

FOCAL STENOSIS IN RIGHT UPPER LOBE BRONCHUS IN A RECURRENTLY WHEEZING CHILD SEQUENTIALLY STUDIED BY MULTIDETECTOR-ROW SPIRAL COMPUTED TOMOGRAPHY AND SCINTIGRAPHY

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Lower respiratory tract infections associated with wheezing are not uncommon in infants and young children. Among the wheezing-associated disorders, allergic etiologies are more commonly encountered than anatomic anomalies. We present a 3-year-old girl with a sudden attack of asthmatic symptoms including dyspnea, cyanosis and diffuse wheezing. Based on a history of choking, and atelectasis in the right upper lobe detected by chest films, flexible tracheobronchoscopy was arranged and incidentally detected a stenotic orifice in the right upper lobe bronchus. Multidetector-row spiral computed tomography and pulmonary scintigraphy subsequently also disclosed the focal stenosis. She suffered from recurrent wheezing, pneumonia and lung atelectasis during 1 year of follow-up. We emphasize the diagnosis, clinical course and management of focal stenosis in the right upper lobe bronchus.

Key Words: flexible tracheobronchoscopy, multidetector-row spiral computed tomography, ventilation and perfusion scintigraphy
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The incidence of tracheobronchial anomalies in the pediatric population ranges from 1% to 12%, and congenital bronchial stenosis is very uncommon [1-3]. The clinical presentation of tracheobronchial anomalies varies, and diagnosis is therefore difficult without the aid of imaging tools such as tracheobronchoscopy or computed tomography [1,4]. This

report describes a 3-year-old girl who presented at our emergency department with shortness of breath, wheezing and cyanosis. Focal stenosis in the orifice of the right upper lobe bronchus was detected incidentally by flexible tracheobronchoscopy (FTB) and confirmed by multidetector-row spiral computed tomography (MDCT).



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CASE PRESENTATION

A 3-year-old girl with a history of intermittent wheezing visited our emergency department because of acute dyspnea. Her body temperature was 37.6°C,

respiratory rate was 64 breaths per minute, oxygen saturation was 89%, heart rate was 192 beats per minute, and blood pressure was 94/70 mmHg. Her family reported that she had developed severe coughing after consuming some nuts the day before admission. Slight subcostal retraction of the chest wall was noted on physical examination. Breath sounds included bilateral wheezing. A chest X-ray revealed upward deviation of the minor fissure and a wedge-shaped opacity in the right upper lobe, suggesting atelectasis in the right upper lobe. A small area of opacity in the right lower lung was also found, and bronchopneumonia was suspected (Figure 1). The patient was admitted to our pediatric intensive care unit with suspected foreign body aspiration, based on the reported choking history.

Laboratory data revealed elevated C-reactive protein (29.1 mg/L) and leukocytosis (white blood cells, 17,900/ μ L; Seg/Lym, 83%/12%). FTB was performed immediately. Severe stenosis of the orifice of the right upper lobe bronchus was detected, which did not allow the 2.0-mm bronchoscope to pass. There was no foreign body or structural anomaly noted in the bronchi. Due to suspected bacterial pneumonia and asthma attack, the patient was given empiric antibiotic therapy with ampicillin/sulbactam (150 mg/kg every 6 hours), bronchodilators, and an inhaled steroid. However, the wheezing persisted and was refractory to bronchodilators. MDCT was performed and revealed narrowing of the orifice of the right upper lobe bronchus, relative to the diameter of the remainder of



Figure 1. Chest X-ray shows right upper lobe atelectasis and mild opacity in the right lower lung, suggesting a possible infectious process.

the right upper lobe bronchus, without external compression (Figure 2).

The patient also underwent ventilation and perfusion scintigraphy (V/Q scans). The perfusion scan showed heterogeneous radiotracer distribution with defects in the bilateral pulmonary fields (Figure 3A). The ventilation scan showed prolonged radioaerosol alveolar epithelial permeability in the right upper lung field (114 minutes), indicating relative obstruction compared with the left upper lung (85 minutes) (Figure 3B). She was discharged 2 weeks later in a relatively stable condition. However, her symptoms recurred 7 months later, in the absence of any choking event. A second chest MDCT scan was performed, which showed the same focal narrowing of the right upper lobe bronchus. The patient suffered from four episodes of recurrent wheezing, pneumonia and lung atelectasis during the following year. Surgical resection of the right upper lobe was therefore suggested, but declined by the patient's parents.

DISCUSSION

Wheezing is one of the main clinical manifestations of small airway narrowing in infants and young children. The causes of airway narrowing are diverse, and include congenital causes, trauma, infection, inhalational burns, and amyloid deposition [5,6]. The presentation of airway abnormalities may mimic other respiratory diseases such as asthma, bronchiolitis, and aspiration pneumonia. FTB is performed if there is a suspicion of foreign body aspiration or persistent wheezing despite treatment [7]. Compared with MDCT, which also plays an important role in the evaluation of tracheobronchial abnormalities in children, FTB can be used to perform therapeutic maneuvers and provide tissue biopsies, show bronchial mucosal morphology, and involves no radiation exposure [8]. However, unlike MDCT, it cannot provide three-dimensional information on adjacent soft tissue structures and the poststenotic airway [9–11].

Lung scintigraphy is another noninvasive tool that can be used for functional assessment of the lung in children [12]. In a study in dogs, artificial occlusion of a sublobar bronchus increased the ventilation to the obstructive lung, induced a shift of blood to the rest of the lung, and increased the ratio between mean ventilation and mean perfusion [13]. The increased

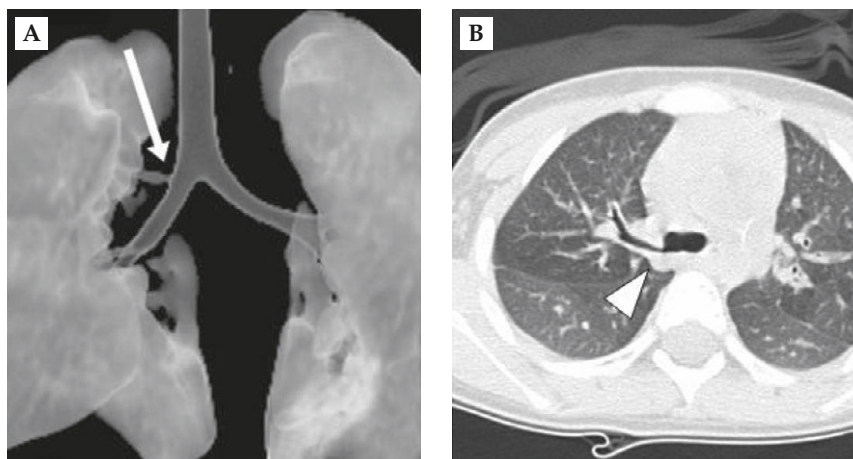


Figure 2. (A) Multidetector-row spiral computed tomography demonstrating that the orifice of the right upper lobe bronchus is narrowed relative to the diameter of the rest of the right upper lobe bronchus (white arrow). (B) The axial view reveals similar findings (arrowhead).

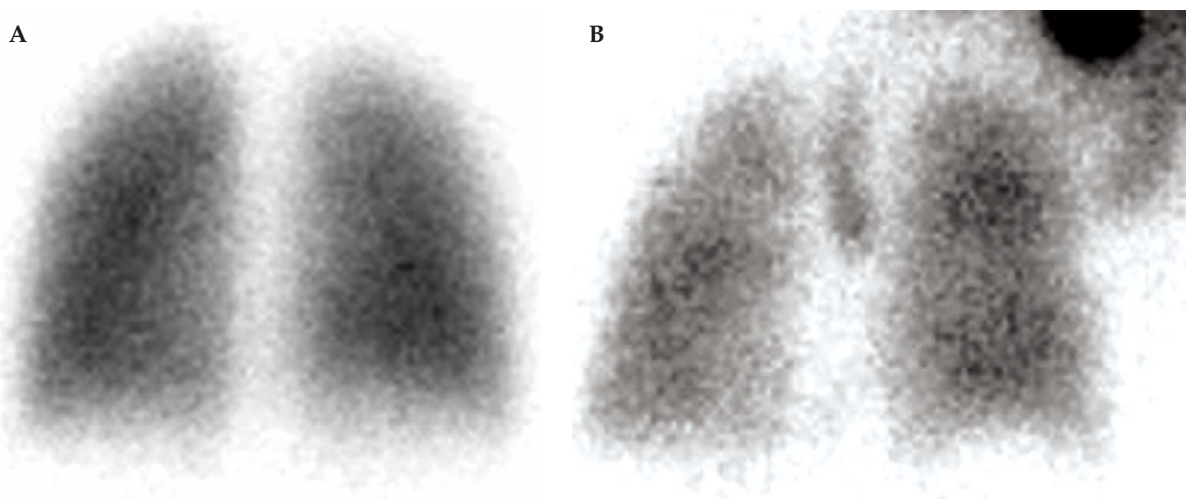


Figure 3. Ventilation and perfusion scintigraphy (V/Q scan) reveals a mismatch in the right upper lobe. The perfusion image (A) appears normal, whereas the ventilation image (B) shows stasis in the right upper lobe.

ventilation to the obstructive lung may have been due to increased communications with collateral ventilation [13]. As in our patient, the stasis of ventilation in the focal stenotic area could possibly be explained by regional mismatching between ventilation and perfusion. In other words, hypoxia vasoconstriction phenomenon can explain the diversion of blood flow to oxygen-rich areas. However, the normal perfusion results in our patient may have been due to the partial, but not total, occlusion of the bronchus. Hauck et al demonstrated a positive correlation among V/Q scans, lung function parameters and gas exchange [14], which prompted us to follow-up the regional lung function in this patient with a V/Q scan. Subsequent

V/Q scans during the asymptomatic period, after anti-inflammatory medication with montelukast and inhaled budesonide, showed improvements in ventilation and V/Q mismatch in the right upper lobes.

The clinical presentation of tracheobronchial anomalies varies from asymptomatic to severe respiratory distress [1,15]. In this patient, recurrent lower respiratory tract infection, lung atelectasis and wheezing were observed at follow-up. Her symptoms could have been caused by stenosis of the right upper lobe bronchus. Frequent wheezing episodes since later infancy suggested that her condition was congenital in nature. To the best of our knowledge, congenital focal stenosis of the orifice in the right upper lobe

bronchus is rare, compared with acquired causes, such as trauma, infection, inhalational burns, and amyloid deposition [5,6]. In addition, chest film abnormalities during the recurrent episodes suggestive of pneumonia and atelectasis were also noted in the surrounding lobes, including the right middle and lower lobes. In addition to mucoid impaction in the distal bronchus which can easily cause recurrent infection and lung atelectasis, some authors have hypothesized that the infectious process can migrate proximally to cause inflammation in the trachea and/or adjacent mainstem bronchus [6].

Management of airway stenosis is complex and depends on the degree, length and location of the stenosis. There are several strategies used to deal with tracheal and bronchial stenosis, including medical treatment, endoscopic dilatation, stent placement, and surgical resection [5,6,16]. Surgical intervention was suggested in this patient, but her parents refused. However, asymptomatic intervals could be achieved through the use of the anti-inflammatory agents montelukast and inhaled budesonide. The effects of montelukast and inhaled budesonide on inflammatory airway narrowing have been proven [17,18], but their effects on congenital focal bronchial stenosis need further study.

In conclusion, this case highlights the importance of considering airway abnormalities in patients with periodic wheezing, recurrent pulmonary infections, or atelectasis on chest films. FTB is the first step in diagnosing airway abnormalities or sputum impaction of isolated lobe bronchus, though other diagnostic workups, including MDCT and V/Q scans, should be used as adjuvant tools for evaluating focal bronchial tree abnormalities.

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右上肺葉支氣管局部狹窄在一反覆出現氣喘聲的 孩童—以多排螺旋電腦斷層與肺部換氣 —血流閃爍攝影追蹤

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下呼吸道感染合併氣喘聲在嬰兒與年幼的孩童身上並不少見。在會出現氣喘聲的疾病當中，呼吸道構造異常比起其他像是過敏引起的原因相對少數。我們報告了一位三歲女童，突然出現類似急性氣喘發作的症狀像是呼吸喘，發紺與廣泛的氣喘聲。根據父母提及有異物吸入的病史與 X 光片上右上肺葉塌陷，我們安排了支氣管鏡檢查，意外的發現在右上肺葉支氣管入口處有狹窄的情形。於是我們安排了多排螺旋電腦斷層與肺部換氣—血流閃爍攝影，都顯示右上肺支氣管入口處有局部狹窄的問題。經過一年的追蹤，病患仍有反覆的氣喘聲，肺炎與肺塌陷。本文著重在討論右上肺葉支氣管局部狹窄的診斷，臨床過程，與治療追蹤。

關鍵詞：軟式氣管支氣管鏡，多排螺旋電腦斷層，肺部換氣—血流閃爍攝影

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