

Posterolateral Thoracotomy Approach for an Ectopic Mediastinum Thymoma: A Case Report

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ABSTRACT

Thymic epithelial neoplasms are commonly aetiology of the anterior mediastinum masses in adults. It represents 20–30% of all mediastinal tumours in adults. Ectopic thymomas usually affect the neck, mediastinal compartments, lung, and pleura, arising from aberrant thymic tissue. For giant thymoma, there is still no consensus on the surgical approach. We herein report a patient with a giant thymoma that was successfully resected through a large right posterolateral thoracotomy. We report a rare case of a giant thymoma in the posterior mediastinal. Our patient underwent a conventional posterolateral thoracotomy to remove the mass. This latter was histologically diagnosed as a WHO classification type AB thymoma and Masaoka stage I. The choice of surgical approach of mediastinal tumor is based on the tumour topography, the assessment of tumour extension and extirpation, the surgeon's experience and clinical symptomatology. A multidisciplinary approach is mandatory to achieve good results.

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Introduction

Thymic epithelial neoplasms are commonly aetiology of the anterior mediastinum masses in adults. It represents 20–30% of all mediastinal tumours in adults (1-3). Ectopic thymomas usually affect the neck, mediastinal compartments, lung, and pleura, arising from aberrant thymic tissue (4-5). A review of the literature confirms that thymoma have slow-growing characteristics and may be asymptomatic for long enough or present with symptoms of local compression or general manifestations such as myasthenia gravis and

paraneoplastic syndrome (1-2). Giant thymomas are an unusual and difficult entity to excise due to tumour size and the involvement of neighbouring organs and vessels directly related to the tumour. However, the most effective treatment modality for thymoma is surgery. Many studies demonstrated different approaches but unanimously the transsternal approach is the current approach for thymectomy of normal sized thymoma (6). For giant thymoma, there is still no consensus on the surgical approach. We herein report a patient with a giant thymoma that was

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successfully resected through a large right posterolateral thoracotomy.

Case Presentation

A 75-year-old man with no significant clinical history was referred to our centre with a six-month history of dyspnea. At admission, the patient's oxygen saturation was 91 % and he was spontaneously breathing (28 breaths/min). He had heart rate 120/min, hemodynamic stable and normal level of consciousness (Glasgow score = 15). Physical chest examination revealed a breath sounds decreased in the right side. He had not suffered any symptoms indicating myasthenia, muscle tone in both upper limbs were normal. Abdominal and other examinations were unremarkable. Blood samples demonstrated only hypoglycaemia and the other laboratory data were normal. The preoperative check-ups were normal and laboratory examination showed normal serum levels of acetylcholine receptor (AChR) antibody. Computed tomography (CT) revealed a sizable well-delineated retrosternal soft tissue mass (13.78x13.85 cm) that occupied the majority of the right hemithorax and compressed the right upper bronchus with atelectasis lung (Figure 1). This mass showed inhomogeneous enhancement after contrast administration. The origin of the mass was uncertain. F18-fluorodeoxyglucose positron emission tomography (FDG-PET) showed FDG uptake with a maximum standardized uptake value of 4, 2 (Figure 2).



Figure 1: Computed tomography (CT) revealed (A) a sizable well delineated retrosternal soft tissue mass (13.78x13.85 cm) that occupied the majority of the right hemithorax and (B) compressed the right upper bronchus with atelectatis lung. This mass showed inhomogeneous enhancement after contrast administration.

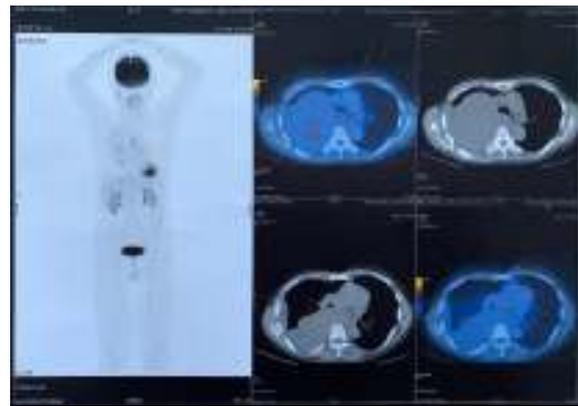


Figure 2: F18-fluorodeoxyglucose positron emission tomography (FDG-PET) showed FDG uptake with a maximum standardized uptake value of 4, 2.

Spirometry results showed forced vital capacity (FVC) of 3.71 l (106%) and forced expiratory volume in 1 second (FEV1) of 2.43 l (104%). An intra-pleural solitary fibrous tumour was suspected. Our patient underwent CT guided percutaneous biopsy of the mass but pathology finding was free of disease. Surgical resection was scheduled based on symptomatology and the radiological findings. Surgical approach to the mass was accomplished with posterolateral thoracotomy. On opening, the tumour occupied approximately half of the right pleural cavity. All surrounding tissue adhesions were removed. A large encapsulated mass was found adhering to the mediastinum pleura and pericardium. Resection of the mediastinal tumour was laborious due to the lack of space between the mass and pleural cavity. The mediastinum pleura was opened and the mass was completely removed with a wide mediastinum pleural excision. The weight of the tumour was 3500g. On macroscopic examination, it was a well-encapsulated mass with a boss elated and hyper vascular surface (Figure 3). The definitive histopathological findings were compatible with thymoma AB (also known as mixed thymoma) according to the World Health Organization classification and diagnosis of Masaoka stage I was made (T1N0M0 according to IASLC/ITMIG). Progress after surgery was uneventful and the patient was discharged on postoperative day 7. He was referred to the oncology-thorax staff for

follow-up. Twenty-four months after surgery, the patient is asymptomatic and he remains free of recurrence.

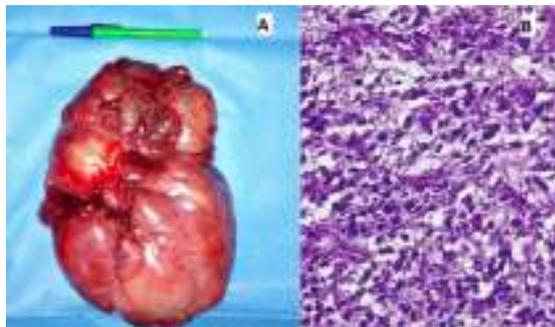


Figure 3: (A) Gross pathologic finding of the tumor (specimen): an encapsulated mass measuring 17 cm × 16 cm × 10 cm after it was removed and weighed 3500 g, tumor revealed a light brown color, an internal lobulated multicystic structure (B): Histological findings of the tumor. The tumor was composed of a lymphocyte associated area and was diagnosed as a WHO type AB thymoma without capsular invasion (Masaoka stage I)

Discussion

Thymomas develop in the thymus and are commonly defined as anterior mediastinum tumours characterized by epithelial cells and lymphocytes (7-8). This report presents a particularly case of posterior mediastinum ectopic thymoma that presented as a giant tumor intra-thoracic. The overall incidence of thymoma is rare, which approximately 0.15 cases per 100,000 of the population per year and account for 20–30% of mediastinal tumours in adults (3, 7). However, giant thymomas in adults are even rarer and comprise about 1% of all mediastinal tumours (1,7) and ectopic thymomas affecting the neck, pleura or the lung account for only 4% of all thymomas (5,9). Most patients are between 40-60 years of age, and there is slight male predominance (2, 7). The advanced age of our patient's diagnosis reflects an indolent and slow growth pattern of thymomas.

Aetiology and pathogenesis

There is no consensus on the pathogenesis of thymomas and the aetiology is not clear. However, various specific autoimmune syndromes, including particularly myasthenia gravis (MG), red cell aplasia, dermatomyositis, systemic lupus erythematosus, Cushing syndrome and syndrome of inappropriate antidiuretic hormone secretion, are present in 5% of

cases in the literature (2, 8). For our patient, the patient had no other disease history, but the disappearance of symptomatic hypoglycemia after complete surgical resection of thymoma suggests an association with autoimmune syndrome.

Clinical presentations:

Thymomas are generally considered to have an indolent and slow growth profile. Although, in rare situations, they should be considered as malignant due to their potential for local invasion and systemic metastasis (7-8). Approximately 70% of patients are asymptomatic and thymoma is generally discovered suddenly on chest imaging performed as part of a routine check-up at autopsy (9, 10). The patient may have nonspecific respiratory symptoms related to mediastinal compression (cough, chest pain, superior vena cava syndrome, dysphagia and dysphonia). Although, some authors described a life-threatening clinical presentation such as haemothorax leading to haemorrhagic shock after spontaneous rupture of a giant thymoma (10) and hemorrhagic pericardial effusion with cardiac tamponade due to malignant giant thymoma (11). The CT scan, the most informative of the imaging procedures, can accurately predict possibly involvement of other mediastinal structures based on the presence or absence of fat planes between the thymoma and surrounding structures, as reported by Keen and Libshitz (12) and guide the practitioner in the tumour removal. Other diagnostic modalities include MRI and PET scans, which can assess the possibility of successful resection (13), determine the stage of the disease, and exclude ectopic localization of thymoma (2).

Several authors reported a significant correlation between the histological subtype of thymomas and FDG-PET accumulation (9). Who type A, AB and B1 tumors have significantly lower SUV as in our observation than that of the other types of tumors (3,9). In addition, the SUV max had strong correlation with Masaoka stages (14). Our case confirms this hypothesis, in our case, the SUV max of the tumor was not very elevated in relation to its size. The tumor was diagnosed to be thymoma AB according to the WHO classification and Masaoka Staging System I.

Diagnosis

In our observation, the tumor protruded into the pleural cavity. It should be differentiated from solitary fibrous tumours of pleura before a large intrathoracic mass associated with hypoglycemia without FDG fixation on the mass and posterior mediastinal mass including neurogenic tumors and sarcoma. The only possibility to obtain a definitive diagnosis is through pathological diagnosis. The biopsy specimen can be obtained by CT guided core biopsy, mediastinoscopy, VATS or open surgical biopsy according to the tumor site and the available technical equipment for diagnosis (2). However, in our case, the tru cut biopsy was not contributive because of some of its disadvantage (small tissue of the tumor, bleeding). In our case, the definitive anatomic-pathological was obtained from the surgical specimen and classified as benign tumours due to the absence of both macroscopic and microscopic capsular invasions.

Surgical approach

As the available literature indicates, adjuvant chemoradiotherapy has no benefit in fully resected stage I thymomas (6). Complete surgical excision is the preferred treatment approach whenever technically feasible. However, randomized controlled trials comparing various surgical approaches to intra-thoracic giant thymoma are lacking in the literature due to the rarity of this particular clinical presentation. For the minimally-invasive approach in the thymectomy; we do not recommend also the use of video-assisted thoracic surgery (VATS) due to the lack of scientific validity and evidence for its applicability in giant thymoma intra-thoracic surgery. Usually, the median sternotomy is considered as the gold standard method for performing thymectomy when thymomas have a normal size (3, 6). Unfortunately, with a sternotomy, the operative exposure of the diaphragm, posterior chest wall, left lower lobe and posterior mediastinum, as in our case, is limited in comparison to the posterolateral thoracotomy approach (15). M. Incarbone et al (15), in a letter to the editor, responded to the Gotte and Bilfinger article and confirmed that even the hemi-clamshell approach (trap door incision) does not permit good exposure of the posterior

chest wall, and a posterolateral thoracotomy may be performed to provide an adequate chest wall resection. In the literature, most authors who advocate the thymectomy on open approaches in the supine position (sternotomy, hemi-clamshell or clamshell, anterolateral thoracotomy) have described either an anterior mediastinal mass (1, 3, 8, 13, 15) or in a hemorrhagic situation with hemodynamic instability (10) justifying their surgical approach. Similarly, Yasushi Shintani et al. (16) report two observations of huge thymoma requiring a posterolateral thoracotomy for thymectomy after unsuccessful initial surgical approach with a median sternotomy. When the thymoma is atypically located, one has to differ from the standard due to bad access. The surgeon should always be well prepared and be aware that the incision must be extended or modified if additional exposure is required and posterolateral thoracotomy can be used for practically all pulmonary, mediastinal or parietal resections. In this observation, we did not choose the median approach based on the posterior localization of the giant thymoma. Well, in our case, a posterolateral thoracotomy seemed to appear a reasonable approach to permit safe dissection. This approach made the complete thymectomy possible to be performed in a single step procedure without any complications in our case. Posterolateral thoracotomy is the historic gold standard of thoracic incisions, offering an excellent exposure for most general thoracic procedures. It provides an excellent access to decrease the risk of uncontrollable haemorrhage, damage to the recurrent laryngeal nerve and incomplete resection of the thymoma. As in our case, thymectomy for mediastinum giant thymomas was conducted through posterolateral thoracotomy in two case reports without complications postoperatively (5). In spite of our patient's old age, we have not encountered cardio-respiratory complications as described in the literature (18) related to posterolateral thoracotomy in the postoperative period. In our opinion, the most important criteria for selecting patients who require posterolateral thoracotomy incision are based on current preoperative staging (tumor size, topography and extension, involved

mediastinal structures (<50 % tracheobronchial obstruction) and resection feasibility of tumor observed on CT scan) like the one we reported. Tomoyoshi Takenaka et al. (9) report that a large tumor size is a poor prognostic factor and the resected giant thymomas tended to be low-grade thymomas (9). Contrasting to our expertise, correlating with Masaoka's classification, we noticed that asymptomatic giant thymomas with a suitable CT resectability criterion have often a favourable outcome.

Conclusion

This case report presented a very rare ectopic giant intra-thoracic thymoma. Surgical resection can be considered for any size of tumour if curative resection is possible. The choice of approach is based on the tumour topography, the assessment of tumour extension and extirpation, the surgeon's experience and the symptomatology presented by the patients. However, we do not recommend our posterolateral approach for patients with compressive syndrome.

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