



A Case Report of *Phialemonium Curvatum* Endophthalmitis

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Abstract

Endophthalmitis caused by *Phialemonium* species are rare. We report a 68-year-old female with unilateral *Phialemonium curvatum* endophthalmitis. She presented with a two day history of right-sided floaters and reduced vision to finger counting, initially treated as iridocyclitis with topical and systemic steroids. Due to worsening, a diagnostic vitrectomy was performed. The culture from the vitreous cavity yielded *Phialemonium curvatum* growth. She was treated with amphotericin B intravitreally once and voriconazole tablets for three months with gain of Best Corrected Visual Acuity (BCVA) to 0.3. Six weeks following discontinuation of voriconazole, the patient developed right-sided eye pain and deterioration of the visual acuity. A second diagnostic vitrectomy was performed and once again the culture demonstrated growth of *Phialemonium curvatum*. She was treated with voriconazole for six months.

Further, she developed eye pain and reduced vision in her right eye about 2 years following the second diagnostic vitrectomy. A third diagnostic vitrectomy was performed which was negative. Two weeks following the third diagnostic vitrectomy, the patient presented with retinal detachment and macular hole in the same eye. Retinal detachment persisted even following surgical repair with silicone insertion. The patient declined further surgeries.

About ten years following initial presentation she developed intolerable right-sided eye pain and phthisis. The eye was eviscerated.

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Keywords

Fundus; Fungal Endophthalmitis; *Phialemonium Curvatum*; Eye Pain

Introduction

The clinical differentiation of fungal endophthalmitis from bacterial, viral and non-infectious causes remains challenging. Fungal endophthalmitis originates exogenously or endogenously. Penetrating trauma and surgeries increase the risk of developing exogenous fungal endophthalmitis. Endogenous fungal endophthalmitis are encountered in immunosuppressed patients or in intravenous drug users. *Candida* species are reported to be the most common cause of endogenous fungal endophthalmitis. Endogenous fungal endophthalmitis is rare and is often associated with disseminated infections caused by *Aspergillus* and *Fusarium* species [1]. *Phialemonium curvatum* is a rare cause of fungal endophthalmitis and few cases of *Phialemonium* endophthalmitis have been reported [2,3]. Immunosuppression remains one of the most common causes of fungal endophthalmitis [4].

Case Presentation

A 68 year old female with diabetes mellitus type 2, bilateral total hip arthroplasty (last operation 8 months prior to the presentation of eye symptoms), psoriasis arthritis and a thyroid gland goiter treated with metformin, methotrexate and levothyroxine presented with a two day history of right-sided floaters and reduced visual acuity to finger counting. It was initially treated as iridocyclitis with topical and systemic steroids. The visual acuity deteriorated to hand movements during the following two weeks, and the intraocular reaction increased with development of vitritis with limited visualization of the fundus. A diagnostic vitrectomy was performed with 23-gauge pars plana vitrectomy with both undiluted and diluted vitreous biopsy and anterior chamber tap. Vancomycin (1 mg/0.1 ml) and ceftazidime (2 mg/0.1 ml) were administered intravitreally. The vitrectomy was complicated by a lens touch and lensectomy was inevitable. Two days following diagnostic vitrectomy she developed hypopyon, and the microbiologist reported growth of *Phialemonium curvatum*. One week following diagnostic vitrectomy an anterior chamber tap was performed to acquire more samples and vancomycin (1 mg/0.1 ml) and amphotericin B (0.0055 mg/0.1 ml) were administered intravitreally. Ten days following the diagnostic vitrectomy her BCVA was 0.05 and hyperpigmented spots and a few round yellow-white exudates, presumably fungal infiltrates, were seen on fundus examination. The culture from the diagnostic vitrectomy was later confirmed by a parallel sample at the Norwegian Reference Laboratory for Medical Mycology as *Phialemonium curvatum* due to morphological characteristics and Internal Transcribed Spacer (ITS) ribosomal Deoxyribonucleic Acid (rDNA) sequencing of the isolate. *In-vitro* susceptibility testing indicated susceptibility to voriconazole with a Minimal Inhibitory Concentration (MIC)

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of 0.25 mg/L and resistance to amphotericin B (MIC 32 mg/L). The culture of the anterior chamber did not reveal growth of any microorganism, but 18S rDNA Polymerase Chain Reaction (PCR) and sequencing on the aspirate identified *Phialemonium curvatum* DNA. She was treated with peroral voriconazole as guided by the antifungal resistance. The voriconazole dose was 600 mg twice a day on the first day of treatment, thereafter 400 mg twice a day for ten days and then she continued with 200 mg twice a day for about three months with gain of BCVA to 0.3.

Six weeks following discontinuation of voriconazole she presented with a 3 day history of right-sided eye pain and deterioration of visual acuity to 0.05. Diagnostic vitrectomy was performed for a second time, and the yellow-white exudates seen on the surface of the retina were removed. The culture demonstrated growth of *Phialemonium curvatum*. The *Phialemonium curvatum* was still considered to be susceptible to voriconazole (MIC 0.064 mg/L) and resistant to amphotericin B (MIC 32 mg/L). Voriconazole was restarted and continued for about half a year (600 mg twice a day on the first day of treatment, 400 mg twice a day for 10 days and 200 mg twice a day for about six months). Intraocular Lens (IOL) implantation was performed 16 months following the second vitrectomy and her BCVA was 0.3 five months following IOL implantation.

Eight months following IOL implantation she presented with eye pain and deterioration of the vision. A third diagnostic vitrectomy was performed, but with negative culture and panfungal PCR (18S rDNA) of the vitreous cavity samples. She developed a retinal detachment and macular hole two weeks following the third diagnostic vitrectomy. Surgical repair was done with vitrectomy, cryotherapy of a retinal tear and silicon oil insertion. At one month follow-up her BCVA was 0.063 oculus dexter and 0.8 oculus sinister. The retina was detached from 3 to 9 O'clock and parafoveally. The patient was offered further surgeries in attempt to improve the condition, but she declined.

About ten years later the patient developed intolerable right-sided eye pain over a period of one month and had vision light perception. On ophthalmological examination a hyphema was seen in her right eye. She did not have diabetic retinopathy in her left eye. She opted for retrobulbar alcohol injection of her right eye. The patient continued to have eye pain and underwent evisceration 6 weeks following retro bulbar alcohol injection. The pathologists reported spores in the eviscerated eye (Fig. 1).

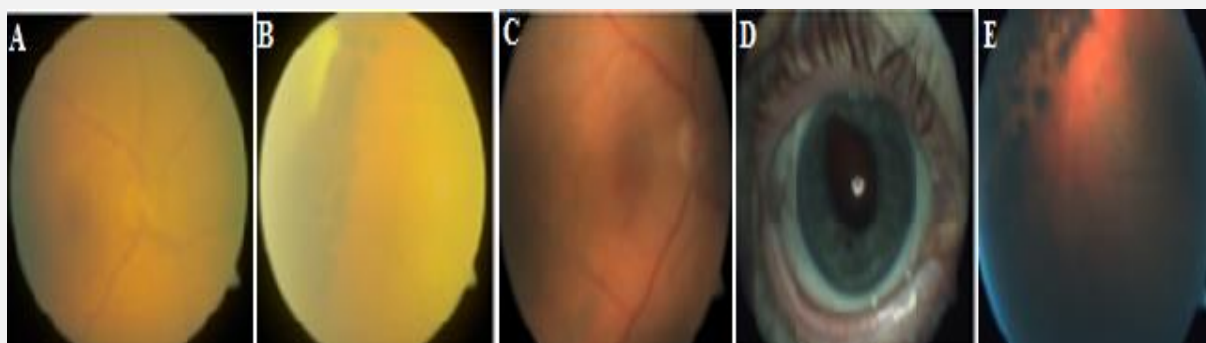


Figure 1: (A-B): Fundus image of the right eye 18 days following first diagnostic vitrectomy. The yellow-white exudation and hyperpigmentation are demonstrated in B; (C): This fundus image was taken one year following the first diagnostic vitrectomy; (D-E): Photo and fundus image of the patient's right eye was taken 19 months following the first presentation. Hyperpigmentation was seen in the periphery.

Discussion

The genus *Phialemonium*, a morphological intermediate between *Phialophora* and *Acremonium*, was first described by Gams and McGinnis in 1983 [5]. These species have been isolated from air, soil, industrial water and sewage [5,6]. *Phialemonium* species belongs to melanized or dematiaceous fungi and are associated with different infectious diseases [7]. In a review of 20 reported cases of *Phialemonium* infection that have been published, the majority of the cases were *Phialemonium* endocarditis (N=7), but also included fungemia/sepsis, peritonitis, spondylodiscitis, arthritis, endophthalmitis, graft infection, lung nodule and mycetoma [5]. The youngest patient with *Phialemonium* infection was a 7-week old premature baby and the oldest patient was aged 84 years (median, 48 years). Fifty percent of the cases were sporadic infections, and the remaining cases were associated with three different outbreaks. Seventy percent of the patients had comorbidities involving immune dysfunction that were mainly terminal chronic renal failure, hematological malignancies and diabetes mellitus. In 15 out of 20 cases, natural trauma of the skin barrier was reported prior to *Phialemonium* infection [6].

Only two case reports of *Phialemonium* endophthalmitis are published [2,3]. The first case of bilateral endogenous *Phialemonium* endophthalmitis was reported in a 71-year-old male in 2005, and this case was part of *Phialemonium* infective endocarditis outbreak in three patients and linked to intracavernous penile injections for treatment of erectile dysfunction. The patient was initially treated with intravenous amphotericin B, but the treatment was changed to intravenous voriconazole when *Phialemonium curvatum* was confirmed in the blood cultures and biopsied heart valve. He underwent bilateral pars plana vitrectomy and cataract extraction for bilateral endophthalmitis and intravitreal amphotericin B was injected. He developed retinal detachment and extensive membranes in the subretinal and subchoroidal space [2]. The second case was reported in a 70-year-old diabetic male patient who was performing intracavernous

penile injections. He presented with symptoms of endophthalmitis in his right eye. He underwent vitrectomy and was treated with intravitreal injections of amphotericin B initially. Eleven months following the initial treatment, he presented with symptoms of endophthalmitis in the same eye and he underwent enucleation [3].

We report the first female endogenous case of *Phialemonium curvatum* endophthalmitis. Although her risk factors for developing *Phialemonium curvatum* endophthalmitis include diabetes mellitus, treatment with methotrexate for psoriasis arthritis and previous total hip arthroplasty, the etiology is uncertain. She did not present with any other focus of *Phialemonium curvatum* infection during the follow-up time of fourteen years. Following discontinuation of voriconazole, the patient presented with *Phialemonium curvatum* endophthalmitis in the same eye. She was further treated with voriconazole for six months with no recurrence. The duration of voriconazole treatment was the main difference between treatment at the first presentation and at recurrence of *Phialemonium curvatum* endophthalmitis.

Conclusion

The visual prognosis in patients with *Phialemonium curvatum* endophthalmitis is uncertain. In our case intolerable pain due to neovascularization and development of phthisis made evisceration necessary, although the infection was cured. The most effective treatment including the duration of treatment for *Phialemonium curvatum* endophthalmitis is uncertain. However, long term treatment with voriconazole and removal of the yellow-white exudates seen on the surface of the retina during vitrectomy may be taken into consideration.

Conflict of Interest

The authors declare no conflict of interest, financial or otherwise.

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