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Case Presentation

An Unusual Presentation of Massive Intrathoracic Schwannoma with Concomittant Pleural Tuberculosis: About a Case and Review of the Literature

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Introduction

Schwannomas are the most common neurogenic tumor of the thorax, and may involve any thoracic nerve [1]. The case of a 49-year old male patient with a massive intrathoracic schwannoma is reported. A thoracotomy resection of the tumor was performed. The presence of adhesions of the tumor to the chest wall pleura created a suspicion of malignancy. This article's aim is to draw attention to this infrequent pathology with concomitant tuberculosis and to discuss different aspects regarding diagnosis and treatment of pleura schwannome.

Observation

A 49-year-old male with no significant clinical history, presented to the emergency department with a 1-month history of cough productive of white sputum, Haemoptysis, unexplained loss of weight and associated with progressive shortness of breath on exertion, which had persisted despite courses of co-amoxiclav. On examination, his vital parameters (temperature; oxygen saturation; blood pressure; respiratory rate; pulse rate) were all within the normal range for a man of his age. Physical examination revealed dull percussion note on right mid chest wall with diminished breath sound with occasional expiratory wheezing at the auscultation of the chest, and syndrome (facial swelling, neck distension). Lymph nodes examination is unremarkable and the rest of the examination was unrevealing. Standard chest X-ray showed a large, well-defined mass in the the right hemithorax, around 18cm in diameter, associated with displacement of the trachea to the opposite side (Figure 1). Initially, this was thought to be a bronchogenic cyst or un lymphoma process. Computed Tomography (CT) scan of thorax demonstrated a large (20x15x13cm) well-circumscribed, heterogeneously enhancing rounded mass in the right hemithorax to whose density was mostly similar to fluid, associated with a solid component in the uppermost part. The mediastinal structures, particularly the trachea and right bronchus, were noted to be compressed and displaced to the left side by this lesion (Figure 2A,B). Examination of the sputum smear samples to identify AFB was positive. Fine needle aspiration was performed, but the specimen was insufficient for diagnosis. Results from percutanous biopsy confirmed a pleural schwannome and has eliminated a lymphoma process. Surgery was planned after ruling out an an intraspinal component of tumor. Complete excision of the well-encapsulated mass was achieved through the right posterolateral thoracotomy (Figure 3A). A 20cm tumor was found in the right chest cavity, adhering closely to the parietal pleura and superior vena cava, which was severely dilated. The mass was found to be benign schwannoma without malignant components and the anatomopathologic examination of pariétal pleura retained it to be tubercular (Figure 3B). The postoperative course of the patient was uneventful. The respiratory and vascular symptoms and facial edema resolved immediately after the surgery. The patient returned to her normal life and recovered gradually through regular chest physiotherapy. He has been followed-up for 1 year with no evidence of recurrence.

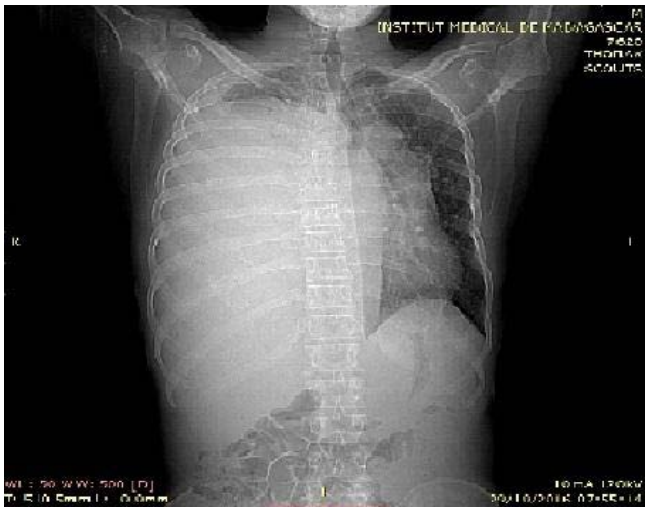

Figure 1: CT topogram shows a large, well-defined mass in the right hemithorax, around 18cm in diameter, associated with displacement of the trachea to the opposite side.

Antitubercular treatment according to the National Tuberculosis Control Program (NTP) standards in our country was started and he

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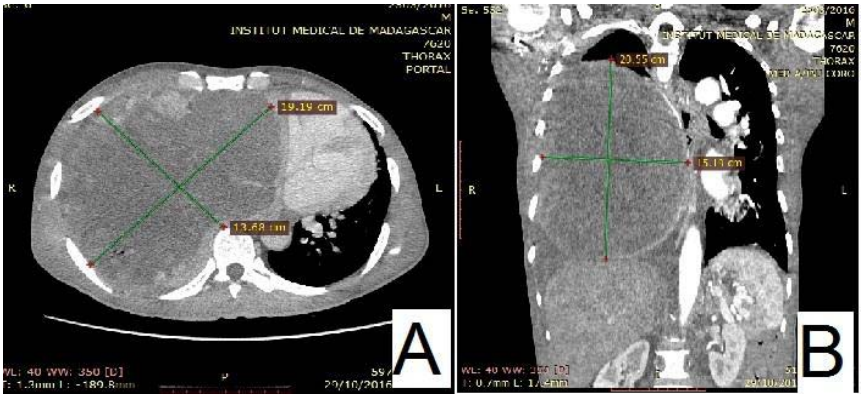

Figure 2: A) Axial Contrast-enhanced computed tomography image of the chest demonstrating well-defined large soft tissue density mass showing peripheral enhancement, **B)** Coronal Contrast-enhanced computed tomography demonstrating he mediastinal structures, particularly the superior vena cava, trachea and right bronchus, were noted to be compressed and displaced to the left side by this lesion.

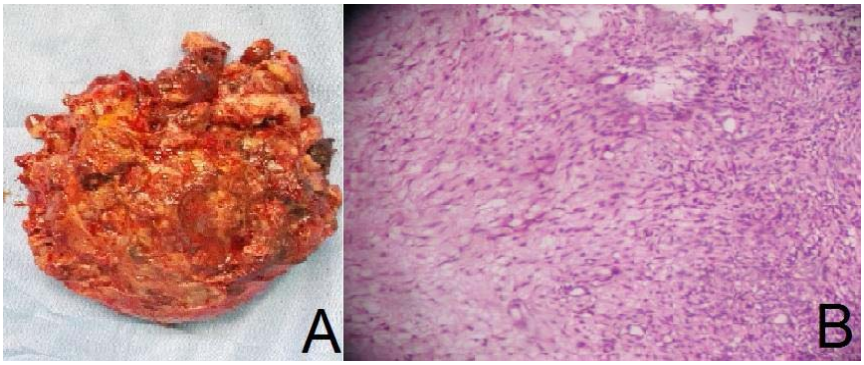

Figure 3: A) A mass around 20cm across was completely excised from the right pleural space, **B)** Histopathologic examination: Microscopic picture (hematoxylin and eosin staining, x400) showing Verocay body, formed by alternating rows of palisading nuclei and intervening nuclei-free stroma. Degenerative changes including hemorrhage, and hyalinization of blood vessels.

performed well and was declared cured and is in our regular follow-up.

Discussion

The primary pleural schwannomas are usually benign [2], yet malignancy has been reported in some cases [3]. They account for 1-2% of all thoracic tumors [3].

They can occur at all ages, though they are uncommon in children. Both genders are equally affected, predominantly in the third or fourth decades of life [4,5].

Shin-ichi Takeda *et al*., analyzed the records of 146 patients with intrathoracic neurogenic tumors who were treated over the past 50 years. 20.5% of the neoplasms were malignant, occurring predominantly in the first 5 years of life. This study concluded that the age of patient seemed to be the most important clinical parameter for distinguishing between histological type and rate of malignancy for neurogenic tumors [6].

Intrathoracic neurogenic tumors occurs predominantly (90%) in the mediastinum and about 10% originate peripherally nerve fibers from the intercostal nerves or pleural nerves [7,8]. Schwannomas (also known as neurilemmomas or neurinomas) are highly vascular nerve sheath benign tumors that arise from the neural crest-derived schwann cells [9].

The risk of malignancy in a nerve sheath tumor is very small (2-5%) frequently associated with von Recklinghausen syndrome (4% of cases), other neurofibromatoses or radiation exposure when the risk increases to 10-20% [7,10,11].

Association of schwannoma with concomitant tuberculosis is a very rare entity. A literature review was performed *via* PubMed, using the MeSH terms 'Benign schwannoma' and 'tuberculosis' and identify two similar case reports reporting cases of intrathoracic schwannoma associated with tuberculosis [7,12]. However, we found only one case of similar size to our observation reported by David D. Odell *et al*., [10].

Up to 80% of cases are asymptomatic, and diagnosis in young and middle-aged adults is generally fortuitous frequently appearing as incidental radiological findings [9,12,13]. In our case, the presentation with superior vena cava syndrome is rare, making our patient atypical reflecting clinical mutility and insidious evolution which can spread over several years and explains the occurrence of the giant forms of this type of tumor. These symptoms are due to compression of neighboring structures [14].

A variety of diagnostic imaging modalities can be utilized to delineate pleural schwannomas. Generally, chest wall neurilemmomas

are evaluated by radiologic examination for planning the surgical approach, including CT and Magnetic Resonance Imaging (MRI).

CT scan remains the diagnostic imaging modality of choice for these neoplasms. CT scan can outline the size, number, and exact location of the lesions. CT scan can also demonstrate cystic and/or solid components of the tumor. Unenhanced CT demonstrates well-marginated soft-tissue mass with possible surrounding fat attenuation ("split fat" sign), may contain areas of low attenuation corresponding to fat or cystic degeneration, calcifiation may be seen in 10% of tumors. Contrast-Enhanced Computed Tomography (CECT) shows a homogeneous enhancement in small tumors and heterogeneous enhancement in larger tumors [15].

Malignant pleural schwannomas have similar features on CT scan; however, they are usually associated with the presence of pleural nodules, pleural effusions, and metastatic pulmonary nodules [15].

In the case of our patient, the presence of dense adhesions and increased vascularity were in favor of malignant tumor component. This could have been secondary to the extension of inflammation response and fibrous reaction from the pleura tuberculous on to the capsule of the tumor and mimicked the features of malignancy.

MRI is the preferred method for the assessment of peripheral nerve sheath tumors [5]. Vessels in neurinomas are usually prominent and their rich vascular supply is reflected in the often intense enhancement of these tumors on imaging studies [5]. Pleural schwannomas are typically hyper/isointense on T1-attenuated images and heterogeneously hyperintense on T2-attenuated images [3,15]. Particularly, MRI should be performed in patients with suspicious posterior mediastinal neurogenic tumors to exclude intra-spinal tumor extension [16].

Thus, preoperative radiologic evaluation alone is enough without needle aspiration biopsy [16].

For giant shapes like our case, intra-thoracic schwannomas should be included in the differential diagnosis of posterior mediastinal masses. They include sympathetic ganglion tumours (neuroblastomas, ganglioneuroblastomas, ganglioneuromas) and paragangliomas (chemodectomas and pheochromocytomas), lymphadenopathy, enteric cysts, bronchogenic cysts, oesophageal tumours, aneurysms and paraspinous abscess [12].

In the absence of MRI, as in our observation, a preoperative percutanous biopsy of the tumor, when feasible, is necessary in order to lead to the definitive diagnosis of benign tumor and to avoid overtreatment [5].

Microscopically, Antoni A and Antoni B areas are revealed in the majority of pleural schwannoma cases. Antoni A represents areas of hypercellularity with verocay bodies. Antoni B areas of myxoidhypocellularity exhibit degenerating changes (i.e. cyst formation, haemorrhage, calcification, xanthomatous infliltration and hyalinisation) [3]. Immunohistochemically, pleural schwannomas typically stain diffusely and strongly positive for S100 protein. On the other hand, they stain negatively for CD-15, CD-30, CD-34, and pan-cytokeratin [3,15].

The standard care of management of pleural schwannomas is primarily surgical resection [14,17].

In the literature, the authors are unanimous that compared to thoracotomy, VATS has better outcomes in terms of duration of surgical procedure, amount of blood loss and length of drainage time. Complete resection can be achieved with low morbidity (smaller incisions, thereby resulting in less pain, fewer lung complications, shorter hospital stays, more rapid recovery (return to normal activities) and less aesthetic damage [5,10,18,19].

A case series literature of 60 consecutive patients with a neurogenic tumor of the chest during 30-year experience concluded that VATS seems feasible as the approach for the thoracic neurogenic tumor since it is less invasive and provides an appropriate view for the operation [20].

A combined Chinese experience reports a thoracoscopic removal successfully of intrathoracic neurogenic tumors on average 3.5cm in greatest diameter [21].

However the video-assisted thoracoscopy may be contraindicated in tumors larger than 6cm [14,22].

David D. Odell *et al*., report a case of intrathoracic schwannoma measuring 27cm in diameter and conclude that the clamshell incision, compared with the median sternotomy offers excellent exposure of the pulmonary hilum, which is especially helpful in the setting of centrally located tumors [10].

In our case, we decided to perform an approach through a right posterior thoracotomy in the 6th intercostal space due to the size of the lesion and a possibility of an intraspinal component of tumor necessitates a combined or staged approach, involving both neurosurgical and thoracic procedures.

Conclusion

This case report highlights the need to be aware of the potential coexistence of tuberculosis and intra-thoracic schwannoma hence the necessity of a systematic identification of an AFB on a sputum smear samples before any thoracic surgery in our country. Conversely, any positivity of AFB test should not conclude for any intrathoracic mass to a tuberculoma to be a tuberculoma without confirmation by examination of tissue through a biopsy or surgical excision.

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