

Acute dacryocystitis: another clinical manifestation of sporotrichosis

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Sporotrichosis associated with exposure to domestic cats is hyperendemic in Rio de Janeiro, Brazil. A review of the clinical records at our institute revealed four patients with clinical signs of dacryocystitis and a positive conjunctival culture for Sporothrix who were diagnosed with Sporothrix dacryocystitis. Three patients were children (≤ 13 years of age) and one patient was an adult. Two patients reported contact with a cat that had sporotrichosis. Dacryocystitis was associated with nodular, ulcerated lesions on the face of one patient and with granulomatous conjunctivitis in two patients; however, this condition manifested as an isolated disease in another patient. All of the patients were cured of the fungal infections, but three patients had chronic dacryocystitis and one patient developed a cutaneous fistula. Sporotrichosis is usually a benign disease, but may cause severe complications when the eye and the adnexa are affected. Physicians, especially ophthalmologists in endemic areas, should be aware of the ophthalmological manifestations and complications of sporotrichosis.

Key words: dacryocystitis - sporotrichosis - *Sporothrix* - cutaneous fistula - Rio de Janeiro

Sporothrix schenckii is a dimorphic fungus that is responsible for cutaneous disease in endemic areas worldwide. Classically, infection is associated with a traumatic subcutaneous inoculation of contaminated soil, plants or organic matter. The most common clinical form of sporotrichosis is cutaneous, lymphatic disease, which accounts for 75% of cases, followed by localised cutaneous forms (20%) (Barros et al. 2011a). Ocular sporotrichosis has rarely been described in immunocompetent patients or in individuals without prior ocular trauma. Intraocular disease has an important association with disseminated disease (Curi et al. 2003, Iyengar et al. 2010, Kashima et al. 2010).

Acute dacryocystitis presents as inflammation of the lacrimal sac and is typically caused by infection. Dacryocystitis is predominantly found in adult women and in young infants. The most common signs and symptoms include erythema, oedema and a painful area of induration that overlies the nasolacrimal sac just below the anatomical boundary of the medial canthal ligament. In addition, epiphora and discharge may be observed. When pressure is applied to the inflamed tear duct, purulent material may be expressed through the lacrimal punctum (Pinar-Sueiro et al. 2012).

Cases of isolated granulomatous conjunctivitis due to *Sporothrix* infection after exposure to cats with sporotrichosis have been reported in Rio de Janeiro (RJ), Brazil (Barros et al. 2004, Schubach et al. 2005). In this study, we evaluated cases of dacryocystitis secondary to *Sporothrix* infection in this hyperendemic area.

This study was approved by the Ethical Committee of the Evandro Chagas Institute of Clinical Research (IPEC)/Oswaldo Cruz Foundation (Fiocruz), RJ, Brazil (0024.0.009.000-10). The authors reviewed the clinical records of patients who were diagnosed with dacryocystitis secondary to *Sporothrix* infection in the dermatology and ophthalmology laboratories of IPEC/Fiocruz from July 2008-July 2010. Patients underwent dermatological and ophthalmological examinations, including visual acuity (Snellen chart), biomicroscopy and ophthalmoscopy. The patients were initially found to be free of chronic stenosis and epiphora. During this period, 2,146 patients were diagnosed with sporotrichosis and sporotrichosis with dacryocystitis was identified in four patients (Table). Three patients were children (≤ 13 years of age) and one patient was an adult (41 years of age). The patients sought medical attention at a median time period of five weeks (3-8 weeks) after the initial manifestations of sporotrichosis. Patients 1 and 4 had a history of contact with cats that had sporotrichosis. Dacryocystitis was present at the outset in all cases. One child had cutaneous nodules and ulcerated skin lesions that were culture-positive for *Sporothrix* (Supplementary data), whereas the other two children had conjunctivitis, but no skin lesions. The adult patient presented only with dacryocystitis (Supplementary data).

The diagnosis of dacryocystitis was established by the presence of swelling at the medial canthus with erythema, epiphora and mucopurulent discharge from the lacrimal punctum. *Sporothrix* infection was confirmed

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TABLE

Clinical characteristics of the patients with *Sporothrix* dacryocystitis, including whether or not there were additional disease manifestations as well as the patients' treatment regimens and complications of their disease

Case	Sex/age	Site of lesion	Treatment (ITC) and length (weeks)	Follow up
1	F/2	Right dacryocystitis + cutaneous nodule-ulcerated lesions on the face	5 mg/kg (13)	Chronic dacryocystitis
2	M/5	Right dacryocystitis + conjunctivitis	100 mg (4)	Chronic dacryocystitis
3	F/13	Right dacryocystitis + conjunctivitis	100 mg (12)	Chronic dacryocystitis
4	F/41	Left dacryocystitis	Up to 400 mg (96)	Cutaneous fistula

F: female; ITC: itraconazole; M: male.

by the isolation of the fungus in culture using the mucopurulent material that was expressed from the lacrimal punctum, which was obtained by swabbing. Cultures were confirmed as positive for *Sporothrix* using previously described methods (Barros et al. 2004). Treatment included itraconazole (ITC) 100 mg/day or 5 mg/kg in children who weighed less than 20 kg. This regimen has been highly successful at our institution as previously described (Galhardo et al. 2008, Barros et al. 2011a, b). Blood count and blood biochemistry tests were conducted at baseline, 12 weeks and when clinically necessary.

A clinical cure was defined as the resolution of inflammation and a negative follow-up culture. Treatment failure was defined as the persistence or worsening of the initial lesion after 12 weeks of treatment, which occurred in the adult patient who received an escalated dose of ITC to 400 mg/day. The treatment of this patient was significantly prolonged (96 weeks). Case 2 was initially lost to follow-up after four weeks of treatment; however, this patient returned seven months later with chronic dacryocystitis and no mycological findings of sporotrichosis. The other children were treated for 12 and 13 weeks. Follow-up was conducted at a minimum of six months after the end of treatment. Despite mycological cures, the three children had chronic dacryocystitis and the adult patient developed a cutaneous fistula. These patients were referred to surgical treatment.

Dacryocystitis secondary to sporotrichosis represented 0.18% of the sporotrichosis cases that were evaluated at the IPEC from July 2008-July 2010. The patients resided in a hyperendemic region for the zoonotic transmission of sporotrichosis and two of the patients had domiciliary contact with cats that had sporotrichosis; however, no specific history of injury was elucidated. Sporotrichosis lesions in cats are rich in parasites and respiratory symptoms can manifest in cats with nasal disease (Schubach et al. 2004, Barros et al. 2011a). Therefore, transmission from cats to humans may occur via respiratory secretions without disruption of the skin barrier when individuals have close face-to-face contact with animals during play (Barros et al. 2004, 2011a).

Fungi have been reported to be present in 4-7% of dacryocystitis cases. The most commonly isolated genus

is *Candida*, followed by *Aspergillus* and *Mucor*. These cases are generally chronic (Pinar-Sueiro et al. 2012). Sporotrichosis with acute dacryocystitis was observed in the cases in this study. Three of the cases were associated with other clinical manifestations (granulomatous conjunctivitis and lymphocutaneous disease). Granulomatous conjunctivitis has been described in 2.2% of patients with cat associated sporotrichosis in RJ (Barros et al. 2004). Notably, dacryocystitis was identified in one of the 81 cases of sporotrichosis in children presented in a previous analysis of patients in RJ (Barros et al. 2008). Dacryocystitis is an unusual manifestation of sporotrichosis; however, these three cases under the age of 13 in the present study represented 2.2% of the paediatric cases of sporotrichosis at the institution, from July 2008-July 2010. Several studies have found that the face is the most frequently affected site of sporotrichosis in children, which is most likely due to the thinner, more delicate skin in this area of the body (da Rosa et al. 2005). Therefore, children are at an increased risk for this clinical form due to the aerosol mode of transmission from nasally infected cats.

The patients in this study responded to treatment; however, each patient had persistent complications that required surgical correction (chronic dacryocystitis and a fistula). Further studies are needed to determine whether dacryocystitis due to *Sporothrix* infection routinely leads to chronic disease. The pathogenesis of this disease is likely due to *Sporothrix* infection through the conjunctiva into the lacrimal sac rather than a haematogenous route. We identified two additional patients in the sporotrichosis cohort in this study who presented with dacryocystitis, but these patients were excluded because their lacrimal cultures were negative. However, both patients developed complications, including a fistula and chronic dacryocystitis.

Recently, *S. schenckii* was found to be a complex of species, including *Sporothrix brasiliensis*, which has been implicated in the hyperendemic transmission of sporotrichosis in RJ (Marimon et al. 2007). The epidemic is associated with the enhanced virulence of the emerging strains of *S. brasiliensis* (Arrillaga-Moncieriff et al. 2009). A molecular analysis of the strains in these four cases was

not performed; however, based on epidemiology, it is likely that *S. brasiliensis* was the species involved.

In conclusion, sporotrichosis is frequently a benign disease; however, extracutaneous manifestations, such as diseases that affect the eye and the adnexa, can lead to severe and chronic complications. Clinicians, particularly ophthalmologists and internists in highly endemic areas, should be aware of the protean manifestations of sporotrichosis.

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