Padmanaban et al. Egyptian Journal of Radiology and Nuclear Medicine (2021) 52:161
https://doi.org/10.1186/s43055-021-00546-6

Egyptian Journal of Radiology and Nuclear Medicine

CASE REPORT Open Access

Pulmonary mucormycosis presenting as central bronchopleural fistula—a case report with review of literature

Elamparidhi Padmanaban*, Preethi Kannan, Umamageswari Amirthalingam, Sudhakar Pitchumani and Padma Rekha

Abstract

Background: Bronchopleural fistula (BFF) is a fistulous communication between the trachea or bronchus and the pleural space. Central type of bronchopleural fistula is usually post-surgical. Infective cause for central bronchopleural fistula is rare. This case report encompasses an infective cause of central bronchopleural fistula, mucormycosis. Pulmonary mucormycosis is a rapidly progressive condition with high mortality. A high index of suspicion and timely intervention is required to alleviate fatal outcome. The present case is discussed in detail about the clinical presentation and diagnostic imaging of pulmonary mucormycosis presenting with central bronchopleural fistula.

Case presentation: Thirty-five years old diabetic male, presented with fever, productive cough, mild haemoptysis and chest pain for 10 days duration. The patient was pale on general examination and had decreased breath sound in the right suprascapular and interscapular areas with coarse crackles in the right infrascapular and infra-axillary areas. The laboratory investigations were unremarkable except for anaemia and raised blood glucose level. Sputum examination on potassium hydroxide (KOH) mount showed broad aseptate hyphae. There was a loculated right hydropneumothorax with collapsed lung in chest radiograph. Multi-detector computed tomography of the thorax revealed central type of bronchopleural fistula with the right main bronchus, consolidation of the middle lobe and superior segment of the right lower lobe with multiple internal thick-walled cavities. Right pneumonectomy was performed as the patient did not improve on medical management and showed worsening of symptoms. Histopathological examination was suggestive of mucormycosis.

Conclusion: Central bronchopleural fistula due to an infective aetiology is uncommon. However, mucormycosis should be considered as a differential diagnosis in cases of central bronchopleural fistula with the destroyed lung, especially in diabetic individuals. Hence, a high index of suspicion is necessary for early diagnosis and management as mucormycosis is a rapidly progressive disease with delay in treatment leading to high mortality.

Keywords: Pulmonary mucormycosis, Bronchopleural fistula, Endobronchial

Background

Bronchopleural fistula (BPF) is a fistulous communication between the trachea or bronchus and the pleural space which is broadly classified as central or peripheral type. Central type is the direct communication between trachea or bronchus and the pleural space whereas peripheral type is an indirect communication between the pleura and airway distal to segmental bronchi or lung parenchyma. Central type of BPF usually occurs post-pneumonectomy. Peripheral type of BPF is usually seen secondary to necrotizing pneumonia, empyema, radiotherapy, bulla/cyst rupture and thoracic interventional procedures. Infectious and rheumatologic conditions

* Correspondence: pepsantosh@gmail.com
Department of Radiodiagnosis, Sri Manakula Vinayagar Medical College and Hospital, Puducherry 605107, India

such as tuberculosis, aspergillosis, granulomatosis with polyangiitis and pulmonary sarcoidosis can also result in BPF [1].

Case presentation

A 35-year-old male presented to the department of pulmonology with complaints of fever, productive cough and two episodes of mild haemoptysis for the past 10 days with associated right-sided chest pain. He is a chronic smoker and alcoholic with uncontrolled type II diabetes mellitus for 2 years and is on insulin replacement therapy. He had no significant past surgical history. On general examination, the patient appeared pale. Respiratory system evaluation revealed decreased bronchial breath sound in the right suprascapular and interscapular area and coarse crackles in the right infrascapular and infra-axillary area.

Routine blood investigation showed haemoglobin of 8.7 g%. Fasting blood glucose level was 162 mg/dl. Complete blood count, total and differential leucocyte count and ESR were within normal limits. Renal and liver function tests were normal. Sputum culture for bacteria was sterile. Sputum examination on potassium hydroxide (KOH) mount showed broad aseptate hyphae.

Plain chest radiograph (Fig. 1) showed a large cavity in the right hemithorax with air-fluid level. The cavity walls appear thick and smooth. No mural nodules were noted within. Part of the collapsed right lung is visualized through the cavity. Bilateral hilum appears normal. Multi-detector non-contrast computed tomography of the thorax demonstrated a fistulous communication between the right main bronchus and pleural space, suggestive of central type of bronchopleural fistula; the right pleural cavity showed hydropneumothorax (Fig. 2) with collapse of the right upper lobe and consolidation of the middle lobe and superior segment of the right lower lobe with multiple internal thick-walled cavities (Fig. 3). Imaging features of central bronchopleural fistula, destroyed right lung with associated uncontrolled diabetes and rapidly worsening clinical course, fulminant angioinvasive fungal disease like mucormycosis was considered as a preliminary imaging diagnosis and was advised bronchoscopic evaluation and bronchioalveolar lavage.

On further evaluation with bronchoscopy, the right secondary carina appeared distorted and destroyed with the right upper lobe bronchus revealing an unhealthy bronchial mucosa with brownish sludge. A large defect is seen in the posterior aspect of the right main bronchus with fistulous communication with pleural space. The posterior wall of the right intermediate bronchus was distorted with brownish sludge adjacent to the right lower lobe superior segment. Bronchoalveolar lavage and culture showed sterile bacterial growth; however, fungal elements and broad aseptate hyphae were seen. The patient did not improve on medical management and showed worsening of symptoms. Right pneumonectomy was done (Fig. 4). Intraoperatively, the right upper and lower lobes were destroyed with exposed right main bronchus communicating with the pleural cavity. A histopathological examination showed anthrocotic pigments and necrotic areas (Fig. 5). Gomori methenamine silver stain (Fig. 6) showed aseptate fungal hyphae which was suggestive of mucormycosis. Even being in the

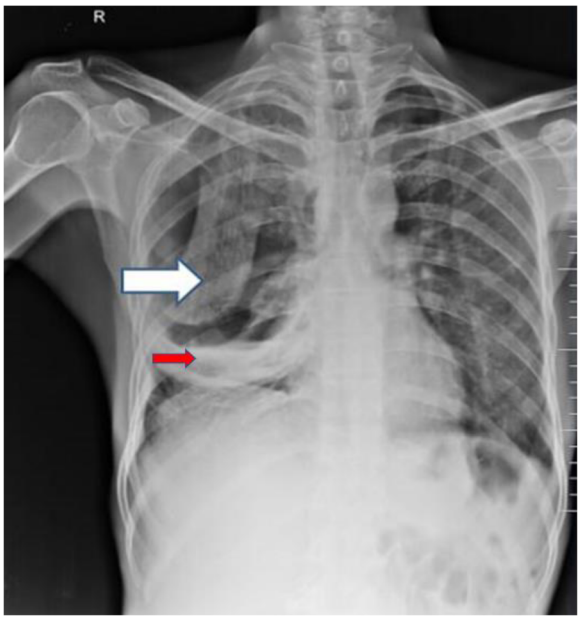

Fig. 1 Plain radiograph of chest posteroanterior view shows loculated right hydropneumothorax (red arrow) with collapsed lung (white arrow)

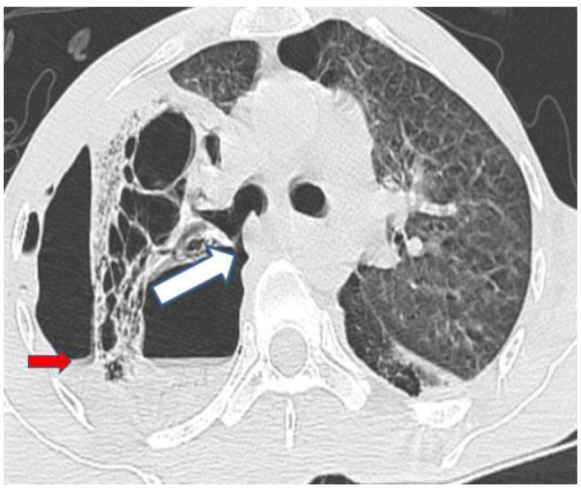

Fig. 2 MDCT axial image shows fistulous communication between right main bronchus and pleural cavity suggestive of central type of bronchopleural fistula (white arrow) with of air fluid level (red arrow) in pleural cavity

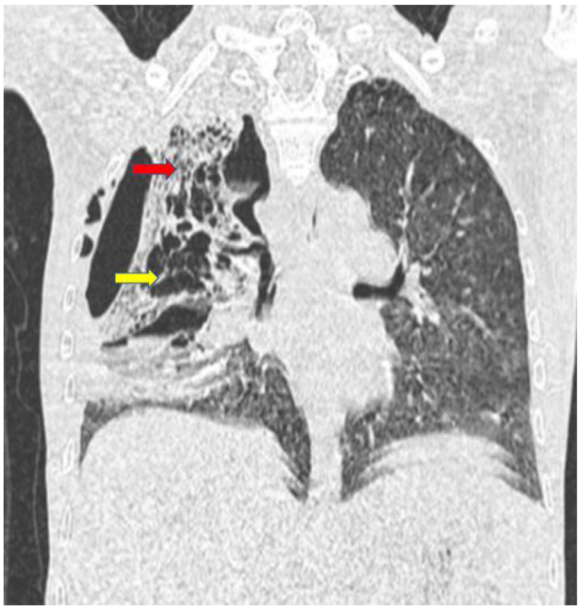

Fig. 3 MDCT with coronal reformation shows collapse of right upper lobe (red arrow) with multiple thick-walled cavities and consolidation of right middle lobe and superior segment of right lower lobe (yellow arrow)

intensive care, the patient succumbed to the disease in the postoperative period.

Discussion

A bronchopleural fistula is a communication with the pleural space and bronchial tree that results in high morbidity and mortality. It is most commonly recognized as a postoperative complication of pulmonary resection; however, other commonly contributing factors include necrotizing pulmonary infection, persistent spontaneous pneumothorax, chemotherapy and radiation therapy [2]. Cases of endobronchial fungal infection (EBFI) are unusual; however, the most common agents causing EBFI are *Aspergillus* species, *Coccidioides immitis*, agents of zygomycosis, *Candida* species, *Cryptococcus neoformans* and *Histoplasma capsulatum* [3]. Angioinvasive type of mucormycosis has a high mortality.

Mucorales species are vasotropic, causing tissue infarctions, and the mucormycosis spectrum ranges from

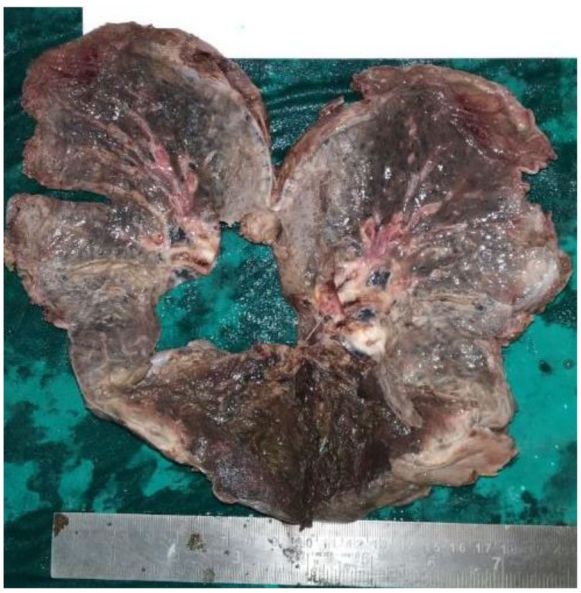

Fig. 4 Cut surface of gross pneumonectomy specimen of the right lung showing a large cavity with necrosis and angioinvasion

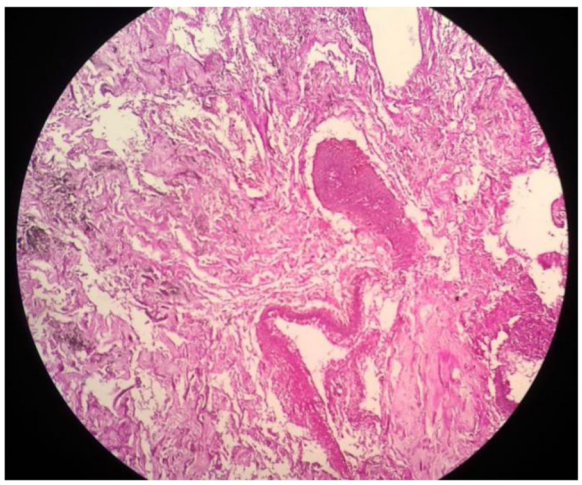

Fig. 5 Haematoxylin and eosin staining of the specimen shows lung tissue with arthrocotic pigments and necrotic areas with angioinvasion

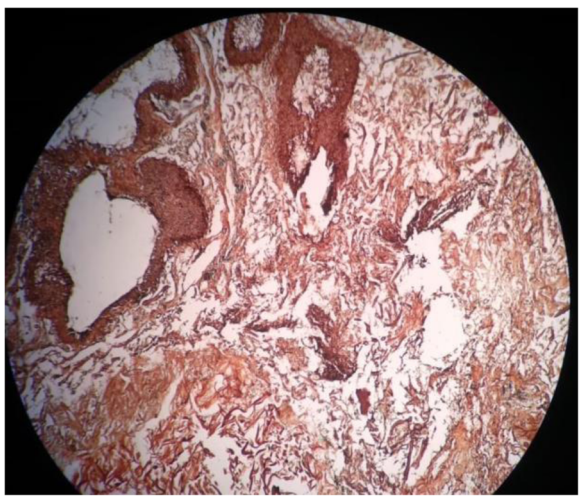

Fig. 6 Gomori methenamine silver staining of the specimen shows broad aseptate fungal hyphae

cutaneous, rhinocerebral and sinopulmonary to disseminated and frequently fatal infections [4]. The best known predisposing factor for mucor infection is diabetes mellitus. These organisms have an enzyme, ketone reductase, that allows them to thrive in a high glucose environment. Hematologic malignancies, prolonged neutropenia, treatment with corticosteroids or deferoxamine, bronchogenic cancer, renal transplantation and acquired immunodeficiency syndrome were other reported risk factors [3]. Patients usually present with prolonged high-grade fever that is unresponsive to broad-spectrum antibiotics. Nonproductive cough is a common symptom, whereas haemoptysis, pleuritic chest pain and dyspnoea are common in angioinvasion [5]. Hemoptysis is reported as the cause of death in pulmonary mucormycosis [6].

The radiological presentation of pulmonary mucormycosis starts with ground-glass opacities that progress to multiple pulmonary nodules. Pulmonary nodules are perivascular in distribution. Mucormycosis can also present with focal or multifocal areas of consolidation. A characteristic reverse halo sign is noted which is defined as central area of consolidation with peripheral ground-glass opacities. These ground glass opacities around the consolidation focus tend to be much larger than the latter. Cavitary lesions, which are usually peripheral in location, are also reported [7]. Fistulas are seen in angioinvasive disease. Mucormycosis is an aggressive infection that crosses fascial planes. They may even extend into the mediastinum and chest wall or even cause septic emboli in pulmonary artery.

Tracheobronchial mucormycosis is less common, accounting for 34% of pulmonary mucormycosis cases. Lobar bronchi are the most frequently involved location, with a predilection for the upper lobes. Mainstem bronchi are the next most common, followed by the trachea. No predilection for the right or left side is observed [8]. The rarity of our case is in the presentation of mucormycosis as central bronchopleural fistulous communication with the right main bronchus.

Bronchoscopy findings include endobronchial mass lesion or a greyish-white fibrinous plug obstructing the bronchus. It also caused sloughing of the mucosa, granular lesions, ulcer, stenosis and pseudomembrane formation and bronchial fistula [3]. The diagnosis is based upon bronchoalveolar lavage examination and biopsy. Biopsy reveals characteristic broad, non-septate hyphae with right-angle branching on calcofluor white or methenamine silver stains [9]. Sputum examination shows fungal hyphae and negative culture for bacteria.

Treatment with antifungal agents in combination with surgical debridement is considered the standard of therapy in invasive mucormycosis. Amphotericin is the approved drug for treatment which on early initiation has a profound impact on survival. Posaconazole, a newer triazole, has shown improvement when used as salvage therapy [5].

Conclusion

Mucormycosis is a rapidly progressive disease with delay in treatment leading to high mortality. Hence, a high index of suspicion is necessary especially in an immunocompromised patient with fulminant destructive lung disease. Imaging plays a key role in identifying central bronchopleural fistulas and evaluating the degree of lung involvement in such cases, which will guide the clinicians in deciding between medical management or pneumonectomy.

Abbreviations

BPF: Bronchopleural fistula; ESR: Erythrocyte sedimentation rate; KOH: Potassium hydroxide; EBFI: Endobronchial fungal infection

Acknowledgements

Nil

Authors' contributions

Dr EP = identification of the imaging findings and diagnosed the condition followed by manuscript preparation. Dr PK = data collection including histopathological slides and equally participated in manuscript preparation. Dr UA = gave expert opinion on the imaging features and contributed in manuscript preparation. Dr SP = helped in refining the manuscript and provided valuable inputs in diagnosing the condition and literature review. Dr PR = provided bronchoscopic findings and case follow up. Helped with literature search and manuscript preparation. All the authors have participated sufficiently in contributing to the content of "Pulmonary mucormycosis presenting as central bronchopleural fistula—a case report with review of literature" and have read and approved the manuscript.

Funding

Nil

Availability of data and materials

Available

Declarations

Ethics approval and consent to participate

Ethical approval is not obtained, since it is a retrospective case report. Informed written consent had been obtained from the participant for participation and publication of the same as a case report.

Consent for publication

Informed written consent had been obtained from the participant for publication of the same as a case report.

Competing interests

The authors declare that they have no competing interests.

Received: 17 May 2021 Accepted: 24 June 2021
Published online: 06 July 2021

References

1. Gaur P, Dunne R, Colson YL, Gill RR (2014 Jul) Bronchopleural fistula and the role of contemporary imaging. J Thorac Cardiovasc Surg 148(1):341–347. https://doi.org/10.1016/j.jtcvs.2013.11.009
2. Lois M, Noppen M (2005) Bronchopleural fistulas: an overview of the problem with special focus on endoscopic management. Chest 128(6): 3955–3965. https://doi.org/10.1378/chest.128.6.3955

3. Karnak D, Avery RK, Gildea TR, Sahoo D, Mehta AC (2007) Endobronchial fungal disease: an under-recognized entity. Respiration. 74(1):88–104. https://doi.org/10.1159/000094708
4. Verma S, Singh J, Kotwal S, Mahajan A, Khajuria R, Singh A (2018) Rhinocerebral mucormycosis: report of a rare case. Int J of Allied Med Sci and Clin Research 6(3):634–638
5. Panigrahi MK, Manju R, Vinod Kumar S, Toi PC (2014 Dec 1) Pulmonary mucormycosis presenting as nonresolving pneumonia in a patient with diabetes mellitus. Respir Care 59(12):e201–e205. https://doi.org/10.4187/respcare.03205
6. Di Carlo P, Cabibi D, La Rocca AM, De Luca D, La Licata F, Sacco E (2010 Dec) Post-bronchoscopy fatal endobronchial hemorrhage in a woman with bronchopulmonary mucormycosis: a case report. J Med Case Rep 4(1):398. https://doi.org/10.1186/1752-1947-4-398
7. Hammer MM, Madan R, Hatabu H (2018 Apr) Pulmonary mucormycosis: radiologic features at presentation and over time. Am J Roentgenol 210(4):742–747. https://doi.org/10.2214/AJR.17.18792
8. He R, Hu C, Tang Y, Yang H, Cao L, Niu R (2018) Report of 12 cases with tracheobronchial mucormycosis and a review. *Clin Respir J* 12(4):1651–1660. https://doi.org/10.1111/crj.12724
9. Chung JH, Godwin JD, Chien JW, Pipavath SJ (2010 Aug) Case 160: pulmonary mucormycosis. Radiology. 256(2):667–670. https://doi.org/10.1148/radiol.10081907

Publisher's Note