

Case report

Chronic necrotizing pulmonary aspergillosis in emphysematous bullae

Saulius Cicėnas^{1,2},

Vladislavas Vencevičius¹

¹Institute of Oncology, Vilnius University, Lithuania

²Institute of Rehabilitation, Sport Medicine and Nursing, Vilnius, Lithuania

Background. A rare case of necrotizing aspergilloma in the right lung emphysematic cavity.

Materials and methods. We are presenting a rare clinical case of fungal pathology: *Aspergillus* localized in lung bullae associated with emphysema. That is why clinical and X-ray appearance differs from classical aspergilloma.

Results. The patient underwent investigations due to the progression of tuberculosis, but it was not found. On chest X-ray the right upper lobe lung cancer was suspected. Haemoptysis was consistent, and the patient underwent right thoracotomy and lung resection (segmentectomy). Histology after the operation was chronic necrotizing aspergilloma. This case is unique and interesting, because necrotizing aspergilloma was localized in emphysematic cavity. Postoperative care was without complications, the patient returned to work in 3 months. In our case, depending on the process stage, lung resections (wedge or segmentectomies) were performed. Patient's performance status and lung functions are of great importance.

Conclusions. 1. Surgery for chronic necrotizing pulmonary aspergillosis is mandatory in cases of: appearance of clinical manifestation of the disease, disease progression – bleeding from lungs, tumour size enlargement during conservative treatment. Surgery is prohibited when fungal disease is widely spread in both lungs. 2. The main surgical operation in case of necrotizing pulmonary aspergillosis is lung resection.

Key words: chronic necrotizing pulmonary aspergillosis, aspergilloma of the lung, tuberculosis of the lung, haemoptysis, thoracic surgery

INTRODUCTION

In 1981, Geftter, Weingrad, Epstein et al. (1) described a small series of semi-invasive cavitary pulmonary aspergillosis expanding the full spectrum of traditional saprophytic, allergic or invasive form of *Aspergillus* – induced respiratory pathology. This variant called chronic necrotizing pulmonary aspergillosis (CNPA) is characterized by local fungal invasion in the underlying lung disease and modest immunosuppression.

In 1997, Verweij and Dennig described the definition of broncho-pulmonary aspergillosis infections in non-immunocompromised patients which still remains vague in a wide range of clinical, radiological and pathological entities and has been described by a variety of names, i. e. simple aspergilloma, semi-invasive aspergillosis, chronic necrotizing pulmonary aspergillosis caused by *Aspergillus*. However, these disease entities share common characteristics suggesting that they belong to the same

group of pulmonary aspergillosis infections. CNPA is an indolent cavitating lesion that occurs due to a localized invasion of the lung tissue by *Aspergillus* (2). Chest radiography showed a patchy infiltrate with cavities. In many cases such a lesion can be mistakenly diagnosed as tuberculosis, histoplazmosis, anaerobic pneumonia or vasculitis. Diagnosis is established by tissue evidence of invasive *Aspergillus* when serum precipitating (IgG) antibodies are present. Treatment includes antifungal agents and surgical resections of the lesion. Prognosis of the disease depends on the underlying lung structure and host's immune response (3).

MATERIALS AND METHODS

Patient S.T. recovered from lung tuberculosis 10 years ago and then received therapy for one year period. Afterwards the physicians followed up the patient, and no recurrence was detected. Recently he started feeling general malaise, coughed up sputum with blood 2 months ago and lost 6 kg of weight. He was treated as an outpatient with antibiotics; however, the clinical state did not change. After the consultation by a TB physician

Correspondence to: Saulius Cicėnas, Department of Thoracic Surgery and Oncology, Institute of Oncology, Vilnius University, Santariškių 1, LT-08660 Vilnius, Lithuania. E-mail: saulius.cicenas@vuoi.lt

progression of tuberculosis was excluded, but a tumour of the superior lobe of the right lung was suspected. There was infiltration of S2 of the superior lobe of the right lung as well as bullous emphysema of S1–3 and paracostal thin wall cavity beside S2 seen on X-ray radiograms. Fibrobronchoscopy revealed traces of blood in the upper lobe and non specific bronchitis. It was then decided to perform an operation, as the status of the patient had improved: he coughed up blood and lung cancer was suspected. But the identification of *Aspergillus fumigatus*

in the sputum as well as in the transtracheal aspiration material and the presence of the precipitating specific serum antibodies were negative. A thoracotomy was performed and the superior lobe of the right lung was found to have adhesions that were dissected extrapleurally. Infiltration in S1–3 and bullous emphysema as well as thick wall bulla in S2 projection were also detected. Superior right lobectomy was performed. Pathohistological diagnosis after the operation revealed chronic necrotizing aspergillosis of the lung. This case has presented atypical findings in X-ray radiograms because of the fungal pathology developed in bullous cavities and necrotic inflammation process.

There were no postoperative complications. Three months later the patient was well and returned to full-time work, reporting a marked increase in working tolerance compared to his former state.

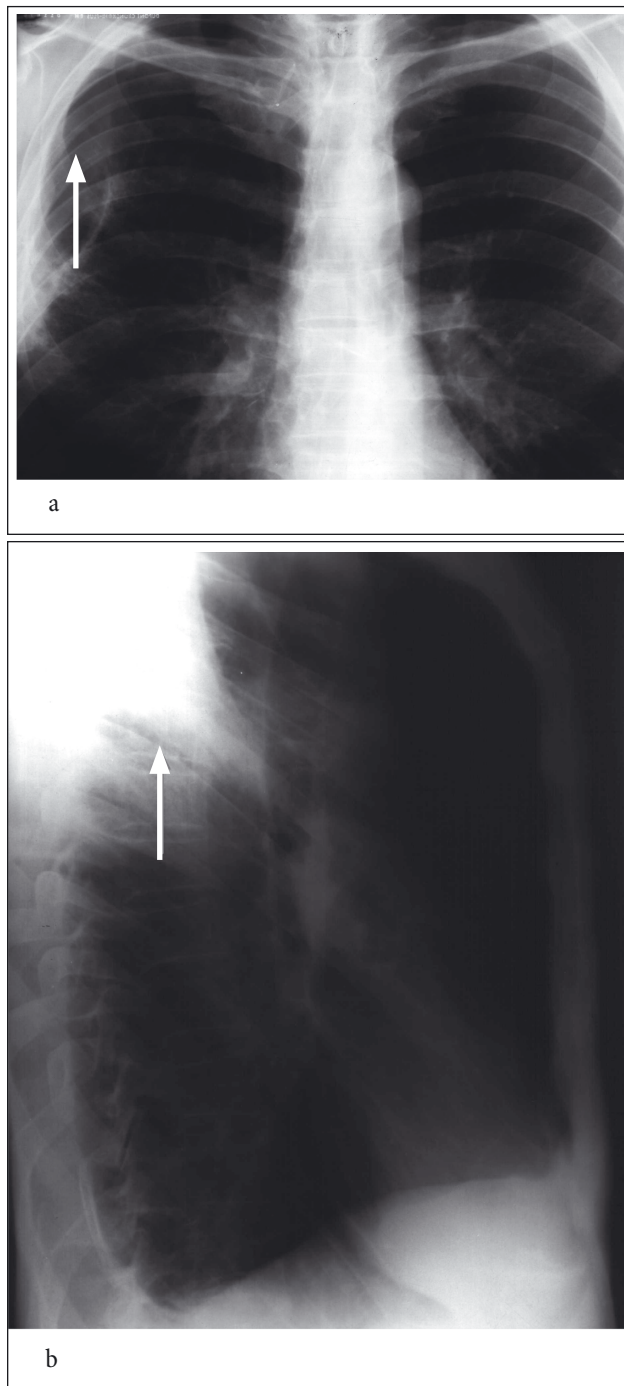


Fig. 1. AP (a) and lateral (b) chest X-ray radiograms show 6.0 x 3.5 cm thin wall bulla with surrounding lung infiltration and hilar reaction in the superior lobe of the right lung

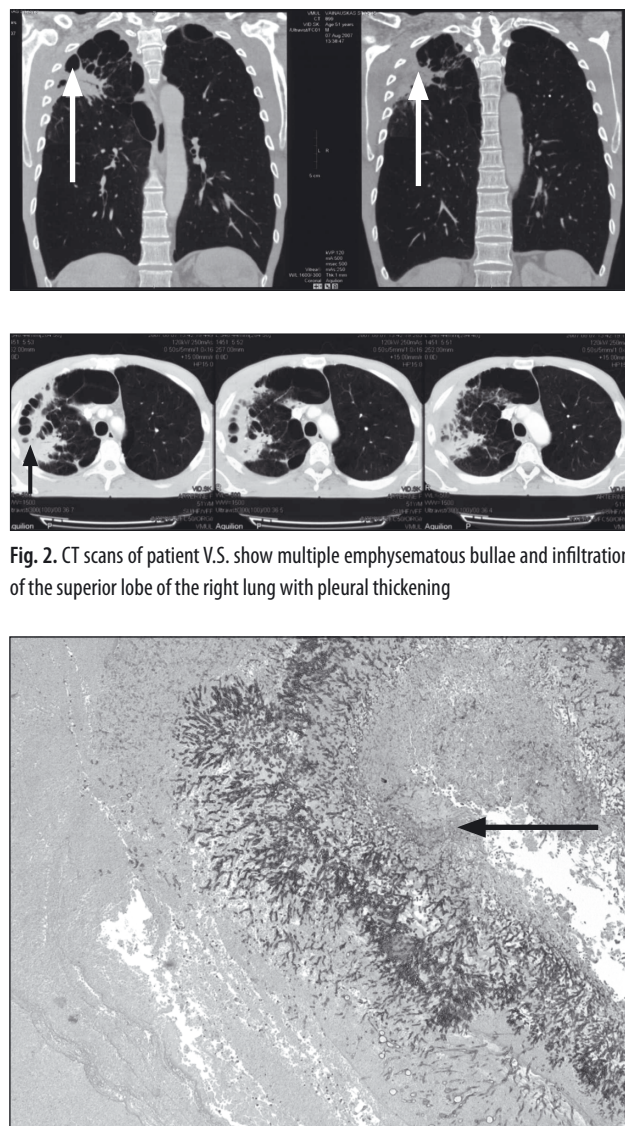


Fig. 2. CT scans of patient V.S. show multiple emphysematous bullae and infiltration of the superior lobe of the right lung with pleural thickening

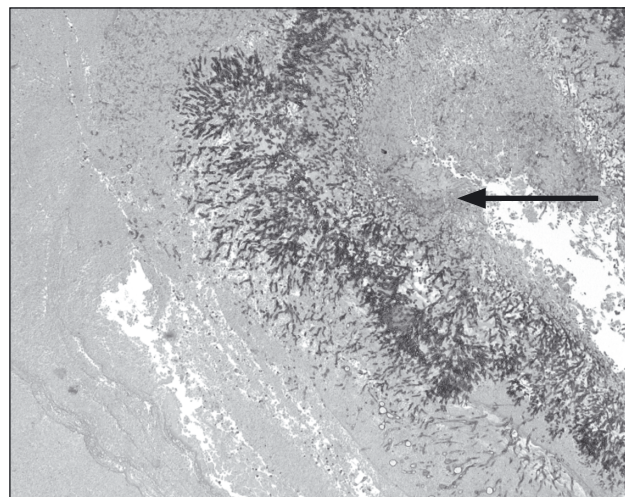


Fig. 3. H&E (hematoxylin and eosin) staining, magnification x40. Fungal hyphae in fibrin

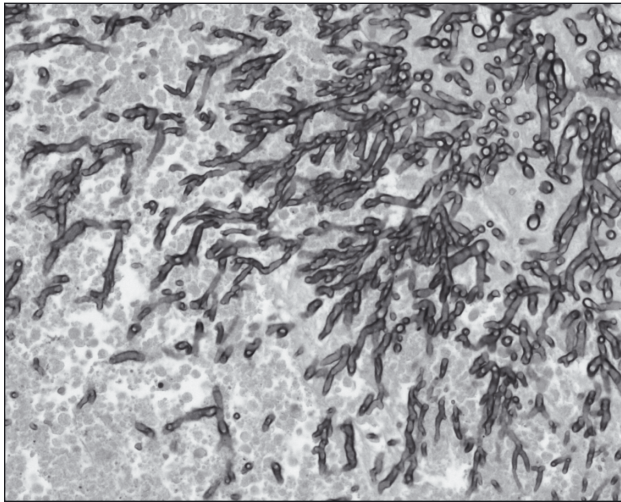


Fig. 4. H&E (hematoxylin and eosin) staining, magnification x200. Fungal hyphae with 45° branches

DISCUSSION

A spectrum of pulmonary pathologies by the ubiquitous *Aspergillus* fungi has been described with the clinical presentation and prognosis of the disease depending on the underlying lung structure and the host's immune response. In the most benign variant, preformed lung cavities such as tuberculosis caverns may be colonized with *Aspergillus*, forming noninvasive mycetoma (4, 5). Chronic necrotizing pulmonary aspergillosis is an uncommon subacute locally invasive form of *Aspergillus* lung infection. (1). This variant called CNPA is characterized by local fungal invasion in the underlying lung disease and modest immunosuppression. It should be observed that invasive pulmonary aspergillosis is often diagnosed only at autopsy because of the difficulty in diagnosing its rapid progression and the restricted options of useful antifungals (6, 7).

Pathologic diagnosis requires demonstration of septate hyphae typical of *Aspergillus* species invading lung parenchyma on biopsy specimens, accompanied by *Aspergillus* culture growth (8, 9). Binder, Faling, Pugatch et al. (10) described a similar locally invasive form of the disease and coined the term chronic necrotizing pulmonary aspergillosis. Most patients who had suffered from an underlying pulmonary disease had accompanying fever, cough and expectoration. Chest radiography showed a patchy infiltrate with a cavity. In many cases such a lesion can be mistakenly diagnosed as tuberculosis, anaerobic pneumonia or vasculitis. Diagnosis is established by tissue evidence of invasive *Aspergillus*. Treatment includes antifungal agents and surgical resections of the lesions (11).

Aspergillus may cause a variety of pulmonary diseases, depending on the immune status and the presence of underlying lung disease. The manifestations range from invasive pulmonary aspergillosis to chronic necrotizing aspergillosis in patients with chronic lung disease and/or mildly compromised immune systems. This review provides a clinical update on the epidemiology, risk factors, clinical presentation, diagnosis and management of the major syndromes associated with pulmonary aspergillosis (12).

The choice of treatment, especially surgery, was selected based on the patients' pulmonary functions and clinical conditions. Generally, surgery was performed for patients in whom conservative treatment had not proven effective (13). These patients were initially given conservative treatment but, as their severe symptoms did not improve, surgery was ultimately performed.

We propose the following criteria for surgery: patients must be symptomatic, the lesion should not be widespread and uncontrollable where superimposed bacterial infection exists and the patient must be assessed as being able to tolerate a surgical procedure. Conservative treatment including the intracavitary instillation of antifungal drugs has proven ineffective.

CONCLUSIONS

1. Surgery for chronic necrotizing pulmonary aspergillosis is mandatory in cases of: appearance of clinical manifestation of the disease, disease progression – bleeding from the lungs, tumour size enlargement during conservative treatment. Surgery is prohibited when fungal disease is widely spread in both lungs.
2. The main surgical operation in case of necrotizing pulmonary aspergillosis is lung resection.

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Saulius Cicėnas, Vladislavas Vencevičius

CHRONINĖ NEKROTIZUOJANTI ASPERGILIOZĖ EMFIZEMATOZINĖJE ERTMĖJE

Santrauka

Aptariamas retas nekrotizuojančios aspergilomos atvejis dešiniojo plaučio emfizeminėje ertmėje.

Prieš 10 metų plaučių tuberkuloze susirgusiam asmeniui apie metus buvo taikytas prieštuberkuliozinis gydymas. Vėliau jautėsi gerai, dirbo tiesioginį darbą. Prieš 2 mėn. ligonis pradėjo jausti bendrą silpnumą, atsirado kosulys su krauju priemaiša, neteko 6 kg svorio. Gydytas antibiotikais (penicilinu, reflinu), tačiau būklė negerėjo. Ligonis buvo ištirtas dėl plaučių tuberkuliozės progresavimo, tačiau progresavimo požymių nerasta. Atlikus rentgeno diagnostiką įtartas dešiniojo plaučio

viršutinės skilties navikas: dešiniojo plaučio S2 projekcijoje stebimas apvalus darinys, kurį supa emfizeminė plonasiene ertmė. Skrepliuose *Aspergillus fungatus* sukėlėjo nerasta. Serumo reakcija į precipitiną taip pat neigiama. Atlikta fibrobronchoskopija: B1,2,3 spindyje aptikti kraujo pėdsakai bei nespecifinis endobronchitas. Kadangi atsikosėjimas krauju kartojosi, būklė negerėjo, ligoniui buvo atlikta dešinioji torakotomija: plautis išdalintas ekstrapleuraliai dėl gausių suaugimų. S1,2,3 projekcijoje plonasieneje ertmėje palpuojamą darinį nutarta pašalinti. Patologo histologinė diagnozė – lėtinė nekrozuojanti aspergiloma. Šis atvejis yra įdomus tuo, kad nekrotizuojanti aspergiloma lokalizavosi emfizematoziniame ertmėje. Pooperacinė eiga normali. Dažniausiai tolimieji rezultatai po tokių operacijų geri, tačiau būtina parinkti adekvatų gydymą. Mūsų nuomone, atliekama kraštinė plaučių rezekcija, lobektomija ar net pulmonektomija turi būti atliekama atsižvelgiant į proceso išplitimą, be to, labai svarbi bendra ligonio būklė ir kvėpavimo sistemos rezervas.

Išvados: 1. Lėtinė nekrotizuojanti plaučių aspergiloma šalinama chirurginiu būdu šiais atvejais: kai liga išlieka nepaisant konservatyvaus gydymo; procesui progresuojant; kraujuojant; rentgenologiškai nustatčius padidėjusį patologinio proceso diametrą konservatyvaus gydymo metu. Chirurginis gydymas netaikomas esant abipusiam procesui.

2. Pagrindinė chirurginė intervencija nekrotizuojančios plaučių aspergilomos atveju – plaučių rezekcija.

Raktažodžiai: lėtinė nekrotizuojanti plaučių aspergiloma, plaučių aspergiloma, plaučių tuberkuliozė, kraujavimas iš plaučių, krūtinės chirurgija