Isolation of unusual bacteria in canaliculitis: A series of four cases

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Access this article online

Quick Response Code:

Website: www.saudijophthalmol.org

DOI: 10.4103/1319-4534.325778

Abstract:

With increased availability of sophisticated microbiological techniques for isolation, growth and identification of micro-organisms the spectrum of organisms is rapidly. Herein we report four cases of canaliculitis with unusual organisms and highlight their clinical significance. To the best of our knowledge, there are no reports of isolation of Brucella melitensis and Leuconostoc species reported in English literature; and only one report of isolation of Myroides species from canaliculitis exists. Sphingomonas paucimobilis, is an uncommon isolate in canaliculitis. Extremes of age and occupational exposure may be possible risk factors for infection with uncommon organisms. Clinical features at presentation do not vary greatly with uncommon or multi drug resistant organisms' hence sampling and microbiological assessment is warranted. The benefit of curettage in canaliculitis is manifold. Unusual organisms and opportunistic pathogens can be multi-drug resistant and determination of antibiotic susceptibility is important to initiate targeted therapy to ensure disease cure and prevent recurrences.

Keywords:

Canaliculitis, pediatric canaliculitis, concretions, Myroides, Leuconostoc, Sphingomona

Introduction

Canaliculitis accounts for 2%–4% of lacrimal diseases and the usual causative organisms include gram-positive organisms such as *Actinomycetes, Staphylococcus, and Streptococcus* species.[1-4] We describe four cases of canaliculitis with uncommon microbiological isolates and elaborate on their clinical relevance in terms of susceptibility to antimicrobial agents. There are no reports of isolation of *Brucella melitensis* and *Leuconostoc* reported in the English literature; and we also report the first case of isolation of *Myroides* species from childhood canaliculitis. *Sphingomonas paucimobilis* has been an uncommon isolate in primary canaliculitis.[3] Extremes of age and occupational exposure may be possible risk factors for infection with uncommon organisms. As there is significant overlap in the clinical presentation of canaliculitis caused by uncommon organisms sampling and microbiological workup are warranted. The benefit of curettage in canaliculitis is manifold in reducing the organismal load, increasing the potential space of antibiotic reservoir, and allowing microbiological assessment. This case series highlights that unusual isolates from canaliculitis can be opportunistic pathogens showing multi-drug resistance and determination of antibiotic susceptibility helps initiate targeted therapy. Patient consent has been obtained for publishing clinical photographs. The study adhered to the declaration of Helsinki.

Case Reports

Case 1

A 49-year-old male came to us with complaints of watering in his right eye of 3 months duration. On examination, there was erythematous swelling of the medial third of the lower eyelid and an inflamed pouting punctum [Figure 1a]. Debris expressed from the lower canaliculus on the application of pressure with cotton-tipped applicator was sent for microbiological examination. Gram's staining of the canalicular contents revealed numerous gram-positive

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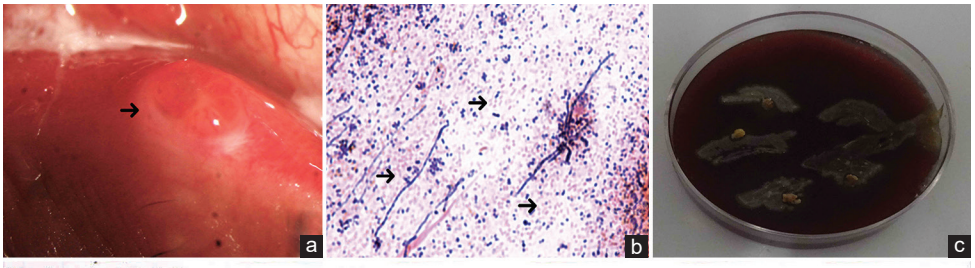
Submitted: 20-Sep-2019
Accepted: 06-Dec-2020
Published: 09-Sep-2021

How to cite this article: Vempuluru VS, Mitra S, Tripathy D, Mohapatra S, Rath S. Isolation of unusual bacteria in canaliculitis: A series of four cases. Saudi J Ophthalmol 2021;35:66-70.

branching filamentous organisms and groups of gram-positive cocci in pairs and short chains among a large number of gram-negative coccobacilli [Figure 1b]. On aerobic culture, confluent pin-point alpha-hemolytic colonies and confluent translucent gray colonies grew on blood and chocolate agar at the site of inoculation [Figure 1c]. Anaerobic subculture from thioglycolate broth grew discrete whitish colonies with a "molar-tooth" appearance. The organisms were isolated as *Streptococcus* species, *Sphingomonas paucimobilis* [Figure 1d], and *Actinomycetes* species. *Streptococcus* and *Sphingomonas* were sensitive to all antibiotics [Table 1], but *Actinomycetes* species showed multidrug resistance.

Case 2

A 9-year-old child was brought by his parents with complaints of discharge from his right eye for 3 months. Careful examination of his right eye revealed upper canalicular edema, pouting punctum, and expression of purulent material on application of pressure over the upper canaliculus. Although rare in children, classical features hinted at the diagnosis of canaliculitis which was confirmed by expression of copious pus and multiple concretions on curettage. Gram's staining of smears from the concretion revealed plenty of polymorphonucleocytes with few gram-negative bacilli per oil immersion field [Figure 2a]. On aerobic culture, growth of discrete, pale yellow colored



Biochemical Details

2	APPA	+	3	ADO	-	4	PyrA	-	5	IARL	-	7	dCEL	-	9	BGAL	-
10	H2S	-	11	BNAG	-	12	AGLTp	+	13	dGLU	-	14	GGT	-	15	OFF	-
17	BGLU	-	18	dMAL	-	19	dMAN	-	20	dMNE	-	21	BXYL	-	22	BAlap	-
23	ProA	+	26	LIP	-	27	PLE	-	29	TyrA	+	31	URE	+	32	dSOR	-
33	SAC	-	34	dTAG	-	35	dTRE	-	36	CIT	-	37	MNT	-	39	5KG	-
40	ILATk	-	41	AGLU	-	42	SUCT	-	43	NAGA	-	44	AGAL	-	45	PHOS	+
46	GlyA	-	47	ODC	-	48	LDC	-	53	IHISa	-	56	CMT	-	57	BGUR	-
58	O129R	-	59	GGAA	-	61	IMLTa	-	62	ELLM	-	64	ILATa	-			

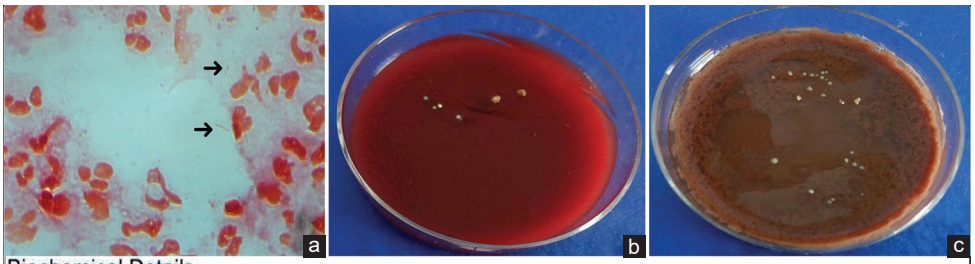
d

Figure 1: (a) Clinical photograph showing eyelid edema and inflamed canalicular mucosa pouting through the punctum (arrow); (b) Smear showing gram-positive branching filamentous organisms, gram-positive cocci in short chains and pairs and numerous gram-negative bacilli (arrows) throughout (Gram stain, ×100); (c) Confluent alpha hemolytic pin point colonies at the site of inoculation on blood agar; (d) Identification of the organism as Sphingomonas paucimobilis by Vitek 2 Compact identification system

Table 1: Clinical and microbiological profile of unusual organisms isolated in our series of patients with canaliculitis

Organism isolated	Patient profile							Coinfection	Antibiotic susceptibility		Comment
	Age/ sex	Duration (months)	OD/ OS	Upper/ lower	Symptoms	Signs	Canalicular contents		Sensitive	Resistant	
Sphingomonas paucimoiblis	49/ male	3	OS	Lower	Watering	Canalicular edema, pouting punctum	Concretions	*Streptococcus species, Actinomyces species*	Ch, Cft, O, Ga, Mo, Ci, Ge, PT, I, Co	-	
Myroides species	9/male	3	OS	Upper	Discharge	Canalicular edema, pouting punctum, discharge	Pus, concretions	*Staphylococcus aureus*	Ch, Cfu, Ak, Ge, PT	AC, O, Ga, Mo, Ci Inter-mediate to Cft, I	Multi-drug resistant
Brucella melitensis	58/ male	12	OD	Lower	Watering, discharge	Canalicular edema, pouting punctum, discharge	Pus, concretions	*Actinomyces species, Staphylococcus epidermidis*	Ch; AC, PT, Cft, Cfu, O, Ge, I, Co	Ga	Possible risk factor: Occupational exposure to cattle
Leuconostoc species	80/ male	1	OD	Lower	Watering, discharge	Canalicular edema, discharge	Debris	-	Ch, Cfa, V, O, Mo, Ci	Inter-mediate to Ga	Possible risk factor: Old age

Ch=Chloramphenicol; Cfa=Cefazolin; Cft=Ceftazidime; Cfu=Cefuroxime; V=Vancomycin; O=Ofloxacin; Ga=Gatifloxacin; Mo=Moxifloxacin; Ci=Ciprofloxacin; Ge=Gentamicin; Ak=Amikacin; AC=Amoxycillin clavulinic acid; PT=Piperacillin tazobacatm; I=Imipenem; Co=Colistin; OD= Oculus dexter; OS= Oculus sinister



Biochemical Details

2	APPA	-	3	ADO	-	4	PyrA	-	5	IARL	-	7	dCEL	-	9	BGAL	-
10	H2S	-	11	BNAG	-	12	AGLTp	-	13	dGLU	+	14	GGT	-	15	OFF	-
17	BGLU	+	18	dMAL	+	19	dMAN	-	20	dMNE	+	21	BXYL	-	22	BAlap	-
23	ProA	+	26	LIP	-	27	PLE	-	29	TyrA	-	31	URE	-	32	dSOR	-
33	SAC	+	34	dTAG	-	35	dTRE	+	36	CIT	-	37	MNT	-	39	5KG	-
40	ILATk	-	41	AGLU	-	42	SUCT	-	43	NAGA	-	44	AGAL	-	45	PHOS	-
46	GlyA	-	47	ODC	-	48	LDC	-	53	IHISa	-	56	CMT	+	57	BGUR	-
58	O129R	-	59	GGAA	-	61	IMLTa	-	62	ELLM	+	64	ILATa	-			

d

Figure 2: (a) Few gram-negative bacilli (arrows) in a background of plenty polymorphs (Gram stain, ×100); (b and c) Discrete, small, pale yellow colored colonies at the site of inoculated concretions on blood and chocolate agar; (d) Identification of the organism as *Myroides* species by Vitek 2 compact identification system

colonies [Figure 2b and c] at the site of inoculation and the organism was identified as *Myroides* species by Vitek 2 Compact identification system [Figure 2d] which was multidrug resistant [Table 1] but sensitive to chloramphenicol.

Case 3

A 58-year-old male, a farmer by occupation presented with persistent watering and intermittent discharge for 1 year. He had had multiple consultations earlier and was treated with topical antibiotics. Ocular examination showed edema of the right lower eyelid localized to the medial aspect with a pouting punctum. Canalicular curettage revealed the presence of pus and concretions which were sent for microbiological evaluation. Gram's stain of the smear showed Gram-positive branching filamentous organisms and Gram-positive cocci in clusters.

Nonhemolytic nonpigmented small gray colonies [Figure 3] grew on blood agar. These were identified as *Brucella melitensis*. *Staphylococcus epidermidis* and *Actinomycetes* were also isolated from culture. The organisms were sensitive to chloramphenicol [Table 1].

Case 4

An 80-year-old male presented with watering and discharge from his right eye for 1 month. On examination, mild lower eyelid edema and matting of lashes with the discharge were noted. There was mild edema at the site of the punctum [Figure 4]. A diagnosis of canaliculitis was made and on curettage, debris was expressed from the lower canaliculus. No pus or concretions were present. Gram-positive cocci were seen in pairs and chains admixed with polymorphonucleocytes on the smear and culture on blood agar revealed small smooth round gray colonies which were identified as *Leuconostoc* species with a favorable sensitivity profile [Table 1].

Management

None of our patients were immunocompromised and all were in good systemic health. Among possible factors predisposing for infection were occupational exposure to cattle in case 3 and old age in case 4. All the patients underwent canalicular curettage and treated with topical chloramphenicol. Microbiological sampling, culture, and interpretation were in accordance with institute laboratory norms. Vitek 2 Compact identification system (Biomerieux, NC, l'Etiole, France) was used for identification of microorganisms in the cases 1–3 and *Leuconostoc* in case 4 was identified with biochemical tests. Kirby Bauer disc diffusion method was used to determine antibiotic susceptibility. All the organisms were susceptible to chloramphenicol and *Myroides* species was found to be multi-drug resistant. All the patients were free of symptoms on follow-up visits at a median follow-up of 6 weeks.

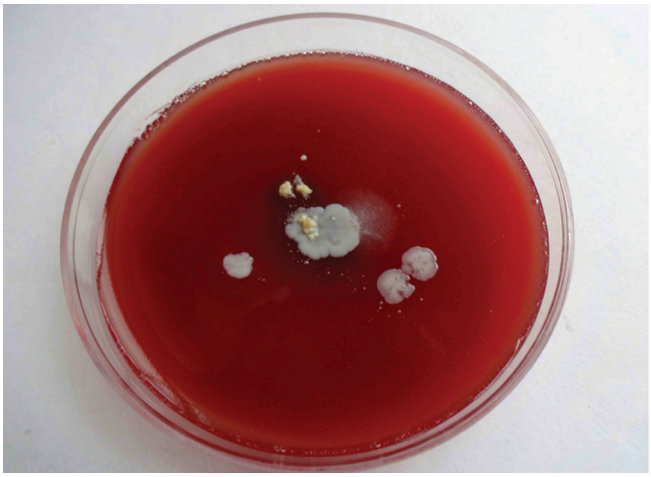


Figure 3: Nonhemolytic non-pigmented small gray colonies of *Brucella melitensis* on blood agar

Discussion

Canaliculitis accounts for about 2% of lacrimal disorders and

Table 2: Epidemiology and clinical relevance of unusual organisms isolated in our series of patients with canaliculitis

Organism isolated	Morphology	Taxonomy	Biochemical properties	Habitat	Human infection	Ocular infection	Reports of canaliculitis in literature
Sphingomonas paucimoiblis	Gram negative coccobacilli	Sphingomonadales	Aerobic, slowly motile	Water	Oppurtunistic pathogen: bacteremia in immunosuppressed	Keratitis Endophthalmitis Canaliculi	Gogandy *et al*. (2015): 4/101 culture positive cases of canaliculitis
Myroides species	Gram negative bacilli	Flavobacteriaceae	Nonmotile Nonfermenter Oxidase	Soil Water	Soft-tissue infections, catheter-related infections, endocarditis	Canaliculitis	Ali *et al* (2015): Only report of myroides isolated from canaliculitis
Brucella melitensis	Gram negative coccobacilli	Brucellaceae	Nonmotile, nonfermenter Oxidase and catalase positive	Infected cattle harbour organisms in mammary glands and reproductive tract	Transmission of infection from infected animals to human through	Kerato-conjunctivitis, uveitis, retinal abscess and vasculitis, endophthalmitis, optic neuritis, dacryoadenitis	None
Leuconostoc species	Gram positive cooci in pairs or short chain	Leuconostococeae	Gas forming	Various foodstuffs; can colonize skin and gastrointestinal tract	Opportunistic pathogen: Bacteremia, catheter-related infections, urinary tract infections, meningitis, osteomyelitis in immunosuppressed	Endophthalmitis	None

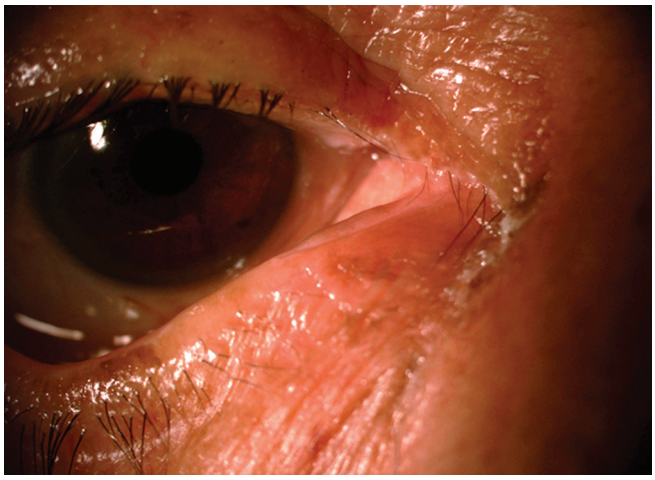


Figure 4: Subtle lower eyelid edema and discharge in the patient with *Leuconostoc* infection

misdiagnosis of the condition ranges from 33% to 100%.[1-6] The most common clinical symptoms include tearing, discharge, swelling and less often pain, or none at all.[1-5] Clinical signs which point at the diagnosis of canaliculitis include canalicular edema, pouting punctum, and expression of discharge on concretions by pressure over the canaliculus.[2,3]

Canaliculitis has a slight female preponderance with the lower canaliculus being more frequently involved.[1] The female preponderance is presumed to be due to a combination of hormonal and lifestyle factors such as the use of cosmetics.[2,3] All the four patients in our series were male. While canaliculitis can affects individuals at any age the mean age at diagnosis lies in the sixth decade.[1,2] Childhood canaliculitis is rare with few reports in English literature.[7] Park *et al*. reported canaliculitis with isolation of *Actinomycetes* in a 5-year-old child on treatment with oral steroids for idiopathic thrombocytopenic purpura but there is not enough evidence implicating immunosuppression as a risk factor for canaliculitis.[7] The minimum age reported in two large series reported by Kaliki *et al*. and Gogandy *et al*. were 8 and 10 years, respectively.[2,3]

There appears to be shift in the microbiological spectrum of organisms in primary canaliculitis with recent large series reported by Kaliki *et al*. and Gogandy *et al*. showing predominance of *Streptococci* and *Staphylococci* species.[2,3] Gram-negative bacilli such as pseudomonas are known to be more common in punctal plug and canalicular stent-related infections.[4]

Sphingomonas paucimobilis as the name implies is a slowly motile gram-negative bacillus that has been isolated infrequently in primary canaliculitis.[4] Systemic and ocular infections with opportunistic pathogens such as *Leuconostoc* species and *Sphingomonas paucimobilis* are known to occur in the setting of immunosuppression.[8-10] Interestingly, we did not find a history of use of punctal plugs or canalicular stents or immunosuppression in the case 1 with *Sphingomonas* infection.

Myroides, isolated in childhood canaliculitis in this series, is not a part of human flora and inhabit water or soil.[11] Ali *et al*. reported an isolated case of *Myroides* associated canaliculitis showing resistance to chloramphenicol, cephalosporins, and penicillin.[11] *Myroides* isolated in our patient was resistant to fluoroquinolones, ceftazidime, imipenem, and penicillins.

Ocular brucellosis has been reported in chronic systemic brucellosis, in the form of kerato-conjunctivitis episcleritis,

uveitis, dacryoadenitis, retinal vasculitis, and optic neuritis.[12] Postulated mechanisms include direct invasion of ocular structures by the pathogen or development of immunological reaction to the bacterium.[12] In either case, the systemic infection is the source and the organism is not known to colonize ocular surface or skin.[12] We herein, report the first case of isolation of *B. melitensis* in canaliculitis. Our patient (Case 3) was a farmer by occupation and exposure to cattle may have been an antecedent factor. No systemic evidence of brucellosis was present.

Leuconostoc is a gram-positive coccus that is an opportunistic pathogen and is known to cause systemic infections such as septicemia and catheter-related infections in immunosuppressed neutropenic individuals.[9,10] Apart from our patient being 80 years old, no systemic predisposing factors were noted. Reports of ocular involvement by *Leuconostoc* are limited to two case reports of endophthalmitis one following uneventful cataract surgery and another following intravitreal ranibizumab injection.[9,10] Both the strains were resistant to resistant to vancomycin and despite early intervention in both cases, one patient had final vision of hand movements and other patient's eye deteriorated to phthisis.[9,10] *Leuconostoc* thus can have a dreadful outcome following intraocular surgery or infection.[9,10] We herein report the first case of canaliculitis with isolation of *Leuconostoc* species and emphasize the importance of complete cure and reviewing microbiological culture in canaliculitis, more so before planning any intraocular surgery.

Concretions that were earlier thought to be specific to *Actinomycetes* have also been reported to occur with, but not limited to *Staphylococcus, Streptococcus, Hemophilus, Pseudomonas, Sphingomonas,* and *Gemella*.[3] Infection from concretions. Ali *et al*. also reported the presence of concretions. *Myroides* and *Brucella* in our patients showed the presence of in canaliculitis with *Myroides* isolation [Table 2].[6]

Management of canaliculitis has been multi-faceted with reasonable success rates reported for each of the procedures described in different case series.[1-5] Large series show curettage is effective in primary canaliculitis and prevents further recurrences.[2,3] The advantages of curettage include decrease in organismal load, improved penetration of antibiotics, and the availability of specimens for microbiological evaluation to identify and determine antibiotic susceptibility. Punctal dilatation and expression as described by Kaliki *et al*. is less invasive procedure but had higher (40%) episodes of recurrent canaliculitis.[2] Conservative management by irrigation with antibiotics, described by Mohan *et al*. was reported to be effective.[13] Thus, conservative and/or punctal dilation and expression may be useful as the first line of therapy in most primary canaliculitis, canalicular curettage is the preferred management modality for recurrent disease.

Culture positivity in canaliculitis has improved over time with recent large case series on canaliculitis showing culture positivity range from 78% to 91%. Freedman *et al*. reported no organisms or non-specific organisms in 30% of the canaliculitis.[1-3] This is likely to be a consequence of advances in microbiological techniques over time.

In summary, this short case series documents for the first time isolation of *Brucella melitensis* and *Leuconostoc* in primary canaliculitis. We also report the first case of *Myroides* species from childhood canaliculitis. *Sphingomonas paucimobilis* remain an uncommon isolate in primary canaliculitis.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patients have given their consent for their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

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