Case reports

Spontaneous regression of a giant pulmonary bulla

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Abstract

Gradual expansion of a lung bulla is common and may be associated with debilitating pulmonary symptoms. The aetiology of bulla expansion is unclear. Spontaneous regression, on the other hand, is rarely observed. The case is presented of a man in whom near complete spontaneous resolution of a giant pulmonary bulla occurred. This event was associated with dramatic improvement in the radiographic picture and pulmonary function.

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Keywords: emphysema, giant bulla, spontaneous regression, pulmonary function.

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Pulmonary bullae occur as isolated abnormalities in otherwise normal lung tissue or, more frequently, in the setting of generalised emphysema.[1] Although long periods of stability are not uncommon, the natural history of pulmonary bullae is often characterised by gradual, progressive enlargement.[2 3] Spontaneous regression, on the other hand, is unusual. We present the case of an elderly man with severe bullous emphysema who experienced dramatic improvement in pulmonary function following spontaneous regression of a giant lung bulla.

Case report

A 70 year old man with bullous emphysema attended the pulmonary clinic for many years with symptoms of fatigue and dyspnoea on exertion. He denied frequent cough, sputum production, or wheezing. He had an 80 pack-year smoking history but had stopped in 1984. Progressive deterioration in his pulmonary function coincided with the gradual enlargement of a large right lung bulla. Diffuse emphysematous changes on computed tomographic (CT) scanning and a low diffusing capacity precluded consideration of bullectomy.

On routine follow up spirometric testing in October 1993 the patient's lung function was noted to be markedly improved (table). Repeat spirometric testing verified a 1·54 l increase in forced expiratory volume in one second (FEV_1). The chest radiograph showed a significant reduction in the size of the right lung bulla as well as a new 2 cm right lower lung nodule. Further evaluation included repeat CT scanning, bronchoscopic examination, and a lung perfusion scan. The CT scan confirmed a remarkable reduction in the size of the bulla compared with previous studies (figure). The 2 cm right lower lobe solitary nodule was not associated with mediastinal adenopathy. No endobronchial lesions were identified at bronchoscopy. The lung perfusion scan showed 52% of total flow to the right lung, significantly greater than the 20% observed in 1989. The patient subsequently underwent resection of a benign lung nodule.

Sequential pulmonary function tests performed during follow up period

	August 1989	*October 1990*	*September 1991*	*October 1993*
FEV_1 (l; % predicted)	1·34 (36)	1·31 (38)	0·72 (21)	2·26 (63)
FVC (l; %)	3·84 (82)	3·18 (71)	1·95 (44)	4·37 (96)
FEV_1/FVC	35	41	37	52
FEF_{25-75} (l; %)	0·40 (11)	0·45 (12)	0·27 (7)	0·89 (25)
TLC (l; %)	6·50 (92)	7·9 (113)	7·83 (111)	8·36 (120)
RV (l; %)	3·30 (125)	4·39 (166)	4·79 (182)	3·99 (148)
FRC (He)(l; %)	3·97 (99)	4·17 (102)	4·00 (94)	4·23 (111)
FRC (P1)(l; %)		4·88 (120)	5·55 (131)	
TLCO (ml/min/kPa; %)	4·09 (53)	4·29 (58)	2·98 (43)	4·59 (58)

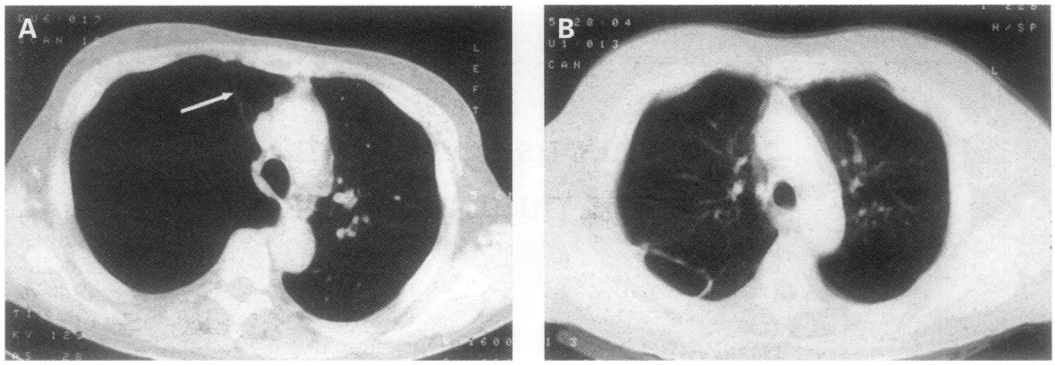
FEV_1 = forced expiratory volume in one second; FVC = forced vital capacity; FEF_{25-75} = forced mid expiratory flow; TLC = total lung capacity; RV = residual volume; FRC(He) = forced residual capacity (helium dilution); FRC(Pl) = forced residual capacity (plethysmography); TLCO = carbon monoxide transfer factor.

Discussion

Pulmonary "bullae" are pathologically dilated air spaces distal to the terminal bronchiole and are more than 2 cm in diameter in the distended state.[4] By convention they are called "giant bullae" when they occupy over one half the volume of the hemithorax.

The mechanism of expansion of giant bullae is uncertain.[5] The most widely held theory is that partial obstruction of the airways acts as a one way valve in an area of emphysematous lung and the resultant air trapping leads to gradual enlargement of the air space. Tension in the bulla may cause compression of neighbouring lung parenchyma, atelectasis, and even mediastinal shift. This theory was recently questioned by Morgan *et al*[6] who measured the pressures in the giant bullae of four preoperative patients and found that the pressures in the bullae mirrored the pleural pressure and were not "under tension". Based on this and other observations they postulated that bullae enlarge because of the relatively greater elastic recoil of adjacent lung parenchyma. In other words, surrounding lung tissue retracts away from the pathologically dilated air space.

The natural history of pulmonary bullae is unpredictable, although gradual enlargement over time is commonly observed.[2 3] Occasionally, rapid symptomatic expansion of bullae occurs for unknown reasons. Although spontaneous regression and even apparent radiographic resolution have been previously described, this is very uncommon. Most reported cases have been associated with infection manifested by cough, sputum production, and an air/fluid level on the radiograph.[7–9] The authors hypothesised that



Computed tomographic scan of the chest from (A) 1991 showing a giant bulla in the right lung with significant shift of the mediastinum to the left and (B) in 1993 showing a dramatic reduction in the size of the bulla. Note that the lung is almost completely re-expanded and the mediastinal shift is resolved.

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. If this mechanism is valid, one might speculate that other known causes of airway obstruction such as tumour, mucous plugging, or blood clot could also lead to shrinkage of the bulla. A retrospective review from Japan reported three cases of lung bulla regression associated with bronchogenic cancer, although no details on individual cases were provided.[10]

Our case is of interest not only because of the rarity with which spontaneous regression has been reported in the literature, but also because it was associated with such dramatic improvements in the radiological picture and pulmonary function. Unlike earlier reports, this occurred in the absence of overt infection or tumour. Re-expansion of compressed lung and reversal of mediastinal shift was accompanied by improved pulmonary blood flow. Substantial increases in FEV_1, as documented in this patient, are sometimes seen following surgical bullectomy in well selected patients. Our patient was not thought to be a good candidate for bullectomy because of diffuse emphysema, which underlines the difficulty in selecting patients for surgery.

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Sternocostoclavicular hyperostosis presenting with thoracic sinus formation

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Abstract

Sternocostoclavicular hyperostosis (SCCH) is a condition which is well described in the Japanese literature but is rare in Western Europe. It is characterised by pain and swelling in the upper anterior part of the chest, which tends to be progressive. A patient is described with bilateral chronic discharging sinuses over the anterior ends of the clavicles in whom the diagnosis appeared to be one of SCCH.

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Keywords: sternocostoclavicular hyperostosis, thoracic sinus, pustular psoriasis.

Case report

A 75 year old woman of Ashkenazi Jewish extraction was referred because of increasing shortness of breath. For five years she had suffered from recurrent clavicular problems. Initially this had been swelling, pain, and stiffness around the medial ends of the clavicles