A large solitary fibrous tumour of the pleura: a case report and review of the literature

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Abstract
We report a clinical case of a solitary fibrous tumour of the pleura (SFTP) in a 67-year-old female patient complaining of chest pain for 2 months. A localized large mass was found in the left inferior hemithorax by computed tomography scan. The patient underwent a thoracotomy at the left side with endotracheal anaesthesia. During surgical resection, the tumour was located in the left inferior hemithorax and was attached to the inferior lobe of the left lung and diaphragmatic pleura by a fibrous pedicle. A wedge resection of the left lower lobe was undertaken to completely remove the tumour. Diagnosis of the SFTP was confirmed by the surgical findings and subsequent histological and immunohistochemical examinations. At the 6-month follow-up, no signs of local tumour recurrence or metastasis were documented. After a 3-year follow-up, this patient remains in good health.

Keywords
Solitary fibrous tumour of pleura, pleura, tumour, case report

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Case presentation
A 67-year-old Chinese woman was admitted to the Department of Cardiothoracic Surgery, First Affiliated Hospital of Yangtze University, Jingzhou, Hubei Province, China in June 2014 complaining of chest pain that had been ongoing for 2 months. A localized large mass was found in the left inferior hemithorax by computed tomography scan. The patient underwent a thoracotomy at the left side with endotracheal anaesthesia. During surgical resection, the tumour was located in the left inferior hemithorax and was attached to the inferior lobe of the left lung and diaphragmatic pleura by a fibrous pedicle. A wedge resection of the left lower lobe was undertaken to completely remove the tumour. Diagnosis of the SFTP was confirmed by the surgical findings and subsequent histological and immunohistochemical examinations. At the 6-month follow-up, no signs of local tumour recurrence or metastasis were documented. After a 3-year follow-up, this patient remains in good health.

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months. A computed tomography (CT) scan revealed a large, well-circumscribed pleural-based mass (cranio-caudal diameter of 10 cm) on the left side (Figure 1A). The tumour contained feeder arteries and was heterogeneously enhanced. The patient was maintained at 90° lateral decubitus position and underwent a thoracotomy at the left side with endotracheal anaesthesia. During surgical resection, the tumour was located in the left inferior hemithorax and was attached to the inferior lobe of the left lung and diaphragmatic pleura by a fibrous pedicle. A wedge resection of the left lower lobe was undertaken to completely remove the tumour. The operation took 110 min and blood loss was 100 ml. Macroscopically, the resected tumour was 13 cm × 12 cm × 9 cm in diameter with a fibrous capsule and extensive vessels (Figure 1B). The histological examination revealed a proliferation of spindle-shaped cells in a patternless or fascicular or partial storiform fashion with thin-walled branching vessels and bands of collagen (Figure 2). The tumour cells were immunohistochemically positive for cluster of differentiation (CD)34 (Figure 3A) and negative for S-100 (Figure 3B). The immunohistochemical evaluation demonstrated positive staining for CD34, melan-A, vimentin and Ki67, whilst pan-cytokeratin (PCK), smooth muscle actin (SMA), S-100, desmin, calretinin (CR) and human melanoma black-45 (HMB-45) were negative. The final diagnosis was confirmed as a solitary fibrous tumour of the pleura (SFTP) without malignant features. Follow-up was performed with 6-monthly chest CT scans in the first year, then once a year. To date, there has been no evidence of recurrence or metastasis.

Discussion

Solitary fibrous tumour of the pleura is a rare neoplasm accounting for less than 5% of primary pleural tumours and the incidence is approximately at 2.8 cases/100 000 per year.1,2 It derives from the submesothelial mesenchymal layer and usually appears to arise from visceral pleura, more rarely from the parietal pleura.3,4 Whereas benign SFTPs are often small pedunculated tumours, most malignant SFTPs may reach more than 10 cm in diameter.2 However, benign fibrous tumours are defined as giants when the diameter is greater than 15 cm or when the tumour occupies more than 40% of the hemithorax.5

Figure 1. A contrast-enhanced computed tomography scan of a 67-year-old female patient revealed a large oval mass located in the left hemithorax with extensive vessels, heterogeneous patchy enhancement and a pseudo-capsule (A). The pathological specimen was 13 cm × 12 cm × 9 cm in size and appeared encapsulated with extensive vessels beneath (B). The colour version of this figure is available at: http://imr.sagepub.com.
in this current case was less than 15 cm in size, so it was described as a large SFTP.

The majority of patients with SFTP are asymptomatic at the time of being diagnosed incidentally by routine chest radiograph.6 Large lesions may cause compression of adjacent structures and patients may present with some symptoms like chest pain, cough or dyspnoea.7,8 Larger tumours are more likely to be symptomatic.9 Some patients present with so-called ‘paraneoplastic syndromes’ including refractory hypoglycaemia, digital clubbing and pulmonary hypertrophic osteoarthropathy.10,11 These paraneoplastic syndromes are possibly due to abnormal expression of hepatocyte growth factor, a massive release of hyaluronic acid or the excessive production of insulin-like growth factor 2; nevertheless, these symptoms always dramatically resolve following resection of the tumour.2,12

Figure 2. Representative photomicrographs of tissue sections taken from a large oval mass located in the left hemithorax of a 67-year-old female patient showing hypercellular (A) and hypocellular areas (B) (haematoxylin and eosin). The tumour comprised of a proliferation of spindle-shaped cells in a patternless or partial storiform fashion with thin-walled branching vessels and bands of collagen. Scale bar 100 μm. The colour version of this figure is available at: http://imr.sagepub.com.

Figure 3. Representative photomicrographs of tissue sections taken from a large oval mass located in the left hemithorax of a 67-year-old female patient showing that the tumour cells were immunohistochemically positive for CD34 (A) and negative for S-100 (B). Scale bar 100 μm. The colour version of this figure is available at: http://imr.sagepub.com.
A thoracic CT scan is a useful diagnostic method, which can clearly identify the location and size of the lesion and help surgeons to assess the possibility of resecting the SFTP. Generally, a thoracic CT scan usually shows a homogeneous well-circumscribed, lobular soft-tissue mass. However, large or giant tumours are frequently viewed as heterogeneous because of tissue haemorrhage, necrosis, or cystic changes. In the current case, the CT scan showed heterogeneous density. In addition, some investigators have suggested that CT-guided aspiration biopsy is not advisable as a reliable tool due to its low diagnostic sensitivity. Magnetic resonance imaging, as a widely used medical diagnostic tool, is occasionally helpful for excluding potential invasion by a sessile tumour into the adjacent structures, because about 28% of tumours abutted the diaphragm and 79% extended into the lower thorax in one case series. The low sensitivity of positron emission tomography scans represents a potential weakness of this technique as reported previously.

In general, SFTP is a firm, well-circumscribed or lobulated lesion, with a smooth encapsulated appearance, and nearly 50% are attached to adjacent pleural surface by a pedicle. The cut surface appears white to yellowish-brown and usually contains areas of haemorrhage and necrosis, which are more common in malignant tumours when compared with benign ones. Microscopically, the tumour is composed of spindle-shaped cells with indistinct borders, scant cytoplasm, and distributed fine-scale chromatin in a round-to-oval nucleus. Bands of collagen and reticular fibres separate the cells into hypocellular and hypercellular areas in a random fashion, and therefore microscopy usually reveals a 'patternless pattern'. Mitoses are barely visible in benign SFTPs, usually less than three mitoses per 10 high-power fields. In malignant tumours, cell division becomes more frequent and the number of mitoses are more than four per 10 high-power fields.

The preoperative differential diagnosis is important to differentiate any lump type in the chest, ranging from lung cancer to various intrapleural sarcomas. Immunohistochemical examinations play a significant role in distinguishing SFTP from mesotheliomas and some other sarcomatous lesions. To confirm SFTP, this current case investigation adopted some specific immunohistochemical stains and the results showed that CD34, melan-A and Ki67, as well as vimentin, were present in the tumour. Whereas S-100 protein, PCK, SMA, desmin, CR and HMB-45 were not present. Immunohistochemical staining helps to exclude a peripheral sarcomatoid carcinoma of the lung, malignant mesothelioma, synovial sarcoma, and other rare tumours.

Complete surgical resection including lobectomy is the procedure of choice for all SFTPs. Generally, if the lump is small, accurate resection of the lesion can be achieved with the help of video-assisted thoracic surgical techniques and most of the resected tumours are benign. Whereas if the lesions are giant tumours or unusually large, such as that shown in Figure 1, an open thoracotomy is necessary to remove the tumour. Some authors have recommended that preoperative embolization of the tumour-supplying arteries should be performed in giant SFTPs due to the risk of haemorrhage. The tumour in the present case was attached to the visceral pleura by a pedicle through which the major feeding vessels entered into the tumour. However, embolization was not undertaken before surgery because the tumour did not show invasive features and the surgeons considered that the tumour pedicle would be easily and safely treated. Despite this, the operation was a little more difficult than expected due to the large tumour size, poor exposure...
and pleural adhesions. To avoid excessive bleeding and ensure en-bloc resection, high-frequency electrocoagulation and an ultrasound knife were used to treat the adhesions and a surgical stapler was used to perform the wedge resection.

Although localized benign SFTP can almost always be cured with complete surgical removal, long-term follow-up of patients after surgery is essential because of the possibility of local recurrence.20,21 After a 3-year follow-up, this patient remains in good health.

In conclusion, this case report describes a rare large solitary fibrous tumour of the pleura that was successfully treated by surgical resection. In this current case, a chest CT scan was an important diagnostic imaging method, which demonstrated the characteristic patterns of the tumour. Nevertheless, the confirmed diagnosis and differential diagnosis still depended on subsequent histological and immunohistochemical examinations. Complete resection is an effective treatment for SFTP, but long-term follow-up of the patient after surgery is necessary for early detection of tumour recurrence.

Declaration of conflicting interests

The authors declare that there are no conflicts of interest.

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