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Case Report

Unilateral Hyper-Translucent Lung: a Rare Case Report of Bronchial Atresia

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Abstract

A young female patient with chronic left side chest pain warranted a work-up. Chest X-ray suggested left hyper-translucency. High resolution computed tomography scan revealed left atretic apicoposterior segmental bronchus with an area of hyper-translucency suggestive of bronchial atresia. Fiberoptic bronchoscopy findings were normal. Spirometry showed moderate restriction with mild obstruction. It is a rare developmental anomaly characterised by interruption of normal bronchial continuity. Most commonly, it involves the apicoposterior segmental bronchus of left upper lobe as seen in our case. Since it is a benign condition, surgical treatment or any aggressive method of treatment is not required. However, intervention may be required in case of life threatening complications.

Keywords

bronchial atresia, congenital anomaly, hyper-translucent lung

INTRODUCTION

Bronchial atresia is a rare developmental anomaly characterised by interruption of normal bronchial continuity. Most commonly, the apicoposterior segmental bronchus of left upper lobe followed by segments within the right upper, middle and lower lobes is affected.[1,2] The segmental bronchus gets sealed off from larger proximal airways. The sealed-off or atretic bronchus becomes distended by bronchial secretions. This results in the formation of a cystic space or mucocele. The mucus-filled, blind-terminating bronchus gives a variety of radiographic images in otherwise healthy individuals. Bronchial atresia is usually asymptomatic incidental finding. It may cause shortness of breath, cough or rarely infection. The etiology of bronchial atresia is not well known. It is a rare diagnosis as most of the patients are asymptomatic.

CASE REPORT

A 22-year-old female normotensive, non-diabetic, and non-smoker presented in the out-patient department of our institute with severe left-sided chest pain of one month duration. Pain was radiating to the back and ipsilateral shoulder. There was no history of expectoration or breathlessness. She had no history of recurrent chest infections or hospitalisations and denied history of any chronic disease. Moreover, she did not give any history suggestive of congenital anomalies in childhood. On examination, no visible swelling or tenderness was noted. However, chest wall movements and air entry were reduced on the left side. Chest X-ray postero-anterior view revealed loss of normal lung markings in the left upper zone ***(Fig. 1)****.*

Her electrocardiogram was normal and the cardiac marker enzymes were within normal limits. The routine blood in-

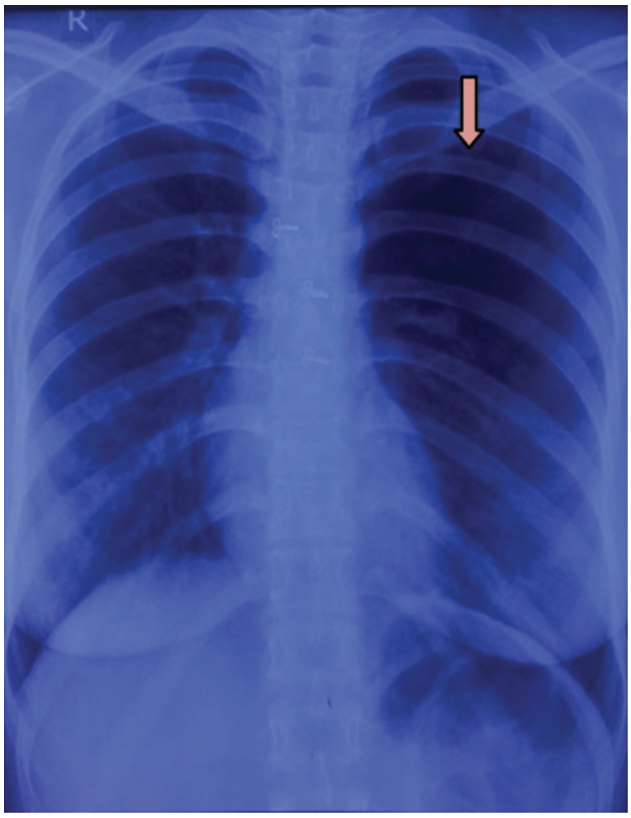

Figure 1. Chest X-ray PA view showing left hyper translucent lung.

vestigations like complete blood counts (Hb: 13 gm%, TLC: 6800 cells/µl, platelets: 3 lakh/mm^3), kidney function test (urea: 21 mg/dl, creatinine: 0.8 mg/dl) and liver function tests (S. bilirubin: 0.4 mg/dl, SGOT: 22 U/L, SGPT: 25 U/L) were also within normal limits.

Further work-up of the patient, including rheumatoid factor, antinuclear antibodies, antineutrophilic cytoplasmic antibodies, alpha-1 antitrypsin, serological tests for human immunodeficiency virus, sputum for acid-fast bacilli, pyogenic culture sensitivity and fungal culture did not reveal any abnormal findings. Spirometry showed a mixed pattern [FEV_1/FVC ratio: 65%, FVC: 2.24 l (69%), FEV_1: 1.45 l (84%)].

The high resolution computed tomography scan of the chest revealed a thinwall cavity in the left upper lobe ***(Fig. 2)****. The lesion was surrounded by an area of hyperlucent lung parenchyma and communicated with a segmental bronchus. The findings were suggestive of bronchial atresia. The patient was subjected to bronchoscopy, but no obvious abnormality was found.*

DISCUSSION

Bronchial atresia is usually a benign condition that is incidentally discovered in the second or third decade of life as seen in this case report. It is a rare anomaly and was first reported by Ramsay et al.[1] in 1953. Their reports documented mucoceles caused by septa which interrupted bronchial

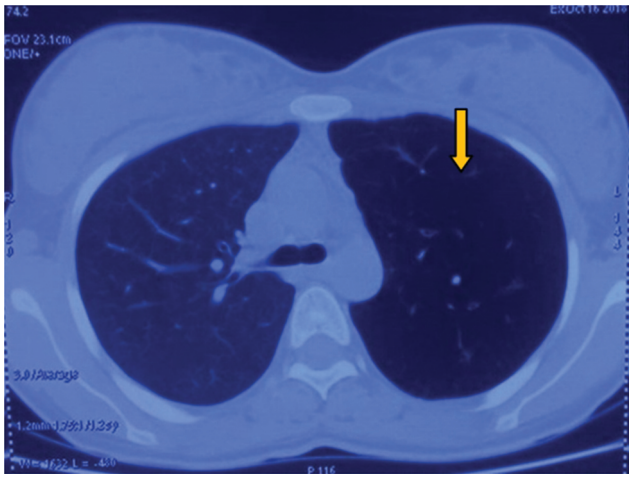

Figure 2. CT scan image showing left atretic segmental bronchus with an area of hyper-translucency in the adjacent lung field.

continuity. Belsey et al.[3] in 1958 reported a third case in a monograph on tension cysts.

It is characterized by a mucocele (or bronchocele) resulting from a mucus-filled, blind-terminating sub-segmental, segmental or lobar bronchus. There is hyperinflation of the isolated lung parenchyma adjacent to it. The etiology of bronchial atresia is unknown; however, focal bronchial interruption seems to occur before birth. The bronchial pattern is entirely normal distal to the site of stenosis. It has been suggested that the atresia is probably not a result of abnormal growth and development, but rather it is secondary to a traumatic event during fetal life.[4] The airway develops systematically, with the lobar bronchi, sub-segmental bronchi, and distal bronchioles appearing in the 5th, 6th, and 16th weeks of fetal development, respectively. One theory is that bronchial atresia is caused by intrauterine ischemia after the 16th week of gestation.[5] Other congenital lung anomalies that are known to develop earlier in embryogenesis occur with bronchial atresia. Another possibility is that the lesion occurs earlier, during weeks 4–6 of intrauterine development.[6]

Contrary to our case report, this disorder seems to have a male predominance, with an estimated prevalence of 1.2 cases per 100,000 males.[7] The majority of patients are asymptomatic and the diagnosis of bronchial atresia is made in young adult life following a chest radiograph that has been requested for some other reason. There are reports of prenatal diagnosis of bronchial atresia.[8] Less than one third of patients presented with symptoms of lower respiratory tract infection. A pneumothorax in a young female with bronchial atresia was reported by Gipson et al.[9]

On radiological analysis, the classic radiographic finding of bronchial atresia is a branching tubular or nodular area of increased opacity that extends from the hilum with surrounding hyperlucent lung parenchyma. The hyperlucent (emphysematous) lung area surrounding the mucocele is suggestive of dilated air space and focal parenchymal oligemia, secondary to intrapulmonary vascular compression and hypoxic vasoconstriction. Distal hyperinflation

is believed to be caused by collateral ventilation through intra-alveolar pores of Kohn, bronchoalveolar channels of Lambert, and interbronchiolar channels.[6] But it has been reported that pores of Kohn are not developed in infants, therefore, interbronchilar channels could be the major connection. Inspiratory and expiratory CXR films do not have much importance and no literature source suggests its role. There could be other radiological presentations. Water attenuation pseudonodular mass with well-defined margins in the parahilar region of the left upper lobe, which appeared to be a pulmonary nodule was reported by Fernandez et al.[10] Another case reported by Psathakis et al. presented as right upper lobe cavity lesion and diagnosed as bronchial atresia.[7]

Bronchoscopy may identify a blind-ending bronchus, but it may be normal as noted in this case. In clinical practice, however, any absence of a segmental or sub-segmental bronchus that is found by chance during bronchoscopy, in the absence of the characteristic radiographic features, may be considered as a normal anatomic variance of the bronchial tree rather than a bronchial atresia. Hence, CT is the most sensitive modality for diagnosing and MRI doesn't play much role. Microscopic finding reflects non-destructive overexpansion of alveoli, a finding that is distinct from the alveolar destruction seen in emphysema.

In this case report, a female patient presented with atypical chest pain with no other obvious findings. Hence, other differential diagnosis should always be kept in mind for thorough investigation. Peripheral hyper-translucency with hilar mass may be produced by internal or external bronchial obstruction: bronchial adenoma or carcinoma, mucoid bronchial impaction, vascular compression, sequestration, or bronchogenic cyst. Hyperlucency without associated mass may result from bronchial neoplasm, a foreign body (very few intrabronchial foreign bodies are radiopaque), abnormal bronchial cartilage or redundant bronchial mucosa, infantile emphysema, Swyer-James or MacLeod's syndromes, or pulmonary embolism. The most common causes of isolated perihilar masses are lymphadenopathy and neoplasia. The presence of a mucocele with adjacent hyperinflation helps narrow the differential diagnosis.

As discussed earlier, the majority of patients are asymptomatic, therefore no treatment is required. Surgical indications include recurrent and severe infection symptoms (such as pneumonias, dyspnea, cough, or hemoptysis) and medical treatment is ineffective. Malignant lesions cannot be excluded without surgery. Lobar resection and segmentectomy are preferred surgical modalities. However, the ultimate goal is to preserve as much normal lung parenchyma as possible to maintain pulmonary function.

CONCLUSIONS

Bronchial atresia is a rare congenital anomaly seen in young patients usually being asymptomatic or presenting with atypical symptoms. CT is the investigation method of choice, which provides the most accurate form of diagnosis. It is a benign condition, therefore surgical treatment or any aggressive method of treatment is not required until it leads to other life threatening complications.

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Одностороннее сверхпрозрачное лёгкое: редкий клинический случай бронхиальной атрезии

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Резюме

Пациентка молодого возраста с хронической болью в левой половине грудной клетки нуждалась в обследовании. Рентгенограмма грудной клетки предположила гиперпрозрачность слева. Компьютерная томография с высоким разрешением выявила атрезированный верхне-задний сегментарный бронх с областью гиперпрозрачности, указывающей на бронхиальную атрезию. Результаты фибробронхоскопии были в норме. Спирометрия показала умеренное ограничение с лёгкой обструкцией. Это редкая аномалия развития, характеризующаяся нарушением нормальной бронхиальной непрерывности. Чаще всего это вовлекает верхушечный сегментарный бронх левой верхней доли, как видно в нашем случае. Поскольку это доброкачественное состояние, хирургическое лечение или какой-либо агрессивный метод лечения не требуется. Однако вмешательство может потребоваться в случае опасных для жизни осложнений.

Ключевые слова

бронхиальная атрезия, врождённая аномалия, гиперпрозрачное лёгкое