CASE REPORT

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Bilateral sequential bacterial dacryoadenitis with abscess- a case report



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Abstract

Background Acute dacryoadenitis is characterized by inflammation of the lacrimal gland, and is typically viral. Bacterial dacryoadenitis is rare, with *Staphylococcus* and *Streptococcus* species being the most common pathogens. The bilateral sequential onset of dacryoadenitis has not been described in the literature to date.

Case presentation A 54-year-old immunocompetent female presented to the emergency department with a 1-week history of left-sided periorbital swelling, erythema, and pain. Orbital computed tomography demonstrated significant soft tissue thickening and swelling surrounding the left orbit and a lacrimal gland abscess. Culture swabs were negative for microorganisms. Despite initial management with intravenous antibiotics and surgical drainage, the condition recurred post-discharge. Three years later, the patient experienced similar symptoms in the right eye. Cultures were repeatedly negative for microorganisms. Despite courses of intravenous antibiotics and surgical drainage, the patient experienced recurrences of abscesses in the right eye, highlighting the challenges in managing this rare condition.

Conclusions We describe a unique case of bilateral sequential bacterial dacryoadenitis with abscess formation. This case highlights the need for prompt diagnosis and management, including surgical intervention, to prevent complications and recurrences.

Keywords Bacterial dacryoadenitis, Orbital cellulitis, Abscess

Background

Acute dacryoadenitis is an uncommon disorder, characterised by the rapid enlargement and inflammation of the lacrimal gland [1]. Dacryoadenitis may be due to autoimmune or idiopathic inflammation, infiltrative neoplasm, or infection [2, 3]. Infectious dacryoadenitis is typically of viral aetiology. Bacterial dacryoadenitis is

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²Department of Ophthalmology and South Australian Institute of Ophthalmology, Royal Adelaide Hospital, Adelaide, South Australia, Australia rare. *Staphylococcus, Streptococcus* and *Haemophilus* species are often responsible [4]. Common clinical features include unilateral erythema, oedema, pain in the superolateral orbit, mechanical S-shaped upper lid ptosis, conjunctival injection and restriction of extraocular movements (EOM) [4]. Rarely, there may be bilateral involvement. Radiologically, contrast-enhanced magnetic resonance imaging is the gold standard. Enlargement and enhancement of the affected lacrimal gland is typical [5]. Histopathological diagnosis is definitive and may be made via lacrimal gland biopsy [6]. Severe cases may be complicated by abscess formation or orbital cellulitis [7]. Antibiotic therapy is the mainstay of treatment. Herein we present a rare case of bilateral sequential bacterial dacryoadenitis complicated by abscess formation.



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Written informed consent for this case report was given by the patient.

Case presentation

A 54-year-old immunocompetent female with no significant past medical history presented to the emergency department in 2021 with a 1-week history of periorbital pain, swelling and erythema. Prior to her presentation to hospital, she had taken oral amoxicillin-clavulanic acid 875 mg-125 mg twice daily for 2 days, with nil response. The patient was systemically well, with no fever, malaise and preauricular, submandibular or cervical lymphadenopathy. Examination revealed left upper eyelid erythema and swelling, and limited abduction of the left eye, with pain. Slit-lamp biomicroscopy revealed left bulbar conjunctival injection. Bloods showed white blood cell (WBC) count 12.56×10^{9} L and C-reactive protein (CRP) 28.3 mg/L. Imaging demonstrated significant soft tissue thickening and swelling surrounding the left orbit and a round rim-enhancing mass concerning for an abscess (measuring $21 \times 18 \times 16$ mm) at the superolateral aspect of the left orbit, intimately associated with the lacrimal gland (Fig. 1a and b). The patient was admitted to hospital and underwent drainage of the left lacrimal gland abscess. A culture swab was negative for microorganisms. Intravenous (IV) ceftriaxone 2 g once daily and topical chloramphenicol eye ointment three times a day was continued for 5 days. Over this period, the patient's symptoms improved significantly and she was discharged on a course of oral amoxicillin/clavulanic acid twice daily for 1 week. The patient represented with worsening periorbital pain and left upper eyelid swelling 1 week post discharge. Orbital computed tomography (CT) revealed a small residual preseptal collection $(10 \times 10 \times 4 \text{ mm})$ and associated preseptal cellulitis (Fig. 1c and d). A 7-day course of IV ceftriaxone 2 g once daily was administered and the patient's symptoms resolved. The subsequent three years were uneventful.

Three years later, the patient presented to the emergency department with a 2-day history of progressive right upper eyelid swelling, painful eye movements and diplopia on right lateral gaze. The patient was systemically well, with no fever, malaise and preauricular, submandibular or cervical lymphadenopathy. Examination revealed best-corrected visual acuity (BCVA) of 6/6-2 in the right eye and 6/9 + 1 in the left eye. Colour vision and intraocular pressure (IOP) were normal and no relative afferent pupillary defect (RAPD) was observed. The right upper eyelid was erythematous, swollen and tender on palpation of the lateral aspect (Fig. 2a). Slit-lamp biomicroscopy revealed right bulbar conjunctival injection temporally. Abduction of the right eye was limited, with pain, worse on lateral gaze (Fig. 2b). EOM were otherwise full. No proptosis was noted. Fundoscopy was unremarkable. Examination of the left eye was unremarkable.

WBC count was 10.95, with neutrophils 7.80 and monocytes 0.93. Erythrocyte sedimentation rate (ESR) measured 17 mm/h and CRP was 116 mg/L. Serum glucose was 4.9mmol/L, thyroid function tests were normal and blood cultures were negative. Conjunctival swabs



Fig. 1 Axial (a) and coronal (b) CT scan of orbits showing soft tissue thickening and swelling surrounding the left orbit and left globe deformation secondary to a lacrimal gland abscess. Axial (c) and coronal (d) CT scan of orbits showing residual small pre-septal collection and pre-septal cellulitis



Fig. 2 Clinical photos showing (a) right upper lid swelling with erythema, (b) limited abduction of right eye



Fig. 3 (a) axial orbital post-contrast CT, (b) axial T1 MRI, (c) axial T2 MRI and (d) coronal T1 MRI showing right lacrimal gland enlargement (T1 isointense, T2 hyperintense), enhancement with contrast, and a small non-enhancing fluid collection (T1 hypointense, T2 hyperintense) representing a complicating abscess (arrow)

of the right eye were negative by nucleic acid amplification testing (NAAT) for viruses, including adenovirus, enterovirus, herpes simplex virus (HSV) and varicella zoster virus (VZV), and bacteria including chlamydia and gonorrhoea. Nasopharyngeal viral swabs were negative for respiratory pathogens (SARS-CoV-2, Influenza A/B, Parainfluenza viruses, rhinovirus, respiratory syncytial virus, Human metapneumovirus and Adenovirus). Viral serologies (HSV, VZV, Epstein Barr Virus (EBV), cytomegalovirus (CMV), mumps, adenovirus) were negative. Extractable nuclear antigens (ENA), antinuclear antibodies (ANA), antineutrophil cytoplasmic antibodies (ANCA) and anti-double-stranded DNA antibodies (anti-dsDNA) were negative. Tear inflammatory cytokines (IL-10, IL-8 and IL-1 beta) were elevated. Contrast-enhanced CT imaging of the orbits showed asymmetric enlargement of the right lacrimal gland with abnormal enhancement, in addition to orbital cellulitis with effusion and soft tissue stranding in the periorbital region (Fig. 3a). High-resolution magnetic resonance imaging (MRI) of the orbits revealed asymmetric bulkiness of right lacrimal gland, with a small abscess within the centre of the gland (measuring $5 \times 5 \times 8$ mm) (Fig. 3b, c and d).

A working diagnosis of right bacterial dacryoadenitis with abscess was made, and the patient was admitted for IV antibiotic therapy with ceftriaxone 2 g once daily and flucloxacillin 500 mg 6 hourly, as advised by the Infectious Diseases Unit who was consulted during the patient's admission. The patient's symptoms worsened over the subsequent 4 days. Lacrimal gland abscess



Fig. 4 (a) Intraoperative photograph showing the lacrimal gland inflammation during the first right lacrimal gland drainage and biopsy procedure, (b) Axial post-contrast T1 MRI scan showing the asymmetric bulky appearance of right lacrimal gland with significant increase in size of the known abscess

incision and drainage, with placement of a vacuum drain, and biopsy of the lacrimal gland were performed (Fig. 4a). Aspirated fluid did not culture any organisms after 5 days (culture media: chocolate agar, horse blood agar, cooked meat medium and CSF broth). Histopathology demonstrated diffuse mixed inflammation including prominent numbers of neutrophils, plasma cells and histiocytes, congested capillaries and activated fibroblasts in fibrous septa, with no microorganisms, granulomas or atypical infiltrate. Postoperatively, the patient experienced symptomatic improvement, and the drain, which was dry, was removed at day 1 post-op. However, 3 days later, she experienced a recurrence of right upper eyelid swelling and erythema. High-resolution MRI orbits showed an interval increase in the size of the lacrimal gland abscess $(17 \times 10 \times 7 \text{ mm})$ (Fig. 4b). Given the progressive worsening of symptoms, the Infectious Diseases Unit advised to broaden antibiotic cover by transitioning the antibiotic regime to IV cefepime 2 g three times daily and IV vancomycin 1.25 g twice daily (for MRSA coverage) for the next 5 days, and after 48 h of therapy, repeat incision and drainage of the lacrimal gland abscess was performed, with placement of a penrose drain. Aspirate fluid again did not culture any organisms after 5 days (culture media: chocolate agar, horse blood agar, cooked meat medium and CSF broth). Postoperatively, the patient's symptoms improved, the drain was removed at day 2 post-op, and she was discharged from the hospital at day 4 post-op on a course of oral amoxicillin/clavulanic acid 875-125 mg twice daily for the next 6 days.

6 days following discharge, the patient represented with a recurrence of pain, redness and swelling of the right upper eyelid. She was readmitted and administered IV cefepime 2 g three times daily and vancomycin 1.25 g twice daily. After 24 h, she was stepped down to oral moxifloxacin 400 mg once daily, as per the Infectious Diseases Unit for a trial of treatment for broader coverage for atypical bacteria. After 48 h of oral therapy, symptoms resolved and she was discharged on a 10-day course of oral moxifloxacin 400 mg once daily and chloramphenicol 0.5% drops four times daily into the right eye. There has been no recurrence after a period of 6 months.

Following recovery from this episode, further bloods were sent to evaluate for a cause, as per immunology, who were consulted during the patient's admissions. Immunophenotyping revealed an increase in the CD4/CD8 ratio (11.50), but there was no immunophenotypic evidence of T cell monoclonality. Other investigations included pneumococcal IgG (50.7 mg/L)- normal immune, and IgG (9.97 g/L), IgA (1.4 g/L), IgM (1.47 g/L), which were normal. The patient did not require any corticosteroids or other immune modulating therapy.

Discussion and conclusions

We present a case of bilateral sequential bacterial dacryoadenitis with abscess complicated by multiple recurrences. A search of the current English-language literature across databases PubMed, CENTRAL and Embase revealed 51 published cases of bacterial dacryoadenitis with abscess [8–33].

Dacryoadenitis is an uncommon condition which affects patients of all ages, and has a reported incidence of 1 in 10,000 [31]. Acute dacryoadenitis occurs usually as a result of infection, most commonly viral. The most common viral etiology is Epstein-Barr virus, with mumps, measles, cytomegalovirus and herpes simplex virus occurring less frequently [34]. Bacterial dacryoadenitis is rare, with Staphylococcus aureus being the most common bacterial pathogen [14]. Among the previously published cases of bacterial dacryoadenitis with abscess, causative organisms were isolated in 39 (76.4%). Of these, methicillin-susceptible staphylococcus aureus (MSSA) was the most common organism (n = 13, 25.4%) [12–14, 18, 22, 26, 30-32], followed by methicillin-resistant *staphylococcus aureus* (MRSA) (*n* = 7) [13, 18, 20, 28, 29]. In 5 cases, mixed skin flora were isolated [14, 24, 28, 32]. In 6 cases, no bacteria grew on culture [11, 22, 25, 27, 28, 32]. In these cases, it is unclear the duration of antibiotics therapy that preceded sample collection.

Dacryoadenitis is typically unilateral. Among cases of bacterial dacryoadenitis with abscess in the existing literature, a single case was bilateral [19]. A 45-year-old female with a history of cocaine misuse and subsequent necrosis of the posterior nasal septum suffered 2 episodes of bilateral dacryoadenitis with abscess across a 6-month period. *Staphylococcus epidermidis* was isolated. In contrast to the case reported here, this patient improved with oral and topical antibiotic therapy alone on both occasions. For our case, the patient's second bacterial dacryoadenitis was particularly aggressive and required more than 3 weeks systemic antibiotic therapy.

Among reported cases of bacterial dacryoadenitis with abscess in the existing literature, only 6 symptoms were reported [8–31]. The most common symptom was upper lid swelling (n=38), followed by pain (n=20), purulent discharge (n=10), blurred vision (n=6), epiphora (n=3) and diplopia (n=3) [8–31]. Various clinical signs have been documented. Extraocular movement (EOM) restriction was the most common of these (n=30), followed by erythema (n=23), chemosis (n=21), lacrimal gland protrusion (n=10), globe displacement (n=9), proptosis (n=8), a palpable mass (n=8), ptosis (n=4), an S-shaped upper eyelid deformity (n=3), and eyelid retraction (n=1). A relative afferent pupillary defect (RAPD) was observed in 2 cases [10, 20].

A large case series of 79 patients found that recurrent disease occurred in up to 15% of patients with idiopathic dacryoadenitis [5]. To the best of the authors' knowledge, a single case of sequential bilateral involvement in bacterial dacryoadenitis with abscess has previously been reported. Boukouvala et al. reported a case of a late contralateral recurrence of acute dacryoadenitis in a 41-yearold female, with a history of Crohn's colitis [35]. The patient initially presented with painful swelling of right upper lid, limited right eye abduction and painful eye movements. Imaging demonstrated right lacrimal gland enlargement with signs of early abscess formation within the centre of the gland. Conjunctival swabs showed no microorganism growth. Full resolution was noted within 2 weeks of antibiotic treatment. However, 8 months later, she presented with signs of acute left dacryoadenitis, which did not respond to oral antibiotics, but resolved after a course of oral prednisolone.

Antibiotics are the mainstay of management for bacterial dacryoadenitis with abscess. No clear guidelines exist on administration route. Similarly, decision to perform incision and drainage is made on a case by case basis. Among the cases of previously published bacterial dacryoadenitis with abscess, 40 (78.4%) received intravenous antibiotics initially, followed by a stepdown to oral antibiotics [8–32]. 34 patients (66.6%) underwent abscess drainage, all of whom received intravenous antibiotics [8, 11–18, 22–26, 28–32]. 28 patients who underwent drainage were administered a course of oral antibiotics following cessation of intravenous medication [8, 11–18, 22–26, 28–32]. The outcomes of 42 patients were documented in the literature, of which, 41 cases (97.6%) resolved [8–10, 12–31]. Time from presentation to resolution ranged from 4 days to 6 weeks. Prolonged antibiotic treatment may be required to achieve resolution. In our patient's case, she required extended courses of antibiotic therapy for both episodes of bacterial dacryoadenitis.

It is unknown why sequential bacterial dacryoadenitis, which was aggressive and difficult to manage, occurred in this healthy patient. In cases of recurrent infection, additional investigations should be performed for possible underlying disease including diabetes mellitus, haematologic or solid malignancies and immunological diseases. Investigation for underlying diabetes and immunodeficiency in this patient was negative, but immunophenotyping revealed an increase in the CD4/CD8 ratio. This pattern has been observed in a range of reactive, inflammatory, or auto-immune conditions [36]. Contributing factors to the patient's recurrence of right lacrimal gland abscess formation may have included early vacuum drain removal, early transition from intravenous to oral antibiotic therapy and potential insufficient antibiotic coverage. It is also possible that anatomical variation of the lacrimal gland ductules may have played a role in her susceptibility to suppurative infection, as this is the space in which lacrimal gland abscess forms.

Given the negative cultures, other differential diagnoses were considered for a patient presenting with upper eyelid oedema, erythema, tenderness, conjunctival injection and painful ocular movements. Orbital cellulitis often results from the contiguous spread of infection from the paranasal sinuses, trauma, or hematogenous dissemination, and tends to have a more diffuse involvement beyond the lacrimal gland [37]. In our patient, there was no sinus involvement seen on CT and MRI imaging, which made orbital cellulitis less likely as the primary diagnosis. Conditions such as lacrimal gland tumours and inflammatory orbital pseudotumours were also considered, however made less likely given the patient's clinical response to antibiotics and presence of pus on incision and drainage.

Bilateral sequential bacterial dacryoadenitis with abscess is a rare entity. Upper lid swelling, tenderness, erythema and conjunctival injection are typical. Imaging and tissue biopsy is required for definitive diagnosis. Antibiotic therapy is first-line management, and source control with incision and drainage may be required to aid resolution and reduce the risk of recurrence.

Abbreviations

ANA Antinuclear Antibodies ANCA Antineutrophil Cytoplasmic Antibodies

Anti-dsDNA	Anti-double-stranded DNA antibodies
BCVA	Best-Corrected Visual Acuity
CMV	Cytomegalovirus
CRP	C-Reactive Protein
CT	Computed Tomography
EBV	Epstein Barr Virus
ENA	Extractable Nuclear Antigens
EOM	Extraocular Movements
ESR	Erythrocyte Sedimentation Rate
HSV	Herpes Simplex Virus
IOP	Intraocular Pressure
MRI	Magnetic Resonance Imaging
MRSA	Methicillin-Resistant Staphylococcus Aureus
MSSA	Methicillin-Susceptible Staphylococcus Aureus
RAPD	Relative Afferent Pupillary Defect
VZV	Varicella Zoster Virus
WBC	White Blood Cell

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Author contributions

All authors (LL, JP, CQ, JS, GD, DS) have made substantive intellectual contributions to this manuscript. LL and JP acquired data and wrote the manuscript. CQ examined the patient, obtained consent of the patient and critically revised the manuscript. GD, JS and DS participated in patient care, and critically revised the manuscript. All authors read and approved the final manuscript.

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Data availability

The datasets used and analysed during the current study are available from the corresponding author on reasonable request.

Declarations

Ethics approval and consent to participate

No ethical approval required.

Consent for publication

Written informed consent was obtained from the patient for publication of this case report.

Competing interests

The authors declare no competing interests.

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