therefore, England (17) favored the term "localized fibrous tumor" of pleura instead of localized mesothelioma.

Most localized benign fibrous pleural mesothelioma were discovered incidentally or routine chest radiographs (17-20). They can occasionally attain such a large size that they cause considerable opacification of a hemithorax in plain radiograph. More often they appear as solitary, often lobulated, well circumscribed, non calcified softtissue masses, either in the periphery of the hemithorax abutting a pleural surface or related to an interlobar fissure. They are often elongated and roughly lenticular, with the greatest dimension in the longitudinal plane. The surface adjacent to the chest wall may be relatively flatter than that abutting the lung parenchyma. Benign mesotheliomas often arise from the visceral pleura and are attached to the pleural surface by a pedicle. In such instances, they project freely into the pleural space, and changes in position and shape can be observed on inspiration and expiration radiographs and/or fluoroscopy (14,21,22). Pleural lesions form obtuse angles with the chest wall (23,24) but a gradually tapering interface of mass and chest wall to be a more reliable sign of pleural origin (17,18).

CT scan has several advantages by showing (1) displacement of adjacent lung parenchyma with

compressive atelectasis and bowing of the bronchi and pulmonary vessels around the mass, (2) a smoothly tapering margin at the junction of the mass with the pleura, (3) in pedunculated lesions, CT can exhibit changes in location and shape in the supine and prone positions, similar to changes observed on fluoroscopy (25), (4) with contrast enhancement, CT scan differentiate a fibrous mesothelioma abutting the mediastinum from an aortic aneurysm. The absence of fat excludes a diaphragmatic herniation that might contain omentum or gastrointestinal viscera, or a pleural lipoma, (5) CT can demonstrate invasion of the chest wall or infradiaphragmatic structures, a rare feature of fibrous mesothelioma.

Ultrasonography may help identify the diaphragm and its location with respect to the mass, when a large mesothelioma fills the inferior hemithorax and CT might not delineate the diaphragmatic interface (26).

Selective inferior phrenic, intercostal, and internal mammary arteriography may localize the mass by determining its blood supply and demonstrate large vascular pedicles and possible sites of vascular adhesion to the diaphragm or chest wall, which can be potential sites of bleeding at surgery (27).

A definitive diagnosis cannot be established by either percutaneous needle aspiration biopsy or cytology from pleural fluid (28). Thoracotomy is necessary for diagnosis, and wide local resection is the treatment of choice (29).

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