
Persistent left upper lobe opacity in middle-aged gentleman

Sir,

We hereby report the case of a 35-year-old gentleman with no medical co-morbidities, who presented with chronic productive cough and recurrent respiratory tract infections with wheeze for the past 5 years. He recalled similar symptoms infrequently from childhood, but the frequency and severity had distinctly increased over the last 5 years. He gave history of smoking for the last 21 years. There was no personal or family history of atopy. He did not have fever, loss of appetite or weight loss. Chest radiograph showed a mass like opacity in the left upper zone [Figure 1]. CT chest revealed a well-defined lobulated lesion with smooth rounded borders measuring 5×4 cm in the left upper lobe. The lesion was septate, had central hypodense (30-40 HU) content and seemed to communicate with left upper lobe bronchus. The walls depicted contrast enhancement with calcifications [Figure 2]. The tomogram also showed a small left upper lobe with focal apical fibrosis and calcification. The left upper lobe bronchus seemed narrowed at its origin and the lesion in question was immediately distal to the narrowed bronchial segment [Figure 3]. The pulmonary vasculature was normal. The differentials entertained included pulmonary tuberculosis with left upper lobe bronchostenosis and bronchocele, allergic bronchopulmonary aspergillosis with secondary mucoid impaction, congenital bronchial atresia

with distal bronchocele, and carcinoma lung arising in the left upper lobe bronchus with central necrosis.

Further work up in the form of fibreoptic bronchoscopy was done which revealed pin hole-sized left upper lobe bronchial orifice, through which scope could not be negotiated. The opening was smoothly narrowed and showed no signs of wall inflammation, infiltration or neoplastic lesion. Mucopurulent secretions were draining through the narrowed left upper lobe opening which was collected and subjected to AFB smear by microscopy as well as bacterial, mycobacterial and fungal cultures. All results were negative. BAL cytology was showing neutrophil-rich inflammatory cells. Spirometry revealed a mild restrictive ventilatory defect with no significant bronchodilator reversibility. Serum Ig E level was within the normal range and *Aspergillus* skin prick test was negative. A diagnosis of left upper lobe hypoplasia with focal bronchial atresia and distal bronchocele was arrived at. He was started on mucolytic, inhaled bronchodilators, chest physiotherapy and postural drainage along with antibiotics. In view of the recurrent infections, thoracic surgery consultation for left upper lobectomy was suggested. An advice for left upper lobe resection was given by the multidisciplinary team involving pulmonologist, thoracic surgeon and radiologist. Patient underwent left upper lobectomy. A 4×3 cm

and 2 × 3 cm cystic mass in the area of upper lobe was identified, which contained multiloculated cystic cavity communicating with bronchi, with dilated bronchioles in adjacent segments. The cavity contained creamy yellow solidified material. The histopathology was suggestive of bronchocele associated with inflammation, most probably related to bronchial atresia. Post-operative chest X-ray showed left upper zone post lobectomy changes with resolution of infection [Figure 4].

Pulmonary underdevelopment includes a wide range of malformations characterized by incomplete development of lungs. Pulmonary underdevelopment has been classified into three groups by Schneider and Schwalbe.^[1,2] According to this classification, group 1 abnormalities are characterized by absence of a lobe or whole of the lung along with its bronchial supply and is termed agenesis of the lobe or lung. The feature of group 2 abnormalities is the presence of a rudimentary bronchus which is limited to a blind-end pouch

without distal lung tissue and is termed aplasia. In the more frequently encountered group 3 developmental disorders, there is bronchial hypoplasia with variable reduction of lung tissue and is termed hypoplasia as seen in our patient. Patients with pulmonary underdevelopment may remain asymptomatic and the diagnosis may not be made until adulthood if the extent is limited and there are no associated major congenital abnormalities.^[3] This was true in our case where the underdevelopment was limited to left upper lobe with normal remaining lung and there were no other congenital anomalies. The diagnosis can be confirmed by multidetector CT which clearly delineates the deformed and stunted pulmonary vasculature and bronchial tree.^[4,5] Bronchoscopy, ventilation-perfusion scanning, magnetic resonance imaging, CT-angiography, pulmonary angiography or bronchography can also reveal absence of bronchovascular structures. There are case reports of agenesis of left upper lobe of lung identified coincidentally in cases of pulmonary

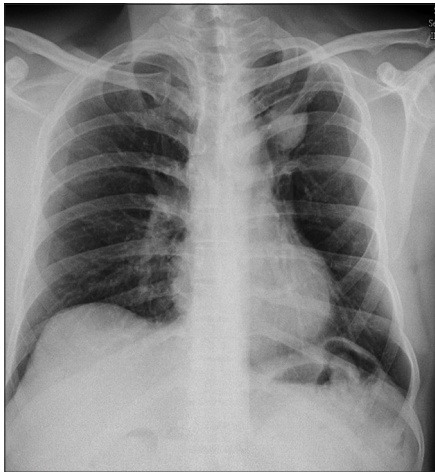


Figure 1: Chest radiograph showing left upper zone homogenous opacity



Figure 2: CT showed well-defined lobulated lesion about 5 × 4 cm in the left upper lobe which was septate, had central hypodense (30-40 HU) content, communicating with left upper lobe bronchus

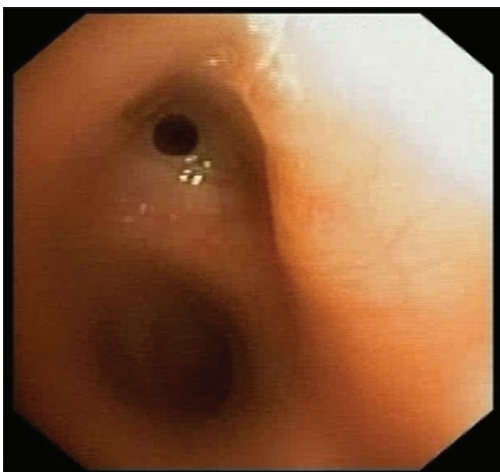


Figure 3: Fiberoptic bronchoscopy image showing left upper lobe apical segment narrowing

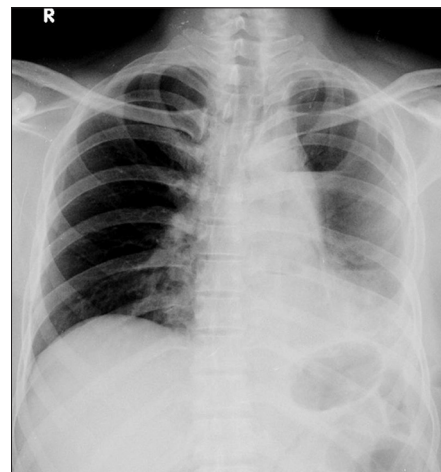


Figure 4: Post-operative chest radiograph showing left upper zone post-lobectomy changes

tuberculosis^[6] and lung malignancy.^[7] In our case, tuberculosis and malignancy were ruled out by appropriate investigations.

Bronchocele stands for dilated bronchus or a group of bronchi filled with fluid. If they contain either mucus or pus, they may be called bronchomucocele or bronchopyocele, respectively. Bronchocele is seen most commonly in congenital bronchial atresia, with accumulation of fluid in the bronchus distal to the atretic segment of bronchus. Bronchoceles are identified in computerized tomography with reasonable certainty, which has become the examination of choice.^[6-8] The branching pattern is characteristic of the lesion, which was evident in our case also. CT, in addition to providing conclusive diagnosis, also shows the parenchymal and airway components of the anomaly, helps in estimating the extent of air trapping and helps in ruling out differential diagnoses such as bronchogenic cyst, bronchiectasis, aspergillosis, tumours, pulmonary aneurysms or arteriovenous malformations.^[8]

Fiberoptic bronchoscopy allows direct visualization of the affected airway and conclusive diagnosis of the airway component of the anomaly and has the added advantage of providing bronchial toilet as well as procuring samples for microbial investigations.^[9] Direct visualization also allows ruling out an endobronchial neoplasm with greater precision and planning interventional procedures like endobronchial stenting. Bronchocele has also to be differentiated from allergic bronchopulmonary aspergillosis, condition will often disappear with treatment, and it will not have an obstructing membrane as in a bronchocele. In equivocal cases, resection for diagnostic purposes may be performed and in such cases, the pathologist has to be highlighted of the possibility. The gross specimen has to be cut in the long axis to demonstrate the membrane or septum, which is characteristic of the lesion.

Although most cases show little change over several years, surgical resection is regarded as the definitive treatment because the lung distal to the bronchocele may act as source of recurrent infection through poor ventilation and atelectasis.^[10] Our patient also underwent surgical resection in the form of lobectomy. We conclude that pulmonary hypoplasia secondary to bronchial atresia causing distal bronchocele can present at any age. A high index of suspicion with characteristic CT findings and confirmative bronchoscopy may clinch the diagnosis in such cases.

Financial support and sponsorship
Nil.

Conflicts of interest

There are no conflicts of interest.

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Access this article online	
Quick Response Code: 	Website: www.lungindia.com
	DOI: 10.4103/0970-2113.184934

How to cite this article: Venkitakrishnan R, Aziz KS, Mehta A. Persistent left upper lobe opacity in middle-aged gentleman. *Lung India* 2016;33:457-9.