Solitary Pulmonary Lymphangioma in an Adult

- A Brief Case Report -

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Lymphangioma can occur in any region of the body in which there is lymphatic drainage. Abnormal proliferation of lymphatic vessels in the lung is seen in lymphangiomatosis and solitary lymphangioma. Among these maladies, solitary pulmonary lymphangioma is extremely rare. Herein, we report on an unique case of solitary pulmonary lymphangioma in a 50-year-old man.

CASE REPORT

A 50-year-old Korean man present with a left pleural based ovoid lesion (6 cm in diameter) on a routine health examination. He was a smoker of 30 pack-years, but he had no previous history of pulmonary disease or trauma. He did not complain of any specific respiratory symptoms. On computerized tomography (CT), there was a well-circumscribed, ovoid lesion with fluid attenuation at the posterobasal segment of the left lower lobe (Fig. 1A). This lesion was suspected to be attached near the pleura rather than to the pulmonary parenchyma. The magnetic resonance imaging (MRI) showed intermediate to low signal intensity on the T2-weighted image and no significant enhancement was seen on the T1-weighted image. The possibility of loculated pleural effusion or a localized pleural mass was proposed based on the radiologic findings. Video-assisted surgical resection was performed. Although the lesion showed focal adhesion to the pleura, the lesion showed continuity from the pulmonary parenchyma on intraoperative inspection. No feeding vessel was identified. The wedge-resected lung specimen showed an ill-defined lesion with multiple microscopic cysts (Fig. 1B, C). Each cyst was lined by a mono-layer of endothelial cells. The cyst walls had loose stroma, and this formed a “common wall” with the nearby cyst. Lymphoid aggregates in the lumen revealed that the channel came from the lymphatics. Immunohistochemical stain for D2-40 (podoplanin, 1:130, DAKO, Glostrup, Denmark) shows positivity in the endothelial cells of the tumor (Fig. 1D).

He was discharged 4 days after the surgery. There has been no evidence of recurrence during 9 months of follow-up.
Localized pulmonary lymphangioma is rare, and it has been found in patients with a wide age range (6 months to 67 years). Especially, different clinical presentations in adults and children have been described.\(^1\) In infants and neonates, pulmonary lymphangiomas often present with pneumothorax and respiratory distress. However, adult patients present with an asymptomatic lung lesion that requires differentiation from primary lung cancer. Two cases of cystic lymphangioma in pediatric patients have been reported in Korea, and both of them also presented with respiratory distress.\(^2,3\) Our case is the first report of solitary lymphangioma in a Korean adult patient, and this case also showed a clinical presentation that was similar to the previously reported cases.

Histologically, most of the solitary lymphangiomas of lung that have been reported in the literature showed features compatible with cystic lymphangioma.\(^1-5\) However, our case showed histologic similarity to cavernous lymphangioma. The mass was mainly composed of microcysts rather than grossly visible large cysts, and the cysts contained thick, but loose stroma.

Making the preoperative diagnosis of solitary pulmonary lymphangioma in adult is difficult. The most common radiologic finding is a solitary, cystic, peripheral lung lesion. However, atypical findings like spiculation and calcification have been also reported and one pediatric case that presented with a large mass has also been reported.\(^1,6\) Therefore, making the radiologic diagnosis of solitary pulmonary lymphangioma is not always possible. Awareness of its occurrence in adults is necessary.

**REFERENCES**

1. Wilson C, Askin FB, Heitmiller RF. Solitary pulmonary lymphan-