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Actinomyces canaliculitis: diagnosis of a masquerading disease

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Abstract *Background:* To review the diagnosis and management of seven cases of *Actinomyces canaliculitis*. *Methods:* Culture of discharge was performed in six of seven patients with *Actinomyces canaliculitis* using a PD Plus/F blood culture bottle. All patients were treated by canaliculotomy with curettage of dacryoliths, followed by treatment with systemic penicillin and Sulphacetamide drops over a period of 3–6 months. Part of the curetted concretions was fixed on a glass slide and part was sent to the laboratory for culture. *Results:* Four patients were women and three men with age ranging between 43 and 90 years. The average time lapse between onset of symptoms until diagnosis was 3 years. All cases presented with epiphora, chronic conjunctivitis, palpably thickened canaliculus, and yellow punctal discharge. Diagnosis was achieved by culture of discharge in three of six cases, culture of concretions in three of five cases, and staining of dacryoliths in all seven cases. Follow-up ranged between 12 and 48 months. The canaliculitis resolved completely and all patients have patent canalicula. *Conclusions:* *Actinomyces canaliculitis* presents with epiphora, chronic purulent conjunctivitis, a palpably thickened canaliculus, and yellow punctual discharge. In suspect cases canuliculotomy and curettage should be performed, although canalicular reconstruction is generally unnecessary. Culture of discharge and concretions using PD Plus/F blood culture medium gave improved results over accepted norms. Fixation of smeared concretions on a slide in alcohol is simple and is diagnostic of the disease. We recommend long-term systemic penicillin treatment in *Actinomyces canaliculitis*.

Introduction

Lacrimal canaliculitis is a relatively rare condition and is commonly undiagnosed for long periods of time [2, 7, 12]. Laboratory culture diagnosis of this disease can be difficult and secondary infection commonly confuses the picture [2, 7, 12]. *Actinomyces* is the most common associated cause of canaliculitis, although several different organisms have been shown to cause the disease [2, 5, 7, 11, 12]. Concretions of lacrimal canaliculus were first recognized by Von Graefe 1854. Israel described human actinomycosis in 1878, and Harz first used the name *Actinomyces* in 1879 to describe the concretions of lesions in cattle [6, 8]. *Actinomyces* are Gram-positive anaerobic bacilli occurring singly, in pairs, or in chains and have a fungal mycelia-like appearance with branched radiating filaments (Fig. 1)

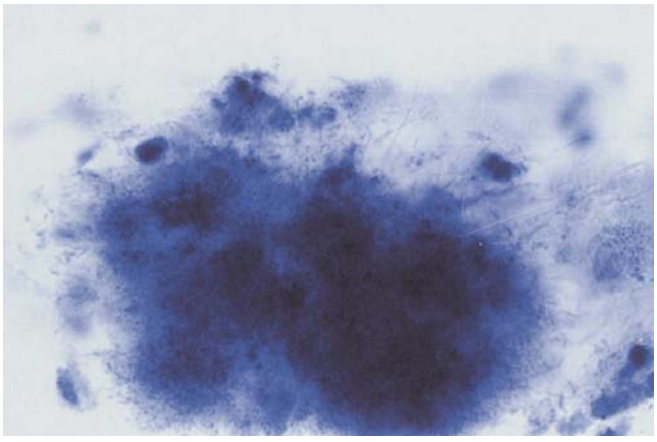

Fig. 1 An actinomycotic granule with numerous radially oriented basophilic filamentous bacteria and peripheral eosinophilic clubs (H&E stain, original magnification, ×1000)

They are a normal commensal part of the bacterial flora of the oropharynx, gastrointestinal tract and female genital tract in humans [17]. The disease occurs when they abandon their commensal properties and become pathogenic. This is characterized by a chronic purulent granulomatous infection with typical sulphur granules which generally spreads directly into adjacent tissues. Actinomycosis involving the eye and adnexae is also known as streptotrichosis, and leptotrichosis [4, 7]. The lacrimal drainage system is the most common site of this infection in the ocular adnexae, although it has been also described as causing keratitis, orbital cellulites, endophthalmitis and orbital implant infection [7, 9, 13, 16, 18]. The classic features of *Actinomyces canaliculitis* are a mucopurulent punctal discharge, epiphora, inflammation of the medial canthus, canalicular swelling, and a red pouting punctum [5]. We found only two reports from the past 25 years [2, 7] discussing the features of a series of *Actinomyces canaliculitis* cases, although some case reports have been published [5, 15]. We therefore set out to discover the frequency of clinical characteristics, the efficacy of modern laboratory diagnostic techniques and to determine the outcome of our treatment regimen, in a series of seven patients.

Materials and methods

We examined the files of all adult patients with canaliculitis who were examined in the Lacrimal clinic of Sapir Medical Center between December 1999 and May 2002. There were seven patients, all of whom came from different referring doctors, having been previously misdiagnosed and treated inappropriately. There was no common source of iatrogenic infection. All cases were examined for concomitant eyelid disease. In six of the seven cases purulent discharge expressed from the punctum was drawn carefully into a sterile syringe. A small amount of sterile saline was then drawn into the syringe and mixed with the discharge, before being injected into a PD Plus/F blood culture bottle (Becktin and Dickinson, N.J., USA).

Canaliculotomy was performed by incising the affected canaliculus over a Bowman lacrimal probe using a number 11 blade. Curettage and removal of all dacryoliths and sulphur granules was meticulously performed using a chalazion currette. The canaliculus was not irrigated with antibiotic in any of our cases. All patients were treated with systemic penicillin for at least 3 months along with Sulphatamide drops four times daily. Four patients initially received home IV treatment with penicillin 20 million units per day for 3 weeks followed by oral therapy of 2 g per day over 3–6 months. The other 3 patients refused IV therapy and were treated by oral penicillin alone 2 g per day over 3–6 months. Follow-up after surgery ranged between 12 and 48 months.

In 6 cases the canaliculus was left open and allowed to heal with no silicone intubation or reconstruction. In one case with a distal canalicular block bicanalicular intubation using Crawford stents was performed and the canaliculus closed using 7.0 polyglactin sutures. In 5 of the 7 cases part of the concretions were sent to the bacteriology laboratory for culture, and in all 7 cases concretions were smeared onto glass slides, fixed in alcohol and left to dry before being sent to the pathology laboratory for staining and examination. The slides were stained using hematoxylin–eosin (H&E), Gram and PAS. The H&E was performed in order to get the general appearance and to examine the type of material present. Gram and PAS stains were used to achieve a specific diagnosis as shown in Fig. 1. In H&E stain actinomycotic granules are basophilic with peripheral club-like eosinophilic structures. The granule typically shows numerous radially oriented Gram-positive filamentous bacteria and peripheral clubs at their edge which are PAS positive.

Results

Four patients were women and three were men (age range between 43 and 90 years). Four patients were affected in the left eye and three in the right eye. The upper canaliculus was affected in two cases, the lower in four, and both upper and lower in one case alone (Table1). The time lapse between the onset of symptoms until diagnosis ranged between 3 months and 15 years (Table2). None of the patients suffered from other eyelid disease.

All seven cases had epiphora and chronic conjunctivitis with secondary infection. Bacteria cultured causing secondary infection are listed in Table 2. Treatment prior to surgery is listed in Table 1. Laboratory results, antibiotic treatment and follow up are shown in Table 4.

On examination, all patients had yellow punctual discharge on forced expression, and a palpable swelling/thickening of the affected canalicula. Five of the seven patients had a red swollen canaliculus and four had a pouting punctum (Table 3). Syringing of the lacrimal system through the affected canaliculus was patent in six of seven cases (Table 2).

One patient whose lacrimal canaliculi were patent on syringing initially developed a canalicular stenosis while being treated for conventional conjunctivitis by her referring ophthalmologist. A second patient who had undergone a repeat DCR with bicanalicular lacrimal intubation due to chronic dacryocystitis with a nasolacrimal

Table 1 Patient data, disease location and treatment received before diagnosis

Patient no.	Age (years)	Gender	Canaliculus	Side	Treatment before diagnosis
1	43	F	Lower	Left	Cloxacillin and Amoxycillin (oral); Chloramphenicol and T obramycin (drops)
2	90	M	Upper	Left	Augmentin (oral) Chloramphenicol, Tobramycin, Ciloxan and Fucithalmic (drops)
3	46	F	Lower	Right	Augmentin (oral), Chloramphenicol and Fucithalmic (drops)
4	70	F	Upper and Lower	Left	Chloramphenicol and Tobramycin (drops)
5	82	F	Upper	Left	Augmentin, Metronidazole, Doxycycline (oral), Chloramphenicol, Tobramycin, Gentamycin, and Fucithalmic (drops)
6	82	M	Lower	Right	Chloramphenicol, Dexamethasone and Tobramycin (drops)
7	89	M	Lower	Right	Chloramphenicol (drops)

Table 2 Time lapse to diagnosis, patency of lacrimal system and laboratory findings in patients examined

Patient no.	Time lapse symptoms to diagnosis	Secondary bacterial infection	Syringing	Discharge culture
1	15 years	*Staphylococcus aureus*	Patent	Negative
2	2 years	*Proteus mirabilis*	Patent	Positive
3	2 months	*Streptococcus*	Canal. block	Negative
4	6 months	*Staphylococcus epidermidis*	Patent	Negative
5	2 years	*Staphylococcus epidermidis*	Patent post-DCR	Positive
6	3 months	*Staphylococcus aureus*	Patent	Not performed
7	1 month	None	Patent	Negative

Table 3 Presence of characteristic findings of *Actinomyces canaliculitis* in patients examined. *YPC* yellow punctual discharge, *PC* palpable thickened canaliculus, *RSC* red swollen canaliculus, *PP* pouting punctum

Patient no.	Epiphora	Conjunctivitis	YPC	PC	RSC	PP
1	+	+	+	+	+	_
2	+	+	+	+	+	+
3	+	+	+	+	+	_
4	+	+	+	+	_	_
5	+	+	+	+	+	+
6	+	+	+	+	_	_
7	+	+	+	+	_	+

obstruction continued to suffer from chronic conjunctivitis despite having a completely patent lacrimal system on syringing. It is noteworthy that no concretions or dacryoliths were reported on dacryocystogram prior to her DCR, and no obstruction or abnormality was felt on preoperative or intraoperative probing of the canaliculus. During follow-up after DCR this patient developed a red swollen canaliculus with punctual pouting. This second patient made an uneventful recovery following canaliculotomy with removal of the concretions and systemic penicillin treatment. She has been followed up for over 3 years.

Anaerobic culture of expressed punctual discharge grew *Actinomyces* in three of the six cases (50%) where it was performed. Anaerobic culture of concretions removed at

Table 4 Laboratory results of concretions removed at canaliculotomy, patient management, and follow up period

	Concretion culture	Concretion Smear	Treatment	Canalicular Reconstruction	F/U
Patient 1	Positive	Positive	IV and oral penicillin, Sulphacetamide (Drops)	No	48 months
Patient 2	Positive	Positive	IV and oral penicillin, Sulphacetamide (Drops)	No	14 months
Patient 3	Negative	Positive	IV and oral penicillin, Sulphacetamide (Drops)	Yes	13 months
Patient 4	Not Performed	Positive	Oral Penicillin Sulphacetamide (Drops)	No	15 months
Patient 5	Positive	Positive	IV and oral penicillin, Sulphacetamide (Drops)	No	44 months
Patient 6	Not Performed	Positive	Oral Penicillin Sulphacetamide (Drops)	No	17 months
Patient 7	Negativ	Positive	Oral Penicillin Sulphacetamide (Drops)	No	12 months

F/U = follow up

canaliculotomy grew *Actinomyces* in three of five cases (60%). Pathology slide staining was diagnostic in all seven cases.

Follow-up after surgery ranged between 12 and 48 months (Table 4). There was no recurrence of disease symptoms and canalicula remained patent to lacrimal syringing in all seven patients.

Discussion

In our series the diagnosis of *Actinomyces canaliculitis* was missed and all patients continued to suffer from chronic infection, with an average duration of 36 months. This disease can masquerade at times to a degree that makes it easy to overlook its diagnosis, as was already described in one of our cases. Actinomycosis has been described in Harrison's Principles of Internal Medicine as being the "most misdiagnosed disease" and that "no disease is so often missed by experienced clinicians."

We found in our series that *Actinomyces canaliculitis* presents with all the following features: epiphora; chronic purulent conjunctivitis resistant to conventional treatment with antibiotic drops; a yellow punctual discharge on digital expression; and a thickened affected canaliculus on palpation. Although most of the literature describes the "red swollen canaliculus" and the "pouting punctum" as a typical features, we found them to be absent in 29 and 43% of cases, respectively, in our series. These are most probably later signs of the untreated disease reflecting our inability to detect the disease early. Five of the seven patients (71%) had patent lacrimal syringing and six of the seven patients (86%) had patent lacrimal canaliculi. Lacrimal syringing was patent in all cases in the series of Hussain et al. [7] and in 10 of 12 patients (83%) of cases in the series of Demant and Hurwitz [2]. The presence of dacryocystitis with nasolacrimal duct blockage is uncommon but can confuse the picture and delay diagnosis. The canaliculitis may sometimes not be determined until after dacryocystorhinostomy when discharge continues despite a patent lacrimal system.

Our series was compatible with the literature in that the disorder affected females more than males, the lower lid more than the upper lid and there was a lengthy duration of symptoms until diagnosis and definitive treatment [2, 4, 7].

The *Actinomyces* bacilli are anaerobic and are very difficult to grow in culture with a yield of less than 30% [1, 10]. Culture is usually performed in anaerobic media with 5% carbon dioxide atmosphere for at least 4–6 days. Conjunctival swab cultures are generally ineffective in growing *Actinomyces* and are not ideal. Hussain et al. [7] achieved a positive anaerobic swab culture of discharge in 25% (1 of 4) of those taken, where culture of *Actinomyces* was specifically requested. Demant and Hurwitz [2] cultured *Actinomyces* in 25% (3 of 12) of his cases.

In our series we used the PD Plus/F blood culture bottle (Becktin and Dickinson, N.J., USA).

We achieved growth of *Actinomyces* in 50% (3 of 6) of cases where discharge was carefully taken and injected into the blood-culture bottles and in 60% (3 of 5) of cases where concretions curetted out of the canaliculus were cultured; therefore, the use of this blood-culture medium was far more effective than those used to date.

Definitive diagnosis of disease was achieved in 100% of our cases where concretions removed at canaliculotomy were smeared onto a slide, fixed in alcohol and sent to the pathology laboratory for staining and examination. Canaliculotomy with the smearing of the concretions removed onto a glass slide, and application of alcohol for fixation, is a simple procedure easily performed in any outpatient clinic. The slides do not have to reach the laboratory urgently, and the materials necessary are readily available. Results of this test in our series were reliable and confirmed the diagnosis in all cases.

Although it has been recommended by some authors to instil aqueous penicillin or povidine iodine, and to meticulously repair the canaliculus, we found this unnecessary [3, 4, 7]. In our series six of seven cases were left to heal without any form of repair and all were patent to syringing at follow-up examination. This is compatible with the results of Demant and Hurwitz [2] where 9 cases underwent canaliculotomy without reconstruction or insertion of silicone stents. They found that the canaliculus and punctum usually return to the pre-inflammation status, even without suturing, and that the post-canaliculotomy dacryocystogram may be normal. They, however, used silver nitrate cauterisation following removal of the stones and this in theory could cause damage to the epithelium of the canaliculus. The patients in two other case reports [5, 12] had no reconstruction performed but made a full recovery with patent syringing following canaliculotomy.

We could not find any standard treatment regimen recommendation with systemic antibiotics for lacrimal canaliculitis in the ophthalmology literature. Treatment with high doses of antimicrobials for long periods with therapy continuing for a significant period after symptoms of this disease have resolved is recommended in other systemic *Actinomyces*. This reduces the risk of relapse which is a hallmark of the disease [14]. There are, however, those who recommend surgery and the use of local antibiotics. Both Demant and Hurwitz [2] and Ellis et al. [5] each in their series had a case where there was recurrence of the disease. In the first case systemic penicillin was given only for a short period following canaliculotomy (2 weeks) and in the second case no systemic penicillin was given at all. It was subsequently found in the second case that the disease had infiltrated the lacrimal sac. We based our treatment regimen on that recommended for oral-cervicofacial actinomycosis. Initial intravenous high-dose penicillin therapy of 18–24 million

units for 2–6 weeks followed by oral administration for 6–12 months is recommended in serious infections. Therapy can be less intensive in less extensive disease particularly in the cervicofacial region [14]. Treatment should be continued for at least 4–6 weeks after clinical resolution of the disease. We therefore chose a regimen of IV penicillin 20 million units administered daily for at least 3 weeks, followed by oral administration of 2 g per day for at least 3 months. We felt that this was necessary given the nature of the disease and would prevent its spread and recurrence.

None of our cases had recurrence of the disease to the time of writing and this may be due to the systemic therapy with high dose penicillin. The intravenous administration of penicillin for long periods, however, is much more costly to the health provider and a controlled study is therefore needed to prove its necessity.

Conclusion

We conclude that in any patient presenting with features of epiphora, chronic purulent conjunctivitis, a palpably thickened canaliculus and yellow punctual discharge on digital expression, exploratory canaliculotomy should be considered to rule out the presence of *Actinomyces*. Smearing the concretions onto a glass slide and fixation in alcohol is a simple, available procedure allowing diagnosis in all cases. Canaliculotomy is harmless, easy to perform, effective and we found that there is generally no need to reconstruct the canaliculus.

We found that culture of discharge using PD Plus/F blood culture bottle (Becktin and Dickinson, N.J., USA) is more effective giving better results than those previously published using anaerobic cultures.

We recommend the use of a standard treatment regimen of prolonged systemic antibiotic therapy with penicillin combined with canaliculotomy and curettage of all concretions, in order to ensure a full recovery and to prevent spread of the disease.

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