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Two-stage Surgical Treatment of a Giant Solitary Fibrous Tumor Occupying the Thoracic Cavity

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A solitary fibrous tumor (SFT) is a mesenchymal fibroblastic tumor inside the pleura, for which complete surgical resection is the standard treatment. For large SFTs, preoperative identification of tumor-feeding vessels using angiography is important for achieving complete resection without unexpected operative bleeding. Extensive adhesions can make resection difficult in a limited operative window, and pulmonary resection may be required to achieve complete SFT resection. Herein, we report successful resection of a large pleural SFT in a 39-year-old man without any complications using a 2-stage approach, in which ligation of the feeding vessels through small another operative window was the first step.

Key words: 1. Tumor, benign
2. Pleura
3. Surgery
4. Complication

Case report

A 39-year-old man presented at the emergency care center of Chonbuk National University Hospital due to recurrent syncope and hypoglycemic symptoms. The patient had no history of trauma or past medical treatment except for a diagnosis of chronic hepatitis B. The patient did not have a history of diabetes mellitus and was not taking any glucose-lowering agents. Upon physical examination, a height of 154 cm, a weight of 50 kg, blood pressure of 117/71 mm Hg, and a heart rate of 84 beats/min were revealed. There were no breathing sounds in the left lung field. Routine clinical laboratory tests were performed, and the findings were within the normal range except for hypoglycemia (35 mg/dL). Tumor markers (alpha fetoprotein, carbohydrate antigen 19-9, carcinoembryonic antigen), serum cortisol secretion, and the adrenocorticotropic hormone level were all normal. Despite marked hypoglycemia, his insulin and C-peptide blood levels were low, at 0.25 μU/mL (normal range, 2.6–24.9 μU/mL) and 0.12 ng/mL (normal range, 1.1–4.0 ng/mL), respectively. His serum somatomedin C level was 66 ng/mL, and his anti-insulin antibody level was 5.9%.

Chest radiography showed that his left chest was nearly filled by a huge mass (Fig. 1). Chest computed tomography (CT) showed a well-defined 27×11×12 cm mixed cystic lesion inside the left pleural space that contained multiple regions showing heterogeneous density with elements of soft tissue plus fat content. Spots of wall calcification were also revealed

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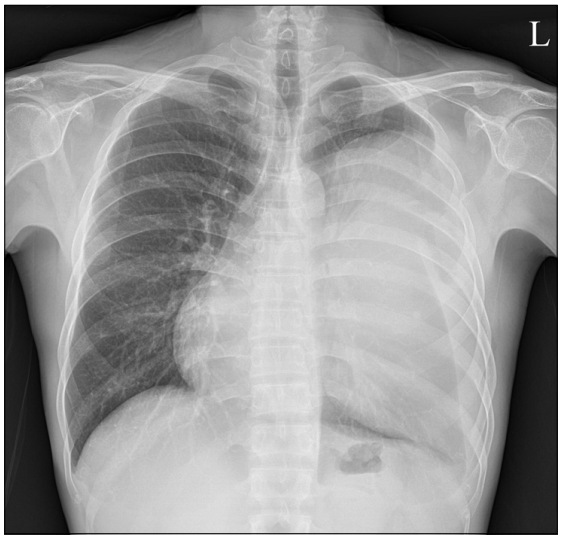

Fig. 1. Preoperative chest X-ray shows a huge mass occupying nearly the entire left hemithorax.

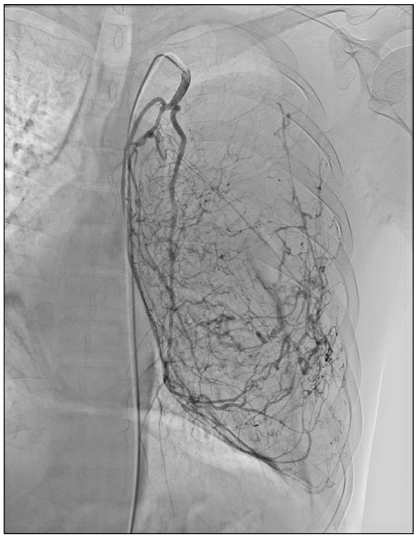

Fig. 2. Angiography shows the vascular pedicle of the solitary fibrous tumor originating from the left internal thoracic artery.

by the chest CT where it is likely that the lesion had originated from the mediastinum; this involved the majority of the left hemithorax. The mass compressed the mediastinum, left pulmonary artery, aorta and left bronchus. The diagnosis was thought to be intrathoracic solitary fibrous tumor (SFT) associated with non-islet cell tumor hypoglycemia (NICTH). In order to preserve the level of glucose in the patient's plasma, glucose supplementation of 10 g/g was needed during preoperative fasting. Selective percutaneous angiography of the left subclavian artery, left thyrocervical trunk, left internal mammary artery, and intercostal arteries was carried out to identify the tumor's feeding vessels. The vascular pedicle originated from the left internal thoracic artery (Fig. 2). Complete resection of the tumor was planned to diagnose and cure the disease. The patient was placed in a right 45° lateral decubitus position for surgery. First, a small anterior thoracotomy was performed on the third intercostal space beside the left sternal margin, the left internal mammary artery was exposed, and the feeding vessels of the tumor were divided (Fig. 3). The tumor was then exposed through lateral thoracotomy at the fifth intercostal space. It originated from the mediastinal pleura and was covered with an encapsulating membrane. The vascular network was well developed. There was dense pleural adhesion at the diaphragmatic surface and mediastinal pleura. The tumor was separated from the mediastinum, chest wall, diaphragm, as well as from all of the structures surrounding it with no injuries occurring or complications with bleeding through the relatively enough operative window. Histopathological examination revealed an SFT with fibrous tissue and hyaline degeneration. The chondroid tissue and adipose tissue showed no specific pathologic findings. Immunohistochemical staining was positive for CD34 and negative for smooth muscle actin. The patient's hypoglycemic symptoms resolved immediately after surgery and he was discharged on postoperative day 7 without any further problems. Two months after

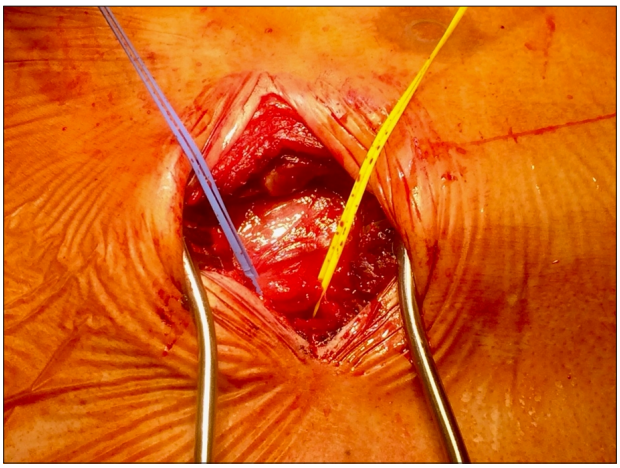

Fig. 3. Ligation of the vascular pedicle through a mini-anterior thoracotomy.

the operation, plasma glucose levels were found to be in the normal range. The present study was exempted from the institutional review board for single, retrospective study. Informed consent was waived.

Discussion

SFTs are rare, representing fewer than 5% of all pleural tumors, and have an incidence of only 2.8 cases per 100,000 [1]. The high incidence of pleural SFT is in those aged between 40 and 70, but these tumors can occur in people of all ages. A number of symptoms including cough, chest pain, and dyspnea are shown in 64% of patients with SFT upon admission. Hypoglycemia is associated with 4% of SFTs [2]. Karl Walter Doege and Roy Pilling Potter first described NICTH in 1930. Reactive hypoglycemia often accompany Large SFTs (>20 cm) and SFTs with a high mitotic rate [3]. A review found that 46% of pleural SFTs that manifested with hypoglycemia were malignant, while 54% were benign [4]. Hypoglycemia was identified and the pathologic findings were benign in the present case. The visceral pleura is most often the origin of pleural SFTs, and just 20% coming from the parietal pleura. It is more probable that SFTs ＞8 cm will originate from the parietal pleura and to have a vascular pedicle [2]. A solitary pedicle attaches about 50% of SFTs with the pleura [3], and up to 47% are encapsulated by a membrane containing a vascular network [3,5]. Notably, a highly vascularized pedicle attaches SFTs in 38% to 46% of patients to the pleura [3], and this is more likely if the tumor is large. SFTs can be diagnosed preoperatively by fine needle biopsy, but the diagnostic accuracy is reported to be low [5]. The diagnosis can be confirmed by surgical resection.

All benign cases of SFT and approximately half of malignant cases are generally cured by resection [3]. Surgical resection is a mainstay of SFT treatment, but SFTs are often difficult to resect completely due to their hypervascular nature and their collateral vessels. Massive hemorrhage can occur due to their hypervascular nature, so it is important to identify tumor vascularity preoperatively in order to perform resection without any bleeding complications. The overall operative mortality is ranged between 1.5% and 12% due to the hemodynamic variations associated with decompressing heart, the great vessels, airway and with bleeding [2]. Preoperative angiography is a valuable tool for identifying major feeding vessels. The blood supply of SFTs has rarely been described, but as reported by England et al. [3], in their series, five of the patients experienced, aberrant vessels, which included a branch of the brnchial artery and a phrenic artery. Guo et al. [6] reported 5 patients with pleural SFTs that had multiple feeding vessels from the internal mammary, bronchial arteries, inferior phrenic arteries, and intercostal arteries. An aberrant blood supply from the celiac trunk has also been reported [7]. Preoperative percutaneous embolization may be of use in lowering perioperative blood loss [8], and surgery should be completed within a 24-hour period to minimize blood loss [6]. Video-assisted thoracoscopic surgery (VATS) could be used for suitable tumors. However, conversion to the open technique is recommended to be used if margin clearance is not achievable with VATS. VATS for lesions ＜5 cm is recommended by certain authors, while larger tumors can be resected through a thoracotomy window. During surgery, a simple and rational way to avoid compression is with the 45° lateral decubitus position. Considerable adhesions around a tumor lead to difficulty in ligating the feeding vessels and to deal with the hilar vessels, particularly in a tumor with a broad-based pedicle [6]. In our patient, the vascular pedicle was identified by preoperative angiography, and ligation was performed first through a small incision. The main incision of the lateral thoracotomy was small enough to excise the huge mass without any bleeding complications. Extended resections, for instance, a chest wall resection, excision of the parietal pleura and pericardium, and lobectomy, may be needed to attain a clear margin. The wholeness of surgical excision is a major prognostic factor for both benign and malignant SFTs.

In conclusion, preoperative angiography is valuable for identifying the vascular pedicle and network of very large pleural SFTs. The vascular pedicle structures must be controlled in the first stage of the operation to ensure safe resection. After the first stage, pleural SFTs can be resected completely, without any bleeding problems, through an appropriately small surgical window.

Conflict of interest

No potential conflict of interest relevant to this article was reported.

Acknowledgments

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