Case Reports

Trans-mediastinal herniation of bulla: Semilunar sign

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ABSTRACT

Trans-mediastinal herniation of lung is seen occasionally but herniation of bulla across the mediastinum is rare. We report two cases with trans-mediastinal herniation of bullae leading to an unusual line appearing near the mediastinum. We propose the name ‘Semilunar sign’ for this radiological sign because of its resemblance to the half-moon shape.

KEY WORDS: Bulla, chronic obstructive pulmonary disease, trans-mediastinal herniation

INTRODUCTION

Bullae of the lung are air-filled spaces distal to terminal bronchiole that are more than 1 cm in size and lined with walls less than 1-mm thick.[1] Compressed lung parenchyma forms the wall of the bullae. In primary bullous disease the underlying lung is healthy and in secondary bullous disease the underlying lung is diseased. The most common cause of secondary bullous lung disease is tobacco smoking and chronic obstructive pulmonary disease (COPD). Other associated risk factors include inorganic dust inhalation, marijuana smoking, intravenous drug abuse, serum alpha-1 antitrypsin deficiency, sarcoidosis, Ehlers-Danlos syndrome[2] and Marfan's syndrome.

Herniation of bulla may occur across various sites including neck,[3] thoracotomy sites[4,5] and mediastinum. Trans-mediastinal herniation of bulla across the mediastinum is rare. Herein, we present two cases with an unusual line appearing near the mediastinum due to trans-mediastinal herniation of bulla. This sign has not yet been reported in literature and we propose the name ‘Semilunar sign’ because of its resemblance to half-moon shape.

CASE REPORTS

Case 1
A 58-year-old male presented with complaints of progressive shortness of breath for 10 years and heaviness of chest since 3 years. He had history of voluntary forceful coughing to bring out expectoration. He was hospitalized 9 months back for pneumonia. He smoked for 30 pack years and quit 8 years back. He was a teacher by occupation and history of travelling on motor-bike for 40 miles per day since 30 years.

His chest X-ray revealed hyperinflated lung fields, left upper zone fibrocalcified lesion and a thin line extending from left upper zone to middle zone [Figure 1]. His computed tomography (CT) of the chest revealed a bulla in the right lung extending across the mediastinum below the sternum [Figure 2]. The compressed pleura produced the thin line on the radiograph and we propose the name ‘Semilunar sign’ for the same. Forceful coughing and daily physical strain were probably responsible for the emphysematous bulla to herniate across the mediastinum.

The patient refused for lung volume reduction surgery and was given bronchodilators for his dyspnea. The possible risk of pneumothorax was also explained to the patient. He responded well to bronchodilators and is in close follow-up.

Case 2
A 56-year-old male presented with progressive left side chest pain for 3 months. He had been receiving antitubercular treatment for left sided pleural effusion since 3 months but did not have improvement with the
same (therapeutic pleural aspiration done twice in past). Past history was significant for history of pulmonary tuberculosis for which he took anti-tubercular treatment. He was an ex-smoker with a 66 pack year smoking history and he quit 3 years back. He was a gem-stone worker (30 yr) and since past 2 years was managing construction sites.

His chest X-ray revealed bilateral upper zone fibrotic lesions, small left hydropneumothorax and a thin line across the left hilum [Figure 3]. CT of the chest revealed a bulla in the right lung extending across the mediastinum below the sternum [Figure 4], left sided hydropneumothorax and a pleural based lung mass in left lower lobe. The compressed parenchyma and pleura of both the sides was responsible for the line appearing on the chest X-ray. Subsequently CT-guided fine needle aspiration (FNAC) of the left lung lesion revealed bronchogenic carcinoma. The patient received chemotherapy for bronchogenic carcinoma. He partially responded to the chemotherapy.

DISCUSSION

Occasional collapse of ipsilateral lung causes contralateral lung to hyperinflate and herniate toward the collapsed lung. However, herniation of bulla has been described rarely. There have been six cases of trans mediastinal herniation of bullae reported till now.[2,6-10] Eveloff et al.[8] had reported trans mediastinal herniation of a bulla after the patient was put on the mechanical ventilator due to intrinsic positive end expiratory pressure (PEEP). Herniation of bulla posterior to the trachea has been reported post-Heimlich maneuver.[10] However, cases reported by Singh et al.[7] and Akhtar et al.[9] did not have any immediate precipitating factors for herniation. Chronic obstructive pulmonary disease (COPD) was the underlying lung disease responsible for the bullae in all except one case. Ehlers-danlos syndrome was the cause of bullous lung disease in the case reported by Safdar et al.[2] Both our patients had predisposing factors for formation of bullae and their subsequent herniation. The first case had history
of forceful cough, smoking, and physical exertion. Second case had history of exposure to inorganic dust and smoking and subsequent coughing.

The presenting complaints include cough, dyspnea, dysphagia or exacerbation in symptoms of COPD. Radiology plays an important role in the diagnosis. Presence of the semilunarsign on chest X-ray may provide a clue to the presence of trans-mediastial herniation of bulla. Though the line was evident in the chest X-ray of a previously reported herniated bulla, it was not elaborated and named. This semilunar-shaped line is probably composed of compressed lung parenchyma and pleura of both sides. The diagnosis is confirmed with the help of CT chest. Azygous fissure on right side, fibrotic bands, and artifacts may also present with similar line near the mediastinum and may mimic this sign of trans-mediastial herniation.

Management may be conservative and involves treatment with bronchodilators. Surgical intervention is indicated in cases with severe dyspnea and bullae larger than 30% compressing the adjacent lung fields. Successful bullectomy was done in three of the reported cases. In case of herniation of bullae in patients on mechanical ventilator, reduction in PEEP may alleviate the symptoms and improve radiological findings.

**CONCLUSIONS**

- A careful look at the chest radiology may reveal yet unknown signs that provide clues to various diseases
- Thin line across the mediastinum called as the ‘Semilunar sign’ may be a clue to trans-mediastial herniation of bullae
- Azygous fissure on the right and fibrotic bands bilaterally may mimic this sign and thus have to be kept in the differential diagnosis.

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