CASE REPORT

Bilateral chronic fungal dacryocystitis caused by *Candida dubliniensis* in a neutropenic patient

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J Clin Pathol 2006;59:1194–1195. doi: 10.1136/jcp.2005.027540

In recent years, candida species other than *Candida albicans* have emerged as causes of human candidiasis, particularly in HIV-infected and other immunocompromised people. *C dubliniensis*, a recently described species closely related to *C albicans*, first isolated from patients with AIDS in Dublin, has been implicated as an agent of oral candidiasis in HIV-positive people. However, it has also been recovered from HIV-negative people, with clinical signs of oral candidiasis and from the genital tract of some women with vaginitis. The first case of bilateral chronic fungal dacryocystitis caused by *C dubliniensis* is described in an HIV-negative woman.

Dacryocystitis is usually the result of lacrimal outflow obstruction and stasis of tears in the lacrimal sac. Outflow obstruction can be secondary to infection or anatomical anomalies. Infection of the lacrimal sac is usually bacterial and in only 1.2% of cases is a fungal organism the causative agent.¹ Only one case with bilateral fungal involvement has been described in a patient who had mid-facial trauma, the causative organism being *Candida albicans*.² We report the first case of chronic bilateral fungal dacryocystitis caused by *C dubliniensis*.

CASE REPORT

A 30-year-old HIV-negative woman with idiopathic chronic neutropenia was referred by the haematology department after having developed a mass at the right medial canthus. She complained of localised tenderness and epiphora (fig 1A), she had no previous history of epiphora or sinonasal disease. A right acute dacryocystitis was diagnosed and she was started on oral co-amoxiclav. Her blood cultures before starting antibiotics were negative. After 2 weeks on oral antibiotics the discomfort and swelling worsened. She was then admitted and started on intravenous gentamicin and tazobactam/piperacillin. On the same day of referral to the eye department, an incisional biopsy and drainage of the abscess was attempted, but a solid tissue mass was found and no pus was evacuated. Cultures of tissue samples from the incisional biopsy and serial blood cultures were negative. After her initial presentation, she complained of epiphora and tenderness over her left medial canthus. Although the right canthal swelling was gradually decreasing, the left canthal swelling continued to develop (fig 1B). Her antibiotic treatment was changed to intravenous meropenem, teicoplanin and metronidazole. A week later she became systemically unwell with respiratory signs, and a swinging pyrexia peaking at 40°C. Cultures of clear fluid expressed at an incisional biopsy specimen of the left canthal swelling were negative. It was believed that she was too systemically unwell to undergo a dacryocystorhinostomy at this stage. Along with a neutropenia of 0.06 ($10^9/l$), she had a lymphopenia of 0.32 ($10^9/l$) and a monocytopenia of 0.03 ($10^9/l$). The microbiologists recommended a change in her treatment to

intravenous ambisone and gentamicin. Her condition slowly improved on this treatment, and 4 weeks later she was well enough to undergo a bilateral dacryocystorhinostomy with silicone intubation. Surgery was uncomplicated and large sulphur-like granules were removed from both lacrimal sacs and sent for microbiological analysis. Tissue culture showed *C dubliniensis*. The patient's clinical symptoms improved; she was discharged 4 days later on oral amoxicillin and returned for intravenous ambisone 150 mg once daily, for 2 weeks. Two months postoperatively she remains well on oral voriconazole 200 mg twice daily, with complete resolution of her dacryocystitis (fig 1C).

Comment

This patient had chronic idiopathic neutropenia, which predisposed her to dacryocystitis. The immune response to fungal infections is associated with the innate (neutrophils and monocytes) and adaptive cell-mediated systems.^{3–5} In patients with neutropenia, the innate pathways are defective. In addition, the prolonged use of broad-spectrum antibiotics facilitates fungal colonisation and fungaemia by depleting mucocutaneous protective bacterial flora.

This is the first reported case of bilateral chronic fungal dacryocystitis in the absence of previous trauma. It is also the first case of dacryocystitis caused by *C dubliniensis*.

Figure 1 (A) Right dacryocystitis. Week 1, prior to ambisone surgery (B) Bilateral dacryocystitis. Weeks 5–6, IV ambisone started. (C) Weeks 16–17. Surgery carried out between weeks 9–10. On IV ambisone until week 12, then voriconazole.

C dubliniensis was first identified as a distinct taxon in 1995⁶ and is rarely found in the oral microflora of healthy people.⁷ This case highlights the need to consider a fungal cause in patients with non-responsive dacryocystitis, particularly in susceptible people with neutropenia, as they are unable to mount an adequate anti-fungal immune response. Although a fungal cause of the disease was suspected early on, it was a surprise that the causative organism was *C dubliniensis*. Dacryocystorhinostomy in cases of dacryocystitis is usually curative. In this patient, dacryocystorhinostomy was performed with silicone intubation to minimise failure of the procedure. This benefit of intubation was believed to outweigh the risk of infection after surgery.

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Competing interests: None declared.

Full patient consent has been obtained.

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Accepted for publication 11 October 2005

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