Spontaneous regression of multiple emphysematous bullae

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The etiology of emphysematous bullae is not well known; thus, the management of bullous lung disease is often difficult because of concomitant underlying emphysema and coexisting cardiac conditions. The present paper reports a case in which near complete resolution of multiple emphysematous bullae in the right lung occurred spontaneously. This case is of interest, not only because of the rarity with which spontaneous regression of multiple emphysematous bullae has been reported in the literature, but also because of the accompanying dramatic improvements in radiological results and pulmonary function of the patients.

Key Words: *Bullous lung disease; Emphysematous bullae; Vanishing lung syndrome*

Régression spontanée de multiples bulles d'emphysème

RÉSUMÉ : On ne connaît pas vraiment l'étiologie des bulles d'emphysème et, par conséquent, la prise en charge de l'emphysème bulleux est souvent très difficile, car il est associé à un emphysème sous-jacent et à des affections cardiaques. Le présent article rapporte un cas où une résolution presque complète de multiples bulles d'emphysème dans le poumon droit est survenue spontanément. Ce cas est d'intérêt clinique, non seulement du fait de la rareté des cas de résolution spontanée de multiples bulles d'emphysème rapportés dans la littérature, mais aussi à cause de l'amélioration spectaculaire des résultats radiologiques et de la fonction pulmonaire que l'on a observée chez les patients.

Gradual expansion of emphysematous bullae (1) is commonly observed, but spontaneous resolution of bullae, on the other hand, is extremely rare. An increase or decrease in the size of bullae may be caused by a varying obstruction of airways; enlargement may be due to a check-valve effect in airways communicating with bullae and shrinkage may be due to absorption of air in the cavity. Giant bullous emphysema or 'vanishing lung' appears to be a distinct clinical syndrome, characterized by progressive development of large bullae usually in the upper lobe, often in young men, most of whom are smokers (2-7). Most cases reported are multiple emphysematous bullae, but a single dominant giant bulla may be seen (8,9). Vanishing lung syndrome includes both multiple emphysematous bullae and single dominant giant bulla occupying at least one-third of the hemithorax and compressing surrounding normal lung parenchyma. The case of a man in whom near complete resolution of multiple emphysematous bullae in the right lung occurred spontaneously is reported. The radiographic resolution of bullae was associated with improved pulmonary function.

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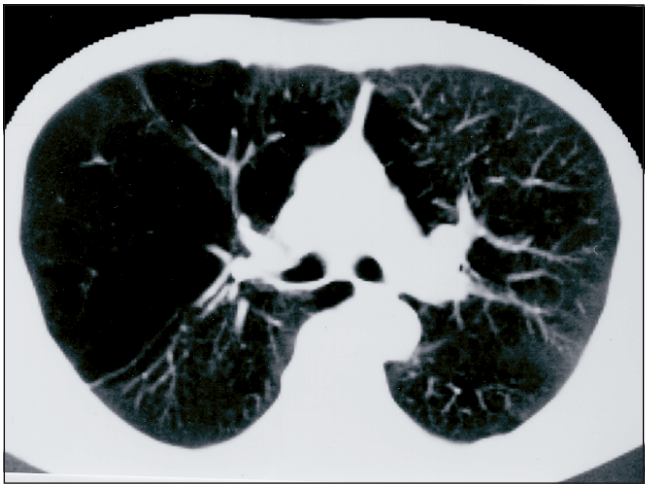

Figure 1) *Chest computed tomography scan on the first admission showing multiple emphysematous bullae in the right lung*

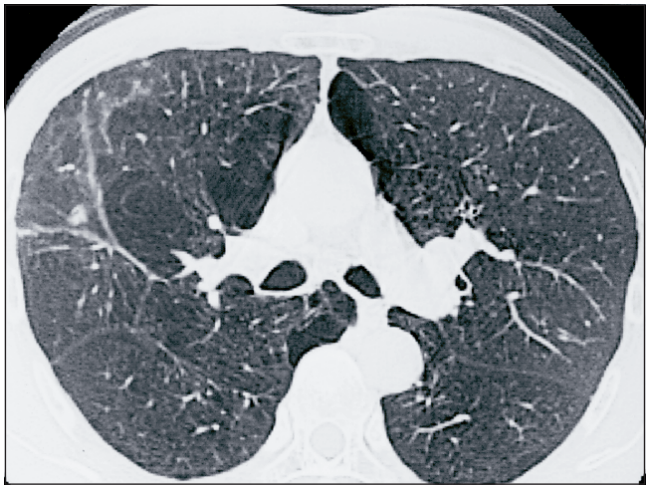

Figure 2) *Chest computed tomography scan of the third admission showing a remarkable resolution of bullous emphysematous change in the right lung*

CASE PRESENTATION

A 54-year-old farmer with a 60 pack-year smoking history presented to Kinu Medical Association Hospital with dyspnea that became progressively worse over a month. Clinical examination of the chest suggested a right pneumothorax, which was confirmed by chest roentgenogram. The rest of the examination was normal. Thoracostomy tube drainage was instituted, with a prompt improvement in symptoms, and chest roentgenogram revealed the lung had re-expanded to the chest wall. Pulmonary function tests demonstrated mild reduction of vital capacity to 2.80 L (predicted value 3.41 L) and severe air flow obstruction with a forced expiratory volume in 1 s (FEV_1) of 1.00 L (predicted value 2.38 L). Diffuse bullous emphysematous changes in the right lung were observed on a chest computed tomographic (CT) scan (Figure 1). The patient was discharged on no medication and returned to daily work on a farm, with minimal symptoms of dyspnea for two years.

However, he again developed increasing dyspnea on exertion. Chest roentgenogram and CT scan showed a recurrent pneumothorax on the right side. Tube thoracostomy was again performed resulting in a resolution of the pneumothorax. Three years after the second episode of pneumothorax, follow-up spirometric testing was obtained that revealed significant improvement in vital capacity to 3.64 L, whereas FEV_1 was 1.22 L. Chest CT scan confirmed a resolution of bullous emphysema compared with previous studies (Figure 2). No endobronchial lesions were identified on bronchoscopic examination.

DISCUSSION

Management of bullous lung disease is often difficult because of concomitant underlying emphysema and coexistent cardiac conditions, and surgical management is rarely indicated. The etiology of emphysematous bullae is not well known. Although many patients with bullae have a history of emphysema or asthma, this is not always the case. The natural history of pulmonary bullae is unpredictable, although gradual enlargement over time is commonly observed (10,11). Occasionally, rapid symptomatic expansion of bullae occurs for unknown reasons. Spontaneous regression in most reported cases has been associated with infection manifested by cough, sputum production and an air/fluid level on radiograph (1,12,13).

Other authors have hypothesized that inflammation may further obstruct already compromised bronchial communications with the bullae resulting in a closed space. Eventually fluid and then air resorption leads to regression of the bulla. Regression of a single dominant giant bulla has also been described in association with lung cancer (8), or rarely spontaneous resolution may occur (9). Our case is of interest, not only because of the rarity with which spontaneous regression of multiple emphysematous bullae has been reported in the literature, but also because of the dramatic improvements in the radiological picture and pulmonary function. Unlike earlier reports, this occurred in the absence of overt infection or tumour. Increases in spirometry, as documented in this patient, are sometimes seen following surgical bullectomy in selected patients (14,15).

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