

Solitary Atypical Adenomatous Hyperplasia in a 12-Year-Old Girl

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Atypical adenomatous hyperplasia is a premalignant lesion reflecting a focal proliferation of atypical cells. These lesions are usually observed as incidental findings in lungs that have been resected due to other conditions, such as lung cancer. We report the youngest case of atypical adenomatous hyperplasia on record in a 12-year-old girl. In this patient, the lesion was found in association with pneumothorax.

Key words: 1. Lung pathology
2. Pneumothorax
3. Thoracoscopy
4. Neoplasm

CASE REPORT

A 12-year-old girl visited the emergency medical care center of Busan Paik Hospital due to chest pain and dyspnea. She had no history of illness and no family history of lung disease. A chest X-ray revealed a large left pneumothorax (Fig. 1A). After the insertion of a chest tube in the left thoracic cavity, she was referred to the department of thoracic and cardiovascular surgery of Busan Paik Hospital for further treatment. After two days, she underwent high-resolution computed tomography (HRCT) to identify the presence of other pulmonary lesions. HRCT demonstrated a 1.5-cm bulla with passive atelectasis (Fig. 1B, C). Due to the high likelihood of recurrence, we decided to resect the lung lesion using video-assisted thoracoscopic surgery. Intraoperatively, a large bulla measuring approximately 1.5 cm with multiple small bullae surrounding it was found (Fig. 2). We resected the bullae using a 60 mm-4.8 mm stapler. After inserting the chest tube (16 Fr), we finished the operation. She was dis-

charged on the fourth postoperative day after removal of the chest tube.

The wedge resection specimen contained the bulla and atypical adenomatous hyperplasia (AAH), which was found incidentally (Fig. 3). It was located below the large bulla and measured 0.2 cm. The resection margin was not involved and the safety margin was 0.7 cm from the resection margin. The patient is still undergoing follow-up, and no evidence has been observed of any other ground-glass opacity lesions.

DISCUSSION

The World Health Organization defines AAH as a focal proliferation of atypical cells lining the involved alveoli or respiratory bronchioles. It is usually 0.5 cm or less and located in the peripheral area of the lung [1]. These lesions are mostly detected as incidental findings in lung tissue resected due to other problems, especially primary lung cancer. The incidence of AAH has been reported to be approximately 9%

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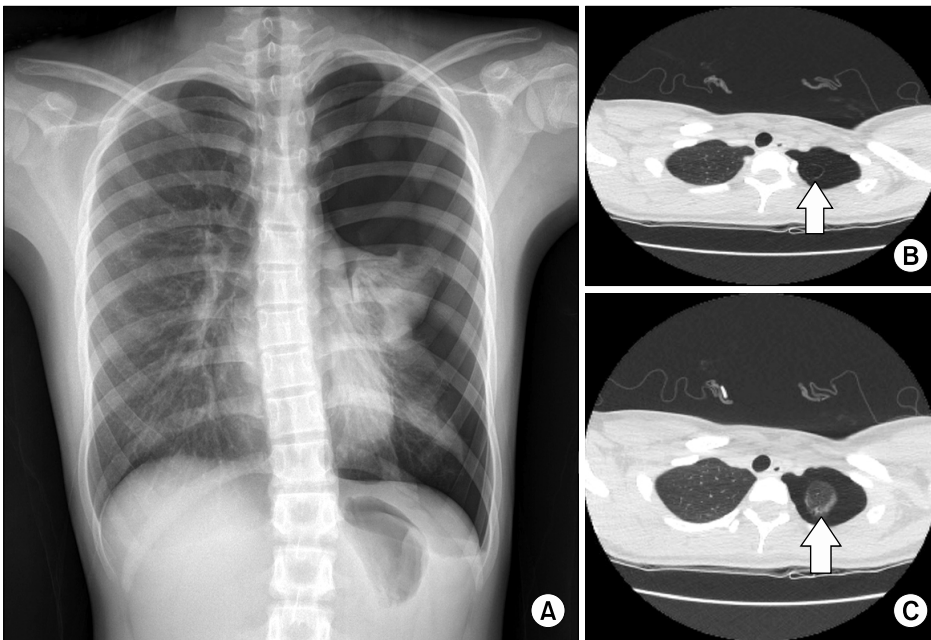


Fig. 1. (A) Initial chest X-ray and (B, C) high-resolution computed tomography. A large bulla and surrounding passive atelectasis (arrow) were found.



Fig. 2. Intraoperative view. A large bulla and small stratified blebs were found on the apex of the left upper lobe.

–21% in patients with primary lung cancer, while the incidence of AAH in patients without lung cancer has been reported to be 4%–10% [2].

Primary lung tumors are an extremely rare condition in children. Most nodules and masses are reactive lesions or congenital malformations, such as bronchogenic cysts, pulmonary sequestration, congenital pulmonary airway malformations, and congenital lobar overinflation. The incidence of primary lung tumors in children has not been established because most of the literature contains individual case reports

and diagnosis-specific case series. In the Texas Children’s Hospital, the total number of surgical pathology specimens was 227,655 during a 25-year period, and only 3,980 surgical specimens (1.7%) were designated as originating from the trachea, bronchus, or lung, including both biopsies and resections. The ratio of primary benign to primary malignant to metastatic malignant tumors was 1.4:1:11.6 [3]. Cohen and Kaschula [4] reported a ratio of primary tumors to metastatic tumors to non-neoplastic lesions of 1:5:60. Previously reported cases of AAH in children occurred in combination with a congenital cystic adenomatoid malformation or metastatic osteosarcoma of the lung [5,6]. However, the number of reported cases is very small and no cases of solitary AAH in a child have been reported. The youngest case of solitary AAH was a 17-year-old male in Japan, reported in 2003 [7].

Histologically, AAH is a focal proliferative parenchymal lesion, and the alveolar septa are lined by rounded low columnar cells with round to oval nuclei. Ciliated and mucin-producing cells are absent. Mitotic figures are rare and cellularity and atypia are variable. Mild to moderately atypical cells, hyperchromatic nuclei, and prominent nucleoli are usually seen, with an increased nuclear-cytoplasmic ratio. Intracellular inclusions are frequent and the alveolar walls are slightly thickened.

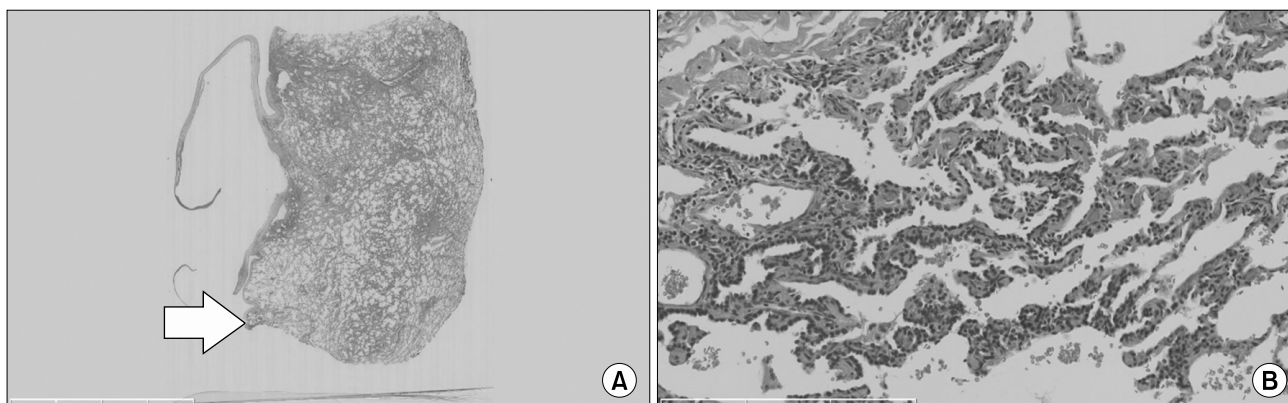


Fig. 3. (A) A low-power view revealed a bulla on the left side. The white arrow indicated a 0.2-cm atypical adenomatous lesion very near to the pleural vesicle (H&E, $\times 1$). (B) High magnification showed the proliferation of atypical pneumocytes with a high nucleus-to-cytoplasm ratio and hyperchromatic nuclei. No mitotic count was noted. The lesion was distinct from the normal lung parenchyma, shown on the right side of this picture (H&E, $\times 200$).

In addition, it is often difficult to differentiate AAH from adenocarcinoma *in situ* (AIS) only based on small biopsy specimens or frozen sections. However, some features are helpful in differentiating between AAH and AIS in specimens. At low power, AIS shows a monotonous cellular proliferation of homogenous columnar cells. Additionally, AIS exhibits a more abrupt transition to the adjacent lung parenchyma, and the presence of goblet cells is a strong indicator of AIS. However, AAH usually measures less than 5 mm in diameter and exhibits polymorphic cellular proliferations of variably sized and shaped cuboidal cells. In the periphery of the lesion, blending with normal alveolar lining cells occurs [2].

No guidelines exist for the surgical or medical treatment of patients without cancer who are incidentally found to have AAH, and no consensus exists regarding the risk of their lesions developing into invasive adenocarcinoma. In this case, the lesion had a sufficient resection margin relative to the nodule size, so we decided that no further resection was necessary.

Recently, imaging techniques have undergone continual improvement, resulting in the increased diagnosis of AAH. Furthermore, most authors have agreed that a genetic correlation is present between AAH and AIS, but this is also controversial [8]. If solitary AAH was not exceedingly rare in childhood, it would be easier to determine whether AAH slowly progresses into AIS or no relationship exists between

AAH and AIS. This case is the youngest patient with solitary AAH who has been reported, and continuous follow-up will be necessary. In the future, we expect appropriate treatment plans to be developed based on the accumulation of more data.

CONFLICT OF INTEREST

No potential conflict of interest relevant to this article was reported.

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