PICTORIAL CME

A Young Adult with Tracheal Bronchus and Congenital Cystic Adenomatoid Malformation

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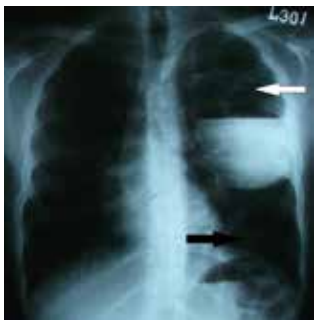

Fig. 1: Chest radiograph PA view showing a bullous lesion with air fluid level in the left upper and mid zones (white arrow) and another bullous lesion in the left lower zone (black arrow)

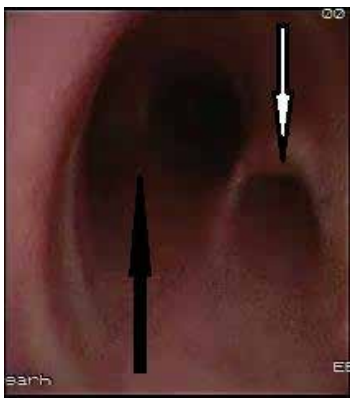

Fig. 2: Fibre-optic video bronchoscopy showing the carina (black arrow), and the opening of an aberrant bronchus (tracheal bronchus) arising from trachea superior to carina (white arrow)

24-yr-old male presented to the Department of Pulmonary Medicine, Government Medical College and Hospital, Chandigarh with history of recurrent sore throat, hemoptysis and generalized weakness since 3 months and fever since 20 days. There was no past history suggestive of tuberculosis or repeated infections. Chest radiograph revealed left sided bullous lesions, one with an air fluid level (Figure 1). Sputum for AFB and gram stain were negative and pyogenic culture and sensitivity was sterile. Complete haemogram showed raised total leukocyte count. Spirometry was

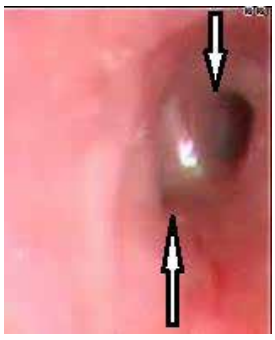

Fig. 3: Negotiation of tracheal bronchus with the bronchoscope revealing two openings (white arrows)

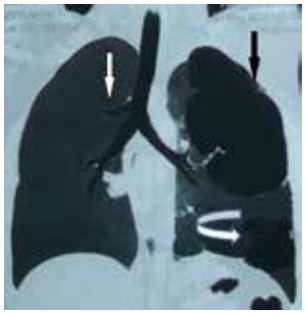

Fig. 4: CECT Chest, coronal minIP image showing displaced tracheal bronchus supplying left upper lobe (white arrow) and lobulated bullous lesions in left lower lobe. (Black arrow and curved arrow)

normal. Patient was managed with antibiotics. After resolution of fever, bullectomy was planned. As a part of routine work-up as per surgical advice, Fibre-optic bronchoscopy (FOB) was done and Contrast enhanced computed tomography (CECT) of the chest was planned. FOB revealed an aberrant bronchus (tracheal bronchus) arising from lateral tracheal wall superior to carina (Figure 2). Negotiation of tracheal bronchus with bronchoscope revealed two openings (Figure 3). Rest of the bronchial tree was normal except that separate right upper lobe bronchus

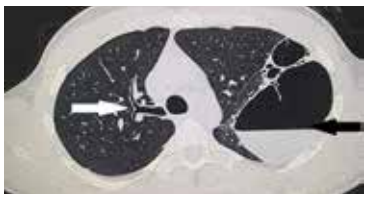

Fig. 5: CECT Chest, axial lung window showing the division of the right upper lobe displaced tracheal bronchus (white arrow) and the bullous lesion in superior segment of left lower lobe with air fluid level (black arrow)

was not seen arising from the right main bronchus.

Our patient was an immunocompetent young adult who presented to the hospital for the first time with infected bullous lesions but otherwise normal intervening lungs. There was no significant past history suggestive of long standing respiratory illness. Hence literature was searched for the associations of the incidental tracheal bronchus with parenchymal involvements, if any.[1,2] Since it was found that tracheal bronchus may coexist with other congenital conditions,[2] a CECT chest was specifically ordered, to find out any such anomaly.

CECT showed lobulated thin walled bullous lesions in the left lower lobe with minimal adjacent fibrosis and the rest of the lung parenchyma was normal. There was no evidence of emphysema or any other sequelae to infective etiology. The bullous lesion was not having any systemic blood supply, so the radiological diagnosis of congenital cystic adenomatoid malformation (CCAM) was made. Thus, it supported the clinical suspicion of associated congenital anomaly post bronchoscopy. CT reconstruction also

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Received: 31.05.2017; Accepted: 20.01.2018

confirmed the findings of a displaced tracheal bronchus supplying the right upper lobe. The displaced tracheal bronchus showed two divisions, one supplying the apical segment, and the other one further dividing and supplying the anterior and posterior segments (Figures 4 and 5).

The patient was taken up for surgery, and the histopathology of the resected bullous lesion confirmed it to be a CCAM. Thus, in our patient, it was found that CCAM mimicked a bulla on initial presentation.

Tracheal bronchus is defined as a bronchus originating from the lateral tracheal wall. It is usually found within 2-6 cms of the carina.[1,2] It may coexist with other congenital anomalies, CCAM being one of them.[2] CCAM is characterized by a multicystic mass of pulmonary tissue with abnormal proliferation of bronchial structures.[3] Most of the congenital abnormalities of the respiratory system are diagnosed in early life. However, some may be diagnosed only in adulthood.[3] CCAM is also one of them, as it usually presents in neonatal period or by the first 2 years of life, rarely can it present in adult life.

Detailed assessment of tracheobronchial tree, keeping in mind the presence of aberrant bronchi/segments, and correlating them with the associated congenital malformations, if any, is of great clinical relevance. Firstly, since the congenital abnormalities are usually present in early age groups, they have to be kept in mind and specifically looked for, in adult populations. Secondly, these entities should be known to the chest physicians and surgeons, as they may pose difficulties in diagnosis due to their mimicry with malignancy or sequestration. Recurrent pneumonias and pneumothorax have also been reported in them.[4] Thirdly, some patients may need surgery to avoid life threatening complications associated with these congenital malformations.[5] And on intubation of such patients during surgery, if the displaced tracheal bronchus is overlooked and an endotracheal tube is placed too distally, gross one lung ventilation may occur, causing further complications and endangering life.[6]

References

1. Rahmanian R, Zheng J, Chadha NK, Kozak FK, Campbell AI, Ludemann JP. False carina: a distinct variant of tracheal bronchus. *Int J Pediatr Otorhinolaryngol* 2015; 79:623-8.
2. Mehta AC, Thaniyavar T, Ghobrial M, Khemasuwan D. Common Congenital Anomalies of the CentralAirways in Adults. *Chest* 2015; 148:274–287.
3. Çay A, Sarihan H. Congenital malformation of the lung. *J Cardiovasc Surg* 2000; 41:507-510.
4. Zylak CJ, Eyler WR, Spizarny DL, Stone CH. Developmental Lung Anomalies in the Adult: Radiologic-Pathologic Correlation. *Radiographics* 2002; 22Issue suppl_1.
5. Mordant P, De Dominicis F, Berna P, Riquet M. Tracheobronchial and pulmonary parenchymatous congenital abnormalities requiring surgical treatment in adults. *Rev Pneumol Clin* 2012; 68:110-6.
6. Moon YJ, Kim SH, Park SW, Lee YM.The implications of a tracheal bronchus on one-lung ventilation and fibreoptic bronchoscopy in a patient undergoing thoracic surgery: a case report. *Can J Anaesth* 2015; 62:399-402.