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CASE REPORT

Solitary bronchial amyloid presenting with haemoptysis

S.M. Tariq, D. Morrison, K. McConnochie

Solitary bronchial amyloid presenting with haemoptysis. S.M. Tariq, D. Morrison, K. McConnochie.
ABSTRACT: Pulmonary amyloidosis can be classified into tracheobronchial diffuse alveolar-septal and nodular parenchymal forms [1]. Tracheobronchial amyloidosis can be further subdivided into diffuse and focal varieties. The latter is rare. We report a patient with a focal intrabronchial deposit of amyloid who presented with haemoptysis. The haemoptysis ceased following bronchoscopic removal of this deposit.
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Case Report

A 67 yr old lady presented with recurrent small haemoptyses in November 1988. She was a life-long smoker of 20 cigarettes daily. Clinical examination, a chest radiograph, and routine blood and urine investigations were normal. Sputum examination was negative. Lung function tests showed minimal airway obstruction (peak expiratory flow rate (PEFR) 301 $l \cdot min^{-1}$, 78% predicted; maximal mid-expiratory flow (MEF50)1.52 $l \cdot s^{-1}$, 43% predicted). A Mantoux test was positive (22mm).

Bronchoscopy revealed an ill-defined swelling with hyperaemic mucosa anteriorly in the bronchus intermedius. An attempt to biopsy this swelling led to brisk haemorrhage and the procedure had to be abandoned. At follow-up, the serial chest radiographs remained normal. However, the patient continued to have small haemoptyses.

Fig.1. – Amyloid demonstrated by Congo Red Stain. The amyloid substance giving a green anisotropic colour under the polarizing microscope (A.G.E. Pearse, Histochemistry, 1985).

A repeat bronchoscopy 4 mths later showed a pale, shiny, raised lesion (10 mm×3 mm approx.) with surrounding hyperaemia, on the anterolateral wall of the bronchus intermedius, which came off the bronchial wall in its entirety when biopsied.

Histological examination showed submusosal deposits of amyloid material confirmed by Congo red staining (fig.1). Further investigations were normal including serum gammaglobulins and protein electrophoresis. Urinary Bence-Jones protein was negative. Erythrocyte sedimentation rate (ESR) was 13 mm in 1 h and titres of rheumatoid and anti-nuclear factor were not raised. A rectal biopsy showed normal mucosa.

Ten months later, no further haemoptysis had occurred, and bronchoscopy showed no abnormality.

Discussion

Localized forms of pulmonary amyloidosis, which may be tracheobronchial or parenchymal, are associated with a good prognosis. The tracheobronchial form is often an incidental finding at bronchoscopy, especially in the elderly [2]. At times, tracheobronchial lesions distinct from the amyloid deposits, such as bronchiectasis or bronchial carcinoma, may be found to account for any respiratory symptoms and signs [3].

Extensive, diffuse tracheobronchial involvement with amyloidosis can mimic asthma [4], and, occasionally, it may be associated with hilar adenopathy simulating pulmonary sarcoidosis [5]. Localized lesions can cause atelectasis and/or infection. Haemoptysis is unusual [6], but can occasionally be life-threatening [7].

Solitary tracheobronchial lesions are rare, and are best treated by piece-meal bronchoscopic removal [8]. It is, however, prudent to follow-up these patients with repeated chest radiographs, especially if they present relatively late in life with a prolonged smoking history. Recurrence of the amyloid lesions has also been reported [9].

Our patient is unusual because the bronchoscopic appearance of her lesion had altered within 4 mths of presentation. It is possible, but unlikely, that the initial pathology of the bronchial lesion was distinct from amyloidosis and, this in turn, led to the disposition of amyloid material during ensuing months. We are unable to attribute her haemoptysis to any cause other than the solitary lesion of amyloid, as we found no other endobronchial pathology. This is also supported by the fact that her haemoptysis stopped following its removal, and she has since completed more than ten mths of follow-up in good health, with normal serial chest radiographs and a normal bronchoscopy.

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Amyloïdose bronchique solitaire se manifestant par des hémoptysies. S.M. Tariq, D. Morrison, K. McConnochie.
RÉSUMÉ: On distingue des formes trachéo-bronchiques, alvéolo-septales diffuses, et parenchymateuses-nodulaires, d'amyloïdose pulmonaire. L'amyloïdose trachéo-bronchique peut être divisée par ailleurs en varientes diffuse et focale. Cette derniére est rare. Nous rapportons l'observation d'une patient avec dépôt d'amyloïde intra-bronchique focal, qui a consulté pour hémoptysie. L'hémoptysie a cessé après ablation bronchoscopique de ce dépôt.
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