

Interesting Localization of a Fungus Ball: Aspergilloma Located in a Tracheal Diverticulum

Coşkun Doğan,¹ Tamer Baysal,² Sevda Şener Cömert,¹
Ayşegül Atalay,² Elif Torun Parmaksız,¹ Dilek Ece İlgici³

¹Department of Pulmonary Diseases,
University of Health Sciences,
Kartal Dr. Lütfi Kırdar Training and
Research Hospital, Istanbul, Turkey
²Department of Radiology,
University of Health Sciences,
Kartal Dr. Lütfi Kırdar Training and
Research Hospital, Istanbul, Turkey
³Department of Pathology,
Istanbul Haseki Training and
Research Hospital, Istanbul, Turkey

Submitted: 30.03.2018
Accepted: 04.06.2018

Correspondence: Coşkun Doğan,
Kartal Dr. Lütfi Kırdar Eğitim
ve Araştırma Hastanesi, Göğüs
Hastalıkları Kliniği, Istanbul, Turkey
E-mail: coskund24@yahoo.com



Keywords: Aspergilloma;
fungus ball; tracheal
diverticulum.

ABSTRACT

Objective: *Aspergillus fumigatus* and *Aspergillus niger* are *Aspergillus* species that generally invade cavitory structures of the lungs and form fungus balls of *Aspergillus* hyphae, fibrin, mucus, blood, and inflammatory and epithelial cells; they are very rarely seen in extra-respiratory organs and systems. Tracheal diverticulum is a rare and mostly asymptomatic entity usually found in the paratracheal region, but its etiology and pathophysiology is not fully known. An interesting and very rare case of aspergilloma located in a tracheal diverticulum is presented in this article.

INTRODUCTION

Aspergilloma as a result of an *Aspergillus fumigatus* infection, generally occurs in cavitory spaces of the lungs, such as a tuberculous or tumoral cavity, or cases of bronchiectasis, though rarely, it may appear in other systems and organs as well. Most often, aspergilloma is due to *Aspergillus fumigatus*, and occasionally *Aspergillus niger*. Aspergillomas, also called fungus balls, consist of *Aspergillus* hyphae, fibrin, mucus, blood, and inflammatory and epithelial cells.^[1,2]

A tracheal diverticulum (TD) is an anatomical structure, the etiology and pathophysiology of which is still uncertain. Diverticula appear in congenital and acquired forms. They are thought to stem from branching anomalies at the time of tracheal bifurcation on the 26th day of gestation. Acquired TD is believed to occur as a herniation in a weak

area of the trachea caused by increased intramural tracheal pressure. TD is typically asymptomatic, and is often only observed on incidentally obtained computed tomograms (CTs). The incidence has been reported as 1% to 2% in autopsy studies.^[3,4]

This is a case of aspergilloma developing in an unusual and interesting location: TD.

CASE REPORT

A 72-year-old female patient in follow-up at the outpatient clinic of chest diseases for a pulmonary nodule had no respiratory complaints. Physical examination did not reveal any finding. An 8 mm nodule had been detected in the left lung 6 months previously. Her family history was unremarkable. The patient did not smoke or use any med-

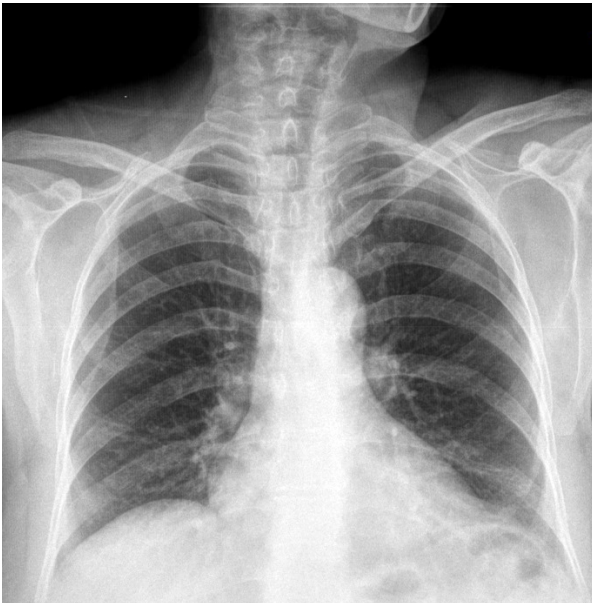


Figure 1. The anteroposterior chest X-ray was normal.



Figure 3. An image from the computed tomography-guided fine-needle aspiration biopsy.

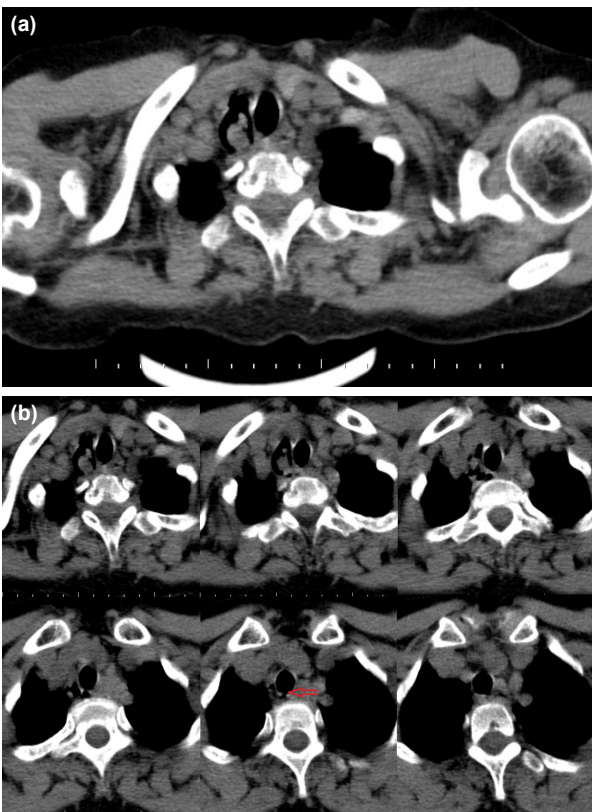


Figure 2. (a) Thoracic computed tomography revealed the presence of a cavitary lesion on the right posterolateral aspect of the trachea containing material with a soft tissue density. (b) Microscopic appearance of serial sections of the lesion localized on the right posterolateral aspect of the trachea. The red arrow indicates the possible trachea-connected portion of the lesion.

ication. Her laboratory test results were as follows: white blood cell count: 9900/uL, platelet count: 212,000/uL, he-

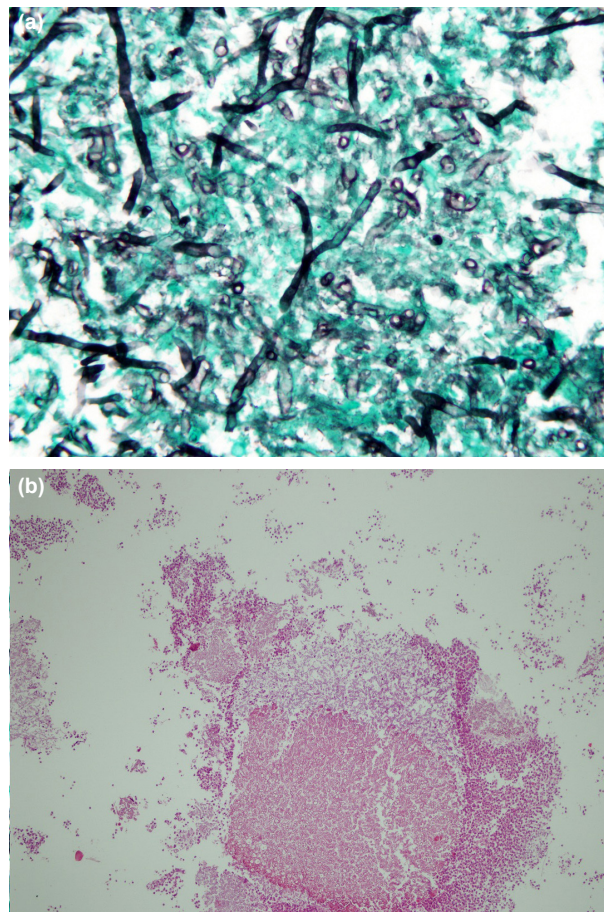


Figure 4. (a, b) Fungal hyphae, some of which demonstrated 45° angulations (Grocott x1000), and a fungal cluster with polymorphonuclear leucocytes (H&E x200).

moglobin count: 13.7g/dL, hematocrit: 41.1%, sedimentation rate: 21 mm/hour, C-reactive protein: 10 mg/L, blood

urea nitrogen level: 36 mg/dL, creatinine level: 1.02 mg/dL, alanine aminotransferase: 28 U/L, aspartate transaminase: 29 U/L, lactate dehydrogenase: 438 U/L, sodium: 140 mEq/L, potassium: 5.18 mEq/L, calcium: 8.9 mEq/dL, and chloride level: 109 mEq/L, without any pathological finding.

The posteroanterior chest X-ray was evaluated as normal (Fig. 1). The thoracic CT scan was requested to examine the nodule detected 6 months earlier. The CT revealed a cavitary lesion with soft tissue density localized in the posterolateral aspect of the trachea. There was no change in the size of the nodule detected in the previous CT, and it was noted that the paratracheal cavitary lesion was also present in the earlier CT image (Fig. 2a, and b). Fiberoptic bronchoscopy revealed no pathological findings. A CT-guided fine-needle aspiration biopsy was performed by the interventional radiology clinic (Fig. 3). The pathology results were fungal aggregates, including hyphae with an angulation of 45°, which suggested aspergilloma (Fig. 4). A serum galactomannan antigen test result was negative (0.215).

Surgery was planned for the patient with a diagnosis of paratracheal aspergilloma; however, she declined to undergo a surgical procedure and instead was scheduled for control visits in the outpatient clinic and 3 months of oral voriconazole treatment was initiated.

DISCUSSION

Aspergilloma can invade almost all systems and organs of the body, but is predominantly seen in cavitary lesions of the lungs. It is rarely located in the paratracheal region. TD is most often asymptomatic and rarely complicated with non-specific infections. A case of paratracheal aspergilloma located in a TD is very rare.

Aspergilloma occurs with the colonization of *Aspergillus* species in preexisting spaces, usually lung cavities. Cavities with insufficient drainage facilitate the formation of aspergilloma. Aspergilloma is usually asymptomatic, but it can lead to life-threatening, massive hemoptysis, which has been reported in 2% to 14% of cases.^[5] Aspergilloma may arise secondary to various etiological and predisposing factors.

In immunosuppressed patients, such as those with hematological malignancies, endobronchial, intracranial, gastrointestinal system, and liver aspergillomas have been reported.^[6-8] Orbital aspergilloma has been cited as a result of paranasal sinus involvement or direct contact.^[9] Cases of cardiac aspergilloma have also been observed in patients who underwent cardiac surgery, and those with a history of intravenous drug dependency or parenteral nutrition.^[10] Furthermore, renal aspergilloma may develop in cases of focal renal abscesses.^[11] These cases, like ours, are rare, and most are seen in immunosuppressed patients. Our pa-

tient was not immunosuppressed and to the best of our knowledge, no case of aspergilloma with a paratracheal localization has been reported in the literature.

Tracheal diverticulum is a rare, benign disease that is the result of an outward bulging of the tracheal wall. There are 2 types: congenital and acquired. Congenital TD differs in size from acquired TD and has a narrower communication with the trachea. Diverticula are frequently localized on the right side of the trachea, a few centimeters above the tracheal carina, and 4 to 5 cm below the vocal cords. They are histologically similar to the tracheal wall. CT is a non-invasive and highly reliable diagnostic tool for the detection of aspergillomas. Although fiberoptic bronchoscopy is another potential diagnostic tool, diverticula and the tracheal connections may not be visible.^[12,13]

Uncomplicated TD is mostly asymptomatic. Complicated TD often presents with a secondary bacterial infection, compression of neighboring organs, or a rupture secondary to trauma. A review of the literature suggests that complicated TD most often presents with a chronic cough and recurrent respiratory infections,^[14] rupture secondary to trauma, mediastinal and subcutaneous emphysema,^[15-17] or hoarseness caused by compression of the laryngeal nerve.

Charest et al.^[18] detected a right-sided paratracheal mass in the CT image of a 74-year-old woman with Waldenström macroglobulinemia who presented at an emergency department after a traumatic event and received a final diagnosis of infected TD. Since malignancy was among the initial diagnoses, positron emission tomography (PET) CT scans were obtained, which indicated that the SUV max value of the lesion was 9.2. In our case, a PET-CT was also performed for the lung nodule and interestingly, the right paratracheal lesion was not observed.

This case of paratracheal aspergilloma as a result of *Aspergillus fumigatus* complicated by TD is believed to be a unique contribution to the literature.

Informed Consent

Written informed consent was obtained from the patient for the publication of the case report and the accompanying images.

Peer-review

Internally peer-reviewed.

Authorship Contributions

Concept: C.D.; Design: C.D.; Data collection &/or processing: T.B.; Analysis and/or interpretation: S.Ş.C.; Literature search: E.T.P., A.A.; Writing: C.D.; Critical review: A.A., D.E.İ.

Conflict of Interest

None declared.

REFERENCES

1. Lee SH, Lee BJ, Jung DY, Kim JH, Sohn DS, Shin JW, et al. Clinical manifestations and treatment Outcomes of Pulmonary aspergilloma. *Korean J Intern Med* 2004;19:38–42.
2. Argento C, Wolfe CR, Wahidi MM, Shofer SL, Mahmood K. Bronchomediastinal fistula caused by endobronchial aspergilloma. *Annals Am Thorac Soc* 2015;12:91–5.
3. Yüce GD, Ulaşlı SS. Chronic cough due to tracheal diverticulum [Article in Turkish]. *Respir Case Rep* 2012;1:62–4.
4. Lee SY, Joo S, Lee GD, Ham SJ, Park CH, Lee S. A case of symptomatic tracheal diverticulum and surgical resection as a treatment modality. *Korean J Thorac Cardiovasc Surg* 2016;49:405–7.
5. Stather DR, Tremblay A, Dumoulin E, MacEachern P, Chee A, Herrgott C, et al. A series of transbronchial removal of intracavitary pulmonary aspergilloma. *Ann Thorac Surg* 2017;103:945–50.
6. Sunnetcioglu A, Ekin S, Erten R, Parlak M, Esen R. Endobronchial aspergilloma: A case report. *Respir Med Case Rep* 2016;18:1–3.
7. Cheko A, Jung S, Teuber-Hanselmann S, Oseni AW, Tsogkas A, Scholz M, et al. Orbital apex syndrome caused by aspergilloma in an immunocompromised patient with cutaneous lymphoma: A case report of a rare entity. *S Afr Med J* 2016;106:46–7.
8. Hadchiti MT, Abdalkader M, Rached L, Mahfound D, Khoury G, Ghanem H. Disseminated intra-abdominal aspergilloma with abdominal wall invasion in a patient with acute myeloid leukemia: A case report. *Clin Lymphoma Myeloma Leuk* 2015;15:94–7.
9. Moreno-Sánchez M, Villanueva AL, Gonzalez GR, Arias JM, Monje F. Intraorbital aspergilloma: a rare cause of orbital apex syndrome. *Br J Oral Maxillofac Surg* 2016;54:1047–8.
10. Soman SO, Vijayaraghavan G, Padmaja NP, Warriar AR, Unni M. Aspergilloma of the heart. *Indian Heart J* 2014;66:238–40.
11. Ullah SR, Jamshaid A, Zaidi SZ. Renal aspergilloma presenting with pelvi-ureteric junction obstruction (PUJO). *J Pak Med Assoc* 2016;66:903–4.
12. Amaral B, Silva S, Feijó S. Infected tracheal diverticulum: a rare association with alpha-1 antitrypsin deficiency. *J Bras Pneumol* 2014;40:669–72.
13. O'Leary CN, Ryan JW, Corbett G, Ridge CA. Barotrauma induced tracheal diverticulum rupture: imaging findings. *BMJ Case Rep* 2016;27:2016.bcr2016217518.
14. Takhar RP, Bunkar M, Jain S, Ghabale S. Tracheal diverticulum: an unusual cause of chronic cough and recurrent respiratory infections. *Tuberk Toraks* 2016;64:77–82.
15. Allaert S, Lamont J, Kalmar AF, Vanoverschelde H. Tracheal diverticulum as a cause of subcutaneous emphysema following positive-pressure ventilation. *Can J Anaesth* 2016;63:1098–9.
16. Gorgas DL, Miller B. Tracheal diverticulum masquerading as pneumomediastinum in a trauma victim. *Am J Emerg Med* 2015;33:310.e1–3.
17. Ceulemans LJ, Lerut P, De Moor S, Schildermans R, De Leyn P. Recurrent laryngeal nerve paralysis by compression from a tracheal diverticulum. *Ann Thorac Surg* 2014;97:1068–71.
18. Charest M, Sirois C, Cartier Y, Rousseau J. Infected tracheal diverticulum mimicking an aggressive mediastinal lesion on FDG PET/CT: an interesting case with review of the literature. *Br J Radiol* 2012;85:17–21.

Fungus Topunun İlginç Bir Lokalizasyonu: Trekeal Divertikül İçinde Aspergilloma

Genellikle akciğerin kaviter yapıları içerisine yerleşerek, kaviter alan içerisinde *aspergillus hifler*, fibrin, mukus, kan, enflamatuvar ve epitelyum hücreleri ile fungus ball'ı oluşturan *aspergillus fumigatus/aspergillus niger* akciğer dışı organ ve sistemlerde çok nadir görülür. Etiyolojisi ve patofizyolojisi hala kesin olmayan trakeal divertiküller genellikle paratrakeal yerleşimli, nadir görülen, çoğunlukla semptomsuz oluşumlardır. Bu makalede trakeal divertikül içerisine yerleşmiş aspergilloma ilginç ve çok nadir bir durum olduğu için sunuldu.

Anahtar Sözcükler: Aspergilloma; fungus topu; trakeal divertikül.