CASE REPORT

Solid cast-forming actinomycotic canaliculitis: Case report*

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SUMMARY

Solid-cast forming actinomycotic canaliculitis is an uncommon cause of unilateral chronic red eye resistant to conventional topical medical therapy. The authors report the history of a 62year old woman who was complaining of mucopurulent discharge from the right lower canaliculus for a period of 12 months. Culture yielded a few colonies of Actinomyces. Magnetic resonance imaging showed a dilation of the right lower canaliculus. The signal was hypointense and heterogeneous on both T_1 and T_2 weighted sequences. Surgery enabled removal of several solid yellowish casts and resulted in resolution of the disease. Histopathologic examination confirmed the presence of dense, basophilic conglomerates of filamentous organisms. Because rhinologists have more and more opportunities to perform surgery of the lachrymal pathway they need to be informed about this clinical entity.

Key words: actinomyces, lachrymal drainage system, canaliculitis, MRI

INTRODUCTION

Actinomyces species are anaerobic, gram-positive bacillus with fungi-like structures. These bacteria are commensal in the oropharynx, particularly in tonsillar crypts and dental plaques. Actinomycosis may occur as an acute pyogenic infection or more commonly as a chronic infection that is both suppurative and granulomatous. The organisms are trapped in concretions adherent to the tissue elements. These concretions are also known as "sulfur granules" (Shah et al., 1971). Bacteriological identification of Actinomyces is usually difficult. Actinomyces species are anaerobic bacteria, not abundant in pus and growing slowly in culture. Therefore histologic examination of the granules is often necessary to make the definitive diagnosis. Actinomyces israelii is the primary cause of chronic canaliculitis in individuals over age 50. In our institution the ophtalmologists refer patients for surgery of the lachrymal drainage system to the rhinologists. That is why we present hereby such a clinical case.

CASE REPORT

A 62-year old woman referred by the ophthalmologists to the ENT outpatient department presented with a 12-month history of persistent unilateral, right-sided epiphora associated with

mucopurulent discharge from the inferior lachrymal punctum resistant to several previous topical medical therapy. Clinical examination revealed an inferior eyelid swollen and inflamed. Some yellowish granulations seemed to be present next by the medial canthus (Figure 1). The superior lachrymal punctum



Figure 1. Macroscopic view: right side: the medial portion of the lower canaliculus is swollen. The upper eyelid looks normal.

appeared normal. A digital manipulation with the fingertip expressed purulent secretions from the inferior lachrymal punctum. Eye swabs taken for culture and sensitivity test did not yield any fungal species nor bacteria.

A magnetic resonance imaging showed a dilation of the lower canaliculus with a low intense and heterogeneous signal on both T_1 and T_2 weighted sequences (Figures 2 and 3).



Figure 2. MRI - coronal cut - T_1 weighted sequence. Hypointense signal into the right dilated lower canaliculus.



Figure 3. MRI - coronal cut - T_2 weighted sequence. Hypointense and heterogeneous signal into the right dilated lower canaliculus due to the presence of several "canaliculiths". (white arrow).

The clinical presentation and the MRI findings were suggestive of a right inferior canaliculitis associated with the development of a granulomatous reaction.

Because previous medical therapy did not improve the symptoms the patient underwent a right endonasal dacryocystorhinostomy associated with seringing of the lower canaliculus. DCR was performed high above the lachrymal eminence in order to expose the lumen of the lachrymal sac and the common canaliculus. The mucosa of the lachrymal sac seemed normal. Pus was expressed from the canalicular system and sent for culture. No concretions were visualized at that time. Intraoperative probing and seringing of the lower canaliculus confirmed the patency of the common canaliculus. A silicone bicanalicular nasal stent was put in place and secured by double knots. Culture vielded a few colonies of Actinomyces susceptible to penicillin and amoxycillin. Amoxycillin (1.5 g per day) was then prescribed. Three weeks later, the patient came back to the ENT office. The swelling of the eyelid was dramatically reduced but the patient was still complaining of purulent

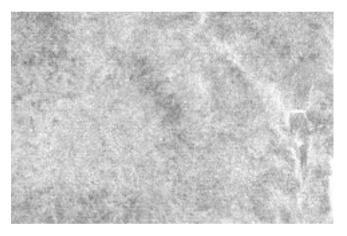


Figure 4. Light microscopy: low magnification. Hematoxylin and eosin-stained section showing a solid cast consisting of dense, dark and basophilic conglomerate of *Actinomyces*.

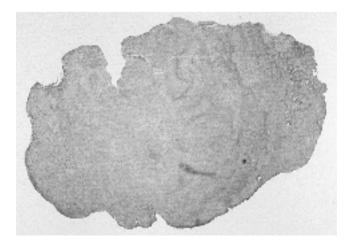


Figure 5. Light microscopy: low magnification. Hematoxylin and eosin-stained section showing clumps of basophilic filamentous organisms.

discharge. The bicanalicular nasal stent was still in place. Fluorescein Dye Disappearance Test (DDT) was negative. A punctoplasty was then performed under local anaesthesia. Unexpectedly, we removed seven solid dehydrated canaliculiths. Histological examination of these "canaliculiths" demonstrated compact conglomerates of *Actinomyces* (Figures 4 and 5). The medical therapy was prolonged. Three weeks later the patient was free of symptoms. DDT was positive. At this moment, after a 7-month follow-up, the patient is still symptom-free.

COMMENTS

Canaliculitis results from an infection of one of the canaliculi. It is a rare condition that accounts for less than 2% of all of the tearing problems (Sullivan et al., 1993, Fulmer et al., 1999). Actinomycotic infection is the first causative agent followed by Propionibacterium propionicus, Nocardia and Bacteroides (Richards et al., 1973; Smith et al., 1980; Brazier et al., 1993; Hussain et al., 1993). Complaints usually consist of a chronic red eye associated with chronic unilateral mucopurulent discharge. Topical medical therapy (eye drops or ointment) typically fails to resolve the disease. Clinical examination reveals an inflamed and outward turned lachrymal punctum. The eyelid and surrounding overlying soft tissue are equally swollen and inflamed. Direct manipulation of the involved canaliculus using a cotton-tipped applicator may express pus. The Fluorescein Dye Disappearance Test (DDT) is negative. Lachrymal probing using a double end Bowman stent reveals a "soft stop" at the level of the common canaliculus. Bacteriological identification is recommended to start the appropriate medical therapy but cultures are routinely negative. Therefore the definitive diagnostic is commonly made by the pathologist. Management of actinomycotic canaliculitis always starts with antibiotic therapy (penicillin G, cephalosporin, lincomycin) administered topically or systemically for one month (Mohr et al, 1970; Martin et al, 1984; Struck et al, 1992; Vecsei et al., 1994). In case of failure to resolve with the medical therapy a surgical exploration of the canalicular system must be performed with complete removal of the adherent concretions (Pavilack et al., 1992). In a very recalcitrant case, adjunctive hyperbaric oxygen therapy could be used successfully (Yaacov et al., 1993). In the present case the medical history, the resistance to several medical therapies and the clinical examination were highly suggestive of a canaliculitis. Bacteriological identification was made late when culture of pus taken during the surgery yielded a few colonies of Actinomyces. If the first bacteriological study had been positive then a high dosage antibiotic therapy should have been prescribed before any imaging or surgery.

Conventional dacryocystography or CT scanning with concomitant dacryography are the most commonly used methods of imaging for evaluation of the lachrymal pathway (Sathananthan et al., 1993; Helies et al., 1995), but in the present case we thought that MR imaging would be better and more adequate to visualize the mucosa and the contents of both the lachrymal sac and the canalicular system (Rubin et al, 1994). In fact, it showed the dilation of the lower canaliculus and a heterogeneous and hypointense signal in both T_1 and T_2 weighted sequences. These findings could have suggested the presence of several pockets of pus at various states of viscosity, or a granulomatous reaction in a dilated canaliculus or the presence of several dehydrated concretions.

As the disease was resistant to previous medical therapy and because we thought we dealt with a primary anaerobic bacterial infection associated with a granulomatous reaction and a subsequent stenosis of the common canaliculus, we performed at first an endoscopic endonasal DCR. This enabled us to expose widely the lachrymal sac and the common canaliculus. No concretions were expressed from the canalicular system at that time. Unfortunately the procedure did not improve the patient. A punctoplasty associated with a complete removal of all the granules was then successfully performed.

Finally we prescribed amoxycillin 1.5 gr per day, since eye drops or ointments containing penicillin are not available in Belgium.

CONCLUSION

Actinomycotic solid cast-forming canaliculitis is an uncommon cause of unilateral mucopurulent discharge with currently negative culture. This condition is usually resistant to any topical or systemic medical therapy. As ENT surgeons have more and more opportunities to perform surgery of the lachrymal pathway they need to be informed about this entity. Careful clinical examination of the medial canthus should alert the physician to that diagnosis. Antimicrobial therapy should be started as soon as the diagnosis is evoked. In case of failure to resolve the disease with the medical therapy, surgical exploration of the canalicular system should be accomplished first. It consists of a punctoplasty associated with removal of all the concretions. Dacryocystorhinostomy is indicated only when stenosis of the common canaliculus persists despite of removal of all the adherent concretions. To our knowledge this is the first time that MR imaging illustrates such a case.

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