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Traumatic Corneal Perforation as a Complication of Neurotrophic Keratitis in the setting of Atopic Keratoconjunctivitis and Rheumatoid Arthritis

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ABSTRACT

Purpose: To report a case of traumatic corneal perforation secondary to neurotrophic keratitis associated with atopic keratoconjunctivitis and rheumatoid arthritis. **Methods:** Case report. **Results:** We present the case of a 34-year-old woman with untreated rheumatoid arthritis (RA), atopic keratoconjunctivitis (AKC) and neurotrophic keratitis (NK) who developed severe bilateral cor-neal ulceration and unilateral perforation as a result of traumatic eye drop application. The patient responded favorably to a multifaceted treatment approach including the application of corneal fibrin glue and bandage contact lens bilaterally, oral steroids, doxycycline and vitamin C. Addi-tional conservative measures to improve the ocular surface were employed, including frequent application of preservative free artificial tears and warm saltwater soaks. Proper technique for eye drop application was discussed and demonstrated to the patient. Lastly, identifying and treating the patient's underlying RA significantly improved the conjunctival inflammation. **Conclusion:** Corneal esthesiometry should be performed routinely in patients with AKC or RA to identify patients with an increased risk factor for severe NK-related complications. A multifaceted ap-proach to treatment including topical and systemic medication is recommended for patients who present with complications of NK associated with AKC or RA. Identifying and treating underly-ing autoimmune disease such as RA can aid tremendously in the treatment of ocular inflamma-tion and prevention of further complications. Despite corneal perforation, no surgical intervention was required and the patient's visual acuity was preserved.

Keywords: Atopic Keratoconjunctivitis, Corneal Perforation, Neurotrophic Keratitis, Rheumatoid Arthritis.

ABBREVIATIONS

RA: Rheumatoid Arthritis; AKC: Keratoconjunctivitis; NK: Neurotrophic Keratitis.

INTRODUCTION

Atopic Keratoconjunctivitis (AKC) is characterized by the combination of chronic inflammatory conjunctival disease and atopic dermatitis [1]. Patients with AKC typically present with intense ocular and periocular pruritus. Exam findings include conjunctival injection, papillae and periocular eczematous skin changes such as scaling, swelling and hyperpigmentation [1,2]. Common corneal exam findings include

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punctate epithelial keratopathy, peripheral neovascular pannus and peripheral ulcerative keratitis [3,4]. Patients can also develop complications such as keratoconus or cataracts over time due to chronic eye rubbing or steroid use [5]. No correlation has been found between AKC severity and serum IgE levels [6].

Neurotrophic keratitis (NK) is a degenerative corneal disease due to reduced or absent corneal sensation]7]. Aqueous tear production in response to stimulus has been found to be reduced in patients with reduced corneal sensation [8]. NK can be associated with herpetic keratitis, previous ocular surgeries, topical anesthetic misuse, contact lens wear and the use of topical medications containing the preservative benzalkonium chloride [7]. NK has also been observed in patients with systemic diseases such as diabetes, rheumatoid arthritis and Grave's disease [7,9]. Diagnosis of NK is often delayed as patients often have a lack of symptoms. Complications of NK range from mild to severe epitheliopathy, corneal ulceration, stromal melting and corneal perforation [7,8].

Corneal sensation has been found to be reduced in patients with AKC [10]. However, there has yet to be a report of complications of NK secondary to atopic disease. Additionally, a causal relationship between RA and NK has yet to be established. Herein, we describe a unique case of corneal perforation caused by repeated traumatic corneal touch during eye drop self-application in the setting of NK associated with AKC and RA.

CASE REPORT

A 34 year old Hispanic woman presented with severe ocular pain, foreign body sensation and itchiness who was diagnosed with nonspecific keratoconjunctivitis of both eyes. There was no previous medical or ocular history. Frequent application of preservative free artificial tears and cool compresses were recommended. At one week follow-up she had developed bilateral inferior corneal ulcerations. External examination of the upper and lower eyelids showed increased skin pigmentation and a thickened, leathery texture suggesting a diagnosis of atopic dermatitis. Slit-lamp examination showed bilateral severe blepharitis, meibomian gland disease, a moderate papillary conjunctivitis, diffuse conjunctival injection, mild chemosis and a ropy mucus discharge. In the right eye, there was a superior sessile bulbar conjunctival lesion consistent in appearance with a pyogenic granuloma. There was bilateral central corneal ulceration with descemetoceles. (Figure 1A.) The right eye had a shallow anterior chamber indicating perforation. (Figure 1B.) The left eye also had an inferior limbal neovascular pannus adjacent to a crescent-shaped area of ulceration and stromal thinning, consistent with peripheral ulcerative keratitis. (Figure 1C-E.) Seidel testing was negative bilaterally. When asked to demonstrate eye drop application technique, corneal touch and aggressive eye rubbing were observed. On further testing with a wicked cotton-tip applicator showed absent corneal sensation bilaterally, indicating a diagnosis of neurotrophic keratitis. Corneal scraping and cultures were positive for Staphylococcus epidermidis. Schirmer I testing was 2 mm bilaterally. MMP-9 was positive. Our primary diagnosis at this stage was bilateral traumatic corneal ulceration in the setting of neurotrophic keratitis secondary to atopic keratoconjunctivitis. Traumatic eye drop administration was deemed as a contributory cause of the ulcerations and eventual perforation in the right eye.

Initial management consisted of the placement of fibrin glue and bandage contact lenses bilaterally. Broad spectrum antibiotics were started, including fortified vancomycin (25mg/mL) and moxifloxacin 0.5% alternating hourly, in addition to cyclopentolate 2% twice daily. Oral medications included doxycycline hyclate 100mg BID and Vitamin C 1000mg TID. Preservative-free artificial tears and cool compresses were also recommended. Proper eye drop application techniques to avoid corneal touch and the impact of eye rubbing were carefully described to the patient.

On week two from initial presentation, no further corneal thinning was observed and anterior chamber of the right eye had deepened. Topical cyclosporine 0.5% and olopatadine 0.2% were then prescribed. Topical cenegermin (Oxervate™) was discussed and recommended, but was deferred by the patient due to cost. On week three, the patient received a 5-day course of prednisone 60mg once daily, which reduced overall ocular inflammation.

A serum allergen IgE panel was performed, but no specific trigger for atopic disease was identified. Additional skin allergen testing was recommended but not performed. Dupilumab was recommended, but deferred by the patient due to cost.

Due to the patient's complaint of diffuse joint pain and morning stiffness in addition to the development of peripheral ulcerative keratitis in the left eye, rheumatology was consulted for the management of suspected rheumatoid arthritis. The patient was found to have elevated rheumatoid factor and anti-cyclic citrullinated peptides (anti-CCP).

Systemic therapy for rheumatoid arthritis was initiated, including 5mg of prednisone daily and 6 mg of methotrexate weekly, followed by 40mg of adalimumab bi-weekly. A significant reduction in ocular burning and itching were noted.

Over the course of eight months, the corneal stroma thickened and the fibrin glue was extruded bilaterally (Figure 1F-G. &

Figure 2.) The pyogenic granuloma of the right eye resolved spontaneously. The patient underwent lacrimal punctal occlusion to aid in aqueous tear retention and continues to have frequent meibomian gland expression during subsequent follow up visits. The patient's best corrected visual acuity recovered from 20/70 and 20/80 in the right and left eyes, respectively, at initial presentation to 20/20 and 20/30 in the right and left eyes, respectively.

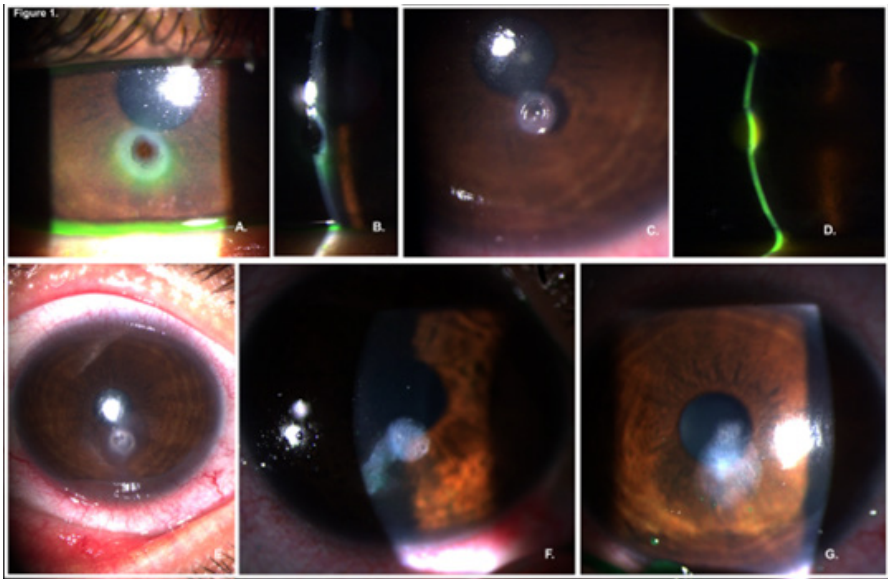

Figure 1. External photographs with slit beam microscopy. A) The right cornea on initial presentation demonstrating traumatic ulceration with descemetocele and B) a shallow anterior chamber indicating perforation. C) The left eye demonstrating corneal ulceration with subtle descemetocele and D) a deep anterior chamber. E) The left eye after fibrin glue application. Inferior neovascular pannus is also present. F) and G) The right and left eyes, respectively, eight months after initial presentation demonstrating central corneal scarring.

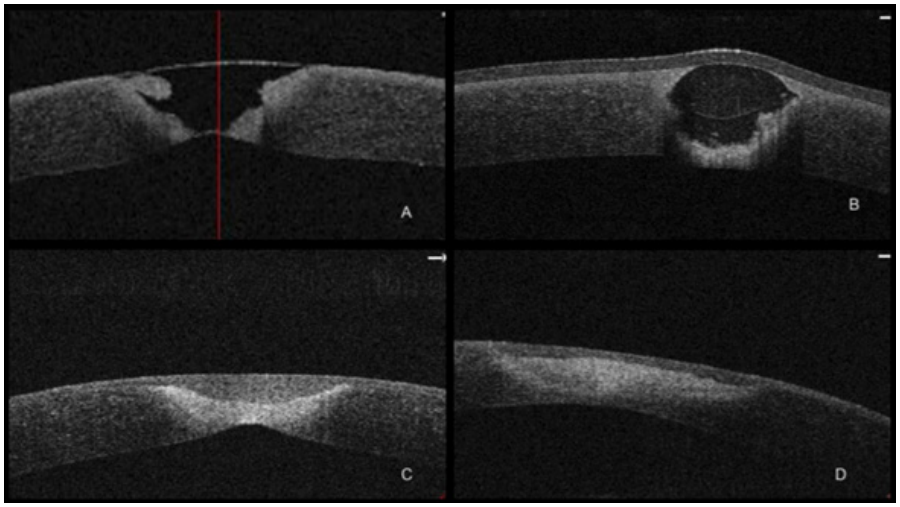

Figure 2. Anterior Segment Optical Coherence Tomography (OCT) imaging of the cornea A) Right corneal ulcer and descemetocele at initial presentation. B) Left corneal ulceration following placement of corneal glue and bandage contact lens. C) and D) Right and left corneal stroma, respectively, following treatment.

DISCUSSION

Hu, et al. described the appearance of the cornea under confocal microscopy in patients with AKC [11]. When compared to the control group, patients with AKC have lower epithelial cell counts, subbasal long nerve fiber counts and nerve branch densities. Morphological changes such as increased nerve fiber tortuosity in patients with AKC are described as well [10]. It is believed that the inflammatory process underlying AKC resulted in the described corneal changes and therefore the reduction of the corneal sensation.

To the best of our knowledge, this is the first reported case of NK associated with AKC or RA that led to corneal perforation secondary to repeated corneal touch with an eye drop applicator tip. We have presented an example demonstrating that frequent application of preservative free artificial tears for patients with undiagnosed NK can lead to severe complications including traumatic corneal ulceration. Patients presenting with AKC or RA should have esthesiometry performed routinely to rule out NK. If NK is identified, the patient can then be counseled about the importance of proper technique of topical eye drop application and more specifically about the dangers of touching the eye directly with the dropper tip.

Of note, our patient had negative serologic testing for IgE. This is consistent with the findings of Tuft et al. who have found that severity of AKC is not correlated with serum IgE levels [6].

There is a higher incidence of rheumatoid arthritis in patients with atopic dermatitis, indicating a possibility of overlapping causative inflammatory processes [12]. The authors believe that in the described case, identification and treatment of aqueous deficient dry eye disease in addition to the underlying RA significantly aided in achieving a positive outcome. It is also important to note that no surgical intervention was required for the treatment of the Descemetocoeles and corneal perforation. Medical treatment targeting the source of the ocular inflammation was sufficient to preserve the patient's vision. Therefore, for patients who develop complications related to NK associated with underlying systemic inflammation a multifaceted non-surgical approach, such as the one described above, may help avoid surgery.

CONCLUSION

Corneal esthesiometry should be performed routinely in patients with AKC or RA to identify patients with an increased risk factor for severe NK-related complications. A multifaceted approach to treatment including topical and systemic medication is recommended for patients who present with complications of NK associated with AKC or RA. Identifying and treating underlying autoimmune disease such as RA can aid tremendously in the treatment of ocular inflammation and prevention of further complications. Despite corneal perforation, no surgical intervention was required and the patient's visual acuity was preserved.

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STATEMENT OF CONFLICT OF INTEREST

The authors have no conflict of interest to disclose

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