

Unresolved pneumonia due to endobronchial lipoma and actinomycosis

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ABSTRACT: We report an unusual case of unresolved pneumonia in a 59 yr old woman due to endobronchial lipoma and actinomycosis, two rare diseases that have never been described before in the same person.

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The differential diagnosis of unresolved pneumonia is wide and calls for complete and thorough investigation. There could be four main aetiologies: 1) infections such as tuberculosis, or due to viruses, or fungi that do not respond to the conventional antibiotics; 2) immune deficiency requiring prolonged antibiotic treatment; 3) lung infiltrate due to benign or malignant tumours; 4) obstructive process such as tumour or foreign body causing a post-obstructive pneumonia.

We present a case of unresolved pneumonia due to an endobronchial lipoma superimposed by actinomycosis.

Case report

A 59 yr old woman was admitted to Hadassah University Hospital because of a cough productive of a purulent sputum.

Four months prior to her admission, the patient began to suffer from a productive cough. At that time, chest X-ray revealed a segmental infiltrate in the anterior medial segment of the left lower lobe. A diagnosis of pneumonia was made and she was treated with erythromycin for ten days. There was a slight improvement but the cough persisted and the patient began to complain of weakness. Two days prior to admission, the patient had three episodes of haemoptysis. Her past history included a hysterectomy 19 yrs previously and a dental procedure four months before.

On admission, physical examination was unremarkable. Laboratory studies showed erythrocyte sedimentation rate of $20 \text{ mm}\cdot\text{h}^{-1}$, the white blood cell count was $5.9 \times 10^9 \cdot \text{l}^{-1}$ with normal differential, the haemoglobin was $13.6 \text{ g}\cdot\text{dl}^{-1}$, and the platelet count was $312 \times 10^9 \cdot \text{l}^{-1}$. Liver and kidney function tests were within normal limits. Blood and sputum Gram stain and culture were negative. Chest

X-ray revealed a segmental infiltrate in the anteromedial segment of the left lower lobe unchanged from the film recorded 4 months previously. Computerized tomography (CT) scan of the chest showed the same finding.

Bronchoscopy was performed and demonstrated an endobronchial yellow-white conglomerate mass in the anteromedial segment of the left lower lobe. The mass almost completely occluded the segmental bronchus. Microscopic examination revealed sulphur granules and a diagnosis of actinomycosis was made (fig. 1). Culture of the biopsies was negative. Penicillin G 12,000,000 units a day was started. After two weeks the cough and the purulent sputum subsided but the patient's chest roentgenogram was essentially unchanged.

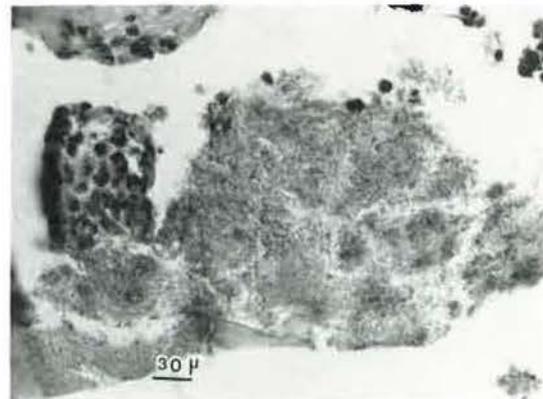


Fig. 1. - Actinomycosis sulphur granules surrounded by neutrophils. Haematoxylin and eosin $\times 470$.

A second bronchoscopy was, therefore, performed. A smooth, shiny endobronchial lesion was seen in the same location as the lesion in the previous bronchoscopy. With repeated bronchial biopsies the yellow material was

removed and a polypoidal, white, shiny, very regular mass was seen. It caused a 50–75% obstruction of the lumen of the involved segment. A bronchoscope was passed adjacent to the lesion and revealed a normal bronchial architecture distal to the mass. Pathological examination revealed an endobronchial lipoma (fig. 2). The patient continued high dose penicillin for a total of two months. At two years follow-up, the patient was completely well and her chest X-rays were normal.



Fig. 2. – Lipoma of bronchus. Note pseudostratified columnar epithelium at luminal surface at the right. Haematoxylin and eosin $\times 90$.

Discussion

Actinomycosis is a chronic suppurative infection caused by anaerobic, Gram-positive, branching, filamentous, non-acid fast bacteria which form sulphur granules. Actinomycosis is often discussed in mycology texts since the disease it causes mimics that of true fungi. However, actinomycoses are indisputably prokaryotic bacteria, lacking the nuclear membrane of eukaryotic fungi, and their growth is inhibited by common antibiotics but not by amphotericin B [1, 2]. The actinomyces are normal inhabitants of the oral cavity. Actinomycosis was relatively common in the early 1900's, but the frequent use of penicillin has reduced the frequency of clinically evident disease [3]. Men are affected 3–4 times more frequently than women [3]. This infection had a mortality rate of 75–90% in the pre-antibiotic era but is today associated with a cure rate of over 90% [3].

There are three main forms of the disease: cervical, gastrointestinal and thoracic. The thoracic disease, which our patient had, accounts for 15–50% of all cases [4–6]. This form of the disease is more common in smoking patients suffering from obstructive lung disease [3, 4, 6] and can also follow aspiration of mouth contents after dental procedures [4, 6]. Patients with thoracic actinomycosis are usually 30–60 yrs of age and have a chronic course, often with no specific complaints. A productive cough, low grade fever, dyspnoea, chest pain, haemoptysis, superior vena cava syndrome, and a mass in the chest wall, may all be presenting manifestations of thoracic actinomycosis. Mild leucocytosis, anaemia, and erythrocyte sedimentation rate up to 100 mm-h⁻¹ are com-

monly encountered [3, 6]. Delays in the presentation and diagnosis from one month to two yrs are common [6, 7]. Our patient presented with productive cough and haemoptysis.

The most characteristic laboratory findings are sulphur granules found in the sputum and excretions from external sinuses formed in the chest wall. These sulphur granules contain yellow to white partially calcified conglomerate masses of actinomyces, often associated with other bacterial organisms [1, 3].

Guidelines for appropriate antibiotic treatment of thoracic actinomycosis remain undefined because of the rarity of the disease and the consequent lack of controlled investigations. Furthermore, the organism is difficult to study *in vitro* because of the requirement for prolonged growth, permitting unstable antibiotics to decay [3, 6]. PEABODY and SEABURY [8], in 1960, emphasized use of intense and prolonged antibiotic therapy combined with a surgical approach for drainage of abscesses or radical excision of sinus tracts [9]. However, occasionally even extensive chronic disease responds to intravenous penicillin alone [4]. Some investigators recommend continuation of therapy for up to one year, since it is necessary to penetrate areas of fibrosis, and suppuration. In most cases, penicillin in adequate and even large doses, given over 2–3 months, is sufficient [1, 3, 10, 11]. In case of failure, sulphadiazine, streptomycin, erythromycin, chloramphenicol and tetracycline may be used [8].

Our patient suffered from actinomycosis of the lung in an area which was partially obstructed by lipoma. Lipomas are ubiquitous tumours, the majority occurring on subcutaneous tissue and being of cosmetic significance only. The retroperitoneum, mediastinum and gastrointestinal tract are the predominant sites of internal lipomas, although other sites, such as the central nervous system, larynx, bronchus or viscera, may be involved. Benign intrathoracic tumours are relatively rare with lipoma being among the least common [12].

Our case is unique in that the lipoma was located relatively peripherally and predisposed our patient to an infection with actinomycosis. This case suggests that when benign endobronchial lesions are complicated by superimposed infections, the judicious use of repeat bronchoscopic evaluations could provide clues for antibiotic treatment that would alleviate the need for thoracotomy with or without pulmonary resection.

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RÉSUMÉ: Nous décrivons un cas inhabituel d'absence de résolution d'une pneumonie chez une femme de 59 ans, par suite d'un lipome endo-bronchique et d'une actinomycose, deux affections rares qui n'ont jamais été décrites précédemment chez le même sujet.

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