Case reports

Spontaneous regression of a giant pulmonary bulla

David A Bradshaw, Kevin M Murray, Dennis E Amundson

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.

(Thorax 1996;51:549-550)

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

Department of Internal Medicine (Pulmonary Division)
D A Bradshaw
K M Murray

Department of Critical Care Medicine
D E Amundson

Department of Clinical Investigation
Naval Medical Center, San Diego, California 92134-5000, USA

Correspondence to: LCDR D A Bradshaw.
Received 11 October 1994
Returned to authors 16 January 1995
Revised version received 22 February 1995
Accepted for publication 28 February 1995

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 (FEV₁). 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.

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

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 (PI)(l; %)		4.88 (120)	5.55 (131)	
TLCO (ml/min/kPa; %)	4.09 (53)	4.29 (58)	2.98 (43)	4.59 (58)

FEV₁=forced expiratory volume in one second; FVC=forced vital capacity; FEF₂₅₋₇₅=forced mid expiratory flow; TLC=total lung capacity; RV=residual volume; FRC(He)=forced residual capacity (helium dilution); FRC(PI)=forced residual capacity (plethysmography); TLCO=carbon monoxide transfer factor.

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 FEV₁, 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.

The Chief, Bureau of Medicine and Surgery, Navy Department, Washington, DC, Clinical Investigation Program sponsored this report #84-16-1968-495, as required by HSETCINST 6000.41A. The views expressed in this article are those of the authors and do not reflect the official policy or position of the Department of the Navy, Department of Defense, or the United States Government.

- 1 Thurlbeck WM. Chronic airflow obstruction. In: *Pathology of the lung*. Stuttgart, New York: Thieme Medical Publishers, 1988:549-50.
- 2 Boushy SF, Kohen R, Billig DM, Heiman MJ. Bullous emphysema: clinical, roentgenologic and physiologic study of 49 patients. *Dis Chest* 1968;54:327-34.
- 3 Stone DJ, Schwartz A, Feltman JA. Bullous emphysema. A long-term study of the natural history and the effects of therapy. *Am Rev Respir Dis* 1960;94:493-507.
- 4 CIBA Guest Symposium. Terminology, definitions, and classification of chronic pulmonary emphysema and related conditions. *Thorax* 1959;14:286-99.
- 5 Murphy DM, Fishman AP. Bullous disease of the lung. In: Fishman AP, ed. *Pulmonary diseases and disorders*. New York: McGraw-Hill, 1988:1219-22.
- 6 Morgan MD, Edwards CW, Morris J, Matthews HR. Origin and behaviour of emphysematous bullae. *Thorax* 1989;44:533-8.
- 7 Rubin EH, Buchberg AS. Capricious behavior of pulmonary bullae developing fluid. *Dis Chest* 1968;54:546-9.
- 8 Rothstein E. Infected emphysematous bullae. Report of five cases. *Am Rev Tuberc* 1954;69:287-96.
- 9 Douglas AC, Grant IW. Spontaneous closure of large pulmonary bullae. A report on three cases. *Br Tuberc Dis Chest* 1958;33:335-8.
- 10 Tsutsui M, Araki Y, Shirakusa T, Inutsuka S. Characteristic radiographic features of pulmonary carcinoma associated with large bulla. *Ann Thorac Surg* 1988;46:679-83.

Thorax 1996;51:550-552

Sternocostoclavicular hyperostosis presenting with thoracic sinus formation

G E Wilson, C C Evans

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.

(*Thorax* 1996;51:550-552)

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

Cardiothoracic
Centre,
Liverpool L14 3PE,
UK
G E Wilson
C C Evans

Correspondence to:
Dr C C Evans.

Received 9 September 1994
Returned to authors
30 November 1994
Revised version received
20 February 1995
Accepted for publication
24 February 1995