

Case Reports

Pulmonary sequestration: a case report and review

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Introduction

Pulmonary sequestration is a relatively rare congenital abnormality. Diagnosis is often delayed and the condition may be confused with tuberculosis in parts of the world where this disease is common. We report a case presenting as recurrent episodes of fever with chest X-ray shadow. The final diagnosis was not made until the fourth hospital admission.

Case Report

A ten-year-old boy presented with 6 months of high grade intermittent fever, sweating and dull left sided chest pain. Since the age of 6 months he had repeated attacks of fever responding to antibiotics and antipyretics. Investigations at a District General Hospital revealed a lymphocytosis (3300 c.m.m.), ESR 60 mm and Mantoux test positive at 12 mm. The chest radiographs showed a soft shadow in the left lower zone. The fever responded within 1 week to Rifampicin, Isoniazid and Ethambutol but recurred 4 weeks later.

On referral to our unit, repeat investigations showed a normal blood count and unremarkable bronchoscopy and lung perfusion scan. The bronchography showed splaying of the left lower lobe bronchi around a uniform density soft tissue mass in the posterior sulcus. Ultrasonography demonstrated a soft tissue mass with fluid filled tubular structures above the left hemidiaphragm, suggesting a pulmonary sequestration. Thoracotomy confirmed an intralobar sequestration with a fully developed fissure and no identifiable blood supply from the aorta. The patient remains well 3 months after resection.

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Discussion

Pulmonary sequestration accounts for less than 7% of all congenital malformations of the lung (1). It is defined as accessory lung tissue of extralobar or intralobar type (1,2). The two forms may co-exist.

The extralobar type usually occurs on the left side of the chest (80% of cases) adjacent to the diaphragm and lower oesophagus (1,2), separated from the normal lung by separate pleura without any bronchial connection. Blood supply is from systemic circulation from aorta or intercostal arteries (1,2). It may remain asymptomatic and may be detected on routine chest radiography or may present as chest infection or its complications. It may present as regurgitation of food or cough related to eating because of the association with foregut communication. Sixty percent of extralobar sequestrations are associated with other congenital abnormalities including diaphragmatic defects, pectus excavatum, lung abnormalities, foregut duplications and congenital heart defects (2). Diagnosis of extralobar as well as intralobar sequestration is confirmed by bronchography. Aortography will reveal any systemic blood supply. The diagnosis may be aided by computed tomography, ultrasound and barium swallow. Elective excision is the treatment of choice.

Intralobar sequestration is more common than extralobar and also occurs more commonly on the left side. It shares a common pleura with the normal lung tissue. There is no bronchial connection but the tissue contains air which is thought to be from adjacent alveoli. Blood supply may be from the systemic circulation. This type may remain asymptomatic until adolescence or may present with a recurrent pneumonia, chest pain, chronic cough and haemoptysis. Incidental findings in both types of sequestration include tuberculosis (12 cases), squamous carcinoma (one case) and nocardial infection (one case) (1).

Aortography has been regarded as mandatory by some authorities to identify anomalous arterial supply and venous drainage pre-operatively (3-5).

References

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