



Pseudobronchial crista-like change in children: A case report

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ABSTRACT

Pseudobronchial crista-like change is an unusual type of inflammatory granulation tissue hyperplasia in the endobronchial membrane caused by chronic retention of bronchial foreign bodies. Here, we report a case of pseudobronchial crista-like change in a 4-year-old boy who required admission for intermittent cough for >10 days and wheezing for 2 days. The main manifestation was persistent and non-healing lobar pneumonia. Initial electronic bronchoscopy showed cristae at the distal left main bronchus, which was misdiagnosed as bronchial opening stenosis. Repeat electronic bronchoscopy was performed after standard antibiotic treatment proved ineffective. Foreign bodies were observed at the opening of the basal branch of the left lower lobe. The left main bronchial cristae were clamped. The cristae appeared to be a pseudobronchial crista-like change caused by long-term retention of bronchial foreign bodies. After CT-confirmation of no abnormal blood supply at the cristae, the bronchial foreign bodies were removed, and the distal cristae of the left main bronchus were cut-off by laser, followed by balloon dilatation. To our knowledge, no similar cases have been reported so far in our review of domestic and foreign literature. Insufficient clinical understanding of Pseudobronchial Crista-like Change increase the risk of misdiagnosis and missed diagnosis. Detailed exploration and careful identification under bronchoscopy are helpful for the timely diagnosis and treatment of Pseudobronchial Crista-like Change.

1. Introduction

Pseudobronchial crista-like change is an unusual type of inflammatory hyperplasia of the bronchial endobronchial membrane, which can be easily confused with normal bronchial tissues. Herein, we present the case of pseudobronchial crista-like change in the left main bronchus of a pediatric patient treated at the Second Hospital of Lanzhou University in 2019. To our knowledge, no similar reports or images have been published yet in either domestic or foreign literature.

2. Case presentation

A 4-year-old boy was admitted on March 22, 2019, because of bronchopneumonia with intermittent coughing for more than 10 days and wheezing for 2 days. History of choking due to foreign bodies and allergy were denied. Physical examination at admission showed a body temperature of 36.4 °C, pulse rate of 118 beats per minute, and respiratory rate of 26 breaths per minute. The child had clear consciousness and was generally active and well. The pharynx was slightly congested, and the tonsils were not swollen. Chest

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auscultation revealed coarse respiratory sounds of the lungs, low respiratory sounds on the upper lobe of the left lung, sputum sounds, and some inspiratory wheezing. No obvious abnormalities were found in other physical examinations. On auxiliary examination at admission, the white blood cell count was 12500/ μ L; neutrophil ratio, 58%; lymphocyte ratio, 34%; C-reaction protein, 32.01 mg/L; serum amyloid protein, 64.19 mg/L; procalcitonin, 0.137 ng/mL; and interleukin-6, 8.93 pg/mL. No recognizable abnormalities were found in blood culture, sputum acid-fast staining, purified protein derivative test, *Mycoplasma pneumoniae* antibody detection, routine urine and stool tests, biochemical tests, coagulant function, or infectious disease detection. Chest computed tomography (CT) revealed stenosis of the left main bronchus, atelectasis of the anterior basal segment in the upper and lower lobes of the left lung, and left lung inflammation combined with partial consolidation of the upper lobe and emphysema of the lower lobe of the left lung (Fig. 1). Pulmonary function examination suggested mild restrictive ventilation dysfunction.

On March 25, 2019, electronic bronchoscopy disclosed stenosis of the left main bronchial lumen and evident hyperemia, edema, and hyperplasia of the local mucosa, with exudation of purulent secretions. After repeated lavage with normal saline, stenosis was found in the two-luminal openings at the distal end, and the cristae widened such that the bronchoscope with an outer diameter of 4.2 mm could not enter (Fig. 2). As the department did not have a bronchoscope with a smaller outer diameter (2.8 mm), further examination was not conducted. Purulent inflammatory changes and bronchial stenosis were diagnosed, and bronchial dysplasia was suspected after tracheoscopy. Bacterial culture of the bronchoalveolar lavage fluid showed negative results. After intravenous infusion of piperacillin sodium, tazobactam sodium, and ambroxol injection; atomization of budesonide; and mechanically assisted expectoration for 10 days, the patient's cough, expectoration, and pulmonary signs did not significantly improve.

On April 1, 2019, the findings of electronic bronchoscopy were similar to those on March 25, 2019. The lumen of the left main bronchus was remarkably narrowed, and the bronchoscope with an outer diameter of 4.2 mm could barely enter, showing that the local mucosa was very congested and inflamed. Moreover, purulent secretions were found at this stage. Two openings were markedly narrowed at the distal end. The bacterial culture of bronchoalveolar lavage fluid was negative. The antibacterial drug was changed to cefoperazone-sulbactam sodium combined with vancomycin for 14 days, and methylprednisolone was administered intravenously for 5 days. A subsequent blood test revealed that levels of C-reactive protein and procalcitonin decreased to normal levels.

On April 15, 2019, electronic bronchoscopy displayed substantial improvement of the left main bronchial lumen stenosis and mucosal congestion and swelling and reduced purulent secretions compared with findings obtained on April 1, 2019. There was a crista at the distal end of the left main bronchus, dividing it into two openings and blocking the entrance of the bronchoscope with an outer diameter of 4.2 mm. An electronic bronchoscope with an outer diameter of 2.8 mm was used to enter from the upper opening, which clearly showed the openings of the left upper and lower lobes and the unobstructed lumen of the left upper lobe. In addition, several white foreign bodies were found at the opening of the lateral posterior basal branch of the left lower lobe, with purulent secretions gushing out. After entering from the lower opening of the distal cristae of the left main bronchus, openings of the left upper and left lower lobes were observed. Microscopic findings were the same as those after entering from the upper opening (Fig. 3). The cristae were hard and could not be pinched off and were hence clamped with biopsy forceps. Because the blood supply to the cristae was not yet confirmed, it could not be cut-off at that time. Therefore, a chest contrast-enhanced CT was performed on April 20, 2019, which revealed the lack of abnormal blood supply to the cristae (Fig. 4). On April 22, 2019, the foreign bodies—several pistachios—were removed under bronchoscopy (Fig. 5). The distal cristae of the left main bronchus were cut-off by laser, followed by balloon dilatation.

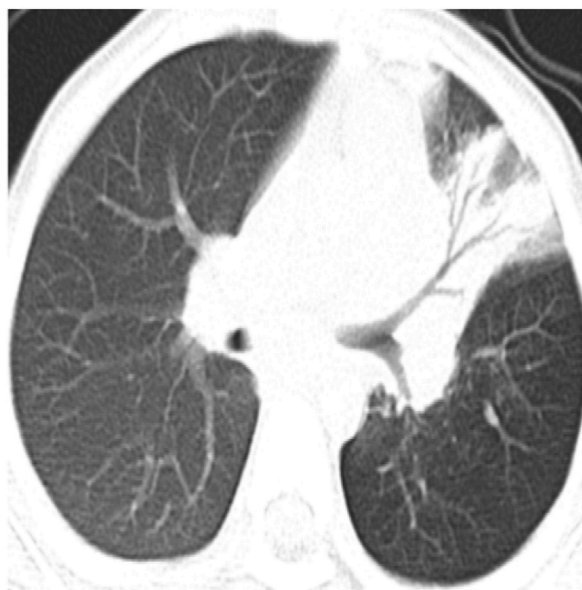


Fig. 1. Computed tomography (CT) on March 22, 2019. Axial CT of the chest reveals left main bronchus stenosis, left upper lobe/lingular atelectasis and left lower lobe hyperinflation (emphysema).

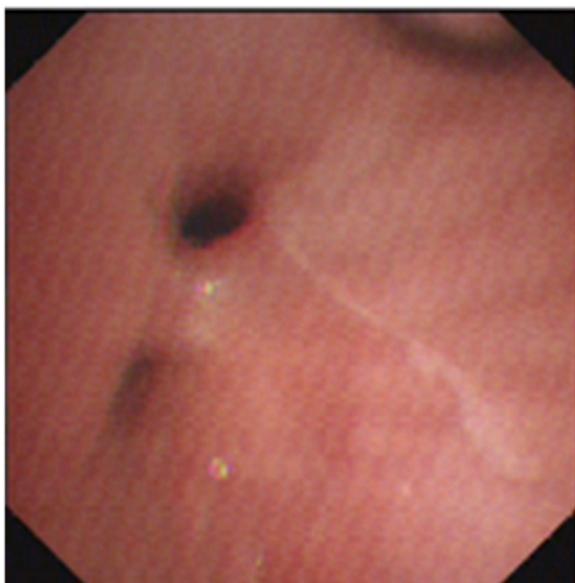


Fig. 2. The electronic bronchoscopy on March 25, 2019. Stenosis was found in the two-luminal openings at the left main bronchus distal end, and the cristae widened.

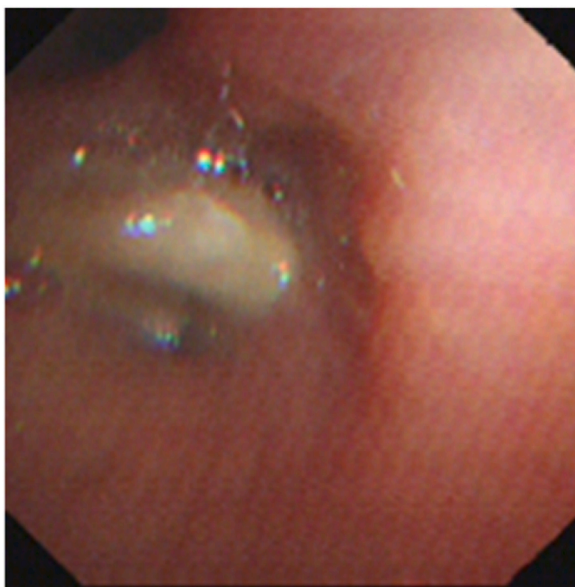


Fig. 3. The electronic bronchoscopy on April 15, 2019. An electronic bronchoscope with an outer diameter of 2.8 mm was used to enter from the upper opening and lower opening of the distal cristae of the left main bronchus, and several white foreign bodies were found at the opening of the lateral posterior basal branch of the left lower lobe.

3. Discussion

Pseudobronchial crista-like change is an unusual type of inflammatory hyperplasia of the bronchial endobronchial membrane caused by chronic retention of bronchial foreign bodies. However, related reports in domestic and foreign literature are rare. The main cause of Pseudobronchial Crista-like Change is the chronic existence of foreign bodies in the bronchus, but the specific mechanism is still not clear [1].

Usually, granulation tissues with bronchial intimal hyperplasia caused by foreign bodies in the bronchus are brittle and fresh and bleed easily. If a foreign body granuloma is formed, it often appears as a new structure embedded with foreign bodies under bronchoscopy, and the pathological manifestation appears as nodular lesions produced by foreign bodies enclosed within macrophages, epithelioid cells, and multinuclear giant cells. Pseudobronchial Crista-like Change induced by foreign bodies in the bronchus are different from granulation tissue hyperplasia in the bronchial-endobronchial membrane. Compared with normal bronchial tissues,



Fig. 4. The chest contrast-enhanced CT on April 20, 2019. The chest contrast-enhanced CT revealed the lack of abnormal blood supply to the cristae.



Fig. 5. The foreign bodies. The foreign bodies were several pistachios that were removed.

Pseudobronchial Crista-like Change has a tough texture, do not bleed easily, and are not easily detected. Unlike foreign body granuloma, Pseudobronchial Crista-like Change is not embedded in foreign bodies. In this case, the Pseudobronchial Crista-like Change was located at the distal end of the left bronchus, and the foreign bodies were located at the opening of the lateral posterior basal branch of the left lower lobe.

Foreign bodies in the trachea/bronchus are common in children. Chronic retention of foreign bodies and foreign plant bodies are risk factors of bronchial foreign bodies complicated with granulation tissue hyperplasia [2–4]. Plant foreign bodies (especially nuts) are rich in fatty acids and can promote the proliferation of fibroblasts and new thin-walled capillaries and activate various inflammatory cells, thus forming granulation tissue [5]. In this case, the bronchial foreign bodies were pistachios, which were among the risk factors of granulation tissue hyperplasia. Therefore, the formation of Pseudobronchial Crista-like Change, in this case, were closely related to some factors after chronic retention of foreign bodies in the bronchus. Foreign bodies retained in the airway can cause chronic inflammation. The foreign bodies repair airway inflammatory injury by means of granulation tissue hyperplasia. If the foreign bodies are found in a timely manner, the disease in the airway can be found as bronchial foreign bodies with granulation tissue

hyperplasia. If the foreign bodies remain in the airway, the inflammatory stimulus will persist, and the granulation tissue will gradually become scar tissue. The scar tissue is composed of a large number of collagen fiber bundles. Fibrous adhesion and hyperplasia of scar tissue may occur in the bronchial lumen. Scar tissue may undergo further extensive fibrotic hyalinization, resulting in fixed and hardened scar tissue. Collagenase can promote the decomposition and absorption of collagen fibers in scar tissue. Collagenase is mainly produced by fibroblasts, neutrophils, macrophages and other cells. The balance of collagen fiber synthesis and decomposition in scar tissue is related to the regulation of collagenase. The hard crista-like change in this case may be related to the abnormal function of the collagen-producing cells. However, the specific mechanism needs further exploration.

Pseudobronchial Crista-like Change is similar to the normal bronchial cristae in morphology and are very likely to be misdiagnosed as bronchial stenosis after bronchial dysplasia, thus neglecting further exploration of the presence of foreign bodies in the bronchus. Moreover, only prolonged antibiotic therapy was used to treat the obstructive pulmonary inflammation, but it was ineffective. Therefore, it is very important to accurately recognize and distinguish Pseudobronchial Crista-like Change. In case of bronchial foreign bodies complicated with Pseudobronchial Crista-like Change, the foreign bodies should be promptly removed, the cristae should be cut-off, and the narrow bronchus should be dilated under the bronchoscope [6]. In this case, the distal cristae of the left main bronchus detected by the initial bronchoscopy were misdiagnosed as bronchial opening stenosis caused by bronchial dysplasia. As there was no electronic bronchoscope with an outer diameter of 2.8 mm, it was only irrigated with normal saline repeatedly without further exploration. It was initially diagnosed as bronchial stenosis, bronchial dysplasia, and lobar pneumonia. Standard antibiotic treatment for 3 weeks showed poor efficacy. Electronic bronchoscopy was performed again, and exploration was conducted through the distal cristae of the left main bronchus, revealing foreign bodies at the opening of the lateral posterior basal branch of the left lower lobe. After removing the foreign bodies, cutting off the cristae, and performing balloon dilatation of the narrow bronchus, the pulmonary inflammation recovered quickly. After inquiring about the medical history, it was seen that the child had an intermittent cough for about 1 year. Thus far, this case suggests that the distal cristae of the left main bronchus were a pseudobronchial crista-like change caused by the chronic retention of foreign bodies rather than the abnormal development of the left main bronchus.

According to the diagnosis and treatment progression of this case, we can summarize this experience as follows: (1) Pseudo-bronchial Crista-like Change is clinically rare; thus, they should be differentiated from abnormal bronchial development, inflammatory stenosis of normal bronchial opening, bronchial dysplasia, and bronchial tuberculosis [7]. (2) If abnormal openings, cristae, and airway stenosis are detected during electronic bronchoscopy in children, they must be explored in detail as much as possible [8]. (3) Young children cannot provide a detailed history of foreign body choking. In the case of persistent and non-healing lower respiratory tract infection, the presence of bronchial foreign bodies should be checked as soon as possible to avoid delaying treatment [9].

Author contributions

NC collected and analyzed the case data and wrote the manuscript. XY collected the case data. FD was involved in the patient's treatment. LS was involved in the patient's treatment. QN participated in the treatment of patient, collected and analyzed the case data, and participated in writing the manuscript.

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Patient consent for publication

The informed consent from the patient's family was obtained.

Declaration of competing interest

All authors of this manuscript declare that there is no potential conflict of interest.

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