

Ocular aspergillosis: Obtaining a specimen is crucial for diagnosis. A report of three cases

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ARTICLE INFO

Article history:

Received 2 May 2012

Received in revised form

18 May 2012

Accepted 4 June 2012

Keywords:

Ocular aspergillosis

Endophthalmitis

Dacryocystitis

Aspergillus

ABSTRACT

Aspergillus species have been implicated in a wide variety of primary ocular conditions, characterized by either slow and asymptomatic infection, or rapid, uncontrollable progression and sometimes death. The study design is a case series including chronic asymptomatic dacryocystitis, dacryocystitis with dacryoliths and endogenous endophthalmitis.

Due to this variable clinical presentation of aspergillosis a high level of suspicion must be maintained as fulminant aspergillosis may lead to a rapidly progressive infection which may result in death.

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1. Introduction

Aspergillus is a fungus, usually considered a harmless saprophyte, which is ubiquitous in the environment [1]. *Aspergillus* infections commonly present as a localized disease of the lungs or paranasal sinuses and mainly affect immunocompromised individuals [2].

Aspergillus dacryocystitis is a rare condition which is mostly due to infection of the paranasal sinuses. Dacryoliths are typically found in the setting of chronic infections with superimposed fungal colonization. They are found in up to 30% of patients with chronic dacryocystitis [3].

Endogenous *Aspergillus* endophthalmitis is most commonly reported in immunosuppressed patients with organ transplants or after valve replacement and in individuals suffering from hematological malignancies [4–6]. It may also be the presenting feature of disseminated aspergillosis [6].

We report here three patients infected with two *Aspergillus* species with different clinical appearances. Species identification of isolates was made by macroscopic and microscopic morphology.

2. Case 1

A 74-year old immunocompetent female with left eye blindness due to glaucoma and bullous keratopathy, had phacoemulsification in the right eye in our clinic one month prior to admission with no signs of dacryocystitis (day-30). During the

one month postoperative follow-up visit, the patient expressed that she was content with her cataract surgery (day 0). However, she stated she had been experiencing tearing and secretion in the operated eye for several years; this information was not divulged to the surgeon prior to the cataract surgery. The lacrimal sac area looked normal; there was no edema, tenderness or regurgitation with pressure (day 0). Syringing was facilitated, and both the inferior and superior canaliculi syringing were negative. However, it was observed that the lacrimal sac was full with no drainage. The patient stated that swelling occurred once every 15-days and she drained it into her eye by pressing it with her finger. Dacryocystography with lipiodol revealed an atonic sac with obstruction below the sac. The patient underwent transcanalicular multidiode laser-assisted dacryocystorhinostomy (DCR) with silicone tube intubation (day 7). Conjunctival cultures were obtained preoperatively from the involved and normal sides of the patient and specimens were obtained from the lacrimal sac perioperatively via the endonasal route after the first burn from the laser was seen in the nasal mucosa. Microscopic examination of the samples showed typical *Aspergillus* hyphae (Fig. 1a). The cultures were positive for *Aspergillus niger* (Fig. 1b). Treatment with Amphotericin-B, -topically and locally by syringing the sac after DCR, provided a cure supported by repeated cultures.

3. Case 2

A 29-year old immunocompetent female with a fistulated lacrimal sac phlegmone with multiple white dacryoliths appearing at the opening of the fistula is presented (day 0) (Fig. 2a). The stones occurred in multiple, and they were 2–3 mm in diameter,

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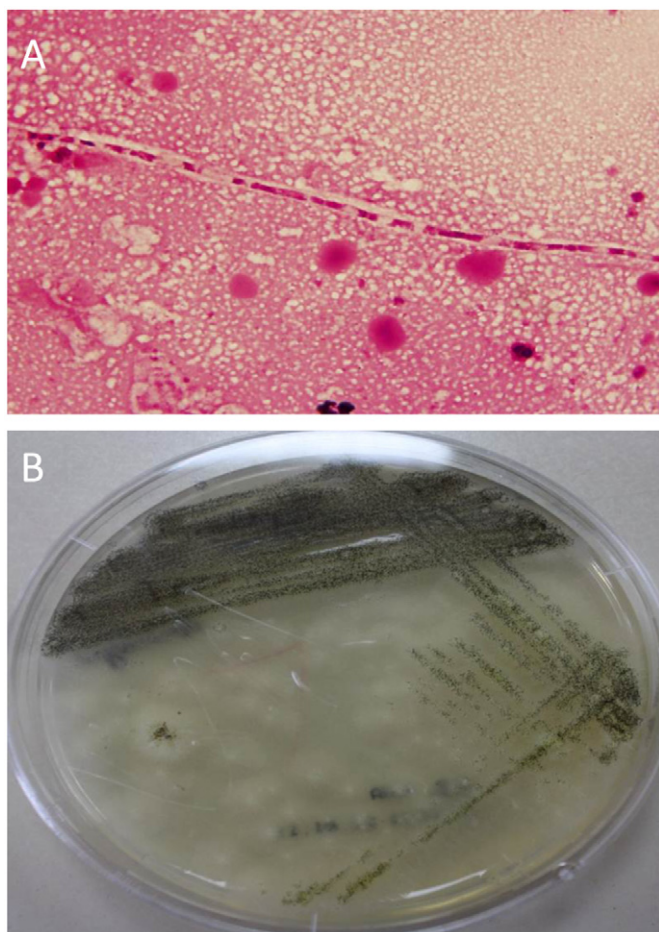


Fig. 1. Patient 1: Gram stain of the lacrimal sac contents revealed septate hyphae (A). Patient 1: Lacrimal sac cultures were positive for *Aspergillus niger* (B).

soft, of a grayish-white color and were able to be mashed easily between the fingers. Pus and mucus filled the remainder of the enlarged sac.

Conjunctival samples were obtained preoperatively from the involved and normal sides of the patient (day +7). Dacryolith removal as well as excision of the fistula opening was facilitated besides transcanalicular multidiode-laser DCR and silicone tube implantation (day +7).

Specimens were obtained from the lacrimal sac via nasal endoscope for microbiological examination at the time of DCR. Cultures obtained from the lacrimal sac grew *Aspergillus fumigatus* (Fig. 2b). Bacterial cultures were negative. Treatment with topical application and local syringing of the sac with Amphotericin-B after DCR, provided a cure as supported by repeated cultures.

4. Case 3

An 11-year old boy diagnosed with acute lymphocytic leukemia, was consulted by the pediatrician with a severe eye infection along with severe pneumonia of an unidentified etiology (day 0). The ophthalmological examination revealed severe endophthalmitis in the right eye with swelling of the eyelids and ocular adnexa, corneal perforation and scleral necrosis with black pigmentations. On admission, the patient was afebrile and had no light perception (day 0). Although macroscopically and clinically aspergillosis was suspected, due to the severity of the pain and lack of light perception, scleral melting and due to the urgency to determine lung disease, the patient's right eye was enucleated (day +2)

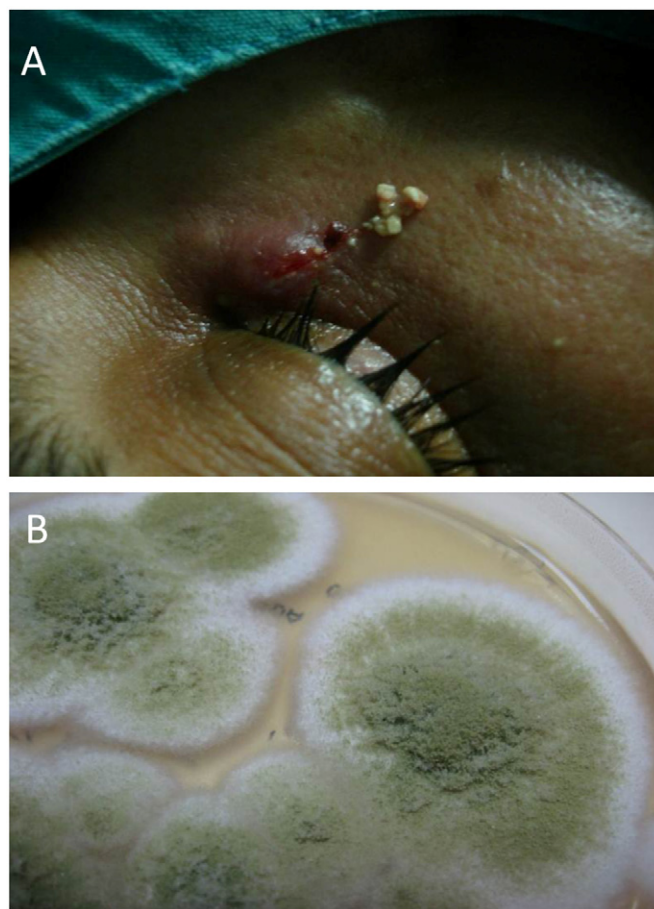


Fig. 2. The opening of the lacrimal fistula with multiple grayish white dacryoliths (A). *Aspergillus fumigatus* colonies on Sabouraud dextrose agar plate (B).



Fig. 3. Enucleation is performed due to massive necrosis and perforation of the globe.

(Fig. 3). Microbiological examination revealed *A. niger*. Despite systemic Amphotericin-B application, septicemia caused death.

5. Discussion

Aspergillus species are usually considered a harmless saprophyte, which are very common in the environment. They rarely

cause invasive or noninvasive infection in immunocompetent hosts.

Acute dacryocystitis is characterized by sudden pain, edema, erythema, and epiphora, in the medial epicanthal area. Infection of the lacrimal sac mostly mimics normal conjunctival flora and in only 1.2% of cases, a fungus is the causative organism [7].

However, in chronic dacryocystitis, *Aspergillus* and *Candida* species may cause a superinfection [8–10]. Dacryocystitis and blepharitis due to *A. niger*, lacrimal sac plugging caused by *A. fumigatus* and chronic dacryocystitis in a healthy 3 year-old boy caused by *Aspergillus flavus* have previously been reported [8–10].

It is a common consensus that dacryocystorhinostomy (DCR) should be performed before cataract surgery in order to minimize the possibility of postoperative endophthalmitis caused by the organisms retained within the lacrimal system. In patients with epiphora, lacrimal sac mucocele and obstruction of the passage, DCR surgery should be performed before cataract surgery. However, Case 1 did not show any signs of epiphora or edema in the lacrimal sac region at the time of cataract surgery due to the regular drainage of the sac by digital pressure performed by the patient. Besides, decreased tear secretion in the elderly may hide the symptoms of epiphora. Because it was the patient's only functional eye, any contamination from the infection in the lacrimal sac might result in sight-threatening infection such as keratitis and endophthalmitis which could have made her completely blind. The lacrimal sac cultures positive for *A. niger* was another surprising finding that we could dismiss if we did not take lacrimal swabs for microbiological evaluation.

Dacryoliths appear as round or oval filling defects on dacryocystography. The etiology of dacryoliths is unclear; however fungi, yeast, and eyelashes have been described as possible niduses [11,12]. Treatment of acute dacryocystitis typically involves treatment with antibiotics in the acute phase, which may or may not be followed by an external dacryocystorhinostomy or other interventional procedure [3]. Adjuvant local irrigation of Amphotericin-B also has been recommended [13]. Complications include abscess and fistula formation as in Case 2.

Endophthalmitis due to *Aspergillus* species is difficult to diagnose because blood cultures, serological tests, and pulmonary radiography may be negative. In Case 3, the etiology of the pneumonia could not be identified because bronchoscopy could not be performed due to the general state of the patient and blood cultures were negative.

In order to identify the agent responsible for endophthalmitis, cultures of pars plana vitrectomy specimens, and examination of Gram- or Giemsa-stained smears are the best diagnostic approaches, because aspergillosis clinically presents with deep retinitis and/or choroiditis [14,15]. Unfortunately we had to perform enucleation due to the scleral necrosis and perforation in the third patient to make a diagnosis.

Aspergillus species have been implicated in a wide variety of ocular involvements ranging from asymptomatic, chronic

infections to rapid, disseminated disease resulting with death. Since the early diagnosis of aspergillosis depends on suspicion, followed by fungal culture findings, we emphasize that obtaining diagnostic material routinely from the appropriate tissues with appropriate techniques for microbiological examination in all eye infections is crucial. Hence, it can be easily facilitated even in laser-assisted DCR patients.

Conflict of interest statement

There are none.

Acknowledgments

There are no acknowledgments for this paper.

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