Letter to the Editor

Acute dacryocystitis as a presenting sign of chronic lymphocytic leukaemia

Acute dacryocystitis is characterized by rapid onset of pain and erythematous swelling predominantly below the medial canthus resulting mainly from an acute infection of the lacrimal sac and perisac tissues. Although rare, involvement of the lacrimal drainage system with leukaemic infiltrates were reported and believed to be facilitated by the presence of lacrimal drainage-associated lymphoid tissue (LDALT).1–5 To the best of the authors knowledge, acute dacryocystitis as a presenting feature of a yet to be diagnosed chronic lymphocytic leukaemia (CLL) has been reported once before but with unclear details. We report the second such case which clearly shows acute dacryocystitis as a presenting feature of a CLL.

A 59-year-old otherwise healthy female presented with swelling and pain on the inner side of left eye of 5 days duration, associated with discharge and epiphora from the left eye. Constitutional symptoms were absent. There was no history of epiphora, trauma or surgery. On examination, there was tender swelling over the left lacrimal sac area with surrounding induration and preseptal cellulitis (Fig. 1a). Ocular examination was otherwise normal except for bilateral aphakia following cataract surgery. Microbiological cultures were negative. The patient was started on systemic broad spectrum antibiotic and anti-inflammatory agents. A week later, there was a suboptimal response (Fig. 1b). Endoscopic evaluation of the nasal cavity was within normal limits (Fig. 1c), and the patient was scheduled for an endoscopic dacryocystorhinostomy. Routine pre-surgical blood counts showed marked leukocytosis (Total leucocyte count ~ 73 100/μl) with lymphocytosis without any anaemia or thrombocytopenia. A peripheral blood smear showed similar picture (Fig. 1d). Bone marrow biopsy showed marked lymphocytosis with predominantly monotonous population of small lymphocytes (Fig. 2a). Flow cytometric analysis showed 60% of atypical B-lymphocytes with bright expression of CD19 and HLADR with moderate expression of CD23, CD200 and CD45. The CD19-gated cells were 0.4% positive for CD38 and 1.9% positive for ZAP-70. The scatter parameters and antigenic expression profile by flow cytometry (Fig. 2b) were suggestive of B-cell chronic lymphocytic leukaemia (B-CLL). Systemic examination did not reveal any lymphadenopathy, hepatomegaly or splenomegaly. The B-CLL was staged as Rai Stage 0 and Binet Stage I with a favourable prognosis. In view of the early stage of the disease, the patient is being closely monitored on a 2-weekly basis.

The patient complained of increasing pain on the left side again after 2 weeks and underwent a dacryocystectomy. Surgery was uneventful except for increased intraoperative bleeding. At the last follow-up (3 months following the surgery), the patient was asymptomatic with resolution of pain and swelling.

Histopathological examination revealed dense sub-epithelial lymphocytic infiltrates (Fig. 2c). Immunohistochemistry showed the cells to be positive to CD20 (Fig. 2d), CD5 (Fig. 2e) and CD23 (Fig. 2f). The histological features were consistent with predominantly B-lymphocytic infiltration of the lacrimal mucosal tissues.

Chronic lymphocytic leukaemia is a common form of leukaemia in adults usually suspected when blood counts show marked lymphocytosis and is confirmed by flow cytometry and target gene analysis. Lacrimal drainage obstructions and subsequent acute or chronic dacryocystitis are believed to be secondary to either a direct leukaemic infiltration or an abnormal stimulation and subsequent overgrowth of pre-existing lymphoid elements.1,4

Wirostko et al.2 documented acute dacryocystitis as a presenting sign of acute leukaemia in pediatric age group, which was successfully managed with antibiotics and chemotherapy. Stokes1 reported a case of bilateral chronic dacryocystitis with a fistula in a 68-year-old woman, where CLL was diagnosed on histological examinations of lacrimal sac following a dacryocystectomy. However, this patient had a history of previous repeated attacks of acute dacryocystitis and lacrimal abscess. One case of acute dacryocystitis in a 69-year-old man, following eight cycles of chemotherapy for CLL was reported by Karesh et al.3 The patient was treated by antibiotics followed by a dacryocystorhinostomos (DCR). Chemotherapy was initiated after surgery and the patient was symptom free at 17 months follow-up. Yip et al.4 reported a case of 84-year-old woman, primarily treated by chemotherapy for CLL and presented with acute dacryocystitis 2 years later, which was successfully managed with a dacryocystorhinostomos. Management options for acute dacryocystitis in a leukemic patient include systemic antibiotics, chemotherapy when indicated, dacryocystorhinostomos or a dacryocystectomy. DCR is a preferred surgical option. Yip et al.4 in their series of CLL, showed successful outcomes in seven of the nine patients (one had acute dacryocystitis). However, restenosis secondary to lymphocytic infiltrations after successful DCR surgeries and dry eyes secondary to graft versus host disease have also been reported.5 Dacryocystectomy can be a viable option if the patient is systemically unwell or unwilling for a DCR.

In conclusion, although rare, CLL can present initially as acute dacryocystitis. Suspicion should be entertained where the severity of leucocytosis does not correlate with the infection.

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Figure 1. External photograph of the patient showing gross left-sided medial canthal swelling with preseptal cellulitis (a). Suboptimal clinical response at 1 week (b). Endoscopic view of the left nasal cavity showing normal lateral wall (c). Microphotograph of the peripheral smear showing lymphocytosis (Lieshman x100, d).

Figure 2. Microphotograph of the bone marrow aspiration showing marked lymphocytosis (Giemsa x400, a). Scatter parameters and antigenic expression profile of flow cytometry (b). Microphotograph of the lacrimal sac showing dense sub-epithelial lymphocytic infiltrates (H&E x100, c). Immunohistochemistry microphotographs of the lacrimal sac showing positivity to CD20 (CD20 x100, d), CD5 (CD5 x100, Fig. 2e) and CD23 (CD23 x400, f).
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