

A modified Tenons sling annular graft for advanced peripheral ulcerative keratitis with an hourglass cornea

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Systemic tuberculosis mostly affects the lungs and ocular involvement is rare. The ocular manifestations are varied, and rarely peripheral ulcerative keratitis (PUK) occurs either due to direct invasion of the microorganism or due to immune reaction. We present a case of advanced PUK in a 28-year-old male patient diagnosed with pulmonary TB in the right eye with hourglass cornea. The ulcer was progressing circumferentially with necrosis of corneal stroma and undermined margins similar to Moorens ulcer, and was not responding to anti-tuberculosis treatment and topical steroids. We describe a modified novel surgical technique using tenons sling annular patch graft to manage the progressive advanced PUK with hourglass cornea to optimize visual and structural outcome in our patient.

Key words: PUK, sling annular patch graft, tenons graft

Peripheral ulcerative keratitis (PUK) is plausibly a devastating disease with juxta limbal, crescent-shaped destructive inflammation at the margins of the corneal stroma, which progresses circumferentially around the cornea, giving it an hourglass appearance in the advanced stage.^[1,2] The clinical features include ulcer, stromal necrosis, and thinning, and leads to perforation in advanced cases.^[2,3] Tuberculosis primarily affects Lungs (80%), and ocular involvement is rare, but occasionally, it can occur as a first clinical manifestation.^[4] The prevalence of presumed ocular TB in India is 0.4%–9.8%.^[4] The ocular disease occurs following the direct invasion of organisms through blood flow or following delayed type IV immunologic reaction.^[4-6] The ocular manifestations are widespread, including panophthalmitis, endophthalmitis, and posterior uveitis with choroidal tubercles, interstitial keratitis, phlyctenular keratoconjunctivitis, scleritis, and very rarely PUK.^[4-6] We present a case of bilateral PUK in a 28-year-old male patient with pulmonary TB and describe a modified surgical intervention with a sling tenons patch graft for the advanced stage of the disease to enhance the structural and visual outcome.

Case Report

A 28-year-old male patient presented to us 2 years back with recurrent episodes of redness in both eyes for 6 months. He was diagnosed to have pingueculitis in both eyes and treated

with topical anti-inflammatory eye drops. He presented after 1 year with left eye redness for 1 week. Vision in both eyes was 6/6 unaided. On examination, the right eye was within normal limits; in the left eye, peripheral ulcerative keratitis (PUK) in the nasal cornea two clock hours from 8 o'clock to 10 o'clock with the adjacent elevated conjunctiva, ulcer with undermined margins, and mild yellowish infiltrate in the base were seen. His blood and other investigations were within normal limits, including the Mantoux test, ESR, random blood sugar, RA factor, ANA profile, and X-ray chest PA view. Thus, he was diagnosed clinically with Moorens ulcer and was treated with topical and systemic steroids concerning Foster's stepladder criteria.^[2,7] As keratolysis was progressing, left eye conjunctival resection with amniotic membrane graft was done. The resected conjunctiva showed sclerosing inflammation with plasma cell granuloma and tissue eosinophilia on histopathological examination.

During the follow-up visits, left eye PUK resolved well [Fig. 1]. However, after 1 year, the patient came back with pain and redness in the right eye. The vision in the right eye was 6/60 and in the left eye was 6/6. On slit-lamp examination, the right eye showed active PUK in the temporal side extending from 10 o'clock to 8 o'clock hours with undermined margins and yellowish infiltrate in the base. He was again advised to take a chest X-ray PA view, which showed a cavity in the left side of the chest in the mid zone. His sputum for AFB stain and

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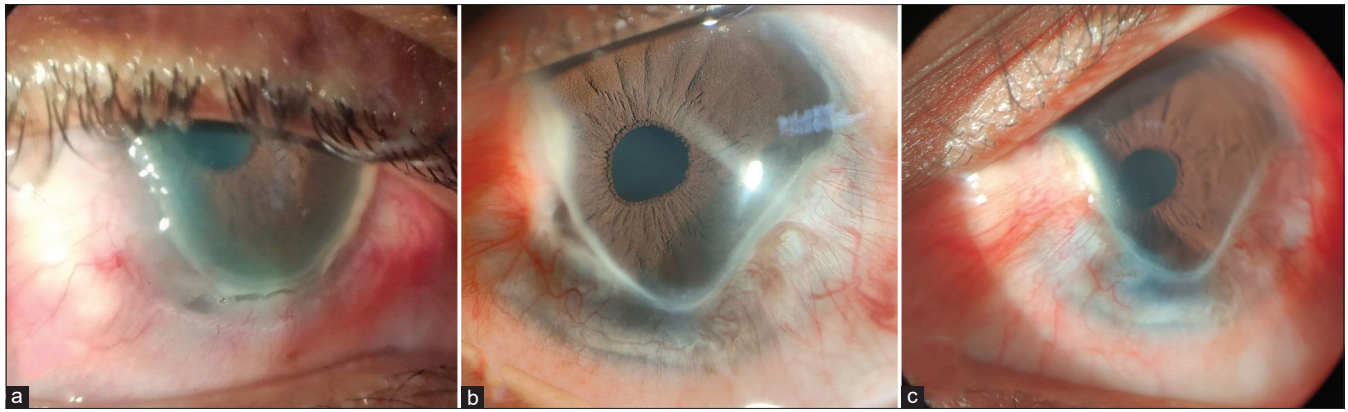


Figure 1: (a) Left eye showing active PUK. (b) Follow-up image showing healing of PUK after conjunctival resection and AMG. (c) Final follow-up image showing stabilized ocular surface

culture was positive for *Mycobacterium tuberculosis* (MBT). He was referred to a pulmonologist and was started on anti-tuberculosis treatment with a multidrug-resistant (MDR) TB regime. Only topical steroids were prescribed in the right eye since systemic steroids were contraindicated.

After three weeks, the patient presented with perforation at 8 o'clock. The anterior chamber appeared formed with iris incarceration at the perforation site — the best-corrected visual acuity in the right eye was 6/18 and the left eye 6/6. As there was no involvement of sclera and we noted the progression of PUK despite initiation of the MDT regime, we made a provisional diagnosis of Moorens ulcer in the right eye and advised conjunctival resection, corneal patch graft, and amniotic membrane graft (AMG). The patient was lost to follow-up and presented after 3 months for the surgery with rapidly progressed PUK with an hourglass cornea, and the vision dropped to 5/60 [Fig. 2a]. We performed modified tenon-sling grafting with an overlay AMG [Fig. 2b]; the postoperative period was uneventful. During the follow-up visits, the PUK in the right eye was stable and vision was 6/24p [Fig. 2c]. In subsequent visits, tenons graft merged with the corneal stroma and the surface was well epithelized in 6 weeks. At 1-year follow-up, he showed a stable ocular surface in the right eye; unfortunately, he was developing cataract, and the vision again dropped to 5/60 at the last follow-up [Fig. 2d].

Surgical Technique

Tenons graft was harvested from the superior bulbar conjunctiva 2 mm beyond the limbus. The surgeon measured the graft size with the sterile caliper from 10 o'clock limbus to 6 o'clock of the ulcer margin. Double the length was taken as the vertical height of the graft and measured horizontal width according to the size of the defect area, and the harvested graft was placed in the saline cup. The recipient bed was prepared by debriding the necrosed stroma, and limbal necrosed edges were cut and removed. Conjunctival resection was done all around the PUK till 4 mm away from the limbus. The peripheral edges of the tenons graft were first sutured to the outermost margins of the PUK at 10 o'clock. Then, the graft was pulled tightly around the ulcer area like an annular sling holding the hourglass cornea [Fig. 2b], and the other end of the graft was sutured to the healthy corneal tissue at 2 o'clock with ten o nylon suture at the superior peripheral margin of the PUK. In this method, the

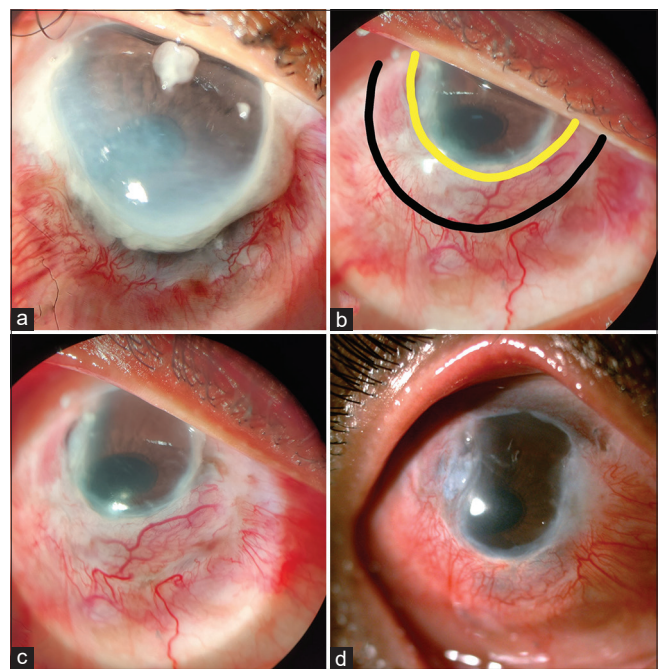


Figure 2: (a) Hourglass cornea in the right eye. (b) Modified sling annular tenons patch graft. (c) Follow-up image showing healing of the ocular surface. (d) Final follow-up image showing well-epithelized surface

tenons were fixed with ten o vicryl sutures in the scleral side of the ocular surface and glue to fix the graft to the base. We aimed to avoid the corneal sutures as the leftover healthy cornea was only 5 mm (horizontal diameter); as such, application of sutures would cause distortion of the corneal contour. The amniotic membrane was used to cover over the tenons and the bare conjunctiva with fibrin glue. The uniqueness in this procedure is the sealing of large area of PUK with tenons alone with the annular sling method, without using sutures over the cornea.

Discussion

The etiologies of PUK are varied. It can be caused by infectious (ocular and systemic) and non-infectious (autoimmune diseases; most commonly, rheumatoid arthritis).^[2,8] The first report of peripheral corneal ulceration was noted by Rafiezadeh

et al.^[9] in 2018 due to autoimmune response against MBT during intensive ATT. Singhal *et al.*^[10] observed bilateral immune stromal keratouveitis in an 11-year-old female with pulmonary TB in 2017. Peripheral corneal melting can occur due to direct invasion of the TB bacillus, as reported by Bayraktutar *et al.*^[6] in a 47-year-old female; he demonstrated TB bacillus in the biopsy specimens of conjunctival nodules. In all these cases, the corneal melt was halted with topical steroids and ATT in contrast to our patient, who did not respond to topical steroids despite starting ATT.^[6,9,10] We provisionally concluded the diagnosis as Moorens ulcer based on the following reasons. 1) There was no TB-associated ocular inflammation such as subconjunctival nodules, scleritis, intraocular inflammations. 2) No response of PUK was noted even after initiation of ATT in our patient. 3) The left eye responded well to conjunctival resection with AMG, although remissions and exacerbations were noted. 4) Blood investigations for autoimmune disorders were normal. Systemic TB was probably an associated finding in our patient. The hallmark of PUK is rapid keratolysis due to the imbalance that exists between collagenases and their tissue inhibitors.^[2,3,8] Moorens ulcer is an idiopathic form of PUK, and perforation is expected in the malignant form of Moorens ulcer in up to 36% in one case series by Young *et al.*^[11]

A perforated corneal ulcer is an emergency, and immediate surgical intervention is needed. Corneal patch grafts are the standard treatment practiced by several corneal surgeons for managing impending cases of large peripheral corneal perforations and descemetocoeles.^[2,3] Most authors have also used conjunctival flaps, corneal patch grafts with the “copy and fix” technique, and scleral grafts to manage peripheral ulcers with impending perforations.^[11-13] Tenons patch grafts also can be used to manage large corneal perforations up to 6 mm, traumatic scleral perforations for plugging corneal fistulas, and leaking trabeculectomy blebs.^[9] In addition, it has a unique property to produce autologous fibroblasts and connective tissue, allowing it to get incorporated into the host’s corneal tissue.^[2,3] Kakizaki *et al.*^[14] noted that the anterior tenons capsule is a dense fibrous tissue comprising an orbital smooth muscle network and regulates the tension of all extraocular muscles. We chose tenons patch graft instead of corneal patch graft for the following reasons. 1) As it is a biological tissue, it gets incorporated easily into the corneal tissue during the healing process. 2) Only minimal sutures were used to anchor the graft (with the annular sling method) to the defect area, and glue was used to fix the rest of the tenons graft to the ulcer bed. 3) It is an autologous tissue and is readily available. Moreover, it offers no immune response; thus, no tissue rejection occurs. 4) No need to depend upon an eye bank for the donor cornea in the recent pandemic situations and a cost-effective procedure. 5) It was an ideal choice to use an autograft because of the systemic disease (TB), which in the future may involve ocular tissues. We adopted this modified technique as most of the cornea was necrosed and the leftover healthy cornea was approximately 5 mm in our patient; the application of corneal grafts and sutures would have distorted the corneal contour and compromised the visual rehabilitation. Moreover, this technique covered the maximum area of the PUK as it had progressed almost Eight clock hours from two’o clock to ten o’ clock circumferentially with an hourglass appearance.

Conclusion

Although our patient presented with the advanced stage of the disease with an hourglass appearance of the cornea, the modified surgical technique of sling tenons graft helped salvage the healthy cornea from distortion of the ocular surface, disease progression, graft rejections, and perforations. Moreover, tenon grafting is a cost-effective procedure compared to corneal patch grafts.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

References

- Mondino BJ. Inflammatory diseases of the peripheral cornea. *Ophthalmology* 1988;95:463-72.
- Sangwan VS, Zafirakis P, Foster CS. Mooren’s ulcer: Current concepts in management. *Indian J Ophthalmol* 1997;45:7-17.
- Sharma N, Singhal D, Maharana PK, Vajpayee RB. Tuck-in tenon patch graft in corneal perforation. *Cornea* 2019;38:951-4.
- Albert DM, Raven ML. Ocular tuberculosis. *Microbiol Spectr* 2016;4:10.1128/microbiolspec.TNMI7-0001-2016.
- Arora T, Sharma N, Shashni A, Titiyal JS. Peripheral ulcerative keratitis associated with chronic malabsorption syndrome and miliary tuberculosis in a child. *Oman J Ophthalmol* 2015;8:205-7.
- Bayraktutar BN, Uçakhan-Gündüz Ö. Ocular tuberculosis with progressive unilateral corneal melting. *Case Rep Ophthalmol* 2015;6:293-7.
- Foster CS. Immunologic disorders of the conjunctiva, cornea and sclera. In: Albert DA, Jakobiec FA, editors. *Principles and Practice of Ophthalmology*. Philadelphia: Saunders; 1994. p. 200-3.
- Riley GP, Harrall RL, Watson PG, Cawston TE, Hazleman BL. Collagenase (MMP-1) and TIMP-1 in destructive corneal disease associated with rheumatoid arthritis. *Eye (Lond)* 1995;9:703-18.
- Rafieezadeh P, Schmack I, Shajari M, Kohonen T. Autoimmune keratitis in mycobacterium tuberculosis. *J Curr Ophthalmol* 2018;30:381-3.
- Singhal D, Maharana PK, Sharma N, Titiyal JS. Immune stromal keratitis: A rare ocular presentation of tuberculosis. *BMJ Case Rep* 2018;2018:bcr2017222571.
- Young RD, Watson PG. Light and electron microscopy of corneal melting syndrome (Mooren’s ulcer). *Br J Ophthalmol* 1982;66:341-56U.
- Deshmukh R, Stevenson LJ, Vajpayee R. Management of corneal perforations: An update. *Indian J Ophthalmol* 2020;68:7-14.
- Jhanji V, Young AL, Mehta JS, Sharma N, Agarwal T, Vajpayee RB. Management of corneal perforation. *Surv Ophthalmol* 2011;56:522-38.
- Kakizaki H, Takahashi Y, Nakano T, Asamoto K, Ikeda H, Ichinose A, *et al.* Anatomy of tenons capsule. *Clin Exp Ophthalmol* 2012;40:611-6.