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A rare solitary and endobronchial pulmonary hyalinising granuloma requiring bilobectomy

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Abstract

Pulmonary hyalinising granuloma is a very rare disease often presenting as multiple smooth rounded nodules within the lung parenchyma and mimicking metastatic disease. Solitary pulmonary hyalinising granuloma is an even rarer subgroup, and to our knowledge, there have been no endoluminal pulmonary hyalinising granulomas reported. A 36-year-old female non-smoker with no significant past medical history presented with a persistent cough and was found to have a right lower lobe bronchial lesion causing lower lobe obstruction. After multiple failed attempts at tissue diagnosis from both percutaneous and endobronchial biopsies, and with worsening haemoptysis, the patient underwent a right thoracotomy and lower bilobectomy. The histopathology was reported as a solitary endobronchial pulmonary hyalinising granuloma. Although benign in nature, tissue diagnosis can be difficult in these lesions, especially when presenting as a solitary mass in a central location. This report demonstrates that these lesions can also be found endobronchially necessitating parenchymal resection for diagnosis and obstructive symptoms.

Keywords

Thoracotomy, lung resection, thoracic surgery, pulmonary hyalinising granuloma

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Introduction

Pulmonary hyalinising granuloma (PHG) is a very rare disease first reported in 1964 by Benfield et al.[1] In 1977, Engleman et al.[2] went on to characterise the histology of these lesions as hyalinised lamellar collagen bundles in the presence of plasma cells, lymphocytes and histiocytes. The disease often presents as multiple smooth rounded nodules within the lung parenchyma and can mimic metastatic disease although the differential diagnosis includes amyloidosis, fungal infection and inflammatory myofibroblastic tumours.[3] Diagnosis is almost always made from surgical histology.[4]

These lesions can be found incidentally; however, they can also cause respiratory symptoms such as cough, haemoptysis, fever, chest pain and shortness of breath.[2] Occasionally patients present with recurrent chest infections. Radiological evaluation demonstrates rounded nodules that can contain calcium. Fluorodeoxyglucose–positron emission tomography (FDG-PET) is thought to demonstrate avidity in around 50% of cases.[3,5]

Few case reports have reported solitary PHGs, and to our knowledge, there have been no reports of endobronchial PHGs. We present the case of an extremely rare solitary and endobronchial pulmonary hyalinising granuloma requiring thoracotomy and bilobectomy to both diagnose and treat the symptoms caused by the lesion.

Case report

A 36-year-old female non-smoker with no significant past medical history presented with a persistent cough, recurrent lower respiratory tract infections and regular episodes of small volume haemoptysis to the thoracic surgical service. Chest radiographs and high-resolution computed

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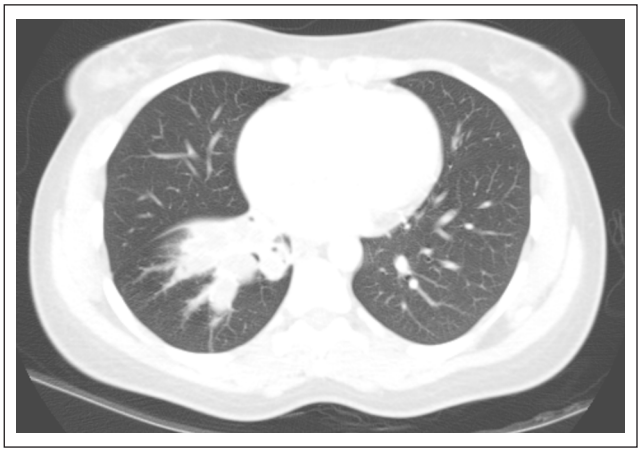

Figure 1. CT scans demonstrating the right lower lobe mass.

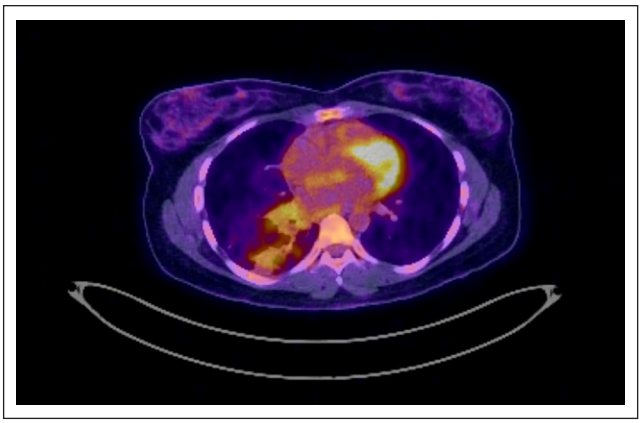

Figure 2. PET scan demonstrating PET avidity within the mass.

tomography (HRCT) demonstrated a soft tissue mass arising from the right lower lobe bronchus and abutting middle lobe bronchus (Figure 1). She underwent multiple rigid bronchoscopic procedures in order to obtain a tissue diagnosis while debulking the endoluminal component of the tumour using ND:YAG laser therapy.

Bronchoscopic and CT-guided biopsies resulted in inflammation and granulation tissue with some evidence of squamous metaplasia. FDG-PET scan demonstrated the 70 mm x 80 mm mass had a standardised uptake value (SUV) of 6.4 and there was a sub-centimetre subcarinal node with an SUV of 4.5 (Figure 2). Lung function results included an FEV1 of 1.93 (63% predicted), forced vital capacity (FVC) of 2.43 (69% predicted) and a transfer factor of the lung for carbon monoxide (TLCO) of 4.72 (52% predicted) with an obstructed right lower lobe.

The patient underwent a right thoracotomy and lower bilobectomy due to the involvement of the middle lobe bronchus and mediastinal lymph node dissection. The post-operative course was uneventful, and the patient was discharged

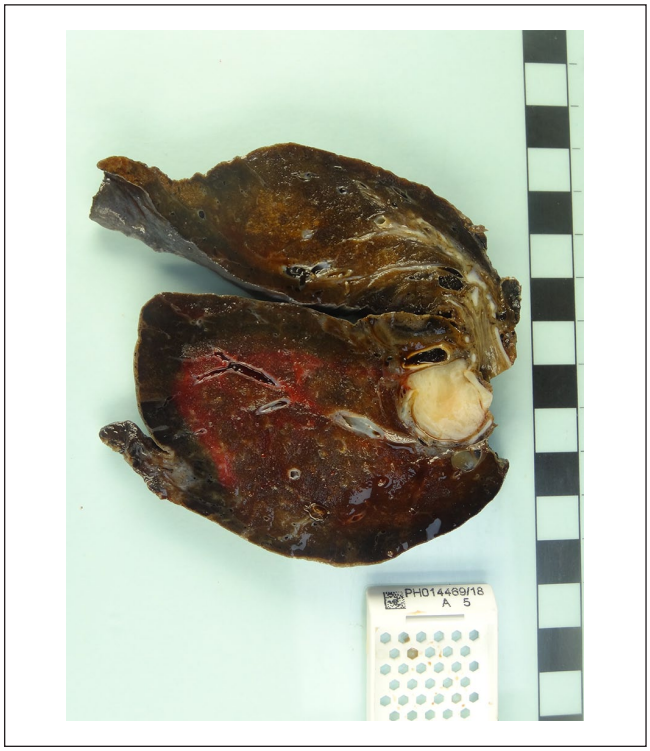

Figure 3. Gross appearance of the right lower and middle lobe with the tumour seen occluding the lower lobe bronchus.

on the fourth day. Two-year follow-up revealed that the patient was fit and well with a performance status of 0 and no long-term complications from her surgery.

Histopathological evaluation of the lung resection revealed a firm 19-mm well-circumscribed grey fibrous tumour beneath the middle lobe bronchus arising from the submucosa of the lower lobe bronchus and sub-totally obliterating the lumen (Figure 3). The background lung showed obstructive changes in the right lower lobe, but was otherwise unremarkable.

The mass lesion comprises plump spindle cells arranged in fascicles and whorls, the latter arranged in an onion-skin pattern around small arterioles (Figures 4 and 5). Scattered plasma cells were seen within the lesional stroma. There was no mitotic activity, low cellularity of the lesion and no positive staining for CK8/18, S100, SMA, EMA, Alk-1, CD99, B-catenin, desmin or Bcl-2, and only equivocal staining for CD34. Grocott-Gomori's methenamine silver stain (GMS) and Congo red stain were not used because the morphology did not suggest either fungi or amyloid. Lymph nodes were found to have reactive feature only and the resection margins were complete.

The appearances were those of a low-grade fibrocollagenous lesion, and an expert second opinion was sought confirming that this was a rare solitary endobronchial pulmonary hyalinising granuloma.

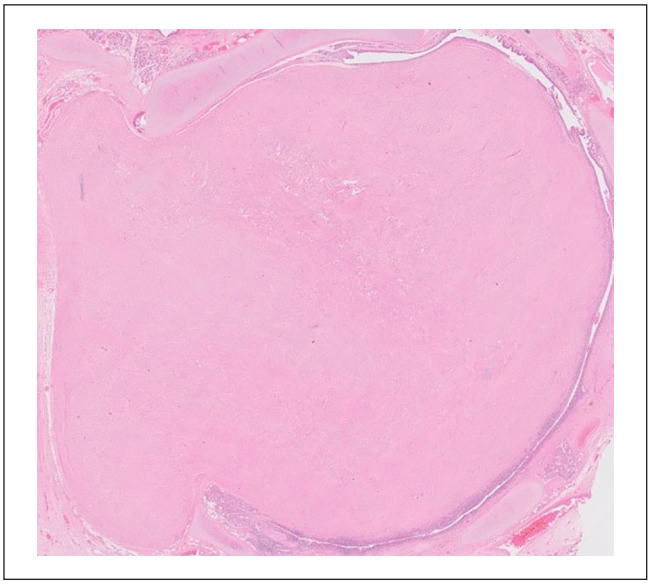

Figure 4. Microscopic overview of the whole lesion at 0.25× magnification demonstrating the compressed bronchial lumen overlying the lesion.

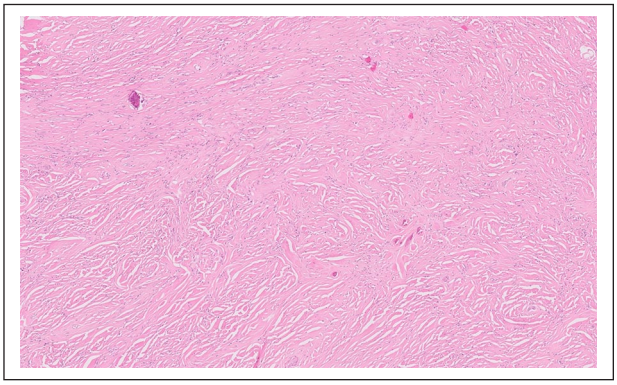

Figure 5. Microscopic appearance at 5× magnification showing the morphology of the lesion, including thick collagen bundles, pseudoangiomatous stromal clefting, interspersed bland spindle cells and foci of punctate calcification.

Discussion

There have been less than 150 case reports of PHG in the literature. A review performed by Lhote et al.[5] describes these well. In this comprehensive review of the available literature, PHG was found in patients between 15 and 83 years of age (mean age at diagnosis 44.6 years). There was no strong evidence of sex discrimination. Less than 30% of the patients described had a solitary lesion, and no mention of endobronchial position was made despite some patient presenting with obstructive symptoms. Furthermore, a solitary nodule extending into an adjacent lobe is also thought to be extremely rare.[5]

The diagnosis of PHG is thought to be associated with an autoimmune aetiology as there is an association with systemic fibrosis and extra-pulmonary autoimmune diseases such as sclerosing mediastinitis, Sjogren syndrome, rheumatoid arthritis, membranous glomerulonephritis and primary biliary sclerosis.[6,7] Positive autoantibody titres have been linked to the disease including anti-nuclear antibody (ANA), rheumatoid factor (RA factor), anti-neutrophil cytoplasmic antibodies (ANCA), anti-smooth muscle antibody (ASMA), anti-microsomal antibody (AMA) and Coombs-positive haemolytic anaemia.[8] The patient described in our study did not demonstrate any symptoms or antibodies suggesting an autoimmune link.

Diagnosis of these tumours is difficult, and in the majority of cases, no diagnosis can be made prior to surgical resection.[4] In this case, the patient required a thoracotomy and lower bilobectomy in order to both diagnose the condition, having had multiple attempts at less invasive diagnostic procedures, and treat the patient for the obstructive symptoms.

The patient we have reported did not suffer from any related condition and was found to have a solitary lesion. This lesion was found to be endobronchial, and the lesion was found to be extending into an adjacent lobe. As this was therefore a novel presentation of a rare disease, a second opinion was sought confirming that this was indeed a pulmonary hyalinising granuloma.

Conclusion

Although benign in nature, tissue diagnosis to rule out malignancy can be difficult in these lesions, especially when presenting as a solitary mass in a central location. This report demonstrates that these lesions can also be found endobronchially necessitating parenchymal resection for diagnosis and obstructive symptoms.

Declaration of conflicting interests

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Ethics approval

Our institution does not require ethical approval for reporting individual cases or case series

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Informed consent

Written informed consent was obtained from the patient(s) for their anonymized information to be published in this article.

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References

1. Benfield JR, Harrison RW, Moulder PV, et al. Bilateral nodular pulmonary granulomas and retroperitoneal fibrosis. *JAMA* 1962; 182: 579–581.
2. Engleman P, Liebow AA, Gmelich J, et al. Pulmonary hyalinizing granuloma. *Am Rev Respir Dis* 1977; 115: 997–1008.
3. Lien CT, Yang CJ, Yang SF, et al. Pulmonary hyalinizing granuloma mimicking multiple lung metastases: report of fluorodeoxyglucose positron emission findings. *J Thorac Imaging* 2010; 25(2): W36–W39.
4. Kawase S, Matsumoto R, Imai S, et al. Pulmonary hyalinising granuloma mimicking primary lung cancer. *Intern Med* 2018; 57: 3615–3617.
5. Lhote R, Haroche J, Duron L, et al. Pulmonary hyalinizing granuloma: a multicenter study of 5 new cases and review of the 135 cases of the literature. *Immunol Res* 2017; 65: 375–385.
6. Yousem SA and Hochholzer L. Pulmonary hyalinizing granuloma. *Am J Clin Pathol* 1987; 87: 1–6.
7. Ussavarungsi K, Khoor A, Jolles HI, et al. A 40-year old woman with multiple pulmonary nodules. *Chest* 2014; 146(6): e198–e203.
8. Schlosnagle DC, Check IJ, Sewell CW, et al. Immunologic abnormalities in two patients with pulmonary hyalinizing granuloma. *Am J Clin Pathol* 1982; 78(2): 231–235.