

Rare conjunctival Rhinosporidiosis: Two cases of superior bulbar involvement, one with associated Scleromalacia

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Case Report

Keywords: ocular rhinosporidiosis, conjunctival mass, scleromalacia

Posted Date: October 11th, 2026

DOI: <https://doi.org/10.21203/rs.3.rs-11049616/v1>

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Additional Declarations: No competing interests reported.

Abstract

Introduction:

Rhinosporidiosis is a chronic granulomatous infection of the mucous membranes caused by the *Rhinosporidium seeberi* that affects both adults and children.

Case presentation:

Two young boys, a 15 years old and a 10 years old, presented with similar history of unilateral painless, gradually progressive reddish lesion in the superior bulbar conjunctiva of 3 to 4 months duration without any blurring of vision. On examination, there were feeder vessels and multiple yellow dots over the lesion in both cases. Both had history of being treated as inflammatory lesions. In the second case, the lesion was associated with scleral thinning. Both underwent complete excision with findings consistent with rhinosporidiosis in histopathology. There was no recurrence, however, the second case came with further bulging of sclera, hence needing donor patch graft for tectonic stability.

Conclusion

Rhinosporidiosis has typical clinical picture. Lesion at superior bulbar conjunctiva may be unrecognised due to silent nature of the disease and may present with complications.

Introduction

Rhinosporidiosis is a chronic granulomatous infection of the mucous membranes (nasal, oral, ocular and rectal) caused by *Rhinosporidium seeberi*. Initially considered fungi, it has been re-classified as an aquatic parasite.¹ The nasal mucosa is most often affected, eye may be affected in 10–18% of cases which manifest in the conjunctiva, lacrimal sac, sclera and eyelids as primary lesion or as secondary dissemination from other mucosal locations.² It usually presents as a slow-growing, reddish friable polyp or sessile mass often with typical pinhead-sized yellowish dots.^{1,2} This disease is endemic across India, Pakistan, Sri Lanka and Nepal. Ocular rhinosporidiosis may mimic a tumour of the eyelid or periorbital skin, hence strong suspicion is needed for its diagnosis.^{2,3} We report two cases of young boys with conjunctival rhinosporidiosis in the upper bulbar conjunctiva, one of them with complication.

Case Report 1

A 15-year-old boy from rural area, living in tropical climate presented with history of pinkish growth on the superior bulbar conjunctiva of the right eye. It was noticed four months back and was slowly progressive. There is no history of itching, pain or any type of discharge. The growth had initially been treated as episcleritis with flurbiprofen followed by topical steroids and moxifloxacin. It seemed to

respond but again increased in size. Then the mass was excised in local eye hospital. After four weeks, it recurred. The mass was excised for the second time again to reappear in few weeks. The excised tissue was not sent for histopathology on both occasions. Thereafter, patient came to our institute. Patient complained of mild discomfort, otherwise he looked comfortable. On examination, he had a pinkish patch measuring 5 x 4mm at the superior bulbar conjunctiva touching the limbus. (Fig. 1A). The surface was irregular with multiple tiny creamy white dots that look studded on the surface of the lesion. It was non tender. Feeder vessels were present. Anterior chamber was quiet. The fellow left eye had normal anterior segment findings. Both eyes' fundus examination was normal. Vision in both eyes was 6/6. Excision biopsy was performed under peribulbar block.

Complete excision of the mass was done with 4 mm of normal looking conjunctiva. Underlying tenons were cleared. Sclera looked normal. Cauterization of the bed was done with cautery forceps (Fig. 1B), the excised tissue was sent for histopathological examination. Post operatively, patient was given fluorometholone and ofloxacin both 4 times a day for 7 days and tapered over four weeks (Fig. 1C). Histopathology reported features suggestive of Rhinosporidiosis.

Case report 2

A 10-year-old boy presented to our hospital with chief complaints of a mass on superior bulbar conjunctiva of the right eye since 2 months. The small mass was first noticed coincidentally by the mother, when she manipulated the eyelid of her son. So, the exact length of time the mass had been present was not known. According to mother, it was gradually increasing in size along bluish discoloration underneath the mass. There was no history of pain, discharge, or trauma to the eye. On examination, the mass in superior bulbar conjunctiva of the right eye was reddish pink with feeder vessels and typical multiple yellowish dots on the surface suggesting possible rhinosporidiosis. Scleral thinning and bulge of the uveal tissue was noted adjacent to the lesion (Fig. 2A). The dilated fundus examination was unremarkable. The patient's uncorrected visual acuity for the right and left eye was 6/36 and 6/6, respectively. Pin hole vision in right eye was 6/9. The intraocular pressure of both eyes was within normal range. Past documents showed that the lesion had been considered as scleritis associated with scleromalacia and received treatment accordingly. Patient had visited several clinics and even had been referred for oncology work up, considering diagnosis of melanoma.

The patient underwent excision biopsy of the lesion and conjunctival autograft under general anesthesia. Conjunctival mass was excised along with 4 mm of normal conjunctiva surrounding the lesion. The underlying tenons were dissected. Significant scleral thinning was noted hence, cautery of the base of the lesion was not performed. Conjunctival autograft was obtained from the inferior site of the same eye and vicryl suture was used to secure it over the thinned sclera (Fig. 2B). The excised lesion was sent for histopathological examination and revealed features suggestive of rhinosporidiosis (Fig. 3A and 3B). Postoperatively, the patient was given a combination of ofloxacin and dexamethasone eye drops 4 times a day and tapered over a month. Other treatments given were ciprofloxacin eye ointment at night, and lubricating eye drops.

We also referred the boy for ENT consultation for any associated nasal lesion but there were none reported. On post-operative day 10, patient was comfortable (Fig. 2B). When patient came for follow-up after two months, although there were no signs of recurrence of infection, the scleral thinning and protrusion was seen more prominent than before (Fig. 2C). Patient was planned for donor patch graft. Thus, we used donor corneal tissue with scleral rim for the graft. The procedure is illustrated and described in Fig. 4A to 4F.

Figure 5 shows 18 months post-operative status where eyeball is tectonically stable with unaided vision of 6/18 correcting to 6/9 with +1 DS /-3.5 Dcyl at 70 degrees.

Discussion

Although globally rare, rhinosporidiosis is endemic in Africa and Asia, especially in India, Sri Lanka, Bangladesh, and Nepal.³ *Rhinosporidium seeberi* is a protistan parasite, found primarily in water sources.^{2,3} It infects the host through transepithelial infection, which requires an injured epithelium; this. Makes trauma a major risk factor for rhinosporidiosis.² Both of our patients reported no known trauma. The infection is most frequently seen in children between eight and ten years of age, with males affected more than females.³⁻⁵ Exposure to polluted water such as bathing in ponds or stagnant water, has been noted as a common factor in these patients.^{1,2,3}

In the study by Alam et al, an initial diagnosis of rhinosporidiosis was considered in only 11.7% of the cases.³ Typical clinical features can facilitate an easier diagnosis, however, similarities with other entities such as pyogenic granuloma, episcleritis, scleritis, or herniated chalazion (when it occur in the tarsal conjunctiva) often delay the correct diagnosis and patient may have to face the financial burden of unnecessary investigations and consultations.³

Alam et al. studied the prevalence of bulbar conjunctival rhinosporidiosis whereas, Shrestha et al. and Mithal et al. showed an increased prevalence of palpebral conjunctival involvement.^{4,5} In the study by Shrestha et al., only 2 of 76 conjunctival rhinosporidiosis cases presented as a patch in the superior bulbar conjunctiva.⁵ In the palpebral conjunctiva, the lesion is typically a friable sessile or pedunculated polyp, whereas the bulbar conjunctiva presents with a strawberry-like sessile lesion.^{3,4,5} All lesions have characteristic small yellowish dots on the surface, representing the sporangia.^{3,4,5} Prior case series have also noted associated scleral thinning and staphyloma formation, as a result of longstanding rhinosporidiosis.^{3,6,7} In a case series of 34 patients by Alam et al., 9(26.4%) had scleral thinning of whom 7 underwent scleral patch grafting.³ Scleral melting occurred in the vicinity of the conjunctival lesion.³ There were no cases of scleral thinning in the study by Shrestha et al. in which most of the lesions were located in the palpebral conjunctiva.⁵

Case 2

was associated with scleral thinning and bulging with normal a retina underneath. The relatively asymptomatic nature of the lesion may result in late presentation, especially when it is located under the eyelid. Our patient did not notice the lesion at first, but as it increased in size, he began to experience foreign body sensations. The mean duration of symptoms in the case series by Alam et al. was 14.8 months.³ Although, the pathologic basis for scleral thinning is unclear, the thinning of the sclera and staphyloma formation may be attributed to enzymatic dissolution of scleral tissues by the pathogen.^{7,8} The retina underneath the scleral thinning may be normal or may be associated with retinal folds, choroidal effusion or retinal detachment.⁸

In endemic countries, the diagnosis of rhinosporidiosis should always be kept in mind. A definite diagnosis is established by histopathology when features of the organism are found, ranging from young trophic forms to sporangia-whether mature, empty, or ruptured.⁴

The first-line therapy for conjunctival rhinosporidiosis is surgical excision with cauterization of the base.^{1,3,9} In our first case, we cauterized the base; in our second case, this was not done due to the presence of scleral thinning. Recurrences may be related to incomplete surgical excision of the lesion at its base. Recurrence of conjunctival rhinosporidiosis has been reported as 2% by Mithal et al. and 14.7% by Alam et al.³ Oral dapsone can be used as additional treatment for ocular rhinosporidiosis postoperatively.⁹ While some studies suggest oral dapsone should only be used in recurring cases or extensive sinonasal involvement, another study suggested the use of oral dapsone in preventing recurrence after excision. Medical therapy has not been proven effective due to unsuccessful organism culture.^{1,4,9} We planned to administer dapsone if recurrence occurred in our second case with scleral thinning. On subsequent follow up, there was no sign of recurrence in either cases, but the patient had marked scleral thinning in 6 weeks postoperatively. This could be due to the loss of support of the tenon's capsule. In our case, we used a corneal graft which offered tectonic stability however, because the cornea is transparent, some hue of the uveal tissue was visible. This may not be cosmetically appealing if a corneal graft is used for staphyloma within the palpebral fissure. In such cases, a scleral patch graft is preferred. Jacob et al., in their case report, also used a corneal patch graft successfully for a superior bulbar staphyloma due to rhinosporidiosis.⁷

Conclusion

The clinical features of rhinosporidiosis are often typical however, the diagnosis is often missed owing to the rarity of the disease. Because the lesion is asymptomatic its early stages routine ocular examination may fail to detect it when it arises in the superior bulbar conjunctiva. Delayed diagnosis may be accompanied by scleral thinning and the risk of perforation. Therefore, a proper clinical approach including eyelid retraction and examination of the fornices is essential, particularly in endemic region and among vulnerable age group with identifiable risk factors.

Declarations

Acknowledgements

Not applicable

Authors contributions

Leena Bajracharya designed, coordinated this research and drafted the manuscript. Kripa Bhattarai prepared figures. Jessica Zarwan obtained photographs. All the authors reviewed and revised the manuscript.

Funding

The authors did not receive any funding grant for this research.

Data availability

No datasets were generated or analysed during the study

Ethics approval declaration

Institutional Review Committee of the Tilganga Institute of Ophthalmology has approved the publication of the case report.

Ethics approval and consent to participate

Not applicable

Consent for publication

Written informed consent for the publication of clinical details was obtained from the parents of both patients. No identifiable clinical images are included in this manuscript.

Competing interests

The authors declare no competing interests.

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Figures

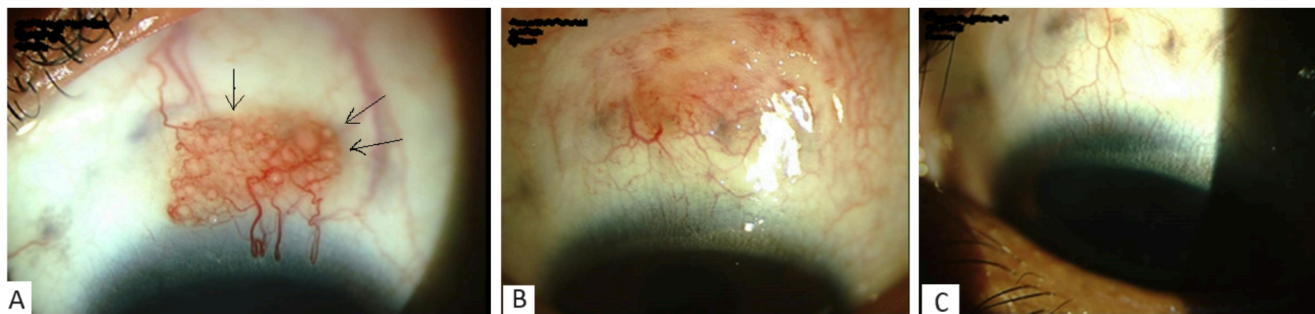


Figure 1

(A) Reddish lesion in the superior bulbar conjunctiva with multiple yellow dots (shown by arrows) and feeder vessels. (B) Complete excision and cautery of the base, day 7 after surgery. (C) Post-operative 3 months showing complete resolution

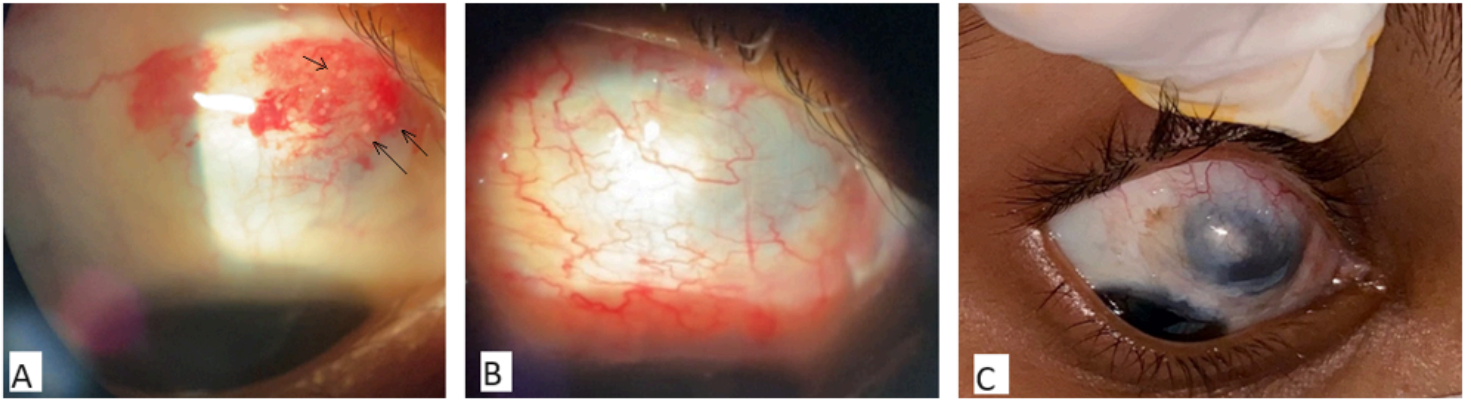


Figure 2

(A) bright red lesion with feeder vessels and multiple yellowish dots (black arrow) at the superior bulbar conjunctiva associated with scleral thinning and bluish hue of underlying uveal tissue. 2(B) Post-operative day-10, after excision of the lesion and conjunctival autograft from inferior bulbar conjunctiva 2(C)Post operative week -10 with increased protrusion of uveal tissue underneath the conjunctival autograft

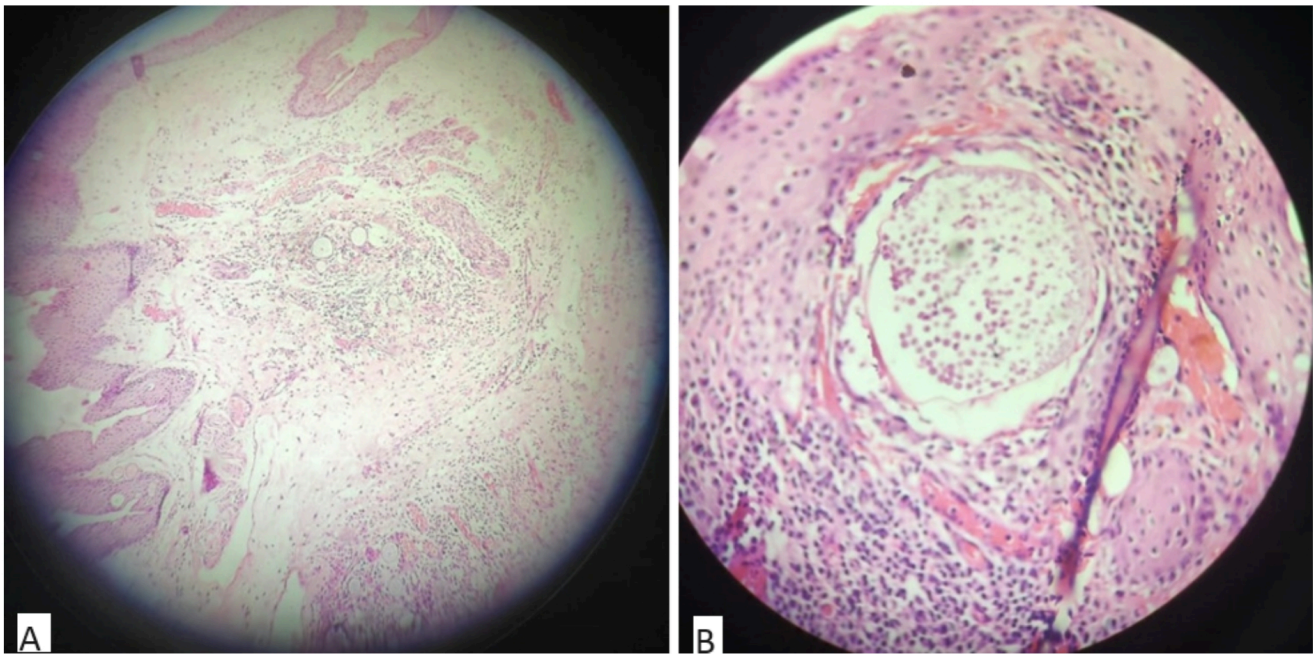


Figure 3

A: conjunctival lining epithelium underlying fibro-collagenous tissue with mild chronic inflammatory cells along with Figure 3B: thick wall sporangia with endospores within it

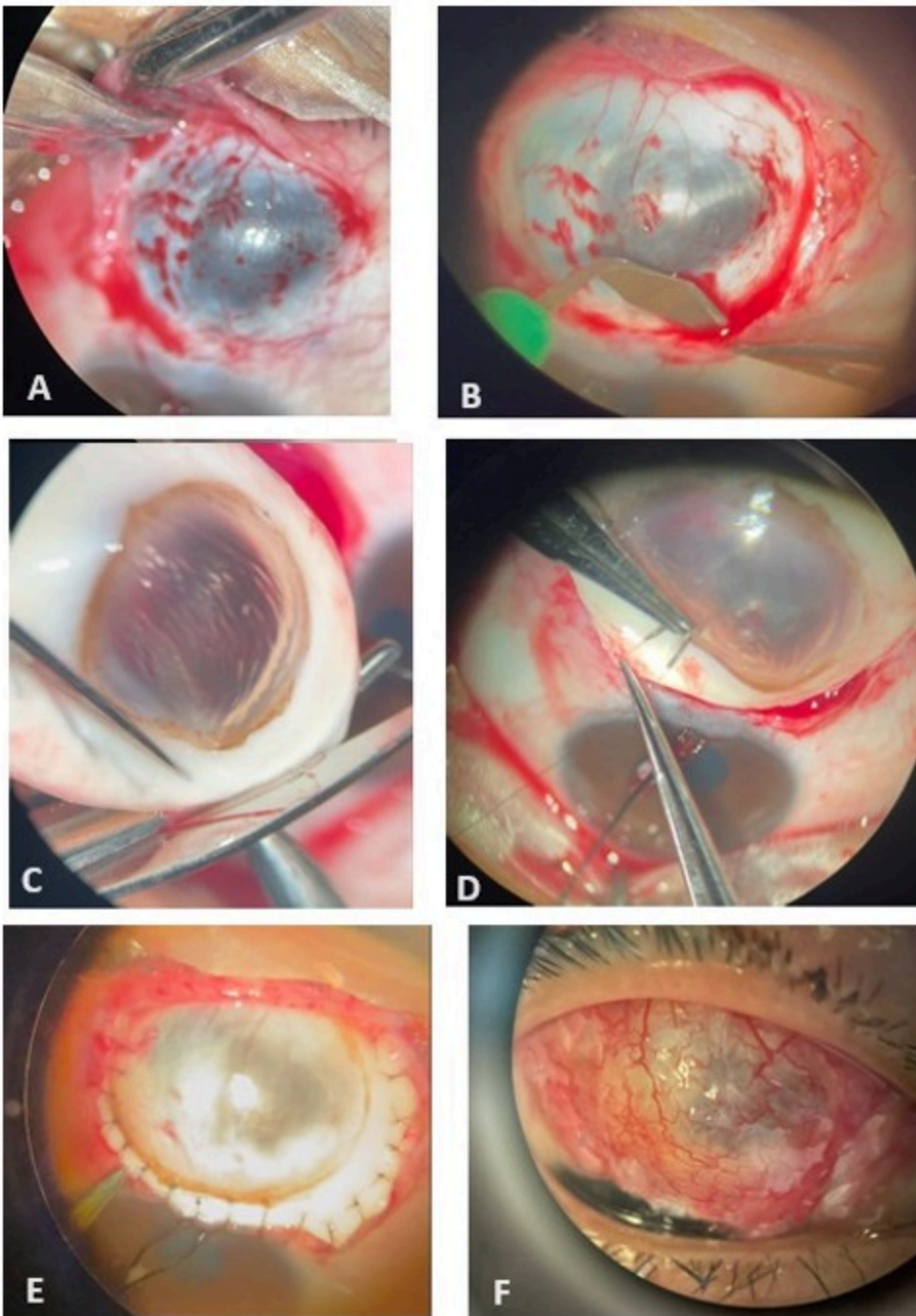


Figure 4

(A to F) Surgical procedure of corneoscleral patch over the scleral thinning. Eye ball was rotated down with the help of silk suture exposing the superior bulbar conjunctiva and steps undertaken as follows:

A: Conjunctiva was retracted from the staphyloma

B: Crescent blade was used to make lamellar flap of sclera tissue circumferentially where sclera resumed its normal thickness.

C: Donor corneo-scleral graft was trimmed to fit the thinned area.

D: The graft was placed over the staphyloma. Pushing the staphyloma back and using 10-0 nylon, the graft was sutured with the lamellar flap created in the normal sclera

E: Conjunctiva was repositioned over the graft.

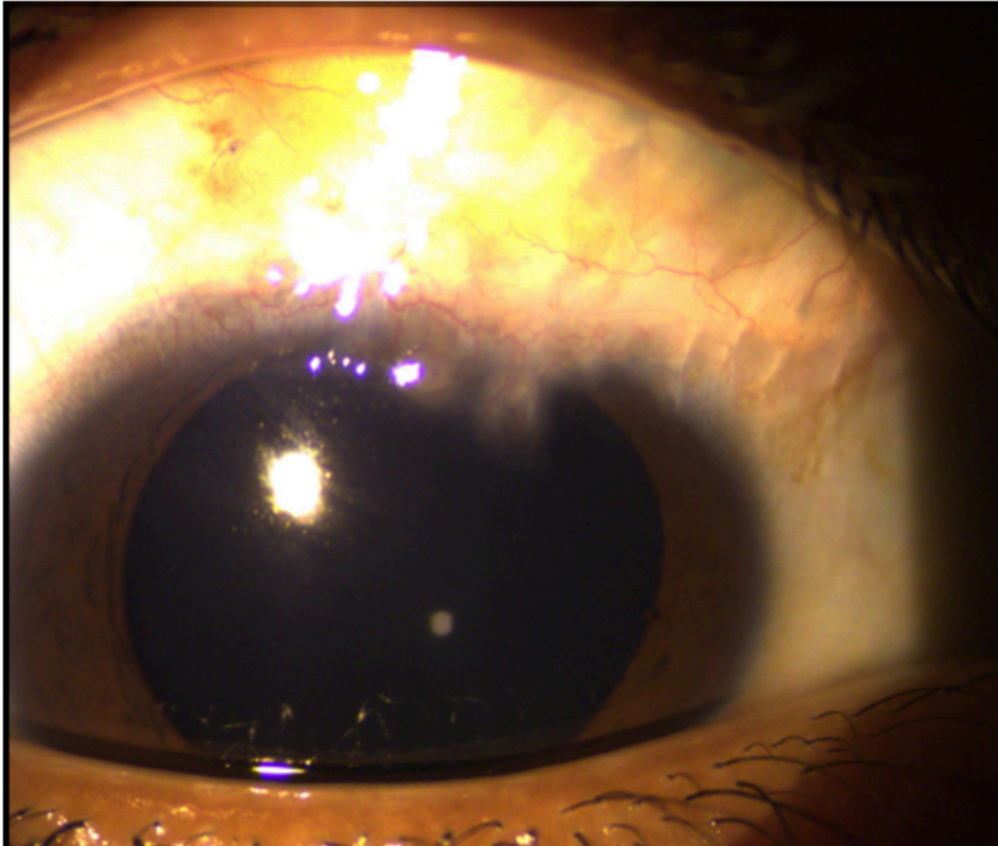


Figure 5

At 18 months post operatively.