Curvularia Dacryoadenitis and Panophthalmitis: A Case Report

Supinda Leeamornsiri[1], Tarawadee Chawarung[1], Pawarit Wanichsetakul[1], Nattapon Wongcumchang[1], Kosol Kampitak[1]

[1]Department of Ophthalmology, Faculty of Medicine, Thammasat University, Thailand

Abstract

Purpose: To examine the clinical course of dacryoadenitis and panophthalmitis from Curvularia infection.

Methods: Retrospective case review. The authors reported a case of a 36-year-old female who presented with dacryoadenitis and panophthalmitis. Clinical features, investigations and management were described.

Results: A 36-year-old female with a history of controlled bilateral pars planitis presented with painful red eye with proptosis in the right eye. Her medication regimen at presentation was prednisolone 5 mg/day and mycophenolate mofetil 250 mg/day. Tissue biopsy was performed and the culture yielded Curvularia spp. Topical and systemic anti-fungal medications were given with a good response.

Conclusion: Curvularia can be the cause of dacryoadenitis with panophthalmitis in immunocompromised hosts.

Keywords: Curvularia, dacryoadenitis, panophthalmitis, fungus

No Author has a financial or proprietary interest in material or method mentioned

EyeSEA 2021;16(2): 1-4

https://doi.org/10.36281/2021020101

Introduction

Curvularia is a genus of dematiaceous fungus or black mold which is commonly found in tropical plants and soil. One main risk factor is an immunocompromised host.[1] However, Curvularia infection can also be found in young and healthy patients.[2] The routes of infection are air-borne spore inhalation, history of trauma and cataract surgery.[1,3] Ocular infection with Curvularia is rare. From literature reviews, ophthalmic presentation of Curvularia infection included conjunctivitis, dacryocystitis, keratitis, sino-orbital cellulitis and endophthalmitis.[1-4] To the best of our knowledge, dacryoadenitis from Curvularia infection has not been reported. We describe here the novel clinical presentation and outcome of a patient with dacryoadenitis and panophthalmitis caused by Curvularia infection.

Correspondence to:
Supinda Leeamornsiri, *Department of Ophthalmology, Faculty of Medicine, Thammasat University, Thailand*
Email: supinda_ta@yahoo.com
Received : August 20, 2021
Accepted : August 20, 2021
Published : December 31, 2021

Case report

A 36-year-old female presented with painful red eye OD for 3 weeks. She had a history of controlled bilateral pars planitis for 3 years. She had history of an uneventful cataract surgery 2 years previously and declined a history of trauma or contact against vegetative material. Her occupation was teaching. At presentation, she was on prednisolone 5 mg/day and mycophenolate mofetil 250 mg/day. Review of systems was unremarkable. No symptoms of sinusitis were detected. Her best corrected visual acuity was 20/40 OD and 20/20 OS. Mild right-sided proptosis and right upper eyelid redness were found with normal extraocular muscle movement. Hertel exophthalmometer measured 18 mm and 16 mm of the right and the left eye respectively. Right ocular examination

showed severe conjunctival injection, clear cornea without keratic precipitate, grade 3 anterior chamber cell, pseudophakia, and grade 2 vitreous haze. Retinitis and shallow retinal detachment were noted inferiorly. No relative afferent pupillary defect (RAPD) was found. The left eye was normal. B-scan ultrasonography of the right eye showed diffuse thickening of choriosclera layer with absent of T sign. The intravitreous cavity showed slightly clumped hyperechogenicity and high echogenic elevated membrane-like lesion at the inferior part of the retina without shifting contour. Orbital computed tomography displayed right-sided proptosis, enhancement of sclera at lateral aspect of right globe and diffuse enhancement of right lacrimal gland. Paranasal sinuses were unremarkable. The diagnosis was dacryoadenitis with panophthalmitis. All immunosuppressive drugs were discontinued. Further investigations included complete blood count (CBC), thyroid function test (TFT), serum immunoglobulin G4 (IgG4), venereal disease research laboratory test (VDRL), Treponema pallidum haemagglutination (TPHA), anti-human immunodeficiency virus (anti-HIV), erythrocyte sedimentation rate (ESR), c-reactive protein (CRP), rheumatoid factor (RF), atinuclear antibody (ANA), antineutrophil cytoplasmic antibody (ANCA). The results were unremarkable except for leukocytosis (white blood cell count 16,000/μL). Upper abdominal ultrasonography showed fatty liver without evidence of abscess. Intravenous vancomycin (1.25 g every 8 hours) and ceftriaxone (2 g every 12 hours) were administered. At 5 days follow-up, intraocular inflammation worsened with a visual acuity of 10/200, worsening of active retinitis, grade 3 vitritis and poorly visualized inferior half of retinal detachment. Rhegmatogenous retinal detachment could not be excluded. The patient underwent a 25-gauge pars plana vitrectomy (PPV), endolaser and heavy silicone oil injection. During surgery, she was found to have inferior exudative retinal detachment. No retinal break was seen. Vitreous was sent for bacterial culture, fungal culture, molecular identification for bacteria, molecular identification for fungus, polymerase chain reaction (PCR) for Tuberculosis (TB), herpes simplex virus (HSV), varicella zoster virus (VZV) and cytomegalovirus (CMV). The results were all negative.

At 1 week follow-up, proptosis worsened significantly, leading to visual acuity at level of hand motion. Hertel exophthalmometer progressed to 22 mm of the right eye. Extraocular muscle movement was limited in all directions. RAPD was positive in the right eye. Ocular examination showed a subconjunctival mass at the lateral part, grade 2 anterior chamber cells with severe plasmoid reaction and total exudative retinal detachment. CT scan showed increased enlargement of right lacrimal gland and mild increase scleral thickness from previous imaging. The right lacrimal gland biopsy and subconjunctival tissue biopsy were performed and tissue was sent for bacterial culture, fungal culture, molecular identification for bacteria, molecular identification for fungus, PCR for TB, gram stain, 10% potassium hydroxide (KOH), acid-fast bacillus (AFB), modified AFB and pathology. Intravenous antibiotics were changed to meropenem (2 g every 8 hours).

At 3 weeks follow-up, right lacrimal gland culture yielded Curvularia spp. Pathology showed lymphoid cell infiltration. Other investigations were negative. Meropenam was discontinued. Intravenous amphotericin B (1 mg/kg/day) 50 mg once daily and oral voriconazole 800mg were prescribed for the first day. Amphotericin B was later decreased to 400mg and topical voriconazole was administered hourly. There were no signs of sinusitis and systemic infection. Proptosis, intraocular inflammation and exudative retinal detachment slowly decreased after antifungal therapy. At 3 weeks of antifungal medications, she was discharged with continuation of topical and oral voriconazole (400 mg/d) which was discontinued after 14 weeks due to drug-induced hepatitis. At 8 months follow-up, visual acuity was improved to counting fingers. No proptosis was detected. Right ocular examination demonstrated no conjunctival injection, quiet anterior chamber. Fundus examination showed optic atrophy and mild tractional retinal detachment inferiorly in the right eye.

Discussion

Fungal infection is extremely common in tropical climates. Although Curvularia is a common type dematiaceous filamentous fungus there were no previous reports of dacryoadenitis from Curvularia. The rarity in both dacryoadenitis

and dacryocystitis could be explained by the antiseptic properties of the lacrimal secretions.[2] Regarding slow growing organism, it may take time before Curvularia could be identified from tissue.[3,4]

Our patients had been taking prednisolone and mycophenolate mofetil which could lead to immunosuppression and predisposition to fungal infections. She did not have a history of trauma with dirt or vegetative matters. In absence of these risk factors, spore inhalation was suspected.[2,3] In the literature reviews, the duration from onset of symptoms to surgical intervention in Curvularia endophthalmitis ranged from 2-5 months and the time from specimen collection to correct diagnosis ranged from 7-24 months.[5]

Computer tomography of orbit is a common modality to investigate for orbital infections such as orbital cellulitis..[4] Concomitant paranasal sinus involvement can also be investigated. Magnetic resonance imaging on the other hand, provides better information on soft tissue lesions.[4,6] In our case, there are signs of lacrimal gland and orbital involvement without paranasal lesion.

There was no standard treatment for Curvularia infection. It is usually susceptible to amphotericin B, miconazole, ketoconazole, and itraconazole in high concentration. Although, some reports mention the failure of treatment and complications of high drug concentration.[2] Oral voriconazole is a good choice due to good ocular penetrance. Intravitreal drug penetration is higher than 90% minimum inhibitory concentration for Curvularia.[7,8] Our patient developed drug-induced hepatitis. Hepatotoxic reaction from antifungal agents were increasingly reported ranged from mild and asymptomatic abnormalities in liver function test results to potential fulminant hepatic failure. The incidence of developing abnormal liver function test results was 12-23% of patients who received amphotericin B therapy, less than 1% stopped treatment due to liver injury. Voriconazole reported the risk of abnormal liver function test results as high as 20%. Although, voriconazole presented a higher risk, it may not lead to treatment discontinuation.[9]

Conclusion

Curvularia can be the cause of dacryoadenitis with panophthalmitis in immunocompromised hosts.

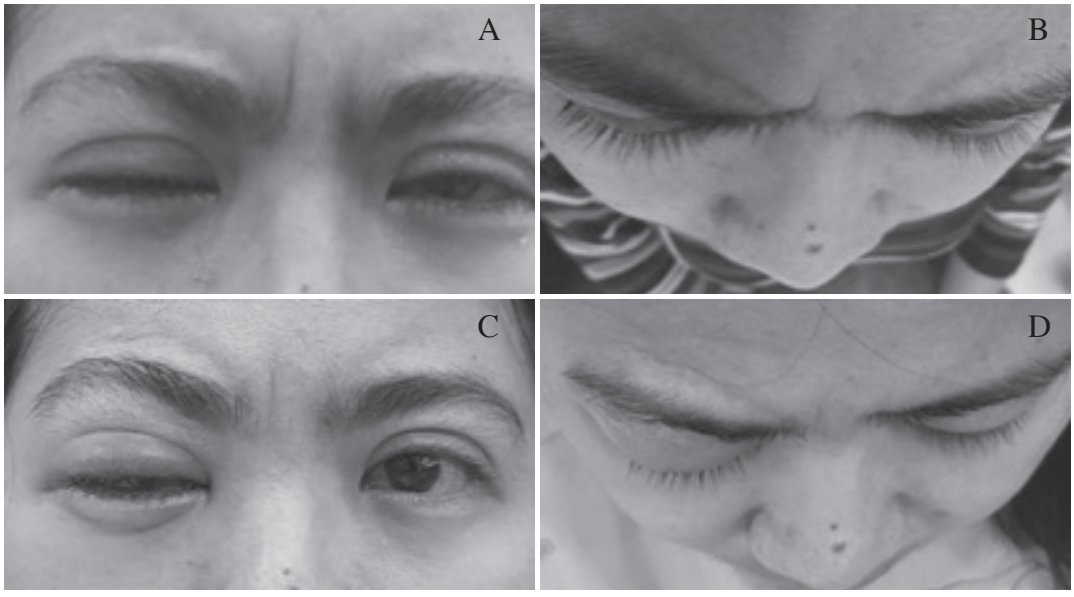

Figure 1. Mild proptosis and erythematous eyelid were noted on the first day of admission (a,b) At 10 day, proptosis increased. (c,d)

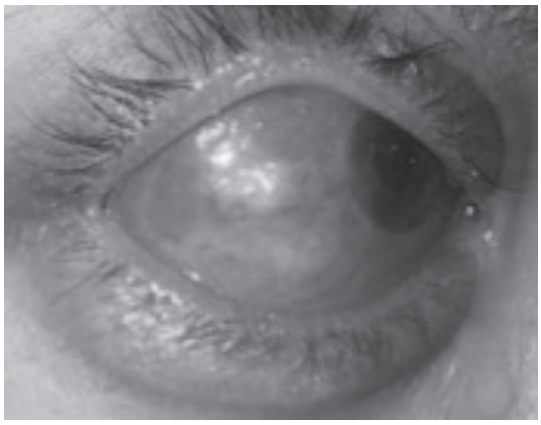

Figure 2. showed a subconjunctival mass at the temporal part of the right eye

References

1. Wilhelmus KR, Jones DB. Curvularia Keratitis. Tr Am Ophth Soc.2001; 99:111-132.
2. Sodhi PK, Kaur R. Curvularia dacryocystitis: report of two cases. Orbit.2000;19:45-50.
3. Rachiskaya AV, Reddy AK, Miller D, Davis J, Flynn JR HW, Smiddy W et al. Prolonged Curvularia endophthalmitis due to organism sequestration. JAMA Ophthalmology. 2014;132(9):1123-26.
4. Heier JS, Gardner TA,Hawes MJ, et al. Proptosis as initial presentation of fungal sinusitis in immunocompetent patients. Ophthalmology 1995;102:713-717.
5. Kaushik S, Ram J, Chakrabarty A, Dogra MR, Gagandeep SB, Gupta A. Curvularia lunata endophthalmitis with secondary keratitis. Am J Ophthalmol. 2001;131:140-142.
6. Adulkar NG, Radhakrishnan A, Vidhya N, Kim U. Invasive sino-orbital fungal infections in immunocompetent patients: a clinic-pathological study. Eye. 2019;33(6):988-94.
7. Xu K, Almeida DR, Chin EK, Mahajan VB; Delayed fungal endophthalmitis secondary to Curvularia. American Journal of Ophthalmology Case Reports 3.2016;1-4.
8. Pathengay A, Miller DM, Flynn HW Jr., Dubovy SR. Curvularia endophthalmitis following open globe injuries. Arch. Ophthalmo. 2012;130:652-654.
9. Wang JL, Chang CH, Young-Xu Y, Chan A. Systematic Review and Meya-Analysis of the Tolerability and Hepatotoxiciy of Antifungals in Empirical and Definitive Therapy for Invasive Fungal Infection. Antimicrobial Agents and Chemotherapy. 2010; 54(6):2409-19.