ENDOBRONCHIAL HAMARTOMA CAUSING OBSTRUCTIVE CHANGES: A CASE REPORT

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Pulmonary hamartoma is a rare, benign, usually asymptomatic tumor of the lung presenting only as an incidental finding on routine chest X-ray. We report a case of an endobronchial hamartoma where the patient had to undergo pneumonectomy due to severe obstructive changes. The patient has been asymptomatic eight months after surgery. Bahrain Med Bull 1996;18(2):

The term "hamartoma" is used to describe certain tumors that result from unbalanced, coordinated, expansile but non-invasive growth of a normal constituent or constituents of an organ. Pulmonary hamartoma is a rare benign tumor of the lung with an incidence of about 0.25 % at autopsy¹. There are two distinct varieties of pulmonary hamartoma. The majority (about 92 %) of cases are "peripheral", situated close to the pleural surface of the lungs^{2,3} and are asymptomatic, being responsible for "coin lesions" on routine chest x-ray. They are thought to be derived from disordered growth of mesenchyme and are composed of undifferentiated mesenchyme and immature alveoli lined by cuboidal foetal-type epithelium⁴. The second variety (about 8 %) are "central" and contain islands of cartilage and cleft-like spaces lined in part by ciliated epithelium but show no attempt at alveolar tissue formation. They are thought to represent a disorder in growth of the bronchial bud⁴.

We report a case of an endobronchial (central) hamartoma causing bronchiectasis. Our case is interesting in that the large size of the tumour and the rapid progression of symptoms caused by it mimicked a malignancy and the severe obstructive changes necessitated a pneumonectomy.

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THE CASE

A 48 year old male was admitted to the MS Ramaiah Medical Teaching Hospital, Bangalore, India, with cough and progressive breathlessness since 6 months. There was no haemoptysis. He was a smoker but had quit smoking 3 years back. There was no history of any facial flushing, skin rashes, urticaria or other ailments.

General examination revealed nothing abnormal. On chest examination, the left side was dull on percussion with diminished breath sounds. Chest X-ray showed deviation of the trachea to the left side with collapse and consolidation of the left lung. Sputum culture showed normal flora of the upper respiratory tract. Bronchoscopy showed a tumour in the left main bronchus. All the haematological and biochemical parameters were within normal limits. A clinical diagnosis of bronchial carcinoid or adenoma was made. A left thoracotomy was done under general anaesthesia. At surgery, a yellow-white mass measuring 3 cm in size was seen occluding the left main bronchus. The left lung was totally collapsed. A left pneumonectomy was done. Post-operatively, the patient was given intravenous fluids, Amikacin (aminoglycosides) and Reflin (cephalosporin). From the second post-operative day chest physiotherapy was started. The patient was discharged on the ninth post-operative day. He has been asymptomatic when followed-up 8 months after surgery.

Pathological findings:

On gross examination, the resected specimen of the left lung showed a yellowishwhite mass measuring 3 x 2 x 1 cm with a smooth surface, occluding the main bronchus. The cut surface of the mass showed soft yellow and firm white areas with gritty foci of calcification. The lung appeared collapsed, measuring 14 x 10 x 3 cm with thickened, opaque pleura. The cut surface of the lung showed dilated bronchi and bronchioles extending up to the periphery (Fig 1). Some of the bronchioles contained pus. Areas of consolidation were seen.

On histopathology, the mass was lined partially by ulcerated bronchial epithelium and showed a mixture of cartilage with foci of calcification, bone with marrow, fat, mesenchyme and lymphoid aggregates (Fig 2, 3). The mass was traversed by a few recognizable bronchioles. The lung showed changes of bronchiectasis and pulmonary hypertension. The hilar lymph nodes showed reactive hyperplasia. The histological diagnosis of lipomatous hamartoma (synonyms: chondroid hamartoma, lipoma with a cartilaginous component, mixed mesenchymoma) was made^{1,2}.

DISCUSSION

In a series of 154 cases of pulmonary hamartoma studied by Van den Bosch et al² only 12 were endobronchial while the rest were peripheral (parenchymal) lesions. They found a male preponderance with the peak incidence in the sixth decade. In 43 out of 49 patients reported by Ramirez-Chavez et al³ the hamartomas were peripheral while only 6 were endobronchial with obstructive changes. Tomashefski⁴ compared 17 benign endobronchial tumors with 147 intraparenchymal hamartomas and concluded that the two were similar mesenchymal neoplasms occurring at different loci within the bronchial tree.

Bronchial derived hamartomas, in most reported cases, have shown a very slow growth over a period of years and remained clinically silent. Most lesions are less than 2.5 cm in diameter and can be treated by surgical removal of the tumour alone. Our case showed a rapid growth of the tumour which attained a fairly large size and caused a rapid progression of symptoms over a short period of 6 months. Thus, it clinically mimicked a malignancy. The location of the tumour and the severe obstructive changes necessitated a pneumonectomy which resulted in a cure of the patient.

CONCLUSION

Pulmonary hamartoma, though a benign tumor, can rarely grow rapidly and cause severe obstructive changes.

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