Journal de Mycologie Médicale (2017) 27, 396—399

Available online at ScienceDirect www.sciencedirect.com

Elsevier Masson France EM|consulte www.em-consulte.com

CASE REPORT/CAS CLINIQUE

Coexistence of aspergilloma and pulmonary hydatid cyst in an immunocompetent individual

F. Aala [a], H. Badali [b], S. Hashemi Fesharaki [b,c], M. Boroumand [c], M. Sotoudeh Anvari [c], H. Davari [d], S. Agha Kuchak Afshari [e], S. Khodavaisy [e,*]

[a] *Department of Medical Parasitology and Mycology, Faculty of Medicine, Kurdistan University of Medical Sciences, Sanandaj, Iran*
[b] *Department of Medical Mycology and Parasitology, Invasive Fungi Research Center (IFRC), School of Medicine, Mazandaran University of Medical Sciences, Sari, Iran*
[c] *Department of Pathology and Laboratory Medicine, Tehran Heart Center, Tehran University of Medical Science, Tehran, Iran*
[d] *General Thoracic Surgeon, Tehran Heart Center, Tehran University of Medical Science, Tehran, Iran*
[e] *Department of Medical Parasitology and Mycology, School of Public Health, Tehran University of Medical Sciences, Tehran, Iran*

Received 21 December 2016; received in revised form 18 March 2017; accepted 7 April 2017
Available online 16 May 2017

KEYWORDS
Coexistence;
Aspergilloma;
Hydatid cyst;
Aspergillus flavus

Summary Echinococcosis is a zoonotic disease caused by *Echinococcus granulosus* sensu lato. The liver and lungs are the most commonly sites of infections, but involvements of other organs were also observed. Recently, the coinfection of pulmonary hydatid cyst with aspergilloma has been reported in the literature. Herein, we report a successful treatment of coinfection of cystic echinoccosis with aspergilloma due to *Aspergillus flavus* in a 34-year-old female. In vitro antifungal susceptibility tests revealed that the MIC values for antifungals employed in this case were posaconazole (0.031 μg/ml), itraconazole (0.125 μg/ml), voriconazole (0.25 μg/ml), and amphotericin B (1 μg/ml). The minimum effective concentration for caspofungin was 0.008 μg/ml. This coexistence of active pulmonary echinococcosis and aspergillosis is being reported because of its rarity and clinical importance for its management.

* Corresponding author. Department of Medical Parasitology and Mycology, School of Public Health, Tehran University of Medical Sciences, Tehran, Iran.
E-mail address: Sadegh_7392008@yahoo.com (S. Khodavaisy).

http://dx.doi.org/10.1016/j.mycmed.2017.04.006
1156-5233/

Introduction

Echinococcosis is a zoonotic disease caused by *Echinococcus granulosus* sensu lato [1]. The liver (50—70%) and lungs (20—30%) are the most commonly sites of infections, but involvements of other organs were observed [2,3]. Pulmonary echinococcosis leads to higher susceptibility to saprophytic fungal coinfection than echinococcosis located in other sites, probably because it is in communication with external environment [3,4]. Pulmonary echinococcal hydatid cysts have been reported coexistent with aspergilloma [4—6]. The latter infection consists of a mass of fungal hyphae, inflammatory cells, fibrin, mucus, and tissue debris and can colonize lung cavities due to underlying diseases, such as tuberculosis, sarcoidosis, bronchiectasis, bullae, cavitary, neoplasms, ankylosing spondylitis, bronchial cysts, and pulmonary infarction [7,8]. Aspergilloma caused by *Aspergillus* species, such as *Aspergillus fumigatus* and *Aspergillus flavus* formed in a residual cavity left after a cystectomy, or active proliferation of fungi in laminated pulmonary echinococcal ectocyst [4,9]. Recently, the coexistence of pulmonary hydatid cyst with aspergilloma has been reviewed in the medical literatures [5,6,9,10]. The majority of reported cases were male (70%), and all patients were immunocompetent individuals [10]. Interestingly, the frequency of cases from India followed by Iran was higher than average [4,10,11]. Herein, we report a successful treatment of coexistence of aspergilloma and pulmonary hydatid cyst in a 34-year-old immunocompetent female.

Case report

A 34-year-old female who suffered from chest pain, dyspnea with non-productive cough was admitted to the Tehran Heart Center, Tehran University of Medical Sciences, Tehran, Iran. The patient had a history of hyperthyroidism without any history of pre-existing pulmonary disease, immunosuppression, or extra-pulmonary symptoms, such as hepatic involvement or anaphylactic reaction. In addition, she never had signs or symptoms in her physical examination. Complete blood count revealed a hemoglobin level of 12.4 g/dL, white blood cells count of 13.4 $\times$ 10^3 μL with 67% polymorphonucleocytes, 3% band formations, 22% lymphocytes, 2% monocytes, and 6% eosinophils. Other systemic examination, laboratory investigation including liver function test and liver function test results were within normal ranges. Contrast enhanced computerized tomography (CECT) showed subpleural consolidation with small cavitation and pleural effusion, thickening and enhancement in left lower lobe and suggested a possibility of hydatid cyst (Fig. 1). Abdominal and pelvic CECT revealed a cystic lesion in caudate lobe of liver (33 mm). Therefore, a diagnosis of disseminated thoraco-abdominal hydatid disease was purposed and a left postero-lateral thoracotomy was performed. An opened out and collapsed cyst, resembling the milky-white glistening and soft coconut kernel, was received (Fig. 2). The cyst wall was an acellular laminated membrane with uniform thickness and showed blackish area on the surface. Also, on cutting the cavity of the cyst contained fungal necrotic material. Histopathological examination showed a proliferation of septate hyphae branching on the outer aspect of chitinous wall of the hydatid cyst (Fig. 2). These fungal hyphae were uniform, acute angle branching septate hyphae conforming to morphology of *Aspergillus* spp., surrounded by mixed inflammatory cells. In addition, the remaining samples were inoculated onto Sabouraud's dextrose agar (SDA; Difco), supplemented with chloramphenicol (50 μg/mL), and incubated at 28 °C for up to one week. No bacteria were detected in cultures of the biopsy specimens and serological test for human immunodeficiency virus (HIV) were negative, but several yellowish fungal colonies yielded repeatedly after 4 days of incubation. *A. flavus* was initially identified based on conventional methods. Subsequently, molecular assay were performed for reconfirmation. Briefly, DNA was extracted from fresh cultures with Ultraclean Microbial DNA isolation kit (Mo Bio Laboratories, USA) according to manufacturer's protocol and stored at −20 °C [11, 12]. Their identity was confirmed by DNA sequencing of the partial *b-tubulin (BTU)* gene, using the primers Bt2a (5′-GGT AAC CAA ATC GGT GCT TTC-3′) and Bt2b (5′-ACC CTC AGT GTA GTG ACC CTT GGC-3′). The PCR amplification setup and sequencing were followed as previously described [12, 13]. Sequencing was performed as follows; 95 °C for 1 min, followed by 30 cycles consisting of 95 °C for 10 s, 50 °C for 5 s, and 60 °C. Sequence data obtained compared with GenBank database (http://blast.ncbi.nlm.nih.gov) and was identified as *A. flavus* having 99.5% sequence identity with the ex-type isolate of that species. Additionally, in vitro antifungal susceptibility testing, involving determination of minimum inhibitory concentrations (MIC) and minimum effective concentrations (MEC, for caspofungin only), of seven antifungal agents was performed according to recommendations stated in the Clinical and Laboratory Standards Institute (CLSI) M38-A2 document [14]. Table 1 summarized the MIC results. The patient was treated as having a ruptured hydatid cyst, with albendazole (10 mg/kg/day) and oral itraconazole (200 mg/day for 3 months). Five months after the operation, she showed good response to treatment and remains asymptomatic with no evidence of aspergilloma or recurrent hydatid cyst. Written informed consent was obtained from the patient. This research was approved by the ethics committee and written informed consent was obtained from the patient.

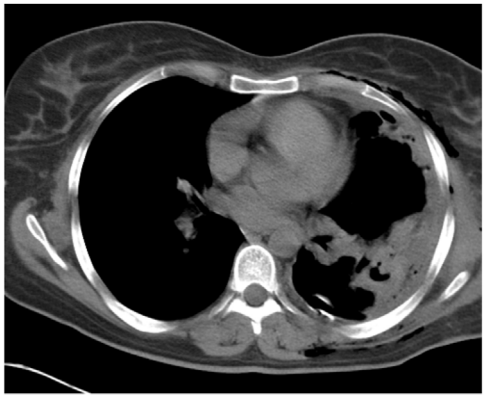

Figure 1 Computerized tomography shows a subpleural consolidation with small cavitation and pleural effusion in left lower lobe.