

# *Mycobacterium chelonae* dacryocystitis after endoscopic dacryocystorhinostomy

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## ABSTRACT

*Mycobacterium chelonae* is a rapidly growing nontuberculous *Mycobacterium* and an uncommon cause of aggressive, treatment-resistant ocular and periocular infection. This is the first known case report of a woman who developed unilateral *M. chelonae* dacryocystitis after undergoing endoscopic sinus surgery and right endoscopic dacryocystorhinostomy (DCR) with Crawford stent placement. We describe our findings and effective methods to manage the infection. Three weeks after undergoing DCR, the patient acutely developed symptoms consistent with dacryocystitis. The patient was treated with broad-spectrum antibiotics followed by incision and drainage of the dacryocystocele abscess, with initial cultures showing no organisms. With continued signs of infection, the Crawford stent was later removed. Cultures eventually grew *M. chelonae* and the patient was treated with 4 months of antibiotic therapy. While receiving antibiotics, the patient developed three abscesses along the inferior lid requiring excision. After 21 months, the patient remains free of infection and has not experienced any other complications. This case serves as a reminder to consider *M. chelonae* as a potential cause of periocular infection, which may be more likely to occur postoperatively with indwelling devices, as well as in patients with sinonasal issues requiring nasal irrigations. This organism can be difficult to treat because of multidrug resistance and biofilm production. Recommended therapy includes surgical debridement, removal of any implanted devices, and a two-drug antibiotic regimen for at least 4 months.

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**D**acryocystitis, or inflammation of the lacrimal sac, is commonly related to distal obstruction of the lacrimal duct with subsequent stagnation of tears and debris leading to infection. Causative bacteria tend to reflect the normal conjunctival flora, with 68.8% caused by Gram-positive organisms (*Staphylococcus*, *Streptococcus*, *Pneumococcus*, and other species), 28.7% due to Gram-negative organisms (*Pseudomonas aeruginosa*), and only 2.5% of cases caused by mycobacteria, according to a national spectrum study.<sup>1</sup>

*Mycobacterium chelonae* is ubiquitous in the environment and tends to cause skin and soft tissue infections but is a very rare cause of dacryocystitis. It is increasingly being recognized as a cause of surgical and ocular infections, particularly those with implantable devices such as prosthetic joints and indwelling catheters or stents.<sup>2</sup> The standard surgery for obstruction of nasolacrimal drainage is dacryocystorhinostomy (DCR), in which the lacrimal sac is connected directly to the nose. In most cases bicanalicular Crawford stents (small tubes made of silicone) are left in place for 6–12 weeks

to allow tears to drain to the nasal cavity. The purpose of this study is to describe a case of *M. chelonae*-associated dacryocystitis after surgical manipulation of the lacrimal duct system with stent placement.

## CASE REPORT

A 49-year-old woman with a long history of chronic rhinosinusitis with nasal polyposis and allergic rhinitis presented to the otolaryngology clinic with continued complaints of nasal congestion and facial pain, despite multiple trials of oral antibiotics and nasal steroid irrigations in the past. She had previously undergone endoscopic sinus surgery (ESS) four times (in January 2008, December 2009, April 2011, and June 2011) and after the most recent ESS developed epiphora on the right. However, her bony nasolacrimal duct appeared intact on computed tomography scan (Fig. 1).

A trial of betamethasone nasal irrigations improved but did not resolve the epiphora or her continued facial pain. She continued to use twice-daily saline nasal irrigations made with distilled water, as well as daily fluticasone and olopatadine nasal sprays. She was not placed on antibiotics or steroid ophthalmic drops at this time, because her sinuses were open and draining and her ophthalmologic exam was otherwise normal. Because of the continued epiphora and residual mild mucosal thickening of the right maxillary and sphenoid sinuses seen on computed tomography, it was decided to undergo revision ESS and right endoscopic DCR with canalization of the right lacrimal system in

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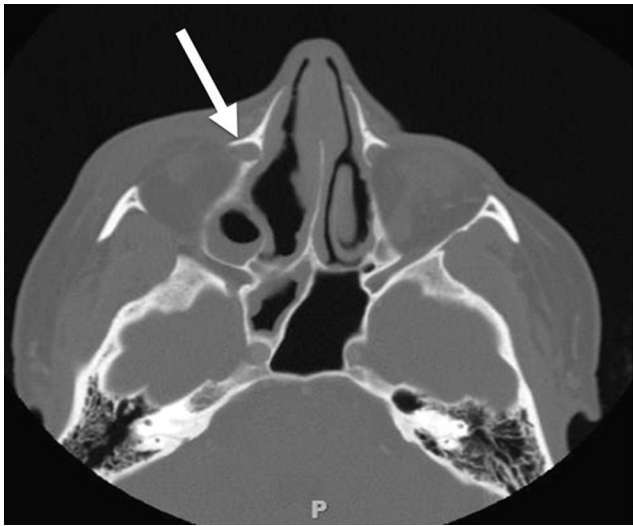
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**Figure 1.** Patent nasolacrimal duct. Computed tomography scan without contrast of the head showing the right bony right nasolacrimal duct (arrow) is patent after, and thus was not injured during the prior 4 endoscopic sinus surgeries (ESSs).



**Figure 2.** Dacryocystocele developing abscess. Computed tomography scan without contrast of the head showing preseptal orbital cellulitis on the right (arrow). At this time, the patient clinically had the start of a right dacryocystocele abscess.

December 2011. During the procedure, extensive polypoid changes were seen along the skull base and posterior ethmoids and along the posterior nasal septum. The lacrimal system was canalized, mucosal flaps were created, a drill was used for the large bony ostium, and a Crawford bicanalicular stent was placed.

Postoperatively, the patient was placed on a 14-day prednisone taper, and nasal irrigations with betamethasone were restarted after the prednisone was completed. She did well in the first 2 weeks, with resolution of her facial pain and epiphora. She then presented on postoperative day (POD) 18 with painful swelling of the right lower lid and was started on amoxicillin/clavulanate. Her facial swelling and pain worsened over the next 2 days and she complained of epiphora and right-sided blurry vision with lateral gaze. She was admitted to the hospital and received intravenous clindamycin and ceftazidime and tobramycin ophthalmic ointment. Ophthalmologic exam revealed a bilateral worsening of her vision from 20/25 on the right and 20/20 on the left preoperatively, to 20/40 and 20/30, respectively. Her right medial canthus remained erythematous with fluctuance over the right lacrimal sac, necessitating incision and drainage of a dacryocystocele abscess at the bedside on POD 24 (Fig. 2); cultures were taken at this time, although no organisms were initially seen on Gram stain. She was discharged from the hospital on oral ciprofloxacin and trimethoprim/sulfamethoxazole and topical bacitracin. She continued to have swelling over the right medial canthus, so the Crawford stent was removed on POD 39. Just over 3 weeks after they were collected, cultures were identified as *M. chelonae* and the patient was started on clarithromycin, which was changed to

azithromycin and doxycycline after she was found to be intolerant of drug side effects. Despite treatment, three abscesses developed along the right inferior lid overlying the lacrimal sac, requiring excision in the office on POD 53 (Fig. 3).

With culture susceptibility results and input from an infectious disease consultant, the patient received 4 months of oral azithromycin, and over 2 months of intravenous imipenem. Her skin lesions, right epiphora, and periorbital pain slowly resolved, and after 3 months her vision returned to the preoperative baseline. After 21 months, the patient remains free of infection and has not experienced any other complications.

## DISCUSSION

*M. chelonae* belongs to the family of nontuberculous mycobacteria (NTM) classified in the rapidly growing mycobacteria.<sup>2</sup> It is considered one of the most drug-resistant of the NTM group.<sup>3</sup> Atypical mycobacteria can grow in distilled and tap water, and infections have been documented in all geographic areas of the United States, although more commonly in the southern coastal states (Texas to Maryland).<sup>4</sup> Skin and soft tissue infections are most common, manifesting as a cellulitis, abscesses, nodules, or sinus tracts, which may drain and appear to diminish only to recur later. Disseminated infection has been well described in immunosuppressed individuals, although it has also been documented in immunocompetent patients.

*M. chelonae* has been linked with all types of surgical infections, including eye, head and neck, cardiovascular and chest, abdominal, cosmetic and reconstructive,



**Figure 3.** Recurrent nasolacrimal abscesses. Development of three abscesses along the inferior border of the patient's right eye overlying the nasolacrimal sac.

and orthopedic procedures. *M. chelonae* has a hydrophobic cell wall that allows it to adhere to multiple surface types, forming biofilms with significant cording.<sup>5</sup> Therefore, procedures involving foreign materials, artificial prostheses, and implantable devices are particularly at risk. Injectable materials are also a concern. There have been reports of surgical wound infections linked to a contaminated stock solution of gentian violet used for skin marking<sup>6</sup> and there have been infections in the public related to contaminated tattoo ink.<sup>7</sup>

There have been very few previously reported cases of *M. chelonae*-associated periocular infections such as dacryocystitis<sup>8,9</sup> and canaliculitis,<sup>3</sup> the majority of which occurred after lacrimal stent or intracanalicular plug placement. We found only four other reported cases of *M. chelonae* dacryocystitis after external DCR with lacrimal stent placement, with similar presentations of painful erythematous nodules over the nasolacrimal sac occurring several weeks after surgery. These patients were all women ages ranging from 44 to 73 years, and also required stent removal, multiple débridements, and several months of antibiotic therapy before resolution of the infection.<sup>8,9</sup> One patient was chronically immunosuppressed with prednisone and methotrexate for rheumatoid arthritis. There was also one sporadic case of *M. chelonae* dacryocystitis in an human immunodeficiency virus-positive male with nasolacrimal duct obstruction that resolved with 2 months of clarithromycin alone.<sup>8</sup> This is the first re-

ported case occurring after endoscopic DCR and the first case in the otolaryngology literature.

Recommended therapy includes surgical debridement of extensive or recurrent infection, removal of any implanted devices, and a long course of antibiotics. Although there have been documented cases of effective treatment with a single antibiotic (clarithromycin), reports also describe the development of drug resistance. Antibiotic therapy with two agents guided by culture sensitivities is preferable in most patients. Rapidly growing NTM are usually sensitive to macrolides, fluoroquinolones, and aminoglycosides.<sup>10</sup> No standard duration of therapy has been established but treatment usually lasts for many months and courses that are  $\geq 6$  months are not unusual.

This study is the only patient with comorbid sinus disease who underwent ESS along with the endoscopic DCR. It is unclear whether our patient had a chronic infection with *M. chelonae* that acutely worsened or if she acquired the infection subsequent to the operation. Along with recent surgery and foreign bodies, nasolacrimal duct obstruction and immunosuppression have been associated with atypical mycobacterial dacryocystitis.<sup>8</sup> Anatomic obstruction of the nasolacrimal duct outlet into the inferior meatus can be caused by nasal pathologies such as nasal polyps, hypertrophied inferior turbinates, a deviated nasal septum, or allergic rhinitis. The patient was using nasal steroids for several weeks before the operation to treat her chronic rhinosinusitis with polyps and had been placed on a prednisone taper postoperatively, which may have induced both a local and systemic immunosuppression that allowed development of this infection. She was also using daily nasal irrigations, and because NTM are commonly isolated from water supplies or solutions,<sup>6,7</sup> the patient's sinus rinse bottle may represent a potential reservoir of mycobacteria if improperly cleaned. Similarly, mycobacteria have been found to persist in footbaths at pedicure salons, causing outbreaks of infection.<sup>11</sup> Our patient's history of chronic rhinosinusitis with polyposis, concomitant sinus surgery, use of steroids, and nasal irrigations with distilled water may have increased her risk of *M. chelonae* dacryocystitis.

## CONCLUSION

*M. chelonae* is an uncommon cause of surgical infections, although the incidence has been increasing over the past 10 years because of increased use of biomaterial and indwelling devices. An infection with painful erythematous nodules or abscesses overlying the surgical site that occurs weeks after operation, particularly when a foreign body is implanted, should arouse suspicion of an atypical mycobacterial infection. Although *M. chelonae* is a rapid-growing *Mycobacterium*, cultures



may be negative for more than a week, thus delaying identification of the causative microbe. Patients benefit from early diagnosis and aggressive treatment with multiple antibiotics and surgical debridement. This case serves as a reminder to consider *M. chelonae* as a potential cause of periocular infection, which may be more likely to occur with indwelling devices, as well as in patients with obstructive sinonasal diseases requiring nasal irrigations and steroids.

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