Thoracoscopic Resection of a Solitary Pulmonary Lymphangioma: Report of a Case

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Abstract: We present herein the rare case of a young man who was found to have a solitary tumor in the right upper lobe of his lung by a routine chest X-ray. The tumor was removed by thoracoscopic surgery, and pathological examinations confirmed the diagnosis of a primary lymphangioma of the lung. A brief review of the available literature on this extremely rare type of benign tumor follows the case report.

Key Words: lymphangioma, lung tumor, thoracoscopic surgery

Introduction

Pulmonary lymphangioma is an extremely rare type of benign tumor and to our knowledge, only seven cases have been documented in the world literature. We report herein the case of a 21-year-old man in whom an asymptomatic pulmonary lymphangioma was incidentally found on a routine chest X-ray.

Case Report

A 21-year-old man was referred to our hospital for further investigations of an abnormal shadow found on a routine chest X-ray. The chest X-ray film showed a solitary, round mass, 2.0 cm x 1.5 cm, in the right upper lung field. Computed tomography (CT) scan revealed a round lesion with uniform density in the right superior dorsal segment (Fig. 1). Physical examination proved unremarkable and the laboratory results, including those of the tumor markers, were also within normal limits. Retrospectively, a fluoro-photograph taken 1 year earlier showed a similar shadow; however, no definite shadow could be detected on the film from 2 years earlier. Fiberbronchoscopy showed no abnormality, and a transbronchial lung biopsy (TBLB) could not be taken. Thus, partial resection of the lung, as an excisional biopsy, was performed thoracoscopically. A purple-colored cystic mass with a smooth surface was observed by imaged thoracoscopic surgery. Intraoperatively, the frozen section of the specimen provided a provisional diagnosis of a hemangioma or lymphangioma. The patient had an uneventful postoperative course and was discharged 5 days after surgery. He has remained well and no signs of recurrence have been detected during 1 year of follow-up.

Pathology

Histological examination showed that the lesion consisted of irregular shaped cystic cavities with thin walls.
These cystic cavities contained weakly eosinophilic fluid-like lymph and a small number of erythrocytes. The cyst walls were lined by flattened endothelium-like cells and contained some smooth muscle. The flattened cells were immunohistologically positive for factor-VIII and Ulex Europaeus Lectin type 1 (UEA-1), which is more sensitive than factor-VIII for recognizing endothelial cells (Fig. 2B). Both the epithelial membrane antigen (EMA) and cytokeratin were negative, and elastica van Gieson staining revealed no internal elastic laminae in the cyst walls. Overall, this lesion was a multi-cystic tumor consisting of endothelial cells, some smooth muscle, and lymph-like fluid, with no elastic lamina, confirming a diagnosis of lymphangioma.

Discussion

Lymphangiomas usually appear in the head, neck, axilla, and occasionally in the mediastinum or retroperitoneum, of newborns, infants, and children. It is not certain whether they represent congenital malformations, hamartomas, or true neoplasms,1,2 but they have been classified into three types, namely: capillary, cavernous, or cystic.1 Our patient was classified as having a cystic-type tumor because of the large size of the lumens.

Primary lymphangioma of the lung is extremely rare, with only seven other cases having been reported in the literature including five men and two women ranging in age from neonates3,4 up to 66 years.2 The lesions were reported in the right upper lobe (2),2 right hilus (1),5 right lower lobe (1),5 lingula (2),7,8 and left lower lobe (2),3,4 and no predominance of any region was found among the total of eight patients.

In our patient, retrospective examination of past X-rays suggested that this mass may have grown within 1 year. The histories of two other cases5,6 indicated that the growth of the mass led to symptoms such as increased fatigability with chest heaviness and hemoptysis. However, it is very difficult to establish a preoperative diagnosis of pulmonary lymphangioma. We believe that resection should be performed to alleviate symptoms if a lung tumor is suspected to be lymphangioma.

In summary, an asymptomatic tumor was discovered in a young man on a routine chest X-ray. The tumor was resected using a minimally invasive procedure, and postoperative histological examination confirmed a diagnosis of primary pulmonary lymphangioma.

References