Placental Transmogrification of the Lung - A Brief Case Report -

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Fax: 02-3410-0025 E-mail: hanjho@skku.edu Placental transmogrification (PT) is an unusual condition in which the alveoli develop a peculiar villous configuration that resembles the placental villi. We report a rare case of pulmonary PT in a 46-year-old man who presented with multiple cystic lesions and nodules on radiography. The patient was treated with a surgical excision. The cut surface of the lung lesion had a villous spongiform manifestation with a partly yellow granular appearance. Microscopically, multiple papillary cores mimicking the villous structures of the placenta were observed within the bullous airspaces. These papillary cores contained many vascular structures, lymphoid aggregates, interstitial clear cells, mature fat and dystrophic calcification. This case was solitary and not associated with other pulmonary or systemic diseases. The etiology is unknown, and further studies will be needed to understand the pathogenesis of the lesion.

Key Words: Pulmonary emphysema; Lung disease; Placental transmogrification

Pulmonary placental transmogrification (PT) is a rare cystic lesion that was first described in 1979. The condition is characterized by the formation of placental villi-like papillary structures in the lung parenchyma. Despite the morphological similarity to the placental villi on lower magnification, this lesion does not contain any biological components of the placenta. It is instead composed of epithelial cells, proliferating vessels, inflammatory cells and fat. Several hypotheses regarding its pathogenesis have been suggested, but none are based on clear evidences. We report a case of placental transmogrification of the lung.

CASE REPORT

A 46-year-old man presented with a small nodular lesion on

chest radiography performed during a routine health examination. He did not complain of any respiratory or systemic symptoms. His prior medical and familial history was unremarkable except for a 20-year-pack smoking history. He began taking empirical anti-tuberculosis medication based on the radiologic findings, but there was no change in his chest lesion. Chest CT revealed multiple cystic lesions and nodules in the right middle lobe of his lung that suggested bronchopneumonia with an underlying congenital cystic adenomatoid malformation (CCAM) (Fig. 1). A right middle lobe lobectomy was performed for histological conformation of nodules and treatment of cystic lesion to reduce dead air-spaces and prevent complication such as pneumothorax and secondary infection. On the macroscopic examination, the resected lobe showed an ill-defined cystic lesion, measuring $7\times4\times4$ cm, which was filled with grape-like gelati-

nous and fatty material, and also showed partially collapsed central emphysema (Fig. 2A). Subpleural bullae were also pre-

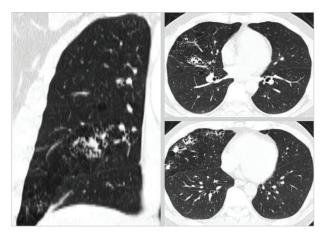


Fig. 1. Computed tomography shows multiple cystic lesions and bronchopneumonia-like consolidation in the right middle lobe of lung.

sent. The microscopic examination revealed papillary structures, resembling placental villi, within the hyperinflated cystic spaces (Fig. 2B, D). These papillary structures were lined with mildly hyperplastic pneumocytes that were positive for TTF-1 (1:50, DAKO, Glostrup, Denmark). The cores of the papillae were sometimes edematous or densely fibrotic.

Of the constituents of central cores, characteristic collections of bland, round to ovoid clear interstitial cells were observed. These cells were positive for CD10 (1:100, Novocastra, Newcastle, UK), but negative for smooth muscle actin (1:100, DAKO, Glostrup, Denmark), S-100 protein (1:2,000, DAKO), and HMB-45 (1:40, DAKO). Focal aggregates of mature fat and smooth muscle bundles as well as small, thin walled vessels, which were positive for CD31 (1:40, DAKO) and D2-40 (1:130, DAKO), were also observed.

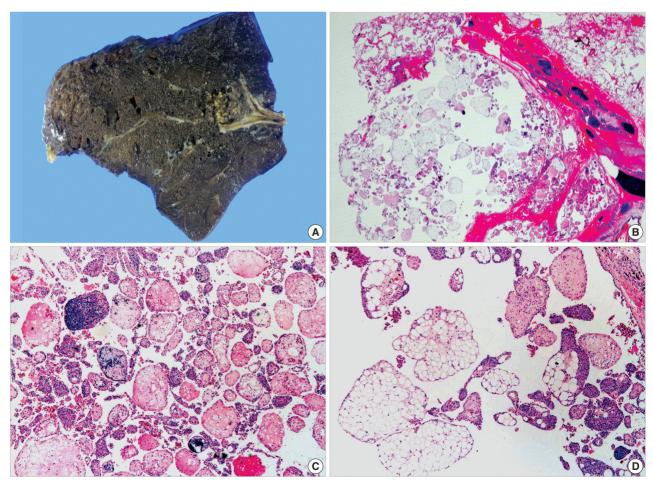


Fig. 2. (A) The cut surface of resected right middle lobe shows an ill-defined cystic lesion, which is filled with grape-like gelatinous and fatty material. (B, C) The microscopic examination shows papillary structures, resembling placental villi, within hyperinflated, cystic spaces. (D) The cores of the papillae show adipose tissue and fibrosis.

DISCUSSION

PT is a rare but unique lesion of the lung that shows two clinical presentations. As in our case, the condition more commonly presents as a cystic lung lesion that is frequently accompanied with other pulmonary cystic lesions. However, this unique lesion can also occur in conjunction with non-cystic lung lesions such as fibrochondromatous hamartoma and pulmonary lipomatosis, and can present as a solitary pulmonary nodule on routine chest images. Therefore, PT can present on radiologic studies as a cystic or emphysematous lesion as well as a mass-like lesion.

PT is not only associated with cystic or emphysematous lesions, but also with hamartomatous conditions. Hence, there is some controversy regarding its pathogenesis i.e. whether this lesion is a reactive change or neoplastic one. Recently, Cavazza et al. focused on aggregates of interstitial clear cells in central cores from their two cases. They described the immunophenotype of these clear cells for the first time; they were positive for CD10, but negative for cytokeratin, actin, desmin, and S-100 protein. Based on these results, they emphasized the immature mesenchymal phenotype of these clear cells. The current case also showed the same immunophenotype. However, we could not conclude that these cells had the immature mesenchymal phenotype because the interstitial cells in the surrounding emphysematous parenchyma also showed the same immunophenotype. In addition, they also reported some microsatellite alterations, which suggested a neoplastic character of PT. However, microsatellite alterations were also reported in the non-neoplastic lesions of the lung. Moreover, it cannot be concluded that PT is a neoplastic lesion based on the results of immunohistochemical and molecular studies.⁶ Therefore, the pathogenesis of PT is still unclear and further studies may provide more information on its pathogenesis.

In spite of the fact that not so many cases have been reported in literatures, all reported cases have done well after surgery, with a follow-up ranging from 2 months to 8 years and complete surgical excision of cystic lesion has been curative. ^{1,2,4,5} Currently, the present case is still alive and well 15 months after surgery.

In conclusion, PT is a unique pulmonary lesion showing morphological similarities to the placental villi on microscopic examination. It can be associated with a cystic emphysematous lesion and other hamartomatous conditions. Hence this unique lesion can have a variety of clinical presentation. Further studies will be needed to clarify the nature and pathogenesis of PT.

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