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Spontaneous pneumothorax followed by reversed halo sign in immunocompromised patient with pulmonary mucormycosis

Majid Moosavi Movahed [a], Hadiseh Hosamirudsari [b,*], Fariba Mansouri [c], Farzaneh Mohammadizia [d]

[a] Department of Nephrology, Baharloo Hospital, Tehran University of Medical Sciences, Behdari Street, Railway Square, Tehran, Iran
[b] Department of Infectious Diseases, Baharloo Hospital, Tehran University of Medical Sciences, Behdari Street, Railway Square, Tehran, Iran
[c] Department of Pulmonary Diseases, Baharloo Hospital, Tehran University of Medical Sciences, Behdari Street, Railway Square, Tehran, Iran
[d] Department of Pathology, Baharloo Hospital, Tehran University of Medical Sciences, Behdari Street, Railway Square, Tehran, Iran

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ABSTRACT

Mucormycosis, an invasive fungus with a variety of clinical presentation, is a devastating infection in immunocompromised host. Here an unusual case of pulmonary mucormycosis is introduced in an immunodeficient patient in which pneumothorax was followed by reversed halo sign (RHS). The clinicians, who visit immunocompromised persons with pneumothorax, should be considerate to take immediate imaging and pathologic measures to confirm or reject mucormycosis.

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1. Introduction

Mucormycosis is a ubiquitous fungus that lives in spoiled vegetation and soil. Human can be infected through inhalation, ingestion and skin inoculation [1,2]. Immunocompromised patients might bear a variety of presentations such as rhino-orbito-cerebral, pulmonary, gastrointestinal, cutaneous, central nervous system and disseminated form [1]. Rhino-orbito-cerebral and pulmonary mucormycosis are the most common forms; the latter progresses rapidly to devastating diseases [1].

Presentation of pulmonary mucormycosis may be confused with several infectious and non infectious diseases; therefore its diagnosis is difficult [3]. Chest CT scan of a patient with pulmonary mucormycosis might demonstrate such presentations as air crescent sign, CT halo sign, solitary or multiple pulmonary nodules or masses, bronchopulmonary fistula and pulmonary artery pseudo aneurysm [1,4].

The occurrence of pneumothorax and its subsequent RHS along with sinusitis, supported by radiology and pathology in a 33 years old patient, is considered as a rare case of pulmonary mucormycosis.

2. Case

Following is the treatment history of a man, 33 years old in July 2014 (day +14), when he died in our hospital as a result of multiorgan failure. He was a farmer, married, with no history of alcohol consumption, illicit drugs use and extramarital relationships.

On day −330: Because of edema he visited a medical center in another city, where he was admitted and membranous glomerulonephritis was diagnosed. Consequently the followings administered; prednisolone and cyclosporine for 2 months, cellcept for 1 month, prednisolone and cyclophosphomide for 7 months.

On day −75: Because of progressive generalized edema he was admitted again. He was discharged under treatment with prednisolone (50 mg/daily) and tacrolymus (1 cap/BD). During the next 2 months he was feeling fine. On day −28, he travelled to village for a few days and he had close contact with sheep. On day −14 the patient reported fever, cough and dyspnea with mild erythema and pain in right eye. Anorexia, weakness, dyspnea and conjunctivitis developed and persisted despite of outpatient antibiotic treatment.

In the emergency department (day 0): The patient suffered from dyspnea, weakness and right eye pain. On examination, he was agitated but completely conscious with diaphoretic skin. The vital signs have been as follows; temperature, 36.7 C; blood pressure, 100/60 mmgh; pulse rate, 99 beats per minute; respiratory rate, 22 beats per minute and oxygen saturation in air

* Corresponding author. Fax: +98 21 5564 8189.
E-mail address: h-hosami@sina.tums.ac.ir (H. Hosamirudsari).

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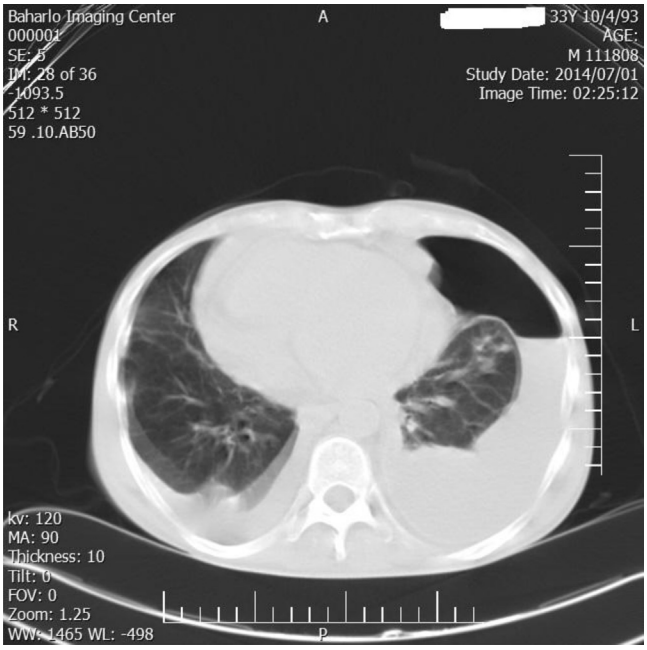

Fig. 1. Axial noncontrast chest computed tomography (CT) image on day 0: showing large pneumothorax with pleural effusion.

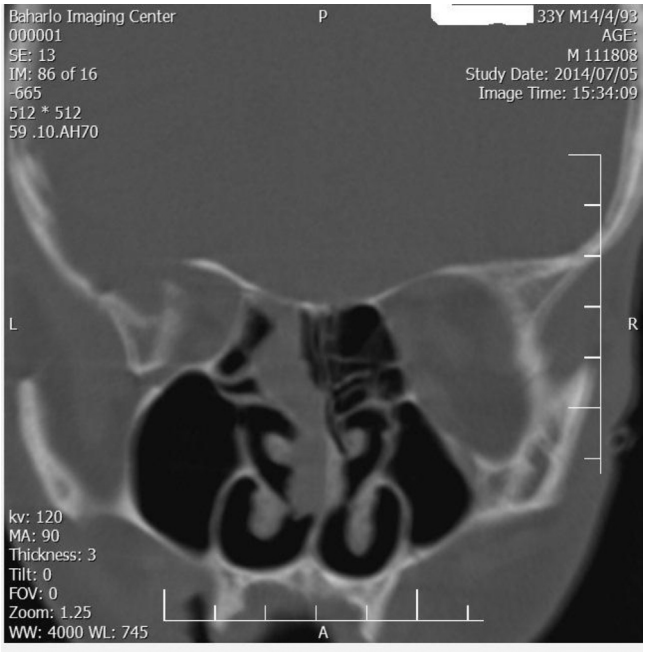

Fig. 3. Coronal PNS CT scan shows soft tissue density lesion in ethmoid sinus with extension to orbit without bone destruction.

room, 99%. He had suprasternal retraction with nasal flaring. In right eye, there was moderate conjunctivitis with eyelid erythema; its movement was normal. The tongue and buccal mucosa were covered with multiple white vegetative lesions. The left lung vesicular sounds decreased with fine rales in both bases. The abdomen was distended without tenderness. There was +3 pitting edema in extremities.

The first day chest CT scan revealed large pleural effusion with pneumothorax in the left side and a shift of mediastinum to the right side. There were also multiple pulmonary nodules with right side pleural effusin (Fig. 1). Consequently drainage with chest tube was administered by the surgeon immediately. Because of the immunocompromised state of the patient, parenteral trimethoprim- sulfamethaxazol (160 mg/IV/TDS) and meropenem (1 g/IV/BD) were administered for pneumocystis jiroveci and multi drug resistant organisms. We also started fluconazole for oral lesions.

On day +3, right eye erythema was extended with small size necrosis in right nasal ala without bleeding (Fig. 2). The tongue's white vegetative plaques turned to brown. With doubt of rhino-orbital mucormycosis, biopsies of tongue and nasal septa were carried out and amphotericin B deoxycholate (1 mg/kg/daily) was administered. On day +4, paranasal sinuses (PNS) CT scan revealed soft tissue density lesion in left ethmoid sinus and nasal passage with extension to left orbit (Fig. 3).

On day +4, he was transferred to Intensive Care Unit (ICU) because of respiratory distress and was intubated after 48 h; his conscious level was 14+T/15. Smear of sputum for acid fast bacilli, stain and culture of sputum for pneumocystis jiroveci were negative. HIV antibody test, D-dimer Enzyme -Linked Immunosorbent Assay (ELISA) and collagen vascular diseases screening tests were negative (Table 1). Pleural fluid cytologic examination and Polymerase Chain Reaction (PCR) for fungi were negative.

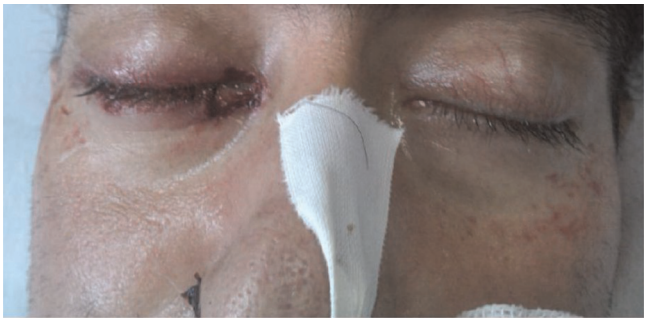

Fig. 2. Right eye conjunctivitis with medial side necrosis and black scar in right nasal ala.

On day +6, laboratory results revealed pancytopenia (Table 1), however in peripheral blood smear there was no schistocyte or immature cells. Bone marrow biopsy which could help in finding the cause of pancytopenia was nevertheless postponed due to the patient's poor condition and low platelet count. Meanwhile his blood pressure was decreased and we started inotropic drugs. Hemodynamic instability of the patient prohibited further attempts such as bronchoscopy and CT guided lung biopsy to identify the causative agent.

On day +9, chest CT-scan showed RHS with multiple nodules (Fig. 4). This was considered as indicating invasive pulmonary mucormycosis. The patient's condition was downgraded on day +11. His level of consciousness suddenly decreased to 4+T/15. Also, in right eye lid, nose and trachea, bleeding appeared. Unfortunately, he died on day +14.

Histopathology of tongue biopsy, revealed mucormycosis (Figs. 5 and 6). Section of tounge biopsy showed ulcerated squamous epithelial mucosa with subepithelial fibro fatty tissue and skeletal muscle fibers of tongue that is on site of ulceration. The nonseptate short and wide hyphae of mucormycetes were present. The inflammatory granulation tissue and inflammatory multinucleated giant cells were also seen in Gimsa staining, but no cyst and trophozoite of pneumocystis jiroveci were identified.

3. Discussion

Pulmonary mucormycosis is a devastating infection when it affects immunocompromised patients such as diabetes mellitus, hematologic malignancies, chronic renal failure, post transplantaion and immunosuppressed persons [5]. Herein, we present a

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