CASE REPORT

Chronic dacryocystitis secondary to *Stenotrophomonas maltophilia* and *Staphylococcus aureus* mixed infection

Arzu Taskiran Comez,¹ Asiye Koklu,¹ Alper Akcali²

¹Department of Ophthalmology, Canakkale Onsekiz Mart University School of Medicine, Canakkale, Turkey

²Department of Medical Microbiology, Canakkale Onsekiz Mart University School of Medicine, Canakkale, Turkey

Correspondence to
Dr Arzu Taskiran Comez,
arzucomez@yahoo.com

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SUMMARY

A 40-year-old woman with a history of recurrent attacks of dacryocystitis for 2 years developed a lacrimal sac abscess. β -Lactam antibiotics, considered the first-line treatment for dacryocystitis, were ineffective. She underwent dacryocystorhinostomy. Cultures from the lacrimal sac demonstrated the presence of *Stenotrophomonas maltophilia* and methicillin-sensitive *Staphylococcus aureus*, both of which are sensitive to trimethoprim-sulfamethoxazole. This rare and antibiotic-resistant bacterial species should be considered in atypical cases of dacryocystitis, and appropriate antibiotics should be started immediately.

BACKGROUND

Stenotrophomonas maltophilia (*S. maltophilia*) is an aerobic, Gram-negative bacillus that is an opportunistic and nosocomial pathogen that causes infection primarily in immunocompromised patients.¹ Although rare, ocular infections, including conjunctivitis, keratitis, endophthalmitis, dacryocystitis and preseptal cellulitis, may be caused by *S. maltophilia*.^{2–10} *Staphylococcus aureus* (*S. aureus*) is a Gram-positive facultative anaerobic microorganism and is mainly found as commensal flora of the human skin and upper respiratory tract.¹¹ *S. aureus* is predominantly found in dacryocystitis specimens with mixed growth.¹²

CASE PRESENTATION

Here we present the case of a 40-year-old woman with a left lacrimal sac abscess. She had five recurrent dacryocystitis episodes over a 2-year period and had developed an apparent lacrimal sac abscess with notable oedema causing upward displacement of the medial portion of the left-lower eyelid over the last 2 months (figure 1, arrow). Although she had experienced some relief with β -lactam antibiotics during the first four attacks, she did not have any relief in the last 2 months, and the mass in the medial canthal area did not resolve.

INVESTIGATIONS

On initial examination, the patient had a 15–20 mm elevated, firm nodule medial to her left-medial canthus that caused upward displacement of the medial portion of the lower eyelid. Although purulent discharge emanated from the punctum on digital pressure, it was observed that the lesion was unmovable, tender and had fibrous components as well as cystic parts. Probing was not carried out and

Figure 1 Clinical aspect at presentation. Note the upward displacement of the medial portion of the left-lower eyelid (arrow head).

an MRI was performed in order to exclude malignancy due to the solid and unmovable nature of the mass. In the MRI, a lacrimal sac abscess with dimensions of 16×13 mm was diagnosed, with mild inflammation in the surrounding tissue (figure 2). No malignancy was suspected. In light of these findings, the patient underwent external dacryocystorhinostomy with silicone stent intubation. During the surgery, the lacrimal sac was found to be very large, and, before incision, the yellow pus was aspirated with a needle and sent for microbiological culture. The remainder of the patient's surgery was uneventful.

Microscopic evaluation of Gram-stained specimen showed leucocytes, Gram-positive cocci and Gram-negative bacilli. *S. aureus* and *S. maltophilia* were grown in culture of the specimen. Antimicrobial susceptibility testing was performed using Vitek2-compact system (bioMérieux, Marcy l'Etoile, France). *S. aureus* was identified as methicillin sensitive, penicillin resistant and trimethoprim-sulfamethoxazole (TMP-SXT) sensitive and *S. maltophilia* was identified as sensitive to TMP-SMX.

Figure 2 Coronal MRI showing large lacrimal sac abscess in the left.

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DIFFERENTIAL DIAGNOSIS

Lacrimal sac tumour.

TREATMENT

As both microorganisms were sensitive to TMP-SXT, bactrim 400/80 mg (Deva, Istanbul, Turkey) was used twice daily for 10 days.

OUTCOME AND FOLLOW-UP

The patient had no symptoms of epiphora after the surgery and antibiotic treatment.

DISCUSSION

The management of *S. maltophilia* infections is often complicated due to the fact that this pathogen is generally not initially suspected, as well as its strong resistance to multiple broad-spectrum antibiotic agents.¹³ Four cases of adult and two cases of infantile dacryocystitis with *S. maltophilia* have been previously described in the literature.^{3–6–10} Penland and Wilhelmus³ documented 15 cases of ocular infections due to *S. maltophilia*, mainly including keratitis, and two cases of infantile dacryocystitis. Briscoe *et al*⁶ reported *S. maltophilia* as the pathogen in 1 (2.5%) of the 39 lacrimal abscess patients' microbiological specimens. Recently, Wladis⁷ reported an 88-year-old man with recurrent dacryocystitis episodes, Hromas and Sokol⁸ reported a post-traumatic lacrimal sac abscess 16 months after multiple facial fractures, in which the same agent grew in microbiological cultures. Marx *et al*⁹ reported a case of *S. maltophilia*-related chronic dacryocystitis with associated coagulase-negative *Staphylococcus* treated with only external dacryocystorhinostomy without intraoperative or postoperative antibiotics. The current case was a 40-year-old healthy woman with no history

of trauma or systemic disease or any risk factor for suppression of the immune system. This case was similar to Marx's case, as it was a mixed infection with *S. aureus* and *S. maltophilia*.⁹ The management of *S. maltophilia* infection is problematic due to its high resistance to multiple broad-spectrum antibiotics as shown by the frequent occurrences of infection and unresponsiveness to antibiotics. Where appropriate cultures can be taken and diligent follow-up guaranteed, a course of monotherapy should be the treatment of choice in patients with dacryocystitis, instead of empirical use of additional antibiotics. TMP-SXT is a reasonable combination antibiotic to consider in these circumstances, particularly since resistance to TMP-SXT has been shown to be uncommon in patients with dacryocystitis.¹⁴

Contributors ATC participated in writing the manuscript, performing the surgery, AK participated in interpreting the data, taking care of the patient and AA participated in microbiological evaluation of the specimen.

Competing interests None.

Patient consent Obtained.

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Learning points

- *Stenotrophomonas maltophilia* should be considered as a possible pathogen in atypical cases of dacryocystitis or in patients with recurrent episodes of dacryocystitis demonstrating resistance to β -lactam antibiotics.
- Trimethoprim-sulfamethoxazole is the treatment of choice for *S. maltophilia* dacryocystitis if susceptible in vitro.
- Microbiological examination and antimicrobial susceptibility testing of the lacrimal sac contents will help to identify the pathogen and the antibiotics to which the bacteria are sensitive, avoiding the cavalier use of multiple antibiotics as this may contribute to microbial resistance.

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