

Trauma to Transplant: Pediatric *Pythium* Keratitis Advancing to Anterior Staphyloma

Abstract

Pythium keratitis is an aggressive, vision-threatening infection that rarely affects children. We report the case of an 11-year-old male who developed progressive corneal opacity and protrusion with vision loss in the right eye, three years after ocular trauma with a cow horn. He was clinically diagnosed with *Pythium* keratitis, which healed with scarring and ectasia, progressing to anterior staphyloma. The patient underwent therapeutic penetrating keratoplasty, and histopathology confirmed sequelae of healed *Pythium* keratitis. Postoperatively, the graft remained clear with improved vision and cosmesis. This case highlights a rare sequela of pediatric *Pythium* keratitis progressing to anterior staphyloma, emphasizing the role of timely keratoplasty in visual and cosmetic rehabilitation.

Keywords: *Pythium* keratitis; anterior staphyloma; cataract; corneal ectasia; penetrating keratoplasty

Introduction

Pythium insidiosum is an aquatic oomycete causing severe, rapidly progressive keratitis, predominantly in tropical and subtropical regions. Its clinical features—including feathery stromal infiltrates, tentacle-like margins, linear extensions, and peripheral guttering—closely mimic fungal keratitis, often delaying diagnosis and appropriate therapy [1,2]. Unlike fungal keratitis, *Pythium* is refractory to standard antifungal agents, and medical management frequently fails to prevent progression [3]. Trauma with vegetative

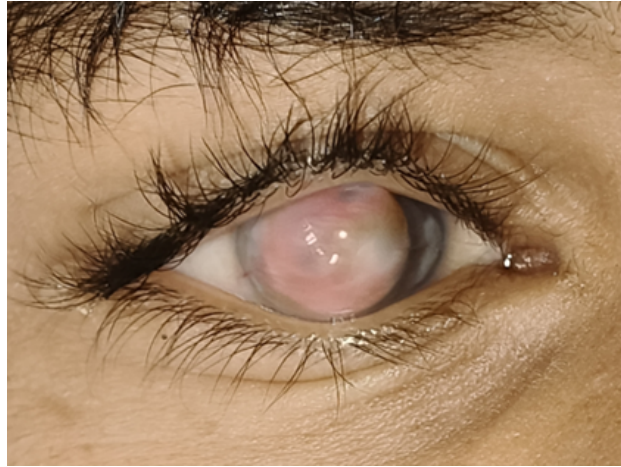
matter or animal exposure is a common predisposing factor, particularly in children [3,4]. Pediatric cases present additional challenges due to delayed reporting, difficulty in examination, and risk of amblyopia from prolonged corneal opacity.

Anterior staphyloma, defined as ectatic protrusion of a thinned, scarred cornea lined by uveal tissue, is a rare but severe sequela of inadequately treated infectious keratitis or trauma, leading to profound visual loss and cosmetic disfigurement. Advanced cases often require therapeutic penetrating keratoplasty (TPK) for anatomical restoration, visual rehabilitation, and cosmetic correction [5]. Pediatric Pythium keratitis progressing to anterior staphyloma is exceedingly uncommon. Here, we report an 11-year-old child with post-traumatic Pythium keratitis who developed anterior staphyloma over three years, successfully managed with TPK.

Case Report

An 11-year-old male presented to our tertiary care ophthalmology department with complaints of progressive whitish opacity and protrusion of the right eye associated with poor vision for the past three years. History revealed ocular trauma to the right eye with a cow horn at the age of eight years, followed by pain, redness, watering, and rapid corneal ulceration. At that time, the child was clinically diagnosed as Pythium keratitis, based on the history of trauma with vegetative matter, rapidly progressive stromal infiltrates, and feathery margins with surrounding tentacle-like extensions. Despite intensive medical management, the infection failed to resolve completely, and the cornea underwent scarring with progressive thinning and ectasia. Over the following years, the affected cornea gradually protruded, leading to the formation of an anterior staphyloma. The parents reported that the protrusion and whitish opacity progressively increased in size, and the child's vision deteriorated significantly. There was no history of prior ocular surgery, systemic illness, or similar complaints in family members.

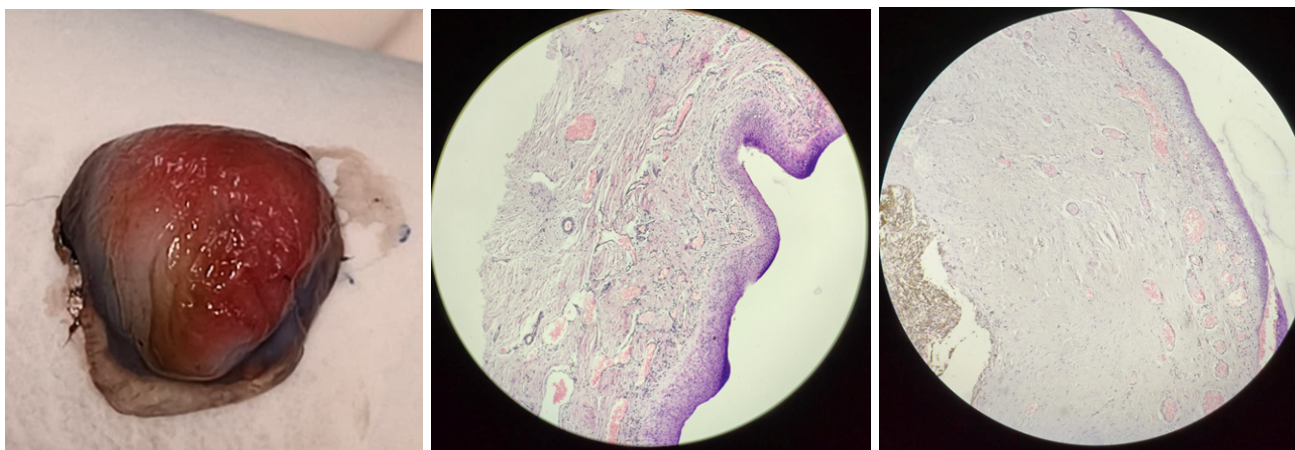
On examination, uncorrected visual acuity (VA) in the right eye (RE) was HM+, PL+, PR4+ not improving further. The left eye (LE) had unaided 6/6 vision. Ocular motility was full in both eyes. Torchlight examination of RE showed an ectatic, irregularly contoured cornea with whitish opacity. Slit-lamp examination revealed a protruding anterior staphyloma with extensive corneal neovascularization, hypertrophic epithelium, and diffuse stromal opacity. (**Figure 1.**) The anterior chamber details were not visualized. Fundus' view was obscured, but B-scan ultrasonography showed cataract formation was present with normal posterior segment. The LE anterior segment, adnexa and fundus were within normal limits.



(Figure 1: Torchlight photograph of the right eye showing anterior staphyloma with whitish corneal opacity, ectasia, and surface neovascularization.)

Given the poor vision and disfiguring anterior staphyloma, the patient was planned for therapeutic penetrating keratoplasty (PKP). Under general anesthesia, a 10.0 mm recipient bed was trephined, and a 10.5 mm donor corneal button was secured with interrupted 10-0 nylon sutures. The excised corneal tissue was submitted for histopathological examination (HPE).

HPE revealed stratified squamous epithelium overlying Bowman's layer, with disorganized stromal collagen bundles, fibrovascular proliferation, and foci of neovascularization, consistent with sequelae of longstanding staphyloma following healed infectious keratitis (Pythium). No viable fungal elements were observed. **(Figure 2A and 2B)**



(Figure 2A: Gross photograph of the excised corneal button, showing an opaque, ectatic specimen consistent with anterior staphyloma. **Figure 2B:** Histopathology of the corneal button demonstrating disorganized stromal collagen with fibrovascular proliferation and neovascularization (H&E, x40))

Postoperatively, the graft remained well-attached and clear with no signs of rejection. (**Figure 3.**) At 4-week follow-up, the best corrected visual acuity (BCVA) in RE improved to counting fingers at 3 meters (CF3m). The cosmetic appearance was significantly better, and the patient and his parents expressed satisfaction with the outcome.



(**Figure 3:** Postoperative photograph showing a clear, well- attached penetrating keratoplasty graft with restored corneal curvature)

Discussion

Pythium insidiosum keratitis is a rare, aggressive infection often misdiagnosed as fungal keratitis due to overlapping clinical features, which can delay diagnosis and worsen prognosis [1,2]. Unlike fungal keratitis, *Pythium* is refractory to conventional antifungal therapy, and therapeutic penetrating keratoplasty (TPK) is often the only definitive management for advanced or non-resolving cases [3,4]. Pediatric presentations are particularly challenging because children often present late, are difficult to examine, and are at risk of amblyopia from prolonged corneal opacity and poor vision [3,5].

Our patient developed keratitis following cow horn trauma, which progressed over three years to anterior staphyloma—a rare chronic outcome in pediatric cases. Most reported pediatric *Pythium* cases either heal with scarring or rapidly progress to perforation, making this prolonged ectatic sequela unusual [5,6].

Pediatric outcomes in the literature are variable. Gurnani et al. reported successful medical management in a 9-year-old using linezolid, azithromycin, and cyanoacrylate glue, achieving BCVA of 6/12 [7]. In contrast, Ranasinghe et al. described a Sri Lankan adolescent requiring multiple keratoplasties with poor visual outcomes [8]. Larger Indian cohorts demonstrate that over 80% of eyes with *Pythium* keratitis ultimately require TPK, with recurrence common and globe loss in up to 25% of cases [9,10].

Histopathology in our patient revealed fibrovascular proliferation and disorganized stromal collagen without viable organisms, consistent with healed infection. TPK with a large graft (10.5 mm) restored globe integrity, improved cosmesis, and provided modest visual improvement. Cosmetic rehabilitation is particularly important in pediatric patients for psychosocial well-being.

Conclusion

Pediatric *Pythium insidiosum* keratitis is a rare, aggressive infection that can progress to severe sequelae such as anterior staphyloma, particularly following ocular trauma. Early recognition and prompt intervention are crucial, as conventional antifungal therapy is often ineffective. Therapeutic penetrating keratoplasty remains the mainstay for anatomical restoration, visual rehabilitation, and cosmetic improvement in advanced pediatric cases.

Informed Consent

Informed consent was obtained from the patient for the publication of this case report, including any accompanying images and details.

References

1. Agarwal S, Iyer G, Srinivasan B, et al. Clinical profile and outcome of medical therapy in *Pythium insidiosum* keratitis in South India. *Cornea*. 2019;38(11):1419-1424. doi:10.1097/ICO.0000000000002089
2. Thomas PA, Kaliyamurthy J. Mycotic keratitis: epidemiology, diagnosis and management. *Clin Microbiol Infect*. 2013;19(3):210-220. doi:10.1111/1469-0691.12126

3. Gaikwad P, Choudhury H, Vaddavalli PK, Garg P. Clinical profile and management outcome of *Pythium insidiosum* keratitis: a review. *Indian J Ophthalmol*. 2022;70(7):2365-2373. doi:10.4103/ijo.IJO_1979_21
4. Sharma N, Sinha R, Vajpayee RB. Therapeutic keratoplasty for microbial keratitis. *Curr Opin Ophthalmol*. 2010;21(4):293-300. doi:10.1097/ICU.0b013e32833b8304
5. Agarwal S, et al. Clinical outcomes of *Pythium* keratitis in India: retrospective multicenter study. *Ocul Immunol Inflamm*. 2020;28(4):609-616. doi:10.1080/09273948.2019.1622931
6. Krajaejun T, Sathapatayavongs B, Prachartam R, et al. Clinical and epidemiological analyses of human pythiosis in Thailand. *Clin Infect Dis*. 2006;43(5):569-576. doi:10.1086/505398
7. Gurnani B, Mishra D, Kaur K, et al. Successful management of pediatric *Pythium insidiosum* keratitis with cyanoacrylate glue, linezolid, and azithromycin: rare case report. *Cornea*. 2021;40(5):646-648. doi:10.1097/ICO.0000000000002591
8. Ranasinghe P, et al. *Pythium* keratitis in Sri Lanka: case report and literature review. *Case Rep Ophthalmol Med*. 2022;2022:6598781. doi:10.1155/2022/6598781
9. Iyer G, Srinivasan B, Agarwal S, et al. *Pythium insidiosum* keratitis: clinical profile and survival analysis of 92 eyes from a tertiary eye care centre in India. *Br J Ophthalmol*. 2018;102(3):329-334. doi:10.1136/bjophthalmol-2017-310162
10. Bagga B, Sharma N, Nagpal R, et al. Meta-analysis of individual patient data on *Pythium* keratitis: factors associated with therapeutic outcomes. *Ocul Surf*. 2023;29:35-44. doi:10.1016/j.jtos.2023.04.004