MAR Oncology & Hematology (2023) 6:1

Case Report

Solitary Endobronchial Plasmacytoma: A Rare and Easily Misdiagnosed Entity

Pooja Phalak[1], Shailee Mehta[2], Sanjiban Patra[2], Priti Trivedi[3], Prerna Walia[1]

1. DM Resident, Dept. of OncoPathology, The Gujarat Cancer and research Institute, Ahmedabad.

2. Assistant Professor, Dept. of OncoPathology, The Gujarat Cancer and research Institute, Ahmedabad.

3. Professor and Head, Dept. of OncoPathology, The Gujarat Cancer and research Institute, Ahmedabad.

***Correspondence to:** Dr. Priti Trivedi. Professor and Head, Department of OncoPathology, , Gujarat Cancer and research institute, New civil hospital campus, Ahmedabad-380016.

Received: 08 June 2023

Published: 01 July 2023

Abstract

Solitary endobronchial plasmacytoma is a rare type of extramedullary plasmacytoma. It can be misdiagnosed as other tumour like neuroendocrine tumour in view of its plasmacytoid and near uniform cytological features. Here, we report a case of a 51year old gentleman who presented with chest pain and dyspnoea. On radiology, an endobronchial mass was detected and biopsy was performed for tissue diagnosis which was inconclusive. Surgical resection was planned along with frozen section. Frozen section was reported as neuroendocrine tumour. But in paraffin section with the help of immunohistochemistry the final diagnosis was rendered as plasma cell neoplasm with extensive amyloid material and involvement of a single hilar lymph node. The workup for multiple myeloma was negative.

Keyword: *Extramedullary plasmacytoma, Amyloid, Solitary endobronchial plasmacytoma.*

Introduction

Extramedullary plasmacytoma (EMP) is a rare type of plasma cell neoplasm without bone marrow involvement or other systemic features of multiple myeloma, comprising of 3-5% of all plasma cell neoplasms, affecting the head and neck including upper aerodigestive tract in 80% of cases. [1,2] Primary endobronchial presentation is very rare.

Case Presentation

A 51year old gentleman with smoking habit, presented with complaints of chest pain, cough and dyspnea at our hospital. Physical examination was normal. A routine laboratory workup and ECG were within normal limits. Chest Xray revealed blunting of left CP angle with homogenous opacity in left middle and lower zone and obliteration of left dome of diaphragm; possibility of left mild pleural effusion appears likely.

A computed tomography (CT) scan of thorax, abdomen and pelvis showed 32 x 28 x 45mm lesion involving mediobasal segment of left lower lobe of lung, which is encasing the bronchus with intrabronchial growth. There were subcentrimetric nodes in preparatracheal, precarinal and subcarinal region. Rest of the CT scan was unremarkable. (Figure A) Bronchoscopy showed polypoidal intrabronchial lesion at left main bronchus with luminal narrowing. Bronchoscopy guided biopsy and fine needle aspiration cytology was performed and reported as non-representative material showing chronic inflammation and haemorrhage. Brochioloalveolar lavage was reported as negative for malignant cells. Subsequent CT guided biopsy was performed and it also showed fibrocollagenous tissue with admixed chronic inflammatory cells. Hence, patient was planned for surgical resection along with diagnostic frozen section. Frozen section showed monomorphic tumour cells arranged in nests and sheets separated by eosinophilic material. Tumor cells showed eccentric nucleus with eosinophilic cytoplasm. It was reported as neuroendocrine tumour on frozen section. (Figure B) Left lower lobectomy and lymphnode dissection (AP window, subcarinal and hilar) was performed. On gross examination, it was a unifocal well circumscribed polypoidal mass arising from left main bronchus measuring 55 x 35 x 30 mm in size which was grey white to brown in appearance on cut surface (Figure C). The lesion was 0.5cm away from pleural surface. Bronchial cut margin was 1cm away and vascular cut margin was 1.2cm away from lesion. Superior and inferior lung parenchymal resection margins were 2cm and 4cm away from lesion. Rest of the lung parenchyma was unremarkable. Three hilar lymphnodes were identified from the resection specimen with largest lymphnode measuring 0.8cm. One hilar, two subcarinal and two AP window lymphnodes were identified from separately sent containers, size of largest lymphnode was 1.4cm. On microscopy, the tumour showed diffuse sheets of plasmacytoid cells with intervening eosinophilic homogenous material. These tumour cells were arranged without any insular / trabecular pattern. Tumour cells showed eccentric nuclei, cartwheel like chromatin and amphophilic cytoplasm. At places, these cells were also showing bi and multinucleation. There was no evidence of necrosis or mitoses. (Figure D, E) On the basis of these findings, differential diagnosis considered were neuroendocrine tumour and plasma cell neoplasm. Immunohistochemistry was done. Tumour cells were immunoreactive for EMA, CD38, CD138 and immunonegative for synaptophysin, chromogranin, AE1, LCA, CD20 and CD3. Tumour cells were positive for lambda chain and negative for kappa chain. (Figure F,G, H, I) Congo red stain was performed for confirmation of amyloid material and it showed apple green birefringence on polarised microscopy.

(Figure J) So the final diagnosis was solitary plasmacytoma with amyloid material. One hilar lymphnode out of total eight submitted lymphnodes was involved by plasmacytoma. Further myeloma workup was advised. Serum light chain assay and kappa: lambda ratio were within normal limits. Serum protein electrophoresis revealed normal pattern with all major fractions within normal limit and absence of monoclonal band. So, the final diagnosis of solitary endobronchial plasmacytoma was considered. The patient had an uneventful postoperative recovery. On follow up, at three months interval, the patient was doing well without any further complaints.

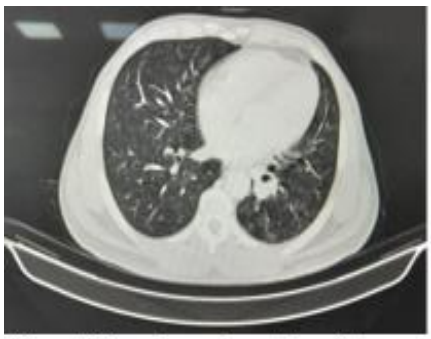

Figure A: CT scan image of an endobronchial mass

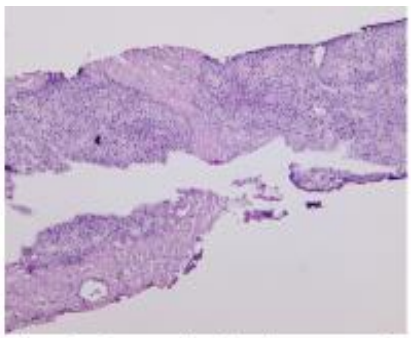

Figure B : Frozen section: Neoplasm composed of cells arranged in large nests separated by eosinophilic material [10x]

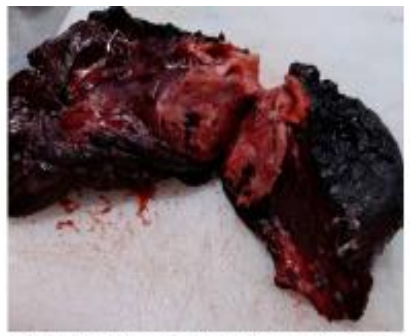

Figure C : Gross image: An Endobronchial mass

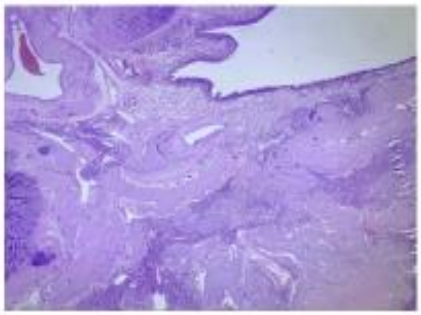

Figure D : Bronchus mucosa with underlying neoplasm composed of sheets of plasma cells with extracellular amyloid like material [10X]

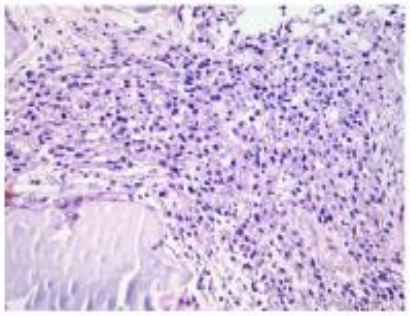

Figure E : Sheets of plasma cells with amyloid material [40X magnification]

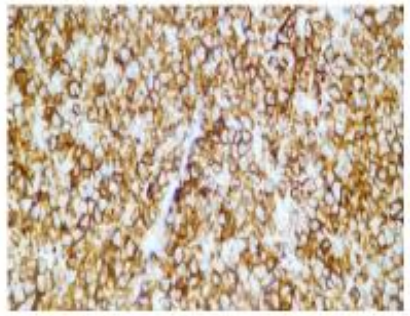

Figure F : CD38 immunohistochemistry

Figure G : CD138 immunohistochemistry

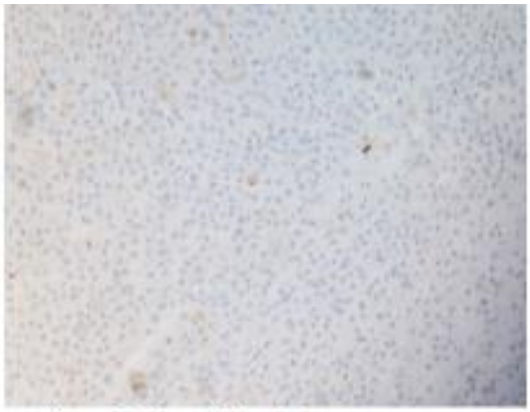

Figure H : Kappa light chain

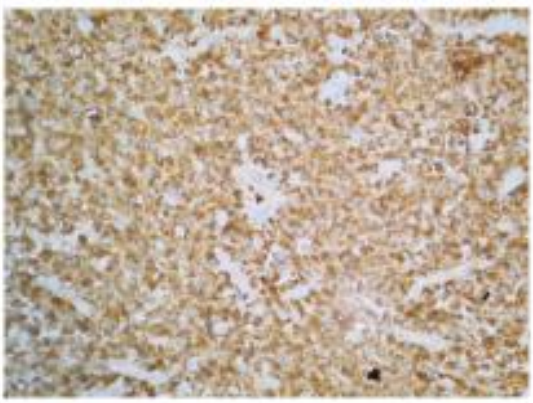

Figure I : Lambda light chain

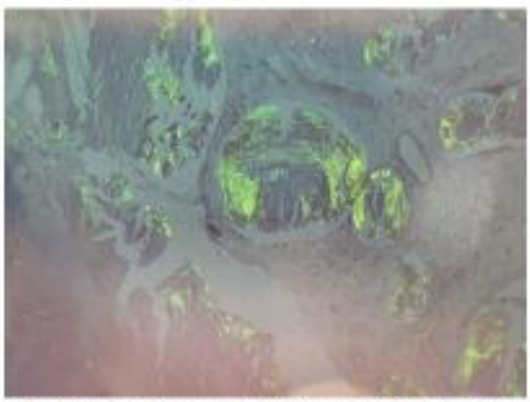

Figure J : Congo red stain on polarising microscopy: Apple green birefringence 200X magnification

Discussion

Extramedullary plasmacytoma is a rare type of plasma cell neoplasm and has two major subtypes, primary and secondary, defined by without and with marrow involvement by plasma cells, respectively. [3] Pulmonary involvement is itself rare and usually confined to the lung parenchyma[4], whereas solitary endobronchial presentation is extremely rare with only a few reported cases so far.[5-9]

A systematic case review of endobronchial plasmacytoma revealed that this tumour has an elderly male predilection with wide age range 17-86 years. Common presenting complaints were cough and dyspnea. In majority of cases, the tumour was predominantly involving right and left main bronchus. Most cases were diagnosed by bronchoscopy. Treatment options were either surgery (resection or endoscopic removal) or radiation or combined surgery and radiation.

On the basis of histomorphology, differential diagnosis considered were neuroendocrine tumour, lymphoma, plasma cell neoplasm and rarely poorly differentiated carcinoma. Neuroendocrine tumour is positive for synaptophysin and chromogranin. Lymphoma cells are positive for LCA and poorly differentiated carcinoma is positive for CK. However, in our case all the above markers were negative. A hilar lymphnode was also showing involvement by plasmacytoma, similar to the case reported by Wei et al [13] and Pierd et al [14].

As per review of literature, 22% cases of extramedullary plasmacytoma had recurred and 16% were progressed to multiple myeloma, but there have been no case reports of primary endobronchial plasmacytoma progressed to multiple myeloma [3]. There is one case report of secondary endobronchial plasmacytoma in patient with a history of multiple myeloma [16]. However, because of high prevalence of recurrence and progression of extramedullary plasmacytoma, patients should be carefully monitored.

Our case indicates that primary endobronchial plasmacytoma, a rare presentation of extramedullary plasmacytoma, should be considered in the differential diagnosis of an endobronchial mass. Radiological studies and optimal histopathological evaluation will confirm the diagnosis. Treatment strategy will depend on the patient's condition and the extent of the disease. Although there have been no reports of primary endobronchial plasmacytoma progressing to multiple myeloma, patients should be carefully monitored, given the possible progression of extramedullary plasmacytoma.

Reference	Age	Sex	Symptom	Location	Histomorphology and Immunohistochemistry	Diagnostic method	Treatment	Prognosis
Edelstein et al.[5]	47	M	Wheezing, shortness of breath	Left main bronchus	Plasma cells with kappa light chain restriction	Bronchoscopy	Endoscopic removal with laser ablation	No disease recurrence 8 months after treatment
Le Noir et al.[6]	54	F	Shortness of breath, wheezing	Right main bronchus	Plasma cells with kappa light chain restriction	Surgery	Surgical resection	No disease recurrence 1 year after treatment
Brackett et al.[7}	68	M	Productive cough, dyspnea	Left main bronchus	Plasma cells with lambda light chain restriction	Bronchoscopy	Endoscopic removal with laser ablation	No disease recurrence
Park et al.[8]	47	F	Blood tinged sputum	Right main bronchus	Plasma cells with lambda light chain restriction	Bronchoscopy	Endoscopic removal followed by radiation therapy	Not reported

Haresh et al. [9]	62	M	Dry cough, hemoptysis	Left main bronchus	Plasma cells with amyloid and lambda light chain restriction	Bronchoscopy	Radiation	No disease recurrence 4.5years after treatment
Kennedy et al.[10], 2 cases	17, 66	M M	Cough, hemoptysis Shortness of breath	Right middle lobar bronchus and Trachea	Plasma cells, no light chain study done	Bronchoscopy	Surgical resection	No disease recurrence 2 years and 10 months after treatment
Terzi et al.[11]	65	M	Dyspnea, non productive cough	Right main bronchus	Plasma cells, kappa light chain restriction. Preoperative biopsies negative	Surgery	Surgical resection	No disease recurrence 5 years after treatment
Zhang et al.[12]	48	M	Dyspnea	Trachea	Plasma cells with kappa light chain restriction	Bronchoscopy	Endoscopic removal with electrocautery snare followed by radiation therapy	No disease recurrence 6 months after treatment
Jong et al. [13]	86	M	Productive cough and dyspnea	Left upper lobar bronchus	Plasma cells with kappa light chain restriction	Bronchoscopy	Radiation therapy	No disease recurrence 6 months after treatment
Wei et al. [14]	42	M	Dyspnea	left lower lobe with mediastinal and hilar lymphadenopathy	Plasma cells with IgG lamba light chain restriction and 5 local lymphnodes metastasis	Surgical resection	Surgery	No disease recurrence 15 months after treatment
Piard et al. [15]	70	M	Dyspnea	Posterior part of the left main stem bronchus	Plasma cells with kappa light chain restriction and local lymphnode metastasis	Bronchoscopy	Surgery	No disease recurrence
Present case	51	M	Chest pain, cough and dyspnea	Left lower lobe bronchus	Plasma cells with lambda light chain restriction, single hilar lymphnode metastasis, amyloid deposition	Surgery	Surgery	No disease recurrence 3 months after treatment

Table 1: Literature review of primary solitary endobronchial plasmacytoma

Reference

1. Wang J, Yang X, Liu X, He T, Liu B, Yang L, Yuan F, Li J. Solitary extramedullary plasmacytoma in the lung misdiagnosed as lung cancer: A case report and literature review. Front Oncol. 2022 Aug 30;12.

2. D. M. Fletcher C, Diagnostic Histopathology of Tumors, Churchill Livingstone, London, UK, 5th edition, 2021.

3. Alexiou C, Kau RJ, Dietzfelbinger H, Kremer M, Spiess JC, Schratzenstaller B, et al. Extramedullary plasmacytoma: tumor occurrence and therapeutic concepts. Cancer. 1999;85:2305-14.

4. Nie S, Peng DC, Gong HH, Ye CL, Nie X, Li HJ. Primary pulmonary plasmacytoma: a case report introduction. World J SurgOncol. 2016;14:205.

5. Edelstein E, Gal AA, Mann KP, Miller JI Jr, Mansour KA. Primary solitary endobronchial plasmacytoma. Ann Thorac Surg. 2004;78:1448-9.

6. LeNoir B, Bruner ET, Denlinger CE, Gibney BC. Extramedullary plasmacytoma of the right main bronchus. Ann Thorac Surg. 2019; 108:e119–e20.

7. Brackett LE, Myers JR, Sherman CB. Laser treatment of endobronchial plasmacytoma. Chest. 1994;106:1276-7.

8. Woo Park C, Kim W, Jae Oh I, Sik Kim K, Duk Choi Y, Soo Kwon Y. Solitary extramedullary plasmacytoma presenting as an endobronchial mass. Intern Med. 2013;52:2113-6.

9. Haresh KP, Sharma DN, Prabhakar R, Naik KK, Mannan AA, Julka PK, et al. Plasmacytoma of bronchus treated by radical radiotherapy-a rare case with four and a half years follow up. JThoracOncol. 2007;2:980-2.

10. Kennedy JD, Kneafsey DV. Two cases of plasmacytoma of the lower respiratory tract. Thorax. 1959;14:353-5.

11. Terzi A, Furlan G, Zannoni M, Adovasio A, Gorla A. Endobronchial extramedullary plasmacytoma. Report of one case. Lung Cancer. 1996; 16:95-100.

12. Zhang X, Su L, Ran YG, Qie S, Liu C, Shi HY. Extramedullary plasmacytoma of the trachea: a case report and review of the literature. Medicine. 2018;97:e9594.

13. Park JI, Lee YY, Lee SS, Ahn JH. A rare case of primary solitary endobronchial plasmacytoma. Thorac Cancer. 2021 Mar;12(6):958-61.

14. Wei S, Li X, Song Z, Zhao H, Qiu X, Gong L, Chen J, Zhou Q. Primary endobronchial plasmacytoma involving local lymph nodes and presenting with rare immunoglobulin G lambda monoclonal gammopathy. Can Respir J. 2012 May-Jun;19(3):e28-30.

15. Piard F, Yaziji N, Jarry O, et al. Solitary plasmacytoma of the lung with light chain extracellular deposits: A case report and review of the literature. Histopathology. 1998;32:356–61.

16. Sunnetcioglu A, Ekin S, Bayram I, Ekinci O, Bugday IB. Endobronchial plasmacytoma in patient with multiple myeloma. ClinRespir J. 2017 Nov;11(6):1057-59.