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Case Report

Defeating Mooren Ulcer: Timely Diagnosis and Strategic Interventions for Swift Recovery

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ABSTRACT

Introduction: Mooren ulcer is a unique and rare manifestation of peripheral ulcerative keratitis of idiopathic origin, triggered by immunological abnormalities, genetic predispositions, and environmental factors. Case Report: We report the successful treatment of a rare case involving a 43-year-old man who presented with a significant worsening of white tissue covering cornea, severe pain, redness, and blurred vision in the left eye. An examination revealed a peripheral ulcer extending centrally with scleral sparing. Ancillary examinations yielded unremarkable findings. The patient was diagnosed with Mooren ulcer and initially treated with topical steroid, which showed no improvement. Therefore, a simultaneous conjunctival resection combined with immunosuppressive therapy was performed. At 1-year follow-up, no pain was reported, vision improved (6/21 to 6/12), and resolved ocular surface inflammation. Discussion: Mooren ulcer is often prone to misdiagnosis due to clinical signs that resemble other differential diagnoses. A meticulous examination is essential to carefully exclude autoimmune and corneal infection. The treatment goals are to arrest the destructive process and facilitate re-epithelization. A stepwise aggressive approach is crucial, starting with topical steroids to control inflammation. However, this single treatment modality may not prevent rapid progression, making conjunctival resection imperative to eliminate the source of collagenase production by cutting off the perilimbal blood vessels' access to ulcer cites. Combining this with an immunosuppressant effectively maintains a stable condition. Conclusion: This case underscores the importance of timely diagnosis and aggressive treatment in preventing the swift progression of Mooren ulcer.

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Introduction

Mooren ulcer stands out as a unique manifestation of peripheral ulcerative keratitis (PUK), characterized by its chronic progressive nature, intense pain and idiopathic origin.[1] It is presumed to be triggered by immunological abnormalities, genetic predispositions, and environmental factors. This rare ulcer is more commonly observed in China, with an incidence of 0.03% and average onset of 48.4 years of age.[2] It often presents unilaterally, with 11-13.3% of cases progressing to corneal perforation. The male-to-female ratio ranges from 1:0.74 to 4.7:1.[2] Diagnosing Mooren ulcer requires ruling out other infectious and systemic causes, which can pose challenges and delays due to its rarity.

Given its aggressive progression and recurrence, management includes various therapeutic modalities, such as local and systemic immunosuppressive agents (both steroid and non-steroid), conjunctival resection, conjunctival autografting, amniotic membrane transplantation, and lamellar or penetrating keratoplasty. Treatment approaches are tailored to individual clinical conditions, response, and disease progression.[3] This case report details a progressive unilateral Mooren ulcer, successfully responding to swift aggressive combinations of therapies.

Case Illustration

A 43-year-old male presented with significant deterioration of the corneal tissue in his left eye (LE), initially manifesting in the limbal region and progressing centrally over the past year. He complained of blurred vision, intermittent redness, and severe pain. Notably, there was no history of trauma, infections, or systemic/autoimmune diseases. Upon examination, his visual acuity without correction was 6/6 right eye (RE) and 6/21 LE, with normal intraocular pressure (RE 18mmHg, LE 10mmHg).

In the LE, a mixed conjunctival injection was observed, along with a grayish peripheral ulcer extending from 6-to-2 o'clock, measuring 9 x 5 mm, with an overhanging edge. Approximately one-third of the stroma was affected, showing superficial vascularization, epithelial defect, and positive staining at the ulcer's border (Figure 1). Anterior chamber examination revealed a deep angle with no detectable cells or flare. A positive fundus reflex was noted, although posterior examination proved challenging. No abnormalities were found in the RE.

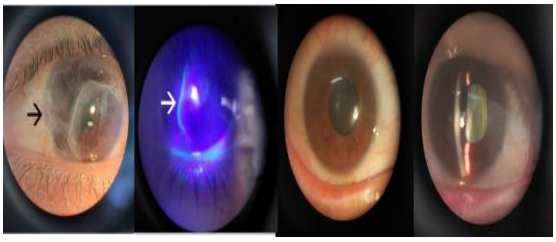

Figure 1. (left) In the LE, a peripheral ulcer extends from 6 to 2 o'clock (black arrow) which is more prominently visible with fluorescence staining (white arrow) (right) No abnormalities were detected in the RE

Laboratory investigations, including tests for infection and immunological markers such as complete blood count, rheumatoid factor, anti-cyclic citrullinated peptide (anti-CCP), antinuclear antibody (ANA), antineutrophil cytoplasmic antibodies (ANCA), complement fixation, circulating immune complexes, liver function, kidney function, and venereal disease research laboratory (VDRL), all returned normal results. A chest X-ray also yielded unremarkable

findings. These results led to a diagnosis of Mooren ulcer in the LE.

Following a week of treatment with prednisolone eye drop (ED), the patient reported reduced redness and pain, but there was no improvement in the appearance of the white tissue and blurred vision persisted.

Consequently, the patient underwent a scheduled conjunctival resection and was given additional therapy, including methotrexate 12.5mg per week, levofloxacin ED 4 times daily, prednisolone acetate ED every 3 hours, and artificial tears every 4 hours. One week after the procedure, there was a slight resolution of grayish lesion with no symptoms of pain (Figure 2).

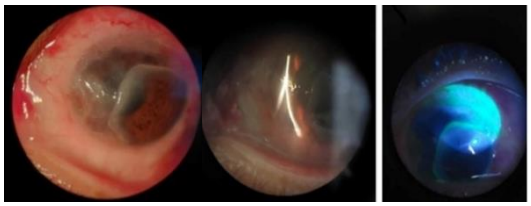

Figure 2. Post-operative condition after conjunctival resection (black arrow) and immunosuppressive therapy with clinical improvement

This comprehensive approach resulted in improvement in the patient's vision, reaching 6/12 within a year of follow-up. The patient's condition remained stable, with no signs of ocular surface inflammation, a clear peripheral surface, resolved superficial vascularization, and an absence of reported pain (Figure 3). Regular monitoring was maintained, with the patient schedule for follow-up visits every three to six months to continually evaluate and manage the condition.

During these visits, detailed examinations were conducted to ensure the absence of any recurrence or new symptoms, thus allowing for early intervention if needed. The patient's adherence to the prescribed treatment regimen and follow-up schedule also played a crucial role in maintaining the stability of the their condition.

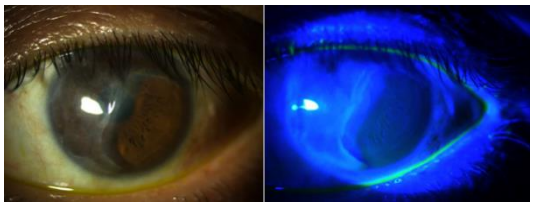

Figure 3. One year after conjunctival resection procedure, there was resolution of the LE's ulcer with no symptoms of pain

Results and Discussion

Mooren Ulcer is a severely painful, chronic, and progressive peripheral corneal ulceration of idiopathic nature.[4] Patients typically presents with unilateral red eyes, severe pain, photophobia, tearing/epiphora, and a distinctive crescent-shaped ulceration that begins at the limbus. This ulceration occurs in the nasal-temporal or interpalpebral region, spreads circumferentially and progresses centrally with an overhanging edge.[5] A unique feature of Mooren ulcer, compared to other PUK is the involvement of the limbus without scleral involvement.[5] Complications may include iritis, glaucoma, cataract, and corneal perforation, which occur in approximately 35-40% of cases.[4] Additional laboratory investigations and radiology are essential to exclude other causes of PUK and confirm a diagnosis of Mooren ulcer.[4,5] In this report, the patient presented with redness, pain and blurred vision in the LE, with the appearance of white tissue progressing from the periphery to the central cornea without scleral involvement. A diagnosis of Mooren ulcer was established after examination revealed the typical characteristic of unilateral Mooren ulcer, with

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laboratory and radiology results showing no abnormalities.

Initial therapy with prednisone drops and artificial tears failed and did not achieve the desired outcomes. Subsequently, conjunctival resection of the LE, combined with a regimen of methotrexate, levofloxacin, prednisone, and artificial tears, effectively halted disease progression, improved vision, and relieved pain, indicating a successful treatment. This approach aligns with the "stepladder" therapy guidelines for Mooren ulcer.[4,6] The initial steps involve topical corticosteroids and cyclosporine, progressing to conjunctival resection in cases that do not respond, often combined with systemic immunosuppression (oral corticosteroids, methotrexate, or cyclophosphamide).[6,7] Recent reports also advocate for anti-tumor necrosis factor agents, such as infliximab or adalimumab to improve outcome with minimal side effects observed.[8] If necessary, alternative surgical interventions such as bandage contact lenses, lamellar keratoplasty/keratectomy, corneal patch graft, or amniotic membrane transplantation can be considered.[4] The primary goal of Mooren ulcer therapy is to mitigate inflammation, support healing through ulcer epithelialization, prevent infection, reduce stromal loss, and repair stromal defects until no fluorescein staining is observed.[4]

Previous research has suggested that the success of conjunctival resection therapy in combination with immunotherapy may indicate an autoimmune etiology involving specific antigens in the corneal stroma associated with HLA-DR17 and HLA-DQ2 genes.[6] Triggered by risk factors, these genes activate cell-mediated and humoral responses, leading to corneal destruction, particularly in the conjunctival area, which is rich in blood vessels, resulting in keratolysis.[8,9] This may explain why ulcers often initiate in the limbus area, the conjunctiva-cornea border, and why immunotherapy and conjunctival resection may help inhibit this process.[9,10] Furthermore, it is essential to recognize that in cases of resistant Mooren ulcer, surgical therapy alone may not achieve optimal healing, as it does not address the underlying immune dysfunction.[4,8]

Conclusion

Timely identification of Mooren ulcer is crucial for initiating prompt and effective interventions to mitigate its progression and prevent sight-threatening complications. Early diagnosis enables clinicians to implement appropriate treatments before the condition worsens, which is essential for preserving visual function. Clinicians should maintain a high index of suspicion for Mooren ulcer, particularly in patients presenting with unilateral ulceration that originates at the limbus while sparing the sclera. A combination of immunotherapy and surgical interventions has demonstrated promising outcomes in managing this challenging condition, underscoring the importance of a multifaceted treatment approach. This integrated strategy not only addresses the local manifestations of the ulcer but also targets the underlying immune mechanisms contributing to its progression.

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