Acta Med. Okayama, 2022
Vol. 76, No. 4, pp. 485–488


Acta Medica Okayama
http://escholarship.lib.okayama-u.ac.jp/amo/

*Case Report*

# A Rare Presentation of Invasive *Aspergillosis*: An Asymptomatic Man with an Abscess Localized to a Parietal Pleura

**Yuya Takahashi, Mototsugu Watanabe*, Shinichi Furukawa, Toshio Shiotani, and Kazuhiko Kataoka**

*Department of Thoracic Surgery, National Hospital Organization Iwakuni Clinical Center, Iwakuni, Yamaguchi 740-8510, Japan*

**Aspergillosis is an infection caused by *Aspergillus* species, and it manifests in various clinical presentations. We describe the case of a 73-year-old man with a small area of thickening on the thoracic wall detected by computed tomography. Surgical resection confirmed the diagnosis of an *Aspergillus* abscess. We report this case in view of the rarity of Aspergillus abscess localized to a parietal pleura without any signs of lung parenchymal involvement. After a thorough literature review, we consider this could be the first report of this manifestation. Accumulation of similar cases will be necessary to help spread recognition of this condition.**

**Key words:** *Aspergillus*, abscess, empyema, fungal infection, pulmonary aspergillosis

Pulmonary aspergillosis refers to fungal infection in the thoracic area caused by *Aspergillus* species, most commonly *A. fumigatus, A. flavus,* and *A. terreus* [1]. Patients with this condition present with a variety of clinical manifestations ranging from space-occupying lesions (SOLs) to systemic allergic reactions to *Aspergillus* antigens [2]. An abscess caused by an *Aspergillus* species is classified as "invasive aspergillosis." Typically, such an abscess will locate in the thoracic cavity, presenting as an empyema, and potentially causing various degrees of respiratory and other systemic symptoms [3, 4]. Here, we describe a case of an asymptomatic man with an incidentally found *Aspergillus* abscess, localized on a small pleural area without any signs of lung parenchymal involvement. To the best of our knowledge, this is the first report of a presentation of this form of invasive aspergillosis.

The patient was an asymptomatic 73-year-old Asian man who was referred to our department of respiratory medicine for the investigation of pleural thickening. The lesion was incidentally found by computed tomography (CT) scan at another hospital when he was admitted for bacterial pneumonia one month prior to the referral. He had a history of hypertension and had undergone appendectomy for appendicitis. He denied any occupational exposure to dust throughout his life. He was a former smoker of 20 cigarettes per day from the age of 20 to 70 years. Spirometry at the first outpatient visit showed a ratio of forced expiratory volume in 1 sec to forced vital capacity of 0.58, highly suggestive of obstructive pulmonary disease.

There were emphysematous changes on CT scan (Fig. 1A). However, he refused to take an inhaled bronchodilator. On clinical examination, contrast-enhanced CT revealed a partially necrotic nodular lesion (3.5 × 1.3 cm) on the right upper thoracic wall, just behind the 3rd costal bone (Fig. 1B). The laboratory workup was negative for tumor markers and $\beta$-D-glucan. We did not perform interferon-gamma release assays. CT scan did not show any change in the lesion or recurrence of symptoms at the 2-month outpatient

Received December 9, 2021; accepted March 23, 2022.
*Corresponding author. Phone: +81827-34-1000; Fax: +81827-35-5600
E-mail: mowatanabe@s.okayama-u.ac.jp (M.Watanabe)

Conflict of Interest Disclosures: No potential conflict of interest relevant to this article was reported.

follow-up. However, during an outpatient visit at 3 months, the 2-[fluorine-18] fluoro-2-deoxy-d-glucose (FDG) uptake on positron emission tomography (PET) showed a peak standardized uptake value of 2.41, raising suspicion of malignancy (Fig. 1C).

The location of the pleural thickening did not allow transbronchial lung biopsy (TBLB). CT-guided needle biopsy was not performed because the patient's lungs showed severe emphysematous changes on imaging studies, and we therefore considered complicating pneumothorax and dissemination of malignant cells throughout the thoracic cavity to be a potential risk in the case that the lesion was malignant. Thus, thoracic surgery was consulted for surgical resection of the lesion.

Video-assisted thoracic surgery (VATS) was performed with a presumptive diagnosis of thoracic wall malignancy. Due to severe adhesion between the right upper lobe and the thoracic wall, the procedure required additional chest opening at the 3rd intercostal margin right above the lesion. A successful right upper lobe partial resection was performed after the lesion was detached with the layer outside the parietal pleura. A small incision of the dissected lesion revealed purulent discharge, and the surgery was completed at that point. The postoperative course was unremarkable, and the patient was discharged on postoperative day 4.

Pathological reports showed the formation of a partially necrotic granuloma, accompanied by colonization of branching hyphae positive for periodic acid-Schiff staining, strongly suggestive of an *Aspergillus* infection (Fig. 2A, B). The surrounding area formed strong fibrous adhesions from the lung parenchyma to the parietal pleura due to the significant inflammatory

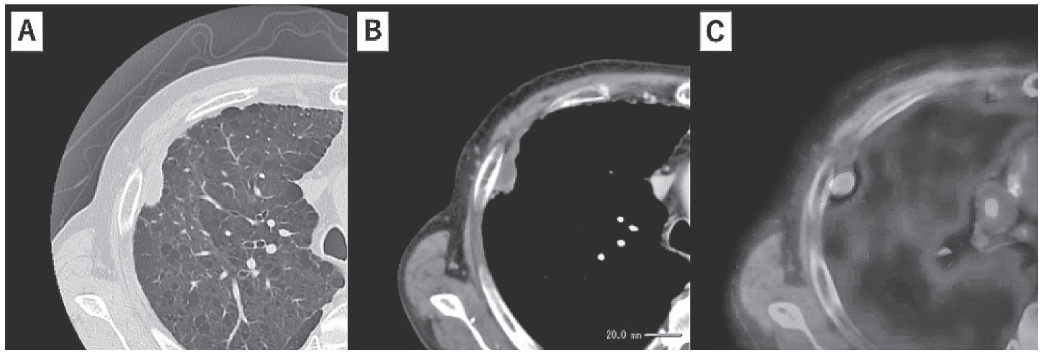


**Fig. 1** (**A**) There were emphysematous changes on CT scan. (**B**) Contrast-enhanced CT scan revealed necrotic space-occupying lesion just behind the 3rd costal bone. (**C**) Positron emission tomography scan showed mild 2-[fluorine 18] fluoro-2-deoxy-d-glucose (FDG) uptake by the lesion.

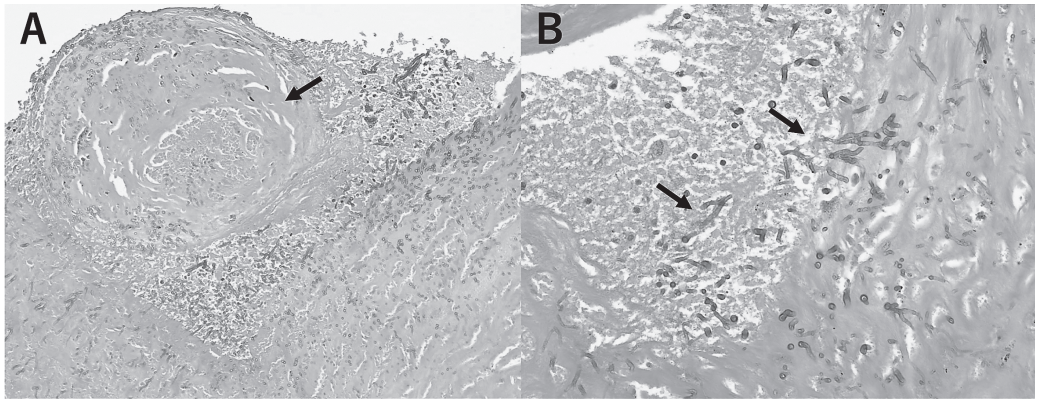


**Fig. 2** (**A**) Pathology showed the formation of partially necrotic granuloma (arrow). (**B**) The colonization of branching hyphae positive for periodic acid-Schiff staining, suggesting *Aspergillus* infection (arrows).

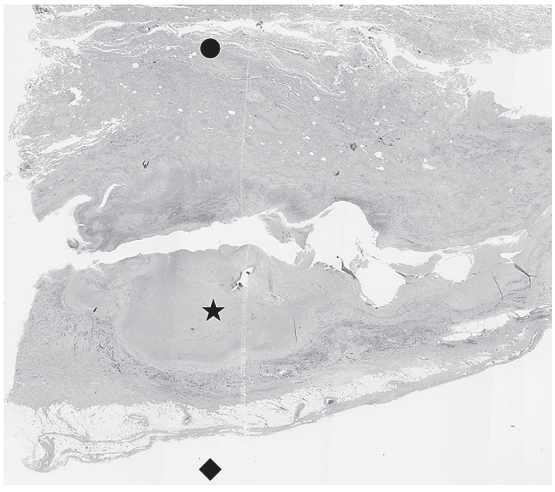

**Fig. 3** Overview of the lesion and the surrounding area. The circle (●) indicates the lung parenchyma, and the rhombus (◆) the side toward the thoracic wall. The star (★) indicates the lesion, which is predominantly located around the parietal pleura. The border between the parietal pleura and visceral pleura could not be determined due to the strong inflammatory response surrounding the lesion.

response. Therefore, the boundary between the visceral pleura and parietal pleura could not be distinguished (Fig. 3). There was only a minor involvement of the inflammatory process in the lung parenchyma. Purulent discharge by a small incision to the lesion during the surgery was taken into consideration, and a confirmatory diagnosis of *Aspergillus* abscess was made. The cultures for general bacteria and Mycobacterium spp. were negative. The patient was doing fine during the outpatient follow-up at 13 months, and no recurrence of a similar lesion was observed by CT scan.

## Discussion

Pulmonary aspergillosis can present with a variety of clinical manifestations. To date, three main spectra of *Aspergillus*-related lung disease have been recognized: chronic pulmonary aspergillosis; allergic bronchopulmonary aspergillosis; and invasive pulmonary aspergillosis [1,2]. *Aspergillus* abscess can be categorized as invasive aspergillosis. There are several different forms of invasive aspergillosis, many of which are associated with strong respiratory and systemic symptoms [2]. In cases with abscess formation, most of them present as empyema [2-4]. They are often refractory to medical and surgical intervention, and can lead to a detrimental course [3-6]. However, in our case, the lesion was localized to an extremely limited area, and the patient was asymptomatic throughout the course. In addition, there were no infiltrative or nodular shadows in the lung fields on imaging studies, which typically could be seen in invasive Aspergillosis. Consequently, total surgical resection alone led to good clinical progress. We did not administer any postoperative antifungal agents because we were able to resect the lesion completely. We continued outpatient follow-up only, in accordance with recommendations for the management of surgically resected *Aspergillus* nodules [7,8].

Patients with high-risk characteristics for Aspergillus infection of the pleura or pleural space are as follows: those who have undergone thoracic surgery; those with an immunocompromised status; and those with a history of tuberculosis infection [3,4]. In our case, the exact origin of the *Aspergillus* infection was unknown. The patient denied any history of the precipitating risk factors mentioned above. Imaging studies did not detect any cavitary lesions in the lungs, which could facilitate colonization of *Aspergillus* species. We assumed that the patient was an immunocompetent host, since he did not undergo any immunosuppressive therapy, have a history of cancer, or lead a lifestyle that included intravenous drug use, multiple sexual partners, or homosexuality. Therefore, we hypothesized that his immunological response towards the local *Aspergillus* infection was sufficient to envelop the abscess by granuloma formation, preventing further expansion around the area.

We herein report a rare case of invasive Aspergillosis, presenting as an abscess localized to a parietal pleura in an asymptomatic patient. Early detection and surgical resection of the lesion produced a favorable outcome. To our knowledge, this is the first case report of this type of manifestation of *Aspergillus* infection. *Aspergillus* abscess should be one of the differential diagnoses when SOL on the thoracic wall is detected by imaging studies. We believe that further accumulation of similar case reports will be necessary to help spread recognition of this condition.

## References

1. Al-Alawi A, Ryan CF, Flint JD and Müller NL: Aspergillus-related lung disease. Can Respir J (2005) 12: 377–387.
2. Kosmidis C and Denning DW: The clinical spectrum of pulmonary

aspergillosis. Thorax (2015) 70: 270–277.
3. Krakowka P, Rowinska E and Halweg H: Infection of the pleura by Aspergillus fumigatus. Thorax (1970) 25: 245–253.
4. Hillerdal G: Pulmonary aspergillus infection invading the pleura. Thorax (1981) 36: 745–751.
5. Hara A, Nagase H, Kuramochi M, Ishida H, Adachi T, Suzuki N and Ohta K: A case of allergic bronchopulmonary aspergillosis complicated with lung abscess which developed into a bronchopleural fistula. Nihon Kokyuki Gakkai Zasshi (2009) 47: 432–437.
6. Rai K, Higo H, Makimoto G, Watanabe H, Itano J, Sato K, Fujiwara K, Shibayama T, Yonei T and Sato T: A case of treatment-refractory Aspergillus Abscess, successfully treated with drainage by percutaneous pleural catheter and occlusion by endobronchial Watanabe spigots. T J Jpn Soc Resp Endoscopy (2015) 37: 388–392.
7. Patterson TF, Thompson GR, Denning DW, Fishman JA, Hadley S, Herbrecht R, Kontoyiannis DP, Marr KA, Morrison VA, Nguyen MH, Segal BH, Steinbach WJ, Stevens DA, Walsh TJ, Wingard JR, Young JH and Bennett JE: Practice Guidelines for the Diagnosis and Management of Aspergillosis: 2016 Update by the Infectious Diseases Society of America. Clin Infect Dis (2016) 63: e1–e60.
8. Muldoon EG, Sharman A, Page I, Bishop P and Denning DW: Aspergillus nodules; another presentation of Chronic Pulmonary Aspergillosis. BMC Pulmonary Medicine (2016) 16: 123.