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CASE STUDY

A REPEATEDLY MISDIAGNOSED CASE OF MUCOEPIDERMOID TUMOR ARISING FROM MAIN BRONCHUS IN A CHILD

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ABSTRACT

Mucoepidermoid tumors of the bronchus are rare tumors. These tumors arise from terminal ducts of proximal tracheobronchial tree. Although mucoepidermoid carcinoma of the salivary gland is relatively common, mucoepidermoid carcinoma arising from the mucous glands of the bronchus is rare. These neoplasms usually exibits slow local growth and rarely metastasize to distant site. Though it is rare in children, it should be thought of when suspecting an intrabronchial mass in children. It usually presents with symptoms of airway obstruction and recurrent respiratory infections. Here we present a 8 year old child who presented with chronic cough, recurrent respiratory infections and occasional hemoptysis since last 3-4 yrs and was repeatedly misdiagnosed for Kochs and Congenital Cystic Adenomatoid Malformation. Post operatively on histopathological examination the diagnosis of mucoepidermoid tumor was made.

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INTRODUCTION

A 8 year old child presented with chronic cough, recurrent respiratory infections and occasional hemoptysis since last 3-4 yrs. He has been admitted on several occasions for respiratory infections and every time treated with a course of an antimicrobials. Initially he was misdiagnosed as Congenital Cystic Adenomatoid Malformation and later for pulmonary Koch's and put on anti-tubercular treatment for 8-9 months however his symptoms persisted. Laboratory blood investigations did not reveal any abnormality. On auscultation there was complete absence of air entry on left side.

IMAGING FINDINGS

Chest radiograph

A frontal chest radiograph revealed complete opacification of the left hemi thorax. Mediastinum was shifted towards left side and there was abrupt cut off of left main bronchus.

Computerised tomoghaphy of chest revealed an intrabronchial growth in left main bronchus 2.5- 3cm away from carina.

On contrast enhanced CT scan, the mass lesion showed homogenous moderate enhancement without any areas of necrosis or calcifications within.

The mass lesion was seen to extend beyond the bronchial walls without involvement of pleura, heart or any major vessel.

The left lung was completely collapsed and converted into multiple cystic areas probably due to chronic collapse and bronchoceles formations.

Few significantly enlarged lymph nodes were also observed in the mediastinum which showed significant enhancement on contrast enhanced scans.

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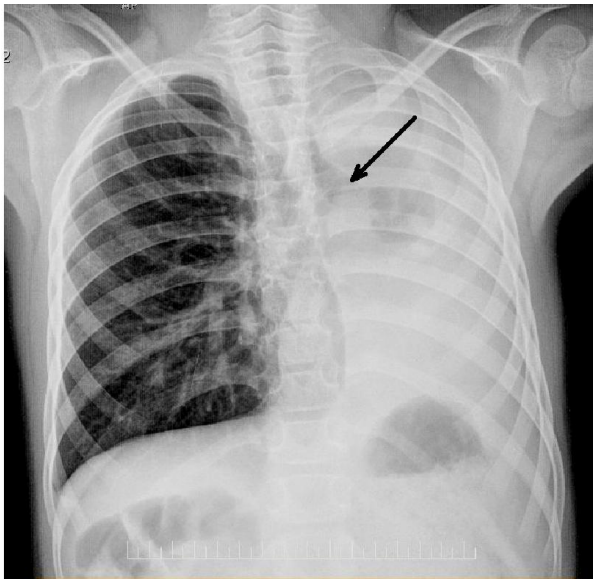


Figure 1. Frontal chest radiograph shows complete opacification of left hemitorax. Mediastinum and trachea shifted on left side. Notethe abrupt cut off of left main bronchus

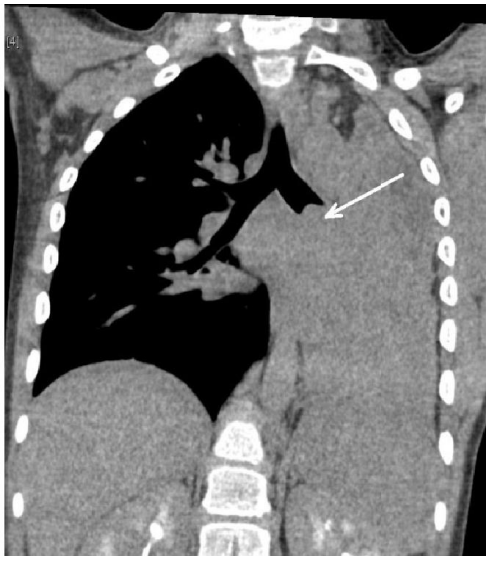
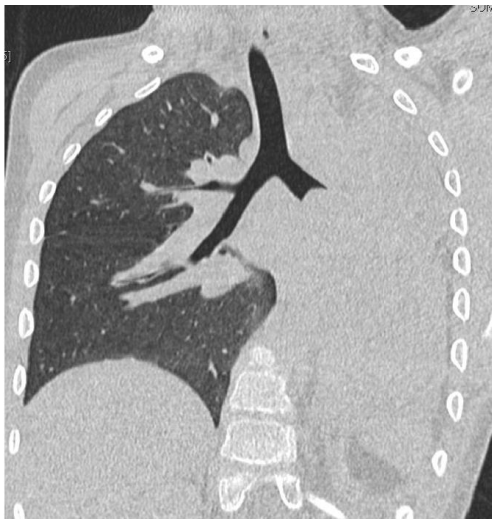


Figure 2 a.



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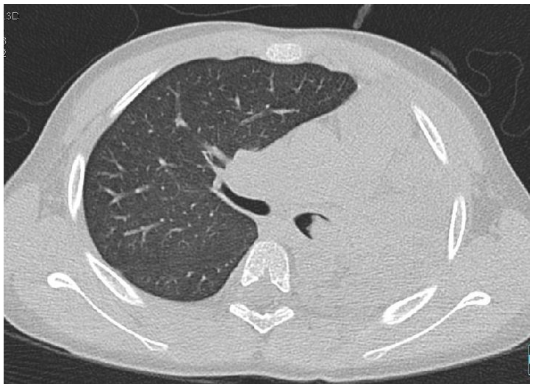

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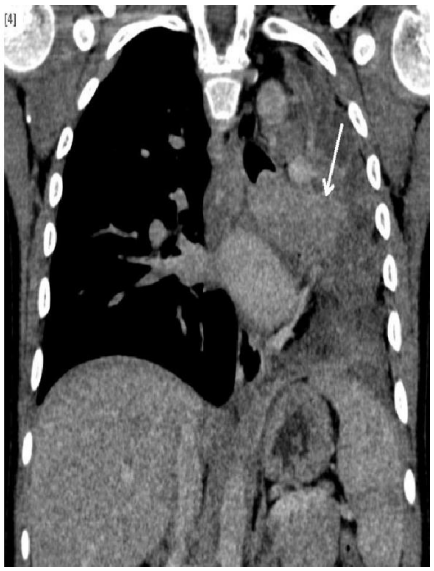

Figure 2 d.

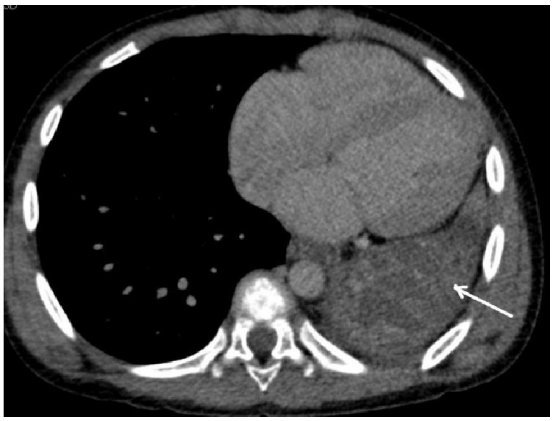

Figure 2 e.

Figure 2: (a). Unenhanced coronal reconstructed CT image shows isodense intrabronchial soft tissue mass lesion causing complete obstruction of its lumen with complete collapse of left lung. (b) Coronal CT image, lung window shows cut off of left main broncus. Normal right lung field. (c) Axial lung window CT image shows intrabronchial mass. (d) contrast enhanced coronal CT image shows homogenously enhancing intrabronchial mass lesion. No areas of calcifications or necrosis are seen within. The mass is seen to extending beyond bronchial walls (arrow) (e) Contrast enhanced axial CT image shows chronically collapsed lung with multiple cystic areas within probably due to bronchoceles formation

Endoscopic Findings

Endoscopy revealed an intrabronchial smooth surfaced polypoid mass lesion in the left main bronchus 2.5 cm away from carina and completely obstructing the bronchus. Core biopsies were taken from the growth and sent for histopathological evaluation.

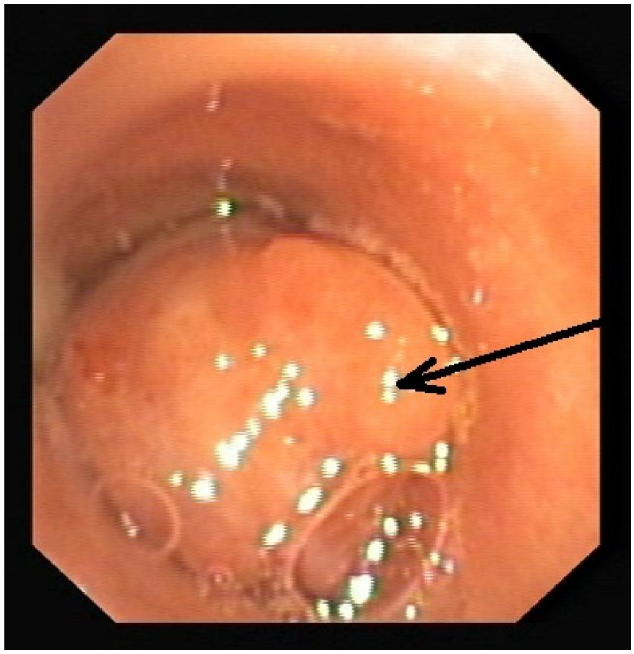

Figure 3a.

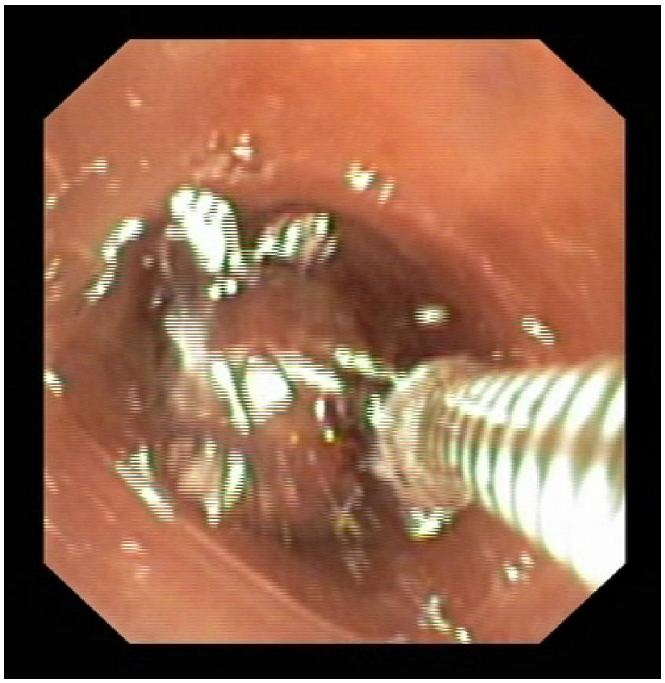

Figure 3b.

Figure 3. (a) endoscopic image shows smooth surfaced, lobulated intrabrochial mass with complete obstruction of the lumen (arrow) (b) endoscopic biopsy was performed and multiple tissue core were obtained and sent for histopathogical examination

Histopathologic correlation

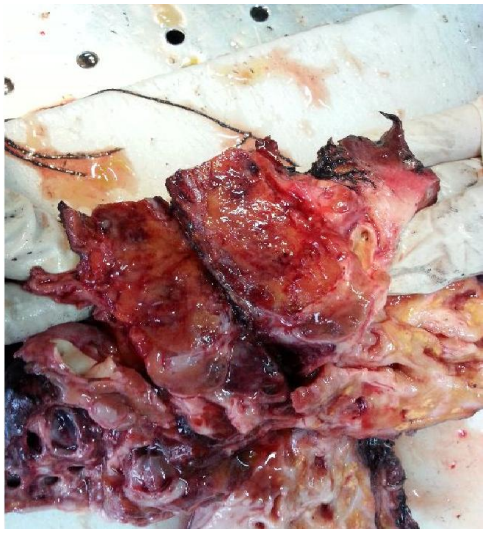

Figure 4a.

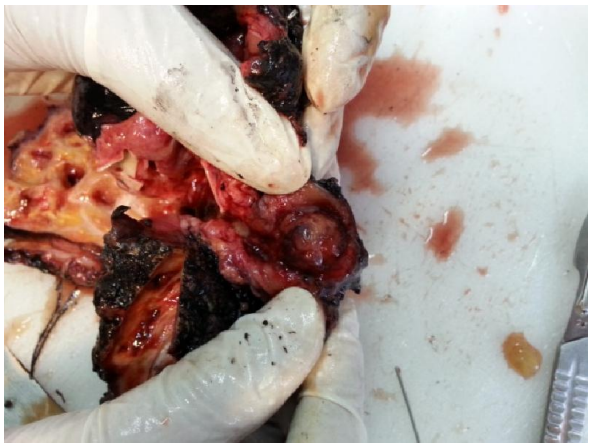

Figure 4b.

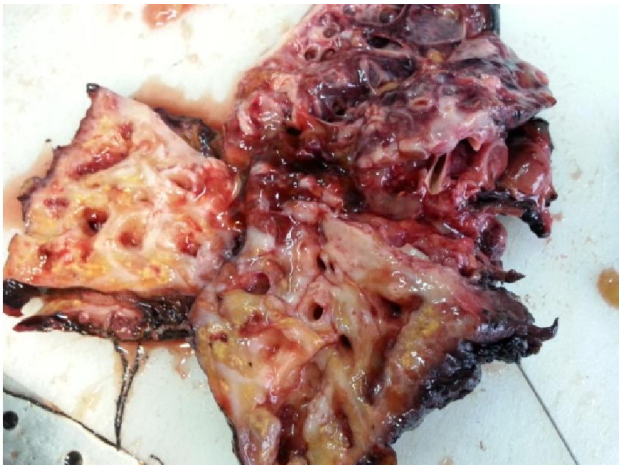

Figure 4c.

Figure 4. (a) Gross pathological specimen shows vertically cut open main bronchus with mass within. Bronchial walls are not seen separate from the mass lesion. (b) specimen shows intrabronchial polypoidal growth (c) Pathologic specimen shows collapsed lung with multiple cystic areas within

Microscopic examination showed unremarkable bronchial epithelium, with a neoplasm beneath, composed of irregular clusters of epithelial cells within a mildly inflamed fibrotic stroma. The cells showed moderately pleomorphic nuclei. Gland formation and mucocytes were seen (mucicarmine positive). Intranuclear cytoplasmic inclusions were present. Mitoses were few. Necrosis was absent.

Immuno Histo Chemistry was done and revealed following characteristics

CK7, CEA- positive
CK20, P63, TTF-1, Napsin A-Negative
Chromogrannin, synaptophysin-Negative

A primary adenocarcinoma and neuroendocrine tumor were excluded. So, histopathological analysis favoured Mucoepidermoid carcinoma of intermediate grade.

Management

After histopathology report of endoscopy guided biopsy, left thoracotomy and radical pneumonectomy was planned and performed. Intercoastal drain was put in left hemi thorax. The child gradually recovered and shifted to the ward.

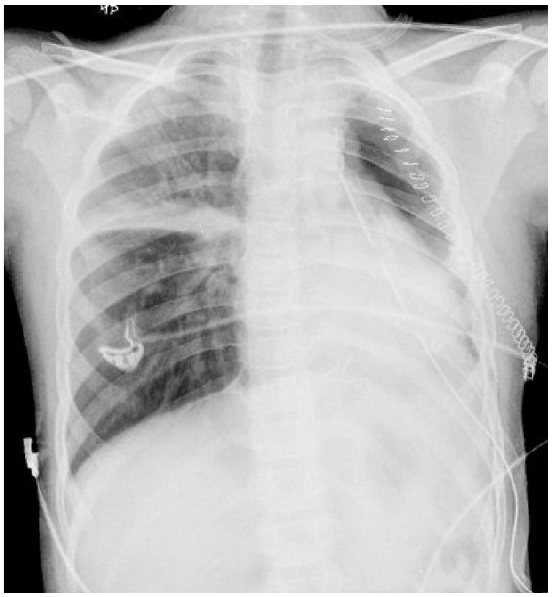

Figure 5. Post operative X-ray shows left radical pneumonectomy with ICD in left hemithorax

DISCUSSION

Although mucoepidermoid carcinoma of the salivary gland is relatively common, mucoepidermoid carcinoma arising from the mucous glands of the bronchus is rare. Mucoepidermoid carcinoma of the bronchus is extremely rare tumor.

Very few cases are reported in literature. The tumor is thought to account for 0.2% of primary lung cancers. (El-Sameed and Al Marzooqi, 2012) These neoplasms usually exibits slow local growth and rarely metastasize to distant site. (Conlan *et al*., 1978; Leonardi *et al*., 1978; Reichle and Rosemond, 1966; Sniffen *et al*., 1958; Wilkins *et al*., 1963) Though it is rare in children, it should be think of when suspecting an intrabronchial mass in children. It usually presents with symptoms of airway obstruction and recurrent respiratory infections.Other primary lung neoplasms, including adenocarcinoma, carcinoid, adenoid cystic carcinoma comprise the major differential diagnoses for mucoepidermoid carcinoma of the bronchus. (Reichle and Rosemond, 1966) The tumor is histologically classified as low or high grade malignancy depending upon nuclear atypia, pleomorphism, necrosis etc. (Reichle and Rosemond, 1966; Colby *et al*., 1995). A bronchoplastic sleeve resection can be done in case of low grade tumors which have excellent prognosis. (Reichle and Rosemond, 1966) Appropriate knowledge of the condition, proper imaging interpretation and high index of suspicion is important for a diagnosis of a mucoepidermoid carcinoma in a children with recurrent pneumonia and chronic lung collapse. As most of the tumors are slow growing early detection is essential for salvaging the lungs. In our case the child was chronically misdiagnosed as Koch' infection and congenital cystic adenomatoid malformation. Early diagnosis would have salvaged his one lung.

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