Pleural and transdiaphragmatic retroperitoneal metastasis developing two and half years after resection of invasive thymoma

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ABSTRACT

We report transdiaphragmatic pleural and retroperitoneal metastasis developing two and half years after resection of invasive thymoma (Masaoka stage III; WHO type B1, lymphocyte-rich) in a 34-year-old man. Post-surgery, he received radiotherapy and chemotherapy. Follow-up computed tomography (CT) one year post-surgery did not reveal any local recurrence or metastasis. He remained asymptomatic throughout. A follow-up CT done two and half years later revealed an enhancing retrocrural-retroperitoneal (posterior pararenal space) soft tissue mass measuring 12 cm x 10 cm x 6 cm. Another enhancing deposit was found in the left pleural space. This lesion was found infiltrating into the subjacent lung. Both these deposits were resected along with wedge resection of the affected subsegment of the lung. Histopathology confirmed these lesions to be metastases from the lymphocyte-rich thymoma.

Keywords: invasive thymoma, lymphocyte-rich thymoma, transdiaphragmatic metastasis

Singapore Med J 2008;49(3):e64-e67

INTRODUCTION

Thymomas are rare tumours, but they represent the most frequently-encountered primary tumours of the anterior mediastinum. They are usually confined to the mediastinum at the time of diagnosis. Most thymomas have a relatively slow-growing indolent course, but have a predisposition for local recurrence after resection. Distant metastasis from invasive thymoma is distinctly rare, but has been reported to occur in the lungs, pleura, diaphragm, liver, bones, kidney, extra thoracic lymph nodes, pelvis, retroperitoneum and nervous system.[1-5] We report a 34-year-old man who developed pleural and transdiaphragmatic retroperitoneal metastasis two and half years following resection of an invasive thymoma (Masaoka stage III; WHO B1, lymphocyte-rich). Pleural and transdiaphragmatic retroperitoneal metastasis occurring in the same patient after resection of invasive
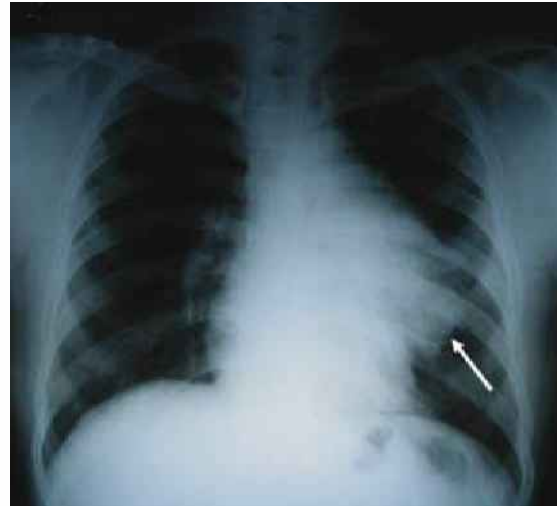

Fig. 1 Chest radiograph (PA view) shows a mediastinal-based mass lesion on the left side (arrow).
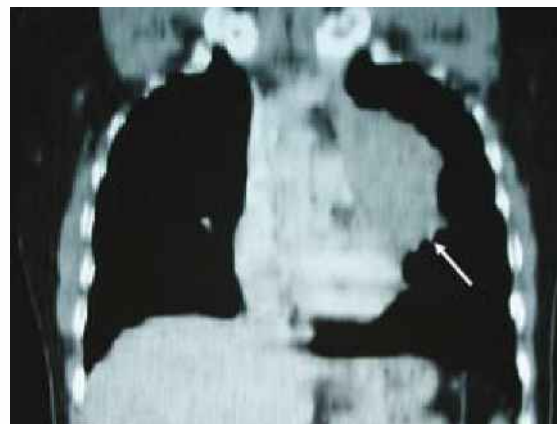

Fig. 2 Contrast-enhanced CT image of the chest (coronal recontruction) shows an enhancing lobulated soft tissue mass with a broad base towards mediastinum.

thymoma has not been described in the literature.

CASE REPORT

A 34-year-old man, a serving military personnel, presented with history of breathlessness on exertion, and cough with scanty mucoid expectoration for a duration of eight months. There was no history of loss of appetite, fever with evening rise of temperature, or significant loss

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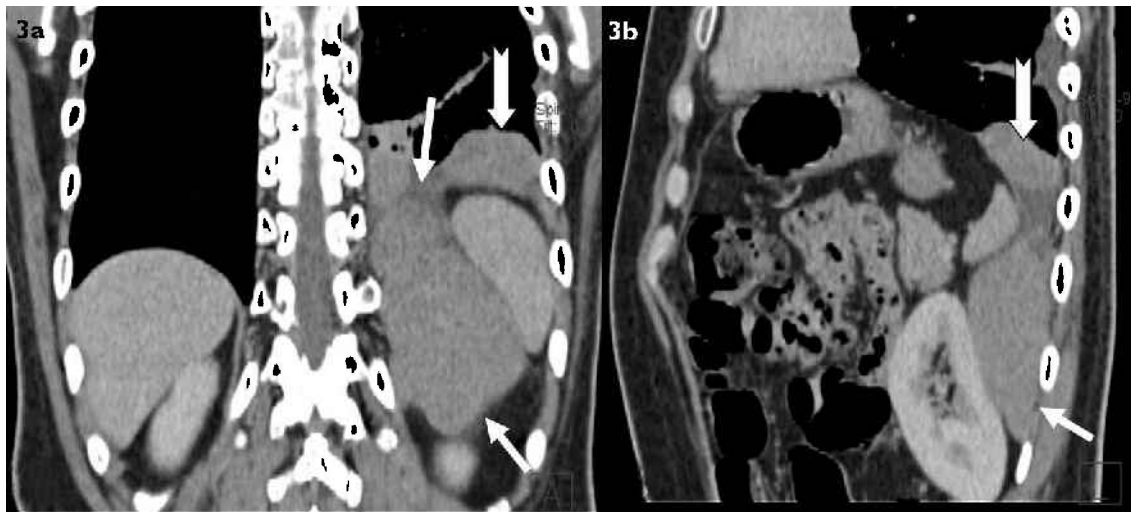Email: jyotindu_
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Fig. 3 Follow-up (a) coronal and (b) sagittal reconstructed CT images (done two and half years later) show a retrocrural posterior para-renal mass lesion (arrows). The pleural space deposit (notched arrows) is also noted.

of weight. On examination, he was well-preserved with normal vital parameters. Routine haemogram was within normal limit, with haemoglobin of 15 gm/dL. A chest radiograph (Fig. 1) revealed a mediastinal mass lesion on the left side. A contrast-enhanced computed tomography (CECT) scan of the chest (Fig. 2) showed a 12 cm × 7 cm heterogeneously-enhancing mass in the left upper hemithorax with a broad base towards mediastinum. Early infiltration into the mediastinum with loss of fat plane with the main pulmonary artery, aortic arch and pericardium was noted. Based on the imaging features, possibility of invasive thymoma was suggested. There was no pleural or focal lung parenchymal lesion elsewhere.

Ultrasonography-guided fine-needle aspiration cytology (FNAC) was done, and the cytology was suggestive of thymoma. At surgery, the mass was found adherent to the left hilum, left pulmonary artery, adjacent parietal pericardium, arch of aorta and left upper lobe (Masaoka stage III). The mediastinal mass was excised via a left posterolateral thoracotomy approach along with partial resection of the left upper lobe, parietal pleura, pericardium and resection of the left phrenic nerve. The tumour needed to be shaved off the pulmonary artery and the arch of aorta, hence the resection was considered marginal with a R2 residue on the above sites. Histopathological examination revealed invasive thymoma, WHO type B1 (lymphocyte-rich) with transcapsular invasion with adherence to pericardium and great vessels. Post-surgery, he received external beam radiotherapy (5040 cGy) and six cycles of chemotherapy (Endoxan + Cisplatin + Adriamycin + Vincristine) and he was doing well. A follow-up CECT done one year after surgery was essentially unremarkable, with no evidence of residual/recurrent disease.

At the recent follow-up, which was two and a half years

post-surgery, CECT of the chest showed a homogeneously-enhancing soft tissue mass lesion measuring 12 cm × 10 cm × 6 cm in the left retrocrural and posterior pararenal space (Fig. 3). There was also an enhancing pleural space nodule. This lesion was seen to infiltrate the adjacent lung parenchyma. FNAC from the retroperitoneal mass lesion revealed metastasis from lymphocyte-rich thymoma. The retrocrural-retroperitoneal mass lesion and pleural deposit were removed via a thoraco-abdominal approach along with a wedge resection of the affected subsegment of the left lower lobe of the lung (Fig. 4). Histopathology confirmed them to be metastasis from thymoma (WHO B1, lymphocyte-rich), having a similar histological pattern as that of the primary tumour (Fig. 5). He had an uneventful postoperative period and has been subjected to six cycles of salvage chemotherapy post-surgery.

DISCUSSION

Thymomas are rare indolent tumours of the anterior superior mediastinum. They can be broadly categorised into two stages: noninvasive and invasive. Invasive thymomas constitute up to 37% of all thymomas.[1] Despite a benign histology, thymomas are known to cause local invasion of nearby structures, such as the pleura, lung, pericardium and major vessels. Complete surgical resection along with resection of the surrounding involved structures, is the standard treatment of choice for the localised form of the disease. The roles of neoadjuvant induction chemotherapy and postoperative radiotherapy as a part of multimodality approach treatment of advanced thymoma (stages III and IVa) are increasingly recognised. There are reports to suggest that preoperative neoadjuvant chemotherapy may help to decrease the tumour size and increase the chances of complete curative resection with improved survival.[6-9]
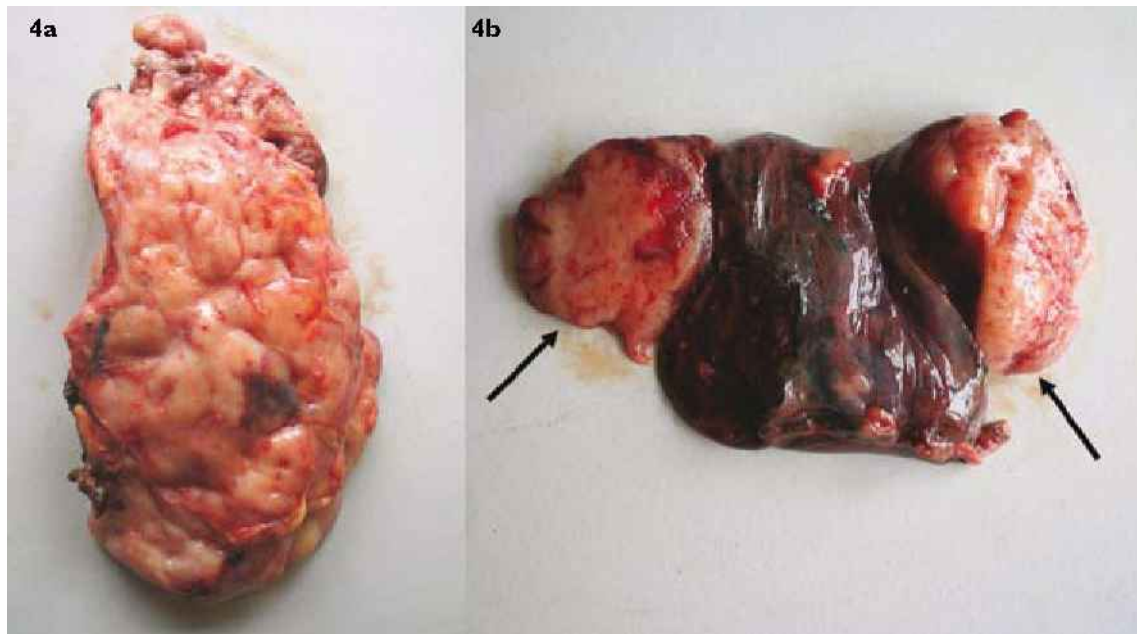
Fig. 4 Photographs of the resected specimen show (a) resected retrocrural retroperitoneal mass, and (b) pleural space deposit (cut open; arrows).
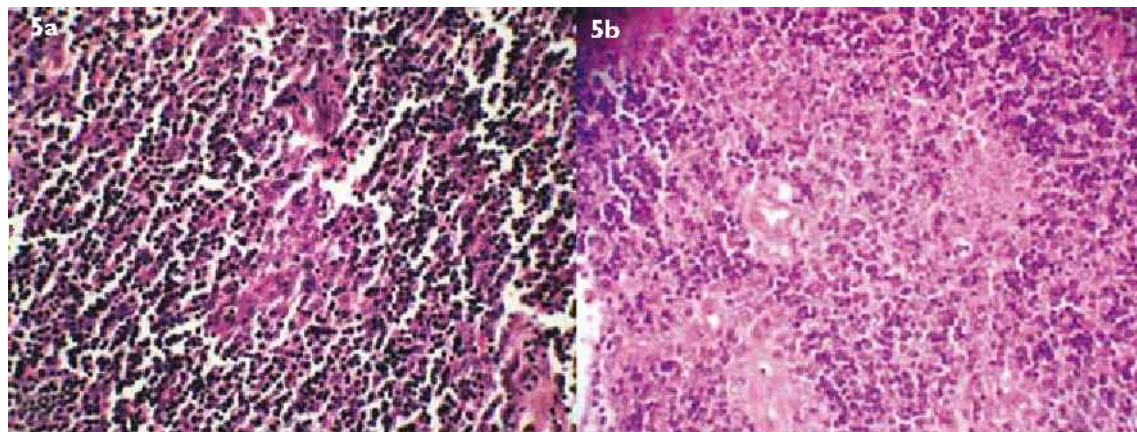

Fig. 5 (a) Photomicrograph of a postoperative specimen of the primary tumour (operated in September 2004) shows predominant population of reactive lymphocytes and a few neoplastic epithelial cells (Haematoxylin & eosin, × 400). (b) Photomicrograph of the metastatic retroperitoneal deposit (resected two and a half years later) also shows similar histological features to those of the primary tumour cells (Haematoxylin & eosin, x 400).

Despite complete resection, local recurrences are not infrequent. Distant metastases from the thymoma, though rare, are known to occur, even several years after complete resection of the primary tumour.[3,4] The most common sites for thymoma metastases reported in the literature are the liver, lung, lymph nodes, and bone. Other rarer sites of distance metastases include the central nervous system, pelvis and retroperitoneum.[1,2,5] There are very few reports of transdiaphragmatic retroperitoneal involvement by invasive thymoma. In a study of 19 patients, Scatarige et al have described transdiaphragmatic extension of invasive thymoma in six (31.5%) cases.[1] However, other than this study, there are very limited reports about retroperitoneal involvement by invasive thymoma. Three potential routes

for the transdiaphragmatic spread of invasive thymoma have been described, namely: (a) through the retrocrural space; (b) through the openings in the anteromedial diaphragmatic origin; and (c) direct invasion of muscular diaphragm and spread into the peritoneal/extraperitoneal space.[1]

Based on our experience at our oncology centre, which generally admits and treats an average of 4–5 cases of thymoma per year, this is the first such case noted in the last ten years. Our case highlights clinically unsuspected, extremely rare combination of pleural and transdiaphragmatic retroperitoneal metastases during follow-up of invasive thymoma. It also emphasises the need for carrying out CT of the chest and upper abdomen

during follow-up of invasive thymoma cases.

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