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Case report

A case of recurrent respiratory infection resulting from a congenital anomaly of the bronchial tree tracheal bronchus

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= Abstract =

The term tracheal bronchus refers to an abnormal bronchus that comes directly off of the lateral wall of the trachea (above the carina) and supplies ventilation to the upper lobe. Tracheal bronchi occur almost exclusively on the right trachea and are associated with other congenital anomalies. In addition, tracheai bronchus may be related to other inflammatory conditions with persistent wheezing, such as recurrent pneumonia, chronic bronchitis and bronchiectasis, which is a result of the relatively poor local drainage of the involved bronchi. An infant with recurrent wheezing is likely to be a challenge for a clinician in the evaluation of the etiology of airway obstruction and in the differential diagnosis of wheezy breathing. The authors report a case of an 8-month-old female infant with a ventricular septal defect, who presented with stridor and recurrent respiratory infection and finally was finally diagnosed with a tracheal bronchus using computed tomography and a bronchoscopy. Therefore, tracheal bronchus should be included in the differential diagnosis of any child who presents with chronic or recurrent respiratory tract symptoms such as coughing, wheezing, stridor and recurrent respiratory infection, particularly in children with other congenital deformities. (Korean J Pediatr 2008;51:660-664)

Key Words : Congenital anomaly, Tracheal bronchus, Stridor

Introduction

Tracheal bronchus is one of the most common anomalies of the bronchial tree, which is an aberrant or ectopic bronchus originating from the right lateral wall of the trachea[1-4].

Most cases of tracheal bronchus are asymptomatic and detected only incidentally by bronchoscopy or radiological examination, and recently it can be easily diagnosed by computed tomography[5-7].

Tracheal bronchus may be associated with other congenital anomalies including congenital heart disease, laryngeal web, rib and vertebra anomalies, and tracheal stenosis[4, 5]. There are several reports claiming these bronchial abnormalities as a source of recurrent infection, persistent cough, stridor, wheezing, hemoptysis, and acute respiratory distress[8, 9]. The main clinical manifestations of tracheal bronchus are hemoptysis, cough, and dyspnea which are resulted from the underlying lung disease regardless of the bronchial anomaly. However in most cases, tracheal bronchus is of no clinical significance[1-3, 9, 10].

Tracheal bronchus may also be responsible for recurrent respiratory infection and wheezy breathing[1-3, 10, 11]. In some cases, children who have symptoms of wheezy breathing and recurrent respiratory infection requiring bronchoscopy for respiratory symptoms are found to have tracheal bronchus, which is frequently thought to be an incidental finding[5-7, 10]. Since these abnormalities predispose to infections and perhaps to malignancies, detection of them still carries importance in a person being worked-up even for another reason and low threshold is required for chest computed tomography (CT) in suspicious cases by chest X-ray. CT-bronchoscopy and CT-bronchography are two complementary techniques to further evaluate the tracheobronchial tree as well as the type of bronchial anomaly and to exclude intraluminal pathologies.

We report a case of 8 month old infant presented with stridor and recurrent respiratory infection who finally was

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diagnosed as tracheal bronchus by computed tomography and bronchoscopy. The bronchoscopy indicated high bifurcation of the carina in our patient, and the direct coronal computed tomography demonstrated concomitant tracheal stenosis and tracheal bronchus.

Case report

An 8-month-old female infant who has a history of recurrent respiratory infections such as pneumonia, bronchiolitis visited in Department of Pediatrics, College of Medicine, Kyung-Hee University, Seoul, Korea. She was presented with persistent loud wheezy breathing and difficulty in respiration and also had congenital heart disease, perimembranous type of VSD. There was associated relatively mild coughing, elevated respiratory rate (tachypnea) and chest retraction which is the evidence of the use of accessory muscles for breathing.

She visited in our hospital very often (6 times) and was hospitalized 4 times during 8 month period since her birth, treated with IV fluids and parenteral antibiotics and nebulization of bronchodilators for each episode.

She was sweating, and looked difficult in respiration. But there were no cyanosis, chronic cyanotic symptoms and signs such as clubbing, peripheral cyanosis or low oxygen saturation. She was tachypneic, but O_2 saturation was 98-99% in room air. Her respiratory examination revealed the trachea in normal position with hyper-inflated chest, wheezy breathing and stridor noted bilaterally. Examination of the chest revealed symmetrical expansion, normal antereoposterior diameter and no spine deformities such as kyphosis or scoliosis.

Auscultation revealed increased intensity of wheezy breath sounds especially right upper lung with widespread crepitations. Rest of the systemic examination parameters were within normal limits.

Complete blood count was as follows. Hemoglobin 11.9 gm%, WBC 13,330 cells/mm^3, differential count: neutrophil 40.6%, lymphocyte 43.5% and platelet count 447,000/mm^3. IgE was 29.7 IU/mL, and pheripheral total eosinophil count was 590/mm^3. Mycoplasma antibody titer was 1:80, and other laboratory findings were within normal limits. Chest radiograph revealed bilaterally hyperinflated lung fields. Clinical diagnosis at admission was upper airway obstruction pneumonia with bronchiolitis and perimembranous ventricular septal defect (VSD).

A dynamic CT was done and revealed diffuse luminal narrowing from low trachea to main bronchus stem, but without other congenital anomalies such as bronchial wall thickening, laryngomalasia, laryngeal web or vascular ring. Tracheal bronchus was distributed to a part of right upper lung and there were calcification of tracheal wall and air trapping of superior segment of left lower lobe (Fig. 1A, 1B). There was diffuse enlarged thymus without mediastinal mass-like lesion. Diffuse tracheal narrowing with tracheal bronchus to right upper lobe was found.

Discussion

Tracheal bronchus, with accessory bronchus, is one of the most common anomalies of bronchial tree[1-3]. Tracheal bronchus is an aberrant accessory or ectopic bronchus oc-

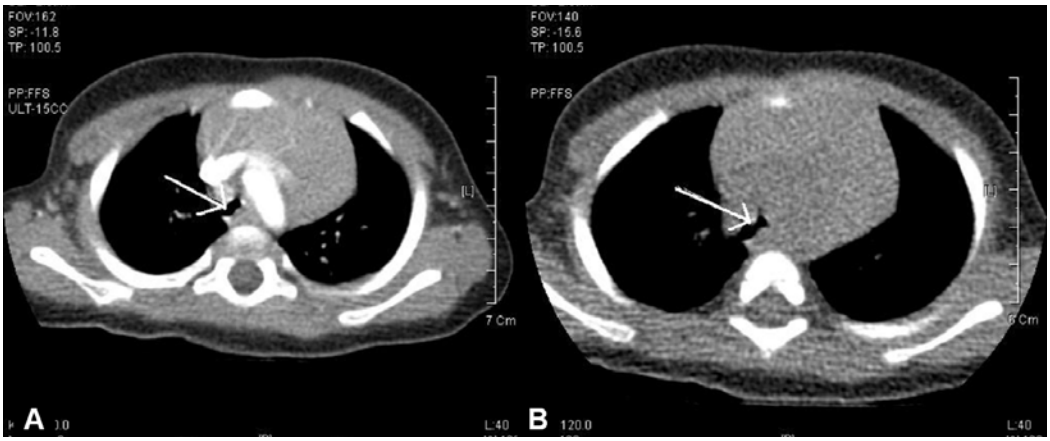

Fig. 1. A dynamic CT scan of the chest showing the presence of the aberrant tracheal bronchus and diffuse luminal narrowing from the low trachea to the main bronchus stem (arrow).

casionally usually originating from the right lateral wall of the trachea less than 2 cm above the level of main carina[1-3, 8, 9, 12]. Contrary to numerous variants of lobar or segmental bronchial subdivisions, tracheal bronchus, abnormal bronchi origination from the trachea or main bronchi, is a rare and usually incidental finding in humans. It can develop from any point above the main carina, but is usually within the 2 cm range. Its diameter ranges from 0.5 to 1.0 cm; its length ranges from 0.6 to 2.0 cm[1-3, 8]. Most cases of tracheal bronchus are asymptomatic and detected only incidentally by bronchoscopy or radiological examination, with reported incidence varies between 1-3% of pediatric endoscopic studies [1, 4, 5, 9]. Tracheal bronchus of our patient is also heading towards right upper lobe and origination from either main bronchus or trachea 1.5 cm above the main carina.

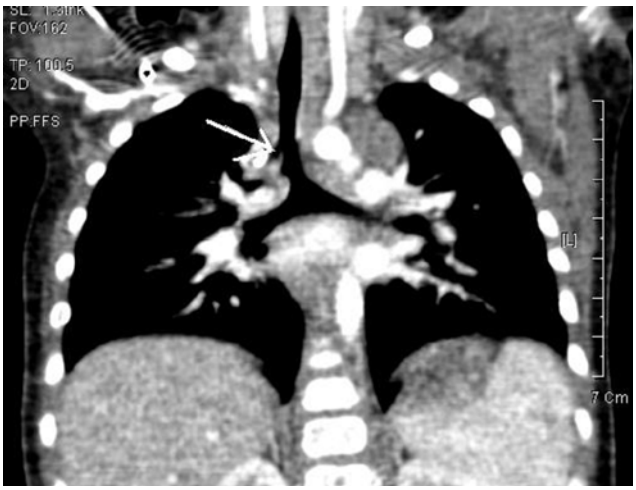

Fig. 2. A dynamic CT scan of the chest, coronal reformat. Accessory tracheal bronchus (arrow). CT scan reveals an abnormal bronchial structure arising from the intermediate bronchus. Note also that there is not any other abnormal lung lobule or abnormality.

Tracheal bronchus can be identified during bronchoscopic examination by the presence of an ectopic opening from the tracheal wall, and CT is the newest modality for evaluating tracheal bronchus[5, 10]. It can be easily diagnosed by CT as a small, round translucency posterolateral opening to the trachea[5, 7, 10]. In our case, tracheal bronchus was supplying the aeration of right upper lobe (Fig. 3A)

Frequently tracheal bronchus is associated with other congenital anomalies[4, 5, 11]. Our patient also has congenital heart disease, VSD. Tracheal bronchus was also related to inflammatory condition, affecting the lung such as recurrent pneumonia, chronic bronchitis and bronchiectasis because of the relatively poor local drainage of the involved bronchi[1, 8,

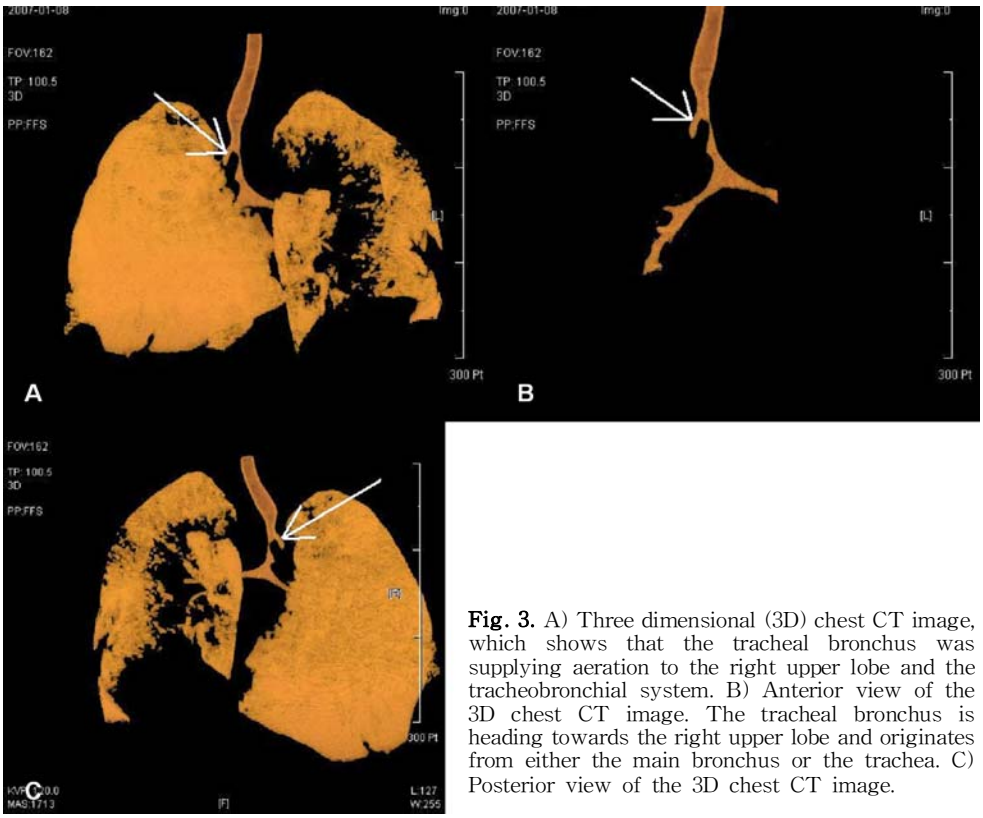

Fig. 3. A) Three dimensional (3D) chest CT image, which shows that the tracheal bronchus was supplying aeration to the right upper lobe and the tracheobronchial system. B) Anterior view of the 3D chest CT image. The tracheal bronchus is heading towards the right upper lobe and originates from either the main bronchus or the trachea. C) Posterior view of the 3D chest CT image.

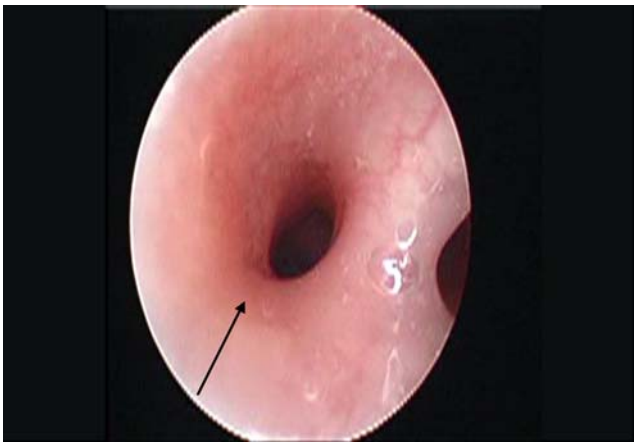

Fig. 4. Fiberoptic bronchoscopic view. The main carina of the trachea was revealed. The tracheal bronchus arises from the right lateral wall of the trachea, and is distributed to a part of the right upper lobe (arrow).

[9, 12]. We experienced recurrent pneumonia with stridor and wheezing in our patient, we could diagnose tracheal bronchus by CT and bronchoscopy. Although tracheal bronchus is usually asymptomatic, incidentally discovered and is not clinically significant, awareness of this condition may be important. Because the presence of a tracheal bronchus may complicate endotracheal intubation such as atelectasis or hypoxia and we can avoid the possible associated clinical complication, including recurrent episode of infection, perhaps malignancies may be anticipated in a small percentage of patients[9, 12].

A substantial number of children have chronic or recurrent respiratory tract symptoms such as coughing, wheezing, and stridor which occur frequently or persist for long periods. These symptoms are common in infants and children, engendered by anatomic and developmental features as well as innate susceptibility to infection. But isolated episodes of acute wheezing, such as may occur with bronchiolitis, are not uncommon. The wheezing that recurs or persists for longer than 4 weeks suggests other diagnoses including congenital structural abnormalities involving the air passage or lower respiratory tract, still further complicating differential diagnosis[11]. Tracheal bronchus can be also responsible for recurrent respiratory infection and wheezy breathing[1-3]. Determining the most likely underlying pathogenic mechanism of this condition, repeated complete physical examination like a carefully taken history may be required to the clinician toward the proper diagnosis and minimize unnecessary laboratory testing. This proper diagnosis must include other causes of airway obstruction (congenital malformations, foreign bodies), infectious bronchiolitis, cystic fibrosis, hypersensitivity pneumonitis, immunologic deficiency disease, and other rarer conditions including tuberculosis and fungal infections[1-3, 10, 11]. Other factors to be considered include environmental allergies, the presence of asthma, coronary artery disease, congestive heart failure or valvular heart problems[11]. A family history of asthma, lung problems, allergies or hay fever must also be considered[5]. Failure of this approach to identify the process responsible or to effect improvement signals the need for more extensive and perhaps invasive diagnostic efforts, including bronchoscopy[6]. Most of these patients underwent bronchoscopy because of respiratory difficulties and these patients have coexisting tracheobronchial stenosis. Bronchoscopy indicated high bifurcation of the carina and direct coronal computed tomography demonstrated concomitant tracheal stenosis and tracheal bronchus.

Most patients with tracheal bronchus can be treated conservatively. Correction of the underlying abnormalities is of paramount importance for the relief of respiratory symptoms[6, 7, 10]. Our patient had recurrent respiratory infection including pneumonia or bronchiolitis associated with wheezing and stridor. And other congenital abnormalities were present including tracheal narrowing and VSD. Recurrent right upper lobe pneumonia and bronchiolitis associated with wheezy breathing were present at 6 times.

The presence of a clinically significant tracheal bronchus should be considered in every child with recurrent right upper lobe pneumonia, especially in children with associated other congenital anomalies. Our case was diagnosed tracheobronchial anomalies, tracheal bronchus, by broncoscopy, which was done to evaluate the chronic or recurrent respiratory tract. Therefore tracheal bronchus may be the causes of chronic or recurrent respiratory tract symptoms such as coughing, wheezing, and stridor and recurrent respiratory infection.

Most patients with tracheal bronchus can be treated conservatively. Correction of the underlying abnormalities is of paramount importance for the relief of respiratory symptoms[6, 7, 10]. If bronchiectasis or bronchial stenosis is found, surgical resection should be performed.

한 글 요 약

반복적인 호흡기 감염을 가진 환아에서 진단된 선천성 기도 기형, 기관기관지 1예

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기관기관지는 선천성 기도기형 중 가장 흔한 질환으로, 기관분기부 상부의 우측 벽에서 기시하는 이소성 부기관지를 말하며, 반복적인 염증성 질환으로 폐렴과 기흉, 기관지 확장증 등을 초래하여 임상적 문제가 되는 경우도 있으나 대부분 기관지내시경검사, 기관지조영술 등을 통해서 우연히 발견된다. 우리는 반복적인 호흡기 감염과 동반되어 나타난 지속적인 천명과 호흡곤란으로 입원치료 중인 환아에서 선천성 기도기형 중 기관기관지를 경험하였으며, 일반 컴퓨터 단층촬영과 삼차원 컴퓨터 단층촬영, 강직형 기관지내시경검사로 이를 확인하였다. 따라서 반복적인 호흡기 감염과, 지속적인 천명을 주소로 내원하는 환아 들에게서 진단의 감별에 있어 선천성 기도기형인 기관기관지의 가능성을 고려해야 하겠다.

References

1) Ghaye B, Szapiro D, Fanchamps JM, Dondelinger RF. Congenital bronchial abnormalities revisited. Radiographics 2001;21:105-19.
2) Ghaye B, Kos X, Dondelinger RF. Accessory cardiac bronchus: 3D CT demonstration in nine cases. Eur Radiol 1999;9:45-8.
3) Bettina C. Hilman BC, Amaro-galvez R. The Wheezing Infant: A challenge in evaluation and diagnosis. Pediatr Asthma Allergy Immunol 2003;16:237-46.
4) Bou-Khalil PK, Aboussouan LS, Mehta AC. Tracheal bronchus. J Bronchol 1996;3:134-6.
5) Shipley RT, McLoud TC, Dedrick CG, Shepard JO. Computed tomography of the tracheal bronchus. J Comput Assist Tomogr 1996;9:53-5.
6) Ritsema GH. Ectopic right bronchus: indications for bronchography. AJR Am J Roentgenol 1983;140:671-4.
7) Wong KS, Lien R, Lin TY. Clinical and computed tomographic features of tracheal bronchus in children. J Formos Med Assoc 1999;98:646-8.
8) Yildiz H, Ugurel S, Soylu K, Tasar M, Somuncu I. Accessory cardiac bronchus and tracheal bronchus anomalies: CT-bronchoscopy and CT- bronchography findings. Surg Radiol Anat 2006;28:646-9.
9) McLaughlin FJ, Strieder DJ, Harris GBC, Vawter GP, Eraklis AJ. Tracheal bronchus: association with respiratory morbidity in childhood. J Pediatr 1985;106:751-5.
10) Won JH, Park JY, Kang TK, Park KS. A clinical experience of tracheal bronchus. Tuberc Respir Dis 1998;46:583?6.
11) Heyer CM, Kagel T, Lemburg SP, Nicolas V, Rieger CHL. Evaluation of tracheobronchial anomalies in children using low-dose multidetector CT. Pediatr Pulmonol 2004;38:168-73.
12) Del Pizzo KR, Joffe DC, Finkel JC. Tracheal bronchus in 10-month-old patient for thoracoscopic resection of congenital cystic adenomatoid malformation. Paediatr Anaesth 2006;16:997-1003.