Medical and Research Publications
International Open Access

Case Report **Journal of MAR Pulmonology (Volume 3 Issue 2)**

There is More to Cough Than Meets the Eye

Dr Anusha.C.M[1], Dr. V. Kesavan Nair[*2], Dr, Rabitha[3]

1,3. Resident.

Corresponding Author: Dr V. Kesavan Nair, Professor, Department of Pulmonary Medicine, Shree Uthradom Thirunal Academy of Medical Sciences, Thiruvananthapuram, Kerala, India.

Received Date: July 05, 2021

Published date: August 01, 2021

Abstract

A 37-year-old nonsmoker male presented with persistent cough in the past 6 months who was previously treated for tuberculosis without relief. Chest X-ray suggestive of right middle lobe consolidation. CECT showed the right "bronchial cut off" sign with a calcified nodule and adjacent overdistended alveoli. There were also remnants of an old intestinal 'pull-through' surgery for stricture esophagus following a corrosive poisoning 15 years ago. Bronchoscopy revealed a smooth rounded endobronchial lesion in the right intermediate bronchus- not biopsied suspecting bronchial adenoma. Bronchial brushings revealed cellular smears consistent with bronchial adenoma. Here we present a diagnostic dilemma due to peculiar clinical, radiological and bronchoscopic pictures.

Introduction

Mucous gland adenoma of the bronchus is a rare, solitary, well-circumscribed, multicystic exophytic bronchial tumor. Mucous gland adenoma arises from the submucosal seromucous gland and ducts of proximal glands. The majority of cases arise from main, lobar, or segmental bronchi.1 Here we present a case of bronchial adenoma in a patient who was thought to have consolidation and who also happened to undergo esophageal pull-through surgery.

Case Presentation

A 37-year-old gentleman came to Respiratory Medicine OPD with persistent, intermittent cough of 6 months duration. He had no history of fever or any other constitutional symptoms. He gave a history of having had smear-positive pulmonary tuberculosis 3 years ago for which he had taken antituberculous treatment for 6 months and declared cured. He had undergone some surgical procedure around 15 years ago as he had dysphagia following a corrosive injury, the details of which were not revealed at the time of admission.

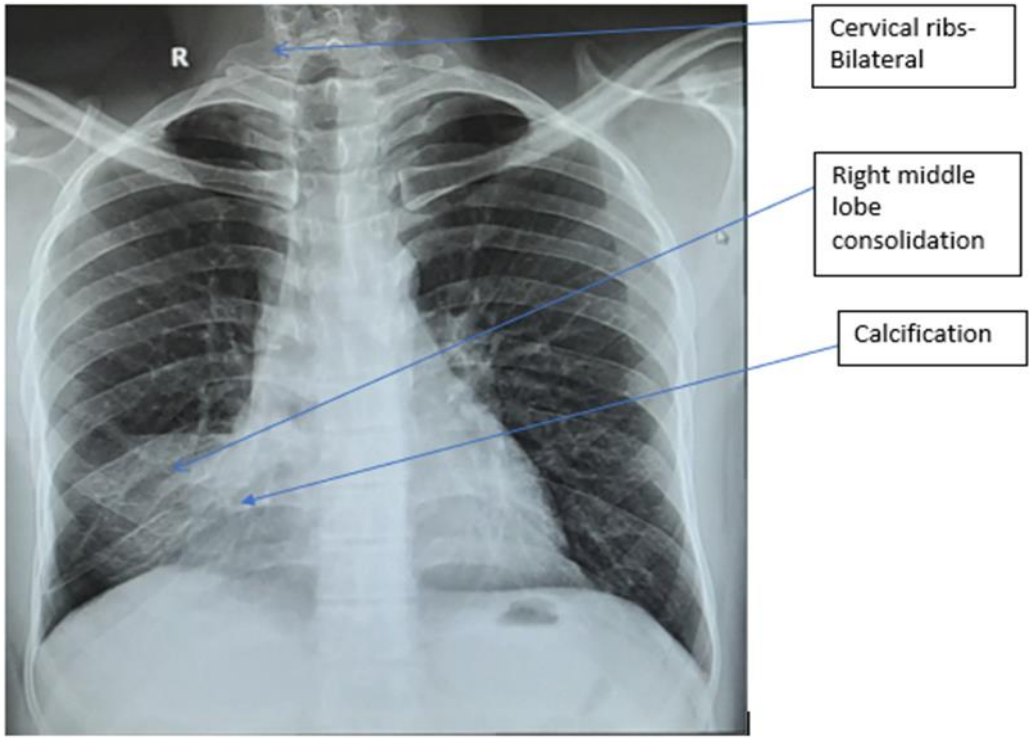


Figure: 1

On examination, he was moderately built and nourished with a hard swelling in the right supraclavicular area. Diminished air entry with crepitations in the right mammary region. Chest X-ray showed a non-homogenous opacity over the right mid and lower zones suggestive of middle lobe consolidation. There was also a calcified nodule seen through the consolidation. (Figure 1) Ultrasound neck showed an expanded anterior end of the first rib. Our probable diagnoses were Lymphadenopathy due to Tuberculosis / Malignancy with Middle lobe consolidation.

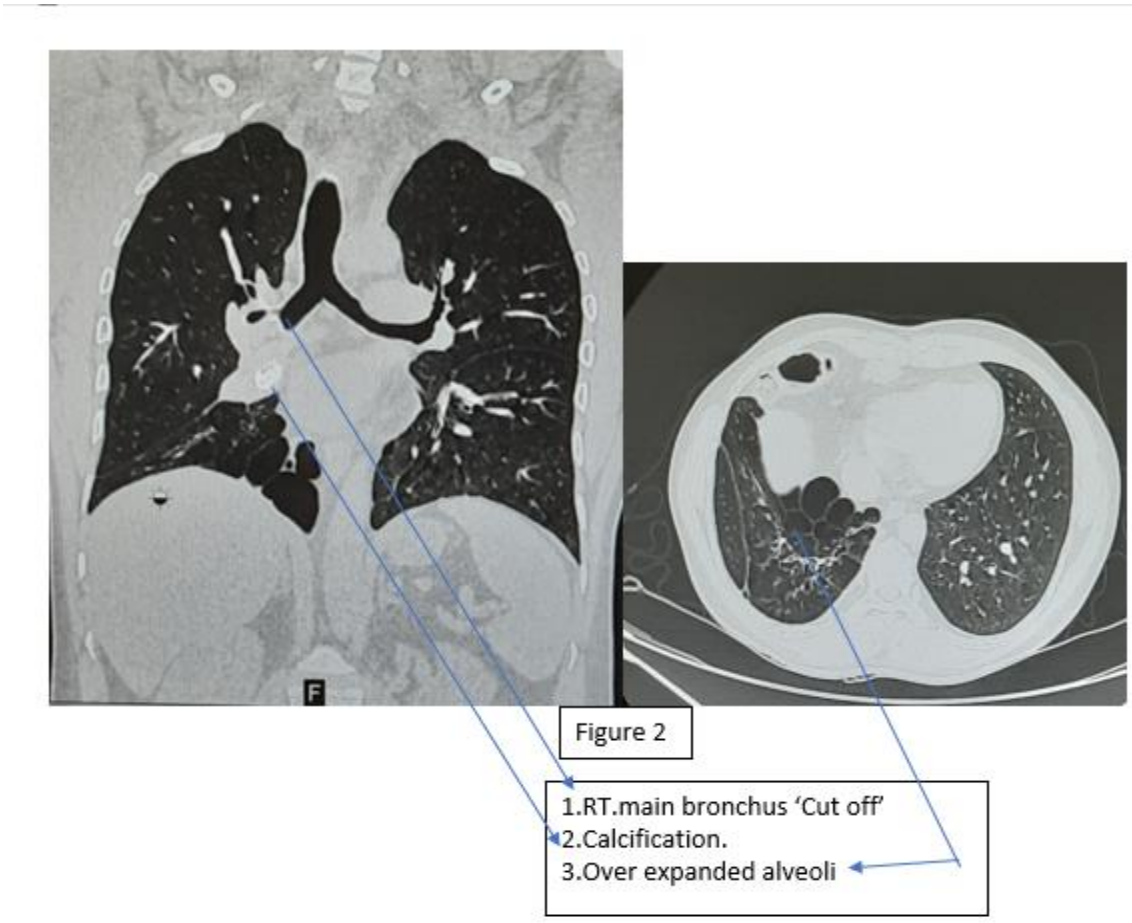


Figure 2

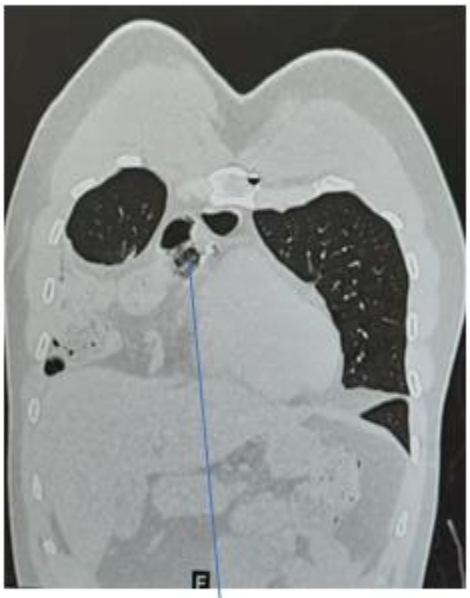


Figure 3

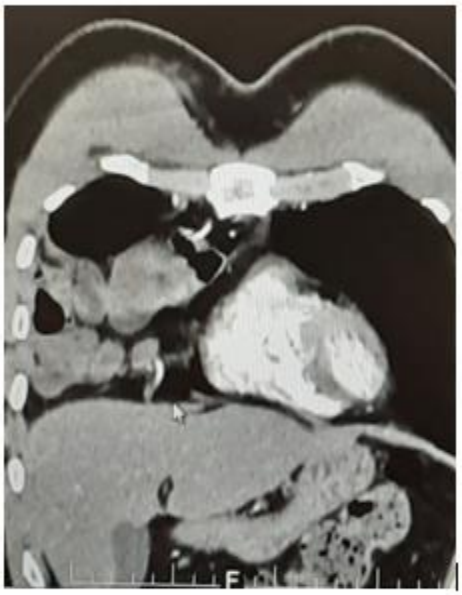


Figure 4

Large Intestinal 'pull through' for stricture oesophagus – old suicidal attempt 15 years back

Routine blood investigation showed mild leukocytosis, ESR was normal.USG neck showed the hard swelling was due to the anterior end of the 1st rib which was swollen. Chest X-ray (Figure 1) shows classic middle lobe consolidation with the calcified nodule. The patient was subjected to CECT thorax, (Figure 2) which showed a calcified, nodular lesion of size 2.5 x 1.5 cm within the right bronchus intermedius extending to the middle lobe bronchus with a distal fluid bronchogram, suspicious of bronchial carcinoid. Colonic interposition was also noted, with anastomosis below the hyoid bone to the distal end of the esophagus. - sequelae of old pull-through surgery following corrosive injury (Figure 3 and 4).

He was admitted for further evaluation. Sputum tests for acid-fast bacilli were negative.

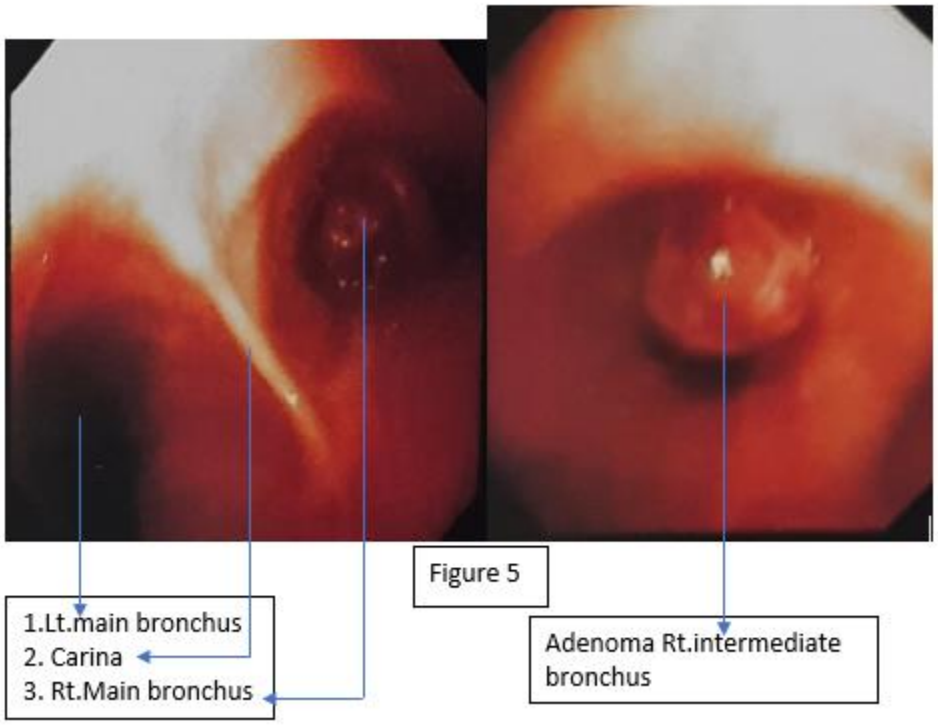
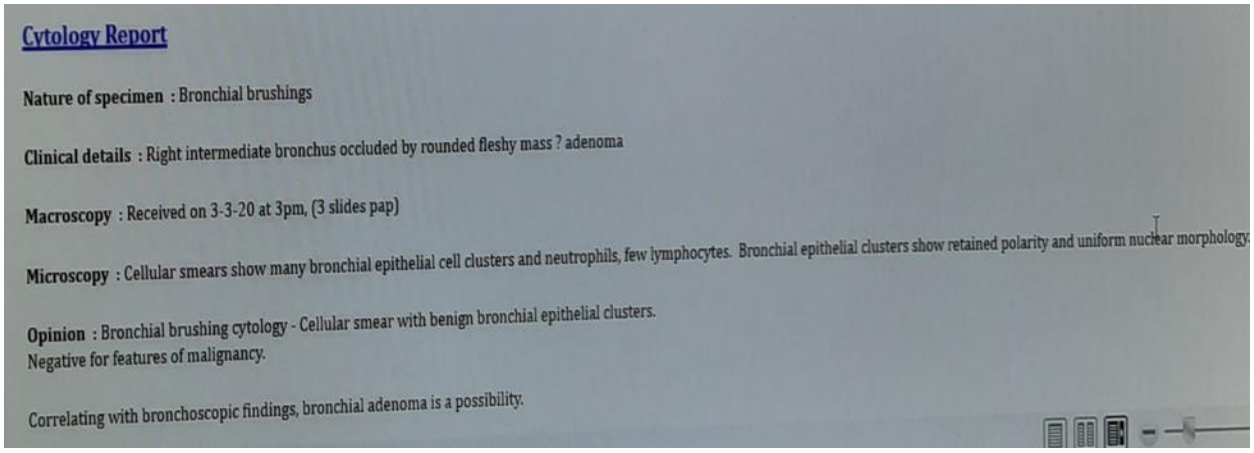


Figure 5



Cytology Report

Nature of specimen : Bronchial brushings

Clinical details : Right intermediate bronchus occluded by rounded fleshy mass ? adenoma

Macroscopy : Received on 3-3-20 at 3pm, (3 slides pap)

Microscopy : Cellular smears show many bronchial epithelial cell clusters and neutrophils, few lymphocytes. Bronchial epithelial clusters show retained polarity and uniform nuclear morphology.

Opinion : Bronchial brushing cytology - Cellular smear with benign bronchial epithelial clusters.
Negative for features of malignancy.

Correlating with bronchoscopic findings, bronchial adenoma is a possibility.

Figure 6

The patient was subjected to bronchoscopy. The right upper lobe bronchus was normal but the intermediate bronchus was almost completely occluded by a rounded fleshy mass. The bronchoscope could not be negotiated further. Plenty of purulent secretion was seen around the lesion. Biopsy was not attempted as the lesion was suspected to be an adenoma. Instead, bronchial brushings (Figure 6)

were taken from the right intermediate bronchus and sent for cytology evaluation which was reported as a possible bronchial adenoma (Figure 5).

The patient consented to have surgery for the removal of the lesion. The cardiothoracic surgeon agreed to proceed with the surgery. He underwent a Ga-68 DOTA TATE PET CT scan for somatostatin receptor imaging. A single densely calcified lesion obstructing the right bronchus intermedius with secondary obstruction to the middle lobe bronchus with partial collapse of the right middle lobe along with compensatory emphysematous changes in the basal segment of the right lower lobe was seen.

Right, bi-lobectomy (surgical removal of right middle and lower lobes) was done. The biopsy report came as a neuroendocrine tumor with extensive heterotopic ossification with reactive hyperplasia of lymph nodes. Immunohistochemistry (M4) was done and the lesion was diagnosed to be a low-grade neuroendocrine tumor. The case was taken for a discussion in the Multidisciplinary tumor board and he was advised to be on regular follow-up as it was a low-grade tumor.

Discussion

A pulmonary adenoma is an extremely rare benign tumor that was first described by Muller in 1882 as a tumoral lesion separate from carcinoma of the lung and first gave the nomenclature of bronchial adenoma arising from mucous glands. **[**3**]** The tumor arises equally in both males and females at any age (mean 52) including children. **[**1**]** Endobronchial lesions in main, lobar, segmental or subsegmental bronchi are common presentation. **[**4**]** Most of the patients present with recurrent cough, fever, unilateral wheezing, recurrent pneumonia which is often misdiagnosed **[**5**].**

In our case, patient had a recurrent cough for almost 6 months, wrongly diagnosed elsewhere and received treatment for tuberculosis. Radiology was pointing towards malignancy with a history of retracing to esophageal pull-through surgery for a previous suicidal attempt. Bronchoscopy showed a small rounded lesion which proved to be bronchial adenoma on brush cytology.

Pulmonary adenomas are classified as alveolar adenoma, papillary adenoma, mucinous cystadenoma and mucous gland adenoma. **[**6**]** Histologically the differential diagnosis includes low-grade mucoepidermoid carcinoma, primary adenocarcinoma, glandular papilloma, papillary adenoma. The mucoepidermoid carcinoma is a rare malignant tracheobronchial tumor but seen more commonly than bronchial mucous gland adenoma. The assessment of squamous and intermediate cells carefully will confirm the diagnosis **[**1,7**]**. Typical features of malignancy such as adenocarcinoma show cytological atypia, mitosis and infiltrative growth pattern along with strong TTF-1 immunopositivity in neoplastic glands **[**8**]**. Endobronchial growth with a fibrovascular core lined by ciliated or non-ciliated columnar cells and varying proportions of cuboidal and goblet cells are seen in glandular papilloma. **[**9**]** The parenchymal lesions are namely mucinous cystadenoma, papillary adenoma and alveolar cell adenoma.

Bronchial mucus adenoma is a benign lesion and similar histologically to mucinous cystadenoma. However, it occurs in the peripheral pulmonary parenchyma and is a true mucin-filled cyst that can be unilocular or multilocular **[**10**]**. A papillary adenoma, however, consists of fibrovascular cores lined by cuboidal or columnar epithelium and it is positive for TTF-1, where were alveolar adenomas are well-circumscribed adenomas unencapsulated multicystic masses with ectatic spaces lined by cytologically bland flattened, cuboidal and hobnail cells that show positivity with broad-spectrum keratins, TTF-1 and CEA **[**11**]**. Mucinous gland adenoma is a benign tumor and does not recur or metastatise. But few authors are of the opinion that it might have malignant proliferative potential **[**12. **]** Diagnosis remains important because the treatment modality varies for example for an adenoma, it is an endoscopic removal or sleeve/wedge resection whereas the malignant forms require more robust management **[**13**]**.

Benign tumors of the trachea bronchial tree remain the spotlight in pulmonary medicine. They can cause obstruction leading to pneumonia and cause recurrent cough as in our case. It may go unnoticed unless such cases are thoroughly evaluated. It can cause a diagnostic dilemma. Thorough work up radiologically, bronchoscopically, and pathologically remains crucial in diagnosis.

References

1. England DM, Hochholzer L. "Truly benign "bronchial adenoma". Report of 10 cases of mucous gland adenoma with immunohistochemical and ultrastructural findings". Am J Surg Pathol. 1995; 19(8):887-99

2. Flieder DB, Thivolet-Bejui F, Popper H. Mucus gland adenoma. In: Travis WD, Brambilla E, Muller-Hermelink HK, Harris CC, editors. "Tumours of the lung, pleura, thymus and heart". Lyon: IARC Press; 2004. pp. 85–100

3. Gilman RA, Klassen KP, Scarpelli DG. "Mucous gland adenoma of bronchus; report of a case with histochemical study of secretion". Am J Clin Pathol. 1956;26:151–4.

4. Merrick SH. "Tumors of the lung other than bronchogenic carcinoma". In: Baum GL, Crapo JD, Celli BR, Karlinsky JB, eds. Texbook of pulmonary diseases, 6th ed. Philadelphia: Lippincott Raven, 1998: 1389

5. Morini F, Quattrucci S, Cozzi DA, Tancredi G, Cicconi AM, Guidi R, et al. "Bronchial adenoma: An unusual cause of recurrent pneumonia in childhood". Ann Thorac Surg. 2003;76:2085–7.

6. Sekine I, Kodama T, Yokose T, Nishiwaki Y, Suzuki K, Goto K, et al. "Rare pulmonary tumours-Review of 32 cases". Oncology. 1998;55:431–434.

7. Gaissert HA, Mark EJ. "Tracheobronchial gland tumours". Cancer Control. 2006;13:286–94.

8. Yatabe Y, Mitsudomi T, Takahashi T. “TTF-1 expression in pulmonary adenocarcinomas”. Am J Surg Pathol 2002;26:767-73

9. Sasikumar S, Arjun P, Aleykutty, Geetha S. “Mucinous cystadenoma of the lung presenting as localised bronchiectasis”. J Assoc Physicians India 2005;53:566-7.

10. Travis WD, Colby TV, Corrin B, Shimosato Y, Brambillia E “Histological typing of lung and pleural tumors”. WHO international Histological Classification of Tumors.3rd ed. Geneva: Springer, 1999; 27

11. Burke LM, Flieder DB. Alveolar adenoma. In: Travis WD, editors. World Health Organization Classification of Tumours. Pathology and Genetics of Tumours of the Lung, Pleura, Thymus and Heart. Lyon: IARC Press; 2004. p. 82

12. Ishida T, Kamachi M, Hanada T, Yamazaki K, Ogura S, Isobe H, et al. “Mucous gland adenoma of the trachea resected with an endoscopic neodymium: Yttrium aluminum garnet laser”. Intern Med 1996;35:890-3.

13.Kwon EJ, Kim TS, et al. “Mucus gland adenoma presenting as a peripheral lung mass;a brief case report”. Korean JPathol. 2004;38:126-8