Case Report

URF PUBLISHERS
connect with research world

World Journal of Surgery and Surgical Case Reports

https://urfpublishers.com/journal/surgery-and-surgical-case-reports

Vol: 1 & Iss: 3

Pulmonary Aspergilloma with DIPNECH; A Rare Case Report

Suleyman Gokalp Gunes[1]*, Baris Gulmez[2], Beniz Irem Ersoy Sigva[2], Kubra Sacar[3] and Inci Bugday[4]

[1]Department of Thoracic Surgery, Ankara University, Turkey

[2]Thoracic Surgery Clinic, Ministry of Health University Van Training and Research Hospital, Turkey

[3]Anesthesiology and Reanimation Intensive Care Unit, Ministry of Health University Van Training and Research Hospital, Turkey

[4]Pathology Clinic, Ministry of Health University Van Training and Research Hospital, Turkey

Citation: Gunes SG, Gulmez B, Sigva BIE, Sacar K, Bugday I. Pulmonary Aspergilloma with DIPNECH; A Rare Case Report. *World J Surg Surgical Case Rep*, 2025;1(3):84-86.

Received: 24 September, 2025; **Accepted:** 30 September, 2025; **Published:** 02 October, 2025

***Corresponding author:** Suleyman Gokalp Gunes, Department of Thoracic Surgery, Ankara University, Talatpasa Blv. Altindag/ Ankara, Turkey, Tel: +90 533 252 595; E-mail: gokalpgunes@yahoo.com

A B S T R A C T

Introduction: Pulmonary Aspergilloma (PA) usually presents with a fungus ball or mycetoma as a result of saprophytic colonization of Aspergillus fumigatus causing parenchymal damage and forming a cavitary lesion. The incidence of pulmonary aspergilloma with tumor lesions is rare. In our study, we aimed to present a patient diagnosed with diffuse idiopathic neuroendocrine cell hyperplasia (DIPNECH) in a patient who underwent anatomical resection for PA.

Case: A 46-year-old woman with no history of comorbidities presented with a history of frequent pneumonia and recurrent hemoptysis. Thorax CT showed a 47x42 mm cavitary lesion with irregular walls in the lower lobe of the right lung, millimetric solid nodular lesions and fungus ball appearance within the cavity, bronchiectatic changes and septal thickening. Spirometry was performed and after 7 days antibiotherapy with clinical improvement, the patient underwent lower lobectomy and mediastinal lymph node dissection via right thoracotomy. Histopathologic examination revealed a 3 cm cavitary lesion in the lobe which was compatible with pulmonary aspergilloma. Histopathologic examination revealed a 3 cm cavitary lesion in the lobe which was compatible with pulmonary aspergilloma. It was observed to be consistent with diffuse idiopathic neuroendocrine cell hyperplasia.

Conclusion: In DIPNECH, multifocal pulmonary nodules with mosaic attenuation are seen, which is rare. It is a generalized intramucosal proliferation of pulmonary neuroendocrine cells clustered in monolayers or small groups that can protrude into the bronchial lumen. DIPNECH was diagnosed in 5.4% of patients operated for carcinoid tumor and the recommendation in the literature is to perform surgical resection in case DIPNECH is detected. PA and DIPNECH are two different entities and their coexistence is rare. DIPNECH, which is also associated with other diagnoses, can often be detected histopathologically in carcinoid tumors and surgical resection is the appropriate treatment method as we have stated in our study.

Keywords: Pulmonary nodule, Pneumonia, Hemoptysis, Tumor

Abbrevations: PA: Pulmonary Aspergilloma; DIPNECH: Diffuse Idiopathic Pulmonary Neuroendocrine Cell Hyperplasia; CT: Computed Tomatoghraphy

1. Introduction

Pulmonary Aspergilloma (PA) usually presents with a fungus ball or mycetoma as a result of saprophytic colonization of Aspergillus fumigatus causing parenchymal damage and forming a cavitary lesion[1]. It is known to affect more than 200,000 people worldwide and can cause recurrent or massive hemoptysis as a result of bronchial artery erosion and can be mortal in 2% to 50%[2]. Conditions such as chronic lung disease, (bronchiectasis, sarcoidosis etc.) immunosuppression, hydatid cyst, malignancy, diabetes mellitus, lupus, hypertension, coronary artery disease, lung abscess, invasive ventilation, postoperative lung injury are frequently involved in the etiology. On computed tomography of the thorax, demonstration of a fungus ball in the cavity and ectatic bronchus are diagnostic[3]. In addition, tumorlet lesions, which present as nodular lesions smaller than 5 mm, are rare neuroendocrine cell hyperplasias and are frequently seen in carcinoid tumors[4]. The incidence of pulmonary aspergilloma with tumor lesions is rare. In our study, we aimed to present a patient diagnosed with Diffuse Idiopathic Pulmonary Neuroendocrine Cell Hyperplasia (DIPNECH) in a patient who underwent anatomical resection for PA.

2. Case Presentation

The patient with no history of comorbidities who is 46 year-old, presented with a history of frequent pneumonia and recurrent hemoptysis. Thorax CT showed a 47x42 mm cavitary lesion with irregular walls in the lower lobe of the right lung, millimetric solid nodular lesions and fungus ball appearance within the cavity, bronchiectatic changes and septal thickening, as well as pathologic hilar and mediastinal lymphadenopathies over 2 cm in size (Figure 1). Laboratory tests on admission did not reveal any features except leukocytosis and elevated C-reactive protein. Further tests were not performed because the Galactomannan Agglutination test was not available in the center where we worked. Spirometry was performed and after 7 days antibiotherapy with clinical improvement, the patient underwent lower lobectomy and mediastinal lymph node dissection via right thoracotomy (Figure 2)

Histopathologic examination revealed a 3 cm cavitary lesion in the lobe which was compatible with pulmonary aspergilloma. Hilar and mediastinal lymph nodes were evaluated as reactive. On microscopic examination, neuroendocrine hyperplasia and tumorlet foci, each 1-2 mm in diameter, were observed in the sections. It was observed to be consistent with DIPNECH. Mitosis was found to be less than 2 per 2 mm^2. Necrosis was not observed. Immunohistochemistry examination revealed CD56 (+), ChromograninA (+), Synaptophysin (+), PANCK (+), CDX2 (-), CD34 (-), D240 (-), CK7 (-), CK20 (-), TTF1 (focal +), NapsinA (-), ER (-), PR (-), PAX8 (-), WT1 (-), GATA3 (-) were detected and Ki67 proliferation index was 1%. (Figure 3). The chest tube was removed on postoperative day 5 and the patient was discharged on postoperative day 6. The patient was followed up with adjuvant voriconazole for 3 weeks as recommended in the literature and no pathology was observed at the 18th month postoperative follow-up[1].

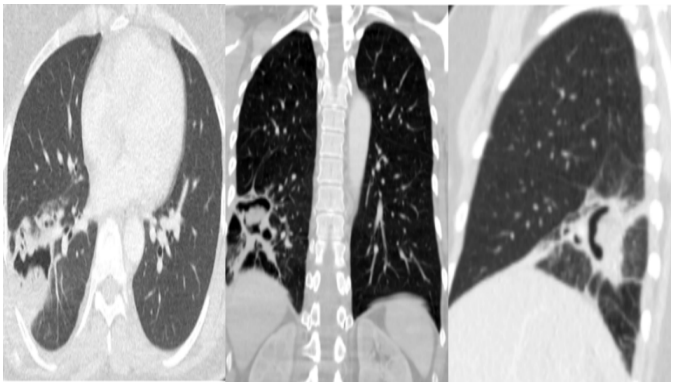


Figure 1: Preoperative computerized thorax CT image (A: axial view B:coronal view C: Sagital view).

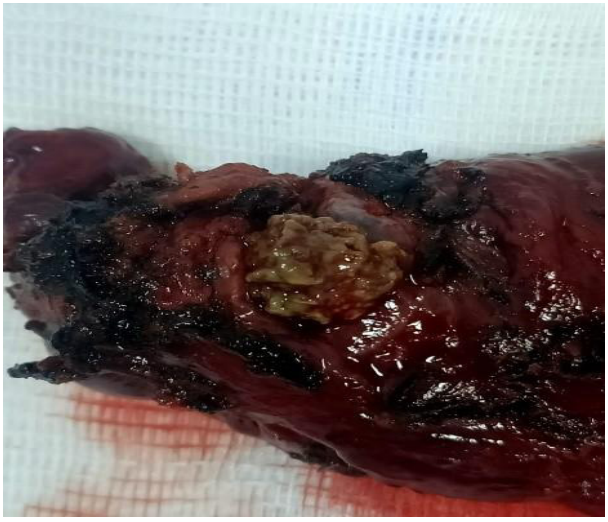
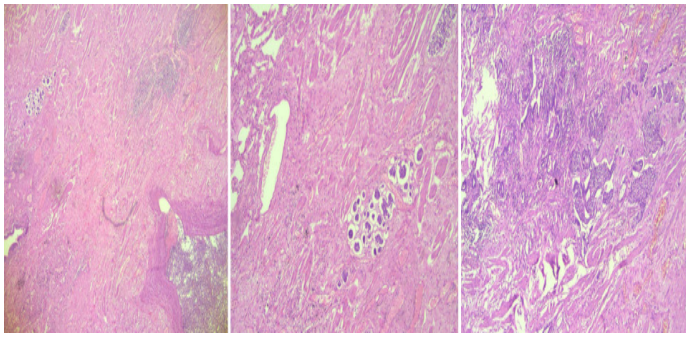


Figure 2: Section image (section was taken for diagnostic purposes after the section was removed from the lesion).



A B C

Figure 3: Histopathologic examination images. A: Cavitary lesion (aspergilloma) with a fungal ball in the lower right corner. B: Chronic inflammation with eosinophils is seen in the cavity wall. C: Bronchiole-limited neuroendocrine cells forming small nodules in the upper left corner (H&E, 40x).

3. Discussion

While medical or surgical treatment was accepted as the standard of care in pulmonary aspergilloma in the 1970s, combined treatment (anatomical resection with adjuvant therapy) has been accepted as the gold standard since 2010[3]. Sublobar resections in peripherally located, <3 cm lesions are controversial due to worse prognosis, more air leakage and risk of recurrence[5]. In DIPNECH, multifocal pulmonary nodules with mosaic attenuation are seen, which is rare[6]. It is a generalized intramucosal proliferation of pulmonary neuroendocrine cells clustered

in monolayers or small groups that can protrude into the bronchial lumen. The cells do not cross the mucosal basal lamina. (If they do, they are called "tumorlets.") The cells are round, oval or spindle-shaped, have moderate amounts of eosinophilic cytoplasm and have round to oval nuclei with salt and pepper chromatin. Histopathologically, the presence of ≥ 5 neuroendocrine cells distributed linearly or in clusters within the basement membrane in ≥ 3 bronchioles and association with ≥ 3 tumorlets is diagnostic.

DIPNECH was diagnosed in 5.4% of patients operated for carcinoid tumor and the recommendation in the literature is to perform surgical resection in case DIPNECH is detected[7]. The coexistence of PA and DIPNECH is a rare condition and in the case report of Yazgan et al.[8], a 67-year-old female patient underwent left lower lobectomy due to fungus ball and the diagnosis of PA and DIPNECH could be shown. In the case report of Moskovljevic, et al.[4], a 71-year-old female patient with a positive galactomannan agglutination test was diagnosed with PA and DPNECH after right lower lobectomy.

If the lesions, which are considered as nodular proliferation of neuroendocrine cells, are <5 mm from the bronchiole wall, they are called tumorlets. If there is a relationship of 3 or more airways, the diagnosis of DPNECH can be indicated radiologically[6]. It has been reported that the use of somatostatin is beneficial in the treatment of these lesions, which are considered premalignant by WHO[9].

In the case report of Inomata et al.[10], in a patient who underwent chemotherapy for bilateral pulmonary nodules and a mass diagnosed as primary adenocarcinoma in the right upper lobe, right upper lobectomy and right lower lobe wedge resection were performed due to regression in the primary tumor but no regression in the nodules and DIPNECH was diagnosed from nodules other than the primary tumor.

4. Conclusion

PA and DIPNECH are two different entities and their coexistence is rare. DIPNECH, which is also associated with other diagnoses, can often be detected histopathologically in carcinoid tumors and surgical resection is the appropriate treatment method as we have stated in our study.

5. References

1. Sagan D, Gozdziuk K. Surgery for pulmonary aspergilloma in immunocompetent patients: no benefit from adjuvant antifungal pharmacotherapy. Ann Thorac Surg. 2010;89(5): 1603-1610.
2. Jiang C, Dai J, Bao Y, et al. Surgical treatment of pulmonary aspergilloma: A 13-year experience from a single clinical center. Ann Thorac Surg. 2022;114(1): 311-318.
3. Denning DW, Cadranel J, Beigelman-Aubry C, et al. European Society for Clinical Microbiology and Infectious Diseases and European Respiratory Society. Chronic pulmonary aspergillosis: rationale and clinical guidelines for diagnosis and management. Eur Respir J. 2016;47(1): 45-68.
4. Moskovljevic D, Colic N, Dimkic-Milenkovic A, et al. Incidental finding of pulmonary tumorlet in a case of surgically treated bronchiectatic cavity superimposed by aspergilloma. Srpski arhiv za celokupno lekarstvo. 2022;150(11-12): 690-693.
5. Lejay A, Falcoz PE, Santelmo N, et al. Surgery for aspergilloma: time trend towards improved results? Interact Cardiovasc Thorac Surg. 2011;13(4): 392-395.
6. Ramirez RA, Cass AS, Das S, et al. A multidisciplinary approach to the work up and management of pulmonary carcinoid tumors and DIPNECH: a narrative review. Transl Lung Cancer Res. 2022;11(12): 2567-2587.
7. Chassagnon G, Favelle O, Marchand-Adam S, et al. DIPNECH: when to suggest this diagnosis on CT. Clin Radiol. 2015;70(3): 317-325.
8. Yazgan S, Gursoy S, Turk F, et al. Conjunction of a fungus ball and a pulmonary tumourlet in a bronchiectatic cavity. Korean J Thorac Cardiovasc Surg. 2018;51(2): 138-141.
9. O'Brien C, Duignan JA, Gleeson M, et al. Quantitative Airway Assessment of Diffuse Idiopathic Pulmonary Neuroendocrine Cell Hyperplasia (DIPNECH) on CT as a Novel Biomarker. Diagnostics (Basel). 2022;12(12): 3096.
10. Inomata S, Matsumura Y, Kobayashi Y, et al. Lung adenocarcinoma coexisting with diffuse idiopathic pulmonary neuroendocrine cell hyperplasia manifesting as multiple pulmonary nodules: A case report. Thoracic Cancer. 2022;13(21): 3076-3079.