

CASE REPORT

Open Access



Aggregatibacter aphrophilus chronic lacrimal canaliculitis: a case report

Marie Boulze-Pankert¹, Cécile Roux¹, Vanessa D. Nkamga², Frédérique Gouriet², Marie-Christine Rojat-Habib³, Michel Drancourt^{2,4*} and Louis Hoffart¹

Abstract

Background: Chronic canaliculitis is often misdiagnosed as conjunctivitis, delaying proper documentation and management. *Aggregatibacter aphrophillus* has not been implicated in chronic canaliculitis.

Case presentation: We report a case of unilateral chronic epiphora associated with chronic lacrimal canaliculitis resistant to prolonged topical antibiotic treatment in a 65-year-old woman without notable medical history. Canaculotomy, curettage with removal of concretions and tubing with silicone stent for six weeks resolved this chronic infection. Culturing lacrimal secretions and concretions yielded *Aggregatibacter aphrophilus* in pure culture. Histological analyses showed elongated seed clusters surrounded by neutrophils. Fluorescence in Situ Hybridization confirmed the presence of bacteria in two distinctive concretions.

Conclusion: This first documented case of *A. aphrophilus* chronic lacrimal canaliculitis illustrates that optimal surgical management of chronic lacrimal canaliculitis allows for both accurate microbiological diagnosis and treatment.

Keywords: Canaliculitis, *Aggregatibacter aphrophilus*, *Haemophilus aphrophilus*, diagnosis, Canaculitis, *Aggregatibacter aphrophillus*, Canaliculotomy, Case report

Background

Chronic canaliculitis is often misdiagnosed as conjunctivitis, delaying proper management. Its diagnosis should include appropriate microbiological investigations based on the analysis of surgical clinical specimens, as treatment should include both surgery of the canaliculus and pathogen-targeted antibiotic treatment. Based on this approach, we here report on a case of *Aggregatibacter aphrophilus* chronic canaliculitis, firmly diagnosed using advanced microbiological methods.

Case presentation

A 65-year-old woman was referred to perform a dacryocystorhinostomy for chronic epiphora with mucopurulent secretions resistant to several topical antibiotic

treatments. The patient had no history of lacrimal plug, palpebral surgery or trauma. This patient had been initially diagnosed with chronic conjunctivitis and dacryocystitis. However, her clinical presentation included a lower eyelid erythema and a red, pouting punctum expressing a mucopurulent discharge after bidigital massage (Fig. 1a). Slit lamp examination showed pericanalicular inflammation without lacrimal sac involvement. Probing and irrigation showed permeability of the lacrimal drainage system. Chronic canaliculitis was finally diagnosed and the patient underwent canaliculotomy under general anaesthesia involving a linear incision into the conjunctival side of the canaliculus, curetting of concretions and tubing with a silicone stent (Mini Monoka silicone stent, FCI Ophthalmics, Paris, France) for six weeks; followed by topical dexamethasone 1 mg/mL combined with tobramycin 0.3 % QID for 15 days. Culture of the secretions and concretions on 5 %-sheep blood Colombia agar incubated under a strict anaerobic atmosphere for seven days remained sterile but culture on a chocolate agar (PolyViteX, bioMérieux, Marcy l'Etoile, France) incubated in a 5 % CO₂-enriched

* Correspondence: michel.drancourt@univ-amu.fr

²Fédération de Microbiologie Clinique et Unité des Rickettsies, CNRS UMR 6020, Aix-Marseille Université, IHU Méditerranée Infection, Marseille, France

⁴Unité de Recherche sur les Maladies Infectieuses et Tropicales Emergentes, Faculté de Médecine, 27, Boulevard Jean Moulin, 13385 Marseille Cedex 5, France

Full list of author information is available at the end of the article



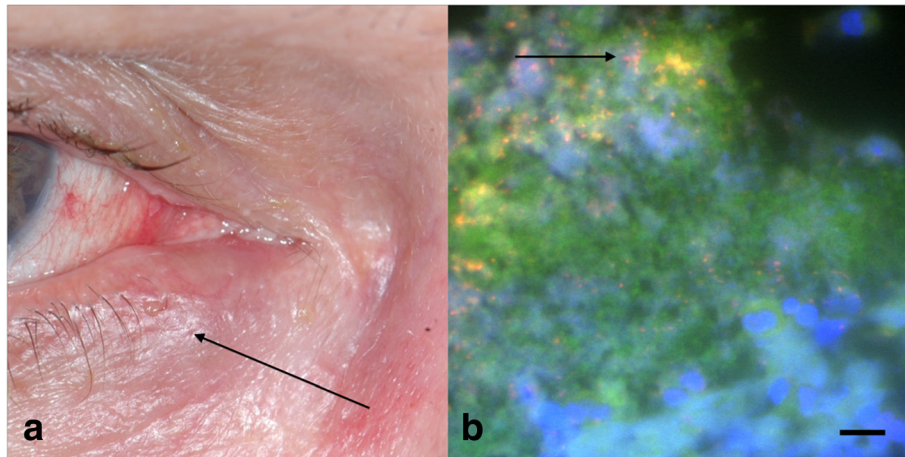


Fig. 1 Chronic *Aggregatibacter aphrophilus* lacrimal canalculitis. **a** Photograph of right eye showing swollen lower canaliculus (arrow), inflamed conjunctiva and mucopurulent secretions **(b)** FISH detection of *A. aphrophilus* in canalculitis concretions. Fluorescent microscopy revealed bacteria labeled by EUB338 probe (red fluorescence) (arrow) when combining non-specific DNA label by DAPI (blue fluorescence) and negative control probe (green fluorescence). Scale bar, 50 microns

atmosphere yielded *Aggregatibacter aphrophilus* identified by matrix-assisted laser desorption ionization time-of-flight mass-spectrometry (MALDI-TOF-MS) with an identification score of 1.737. Using the disk diffusion method, the isolate tested susceptible to amoxicillin (minimum inhibitory concentration (MIC), 0.5 mg/L), ceftriaxone (MIC <2 g/L), gentamicin (MIC, 0.25 mg/L) and rifampicin (MIC <2 g/L). The microbial community structure of the canalculitis was studied by Fluorescence in situ hybridization (FISH) incorporating probe EUB338 5'-GCTGCCTCCCGTAGGAGT-3' labeled with Alexa fluor-546, specific for Eubacteria 16S rRNA gene and probe ARC915 5'-GTGCTCCC CCGCCAATTCCT-3' labeled with Alexa fluor-488, specific for Archaea 16S rRNA gene. FISH revealed cocci detected by EUB338 probe (red fluorescence), while the ARC915 probe (green fluorescence) remained negative (Fig. 1b). Histological analysis after hematoxylin and eosin staining showed clusters of elongated microorganisms surrounded by neutrophils. After ablation of the silicone stent at six weeks, the four-month follow-up showed no sign of infection and the patient was judged cured.

Conclusions

Chronic lacrimal canalculitis is rarely detected in clinical practice, accounting for 2 % of lacrimal duct diseases. This inflammation is caused by infection or punctual plug insertion. Generally, canalculitis is a primitive unilateral condition caused by *Streptococcus* spp., *Staphylococcus* spp. or *Actinomyces* spp. [1]. In the patient here reported, *A. aphrophilus*, formerly *Haemophilus aphrophilus*, a fastidious Gram-negative inhabitant of the oropharyngeal microbiota, was detected by FISH in two distinct

concretions, cultured and firmly identified by mass spectrometry. Additional next-generation sequencing is a more research-oriented method, which can also be used in selected cases to disclose microorganisms. Only four cases of *A. aphrophilus* ocular infection have been previously reported [2–4] including two cases of endophthalmitis, one case of trabeculectomy bleb infection and one cited and as yet undescribed case of canalculitis [3]. Other infections mainly include brain abscess [5] and endocarditis [6].

Topical antibiotics are ineffective for curing chronic canalculitis due to chronic colonized concretions [7], as illustrated by the case here reported in which antibiotics failed, despite an exquisitely antibiotic-sensitive strain of *A. aphrophilus*. We therefore recommend surgical treatment, canalculotomy with incision of the punctum and curetting of the concretions, as the standard treatment of chronic canalculitis.

Abbreviations

FISH, fluorescence in situ hybridization; MALDI-TOF-MS, matrix-assisted laser desorption ionization time-of-flight mass-spectrometry; MIC: minimum inhibitory concentration

Availability of data and materials

The dataset supporting the conclusions of this article is included within the article.

Authors' contribution

MBR, CR, LH managed the patient. VN, FG and MD performed microbiological analyses including FISH. MRH performed histological analyses. MBP drafted the report. All authors wrote the report and approved its final version. Consent to publication was obtained.

Competing interests

The authors declare that they have no competing interests.

Consent for publication

Written informed consent was obtained from the patient for publication of this case and any accompanying images. A copy of the written consent is available for review by the editor of this journal.

Ethics approval and consent to participate

Not applicable.

Author details

¹Service d'Ophtalmologie, Hôpital de la Timone, Aix-Marseille-Université, Marseille, France. ²Fédération de Microbiologie Clinique et Unité des Rickettsies, CNRS UMR 6020, Aix-Marseille Université, IHU Méditerranée Infection, Marseille, France. ³Service d'anatomopathologie, Hôpital de la Timone, Marseille, France. ⁴Unité de Recherche sur les Maladies Infectieuses et Tropicales Emergentes, Faculté de Médecine, 27, Boulevard Jean Moulin, 13385 Marseille Cedex 5, France.

Received: 31 March 2016 Accepted: 28 July 2016

Published online: 02 August 2016

References

1. Freedman JR, Markert MS, Cohen AJ. Primary and secondary lacrimal canaliculitis: a review of literature. *Surv Ophthalmol*. 2011;56:336–47.
2. Macken PL, Boyd SR, Campbell I, Chang D, Rootman DS, Trope GE. *Haemophilus aphrophilus* bleb infection after a mitomycin trabeculectomy. *Aust N Z J Ophthalmol*. 1995;23:323–5.
3. Huang S-T, Lee H-C, Lee N-Y, Liu K-H, Ko W-C. Clinical characteristics of invasive *Haemophilus aphrophilus* infections. *J Microbiol Immunol Infect Wei Mian Yu Gan Ran Za Zhi*. 2005;38:271–6.
4. Shum JWH, Tsang FCW, Fung KSC, Li KKW. Presumed *Aggregatibacter aphrophilus* endogenous endophthalmitis. *Int Ophthalmol*. 2015;35:269–73.
5. Maraki S, Papadakis IS, Chronakis E, Panagopoulos D, Vakis A. *Aggregatibacter aphrophilus* brain abscess secondary to primary tooth extraction: Case report and literature review. *J Microbiol Immunol Infect*. 2016;49:119–22.
6. Nørskov-Lauritsen N. Classification, identification, and clinical significance of *Haemophilus* and *Aggregatibacter* species with host specificity for humans. *Clin Microbiol Rev*. 2014;27:214–40.
7. Kaliki S, Ali MJ, Honavar SG, Chandrasekhar G, Naik MN. Primary canaliculitis: clinical features, microbiological profile, and management outcome. *Ophthalm Plast Reconstr Surg*. 2012;28:355–60.

Submit your next manuscript to BioMed Central and we will help you at every step:

- We accept pre-submission inquiries
- Our selector tool helps you to find the most relevant journal
- We provide round the clock customer support
- Convenient online submission
- Thorough peer review
- Inclusion in PubMed and all major indexing services
- Maximum visibility for your research

Submit your manuscript at
www.biomedcentral.com/submit

