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Case Report

Unilateral Painful Red Eye with Corneal Cysts Assessed with Corneal Confocal Microscopy: A Case Mimicking *Acanthamoeba* Keratitis

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Abstract

A 28-year-old woman with a history of trauma to her right eye 2 months prior reported experiencing a stinging sensation and tearing in the morning since the injury occurred and the need to occasionally use an eye patch. Three days before presentation she had been prescribed a therapeutic contact lens (CL) with the diagnosis of a corneal epithelial defect. She described significant pain despite the CL. There was a corneal lesion with haze at the base surrounded by corneal edema. Corneal confocal images revealed hyperreflective cystic lesions that suggested *Acanthamoeba* keratitis (AK). However, the lesion healed within 10 days and the results of cultures taken before the initiation of treatment proved to be negative. The history of trauma and CL wear, the presence of severe pain, corneal findings, and the confocal microscopy detection of cysts led to a suspicion of AK in a differential diagnosis, but the final diagnosis was recurrent epithelial erosion based on the negative culture results, quick response to treatment, and the possibility of similar confocal findings in a healing epithelium. Since AK may cause loss of vision, suspicion should require that samples be obtained for microbiological study and close follow-up of the clinical course until a final diagnosis can be achieved.

Keywords: *Acanthamoeba* keratitis, corneal confocal microscopy, corneal epithelium, recurrent epithelial erosion.

Introduction

Since its description as a dangerous ocular pathogen in 1974, the recognition and incidence of *Acanthamoeba* keratitis (AK) has grown with the widespread use of contact lenses (CL) (1). Trophozoites can adhere to CL, which can act as a vehicle for access to the corneal surface. CL can also cause microtrauma that facilitates invasion by protozoa (2). Although difficult, early diagnosis of AK is crucial. A history of CL use, symptoms of a disproportionally painful eye, findings of punctate epithelial keratopathy, pseudodendrites, epitheliitis with or without radial neuritis. and cysts observed on in vivo corneal confocal microscopy (CCM) are diagnostic clues in the early phase of the disease (3). Although CL use is the main risk factor, trauma and contact with contaminated water have also been described as causes (3, 4). AK is a diagnostic challenge and other forms of keratitis and keratopathies should be part of the differential diagnosis (5).

In cases of recurrent corneal erosion, there are repeated episodes of corneal epithelial defects that may be associated with mechanical trauma, corneal dystrophy, or diabetes mellitus (6).

This case report describes an instance of recurrent epithelial erosion mimicking AK.

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Case Report

A 28-year-old woman was referred to the cornea unit with complaints of pain, photophobia, redness, and blurred vision in the right eye. She had a history of a flower prick trauma to the right eye 2 months earlier. A patient anamnesis indicated no systemic disease. She reported a mild stinging sensation in the morning and the need to occasionally cover her eye with a patch for a few hours. Three days before presentation, a therapeutic CL had been prescribed with the diagnosis of a corneal epithelial defect; however, her symptoms of pain, photophobia, and eye redness had not improved.

An ophthalmologic examination indicated a visual acuity in the right eye of 0.8. There was edema present in the right eyelid and conjunctival hyperemia with ciliary injection. The lesion was located in the paracentral cornea with loose, irregular corneal epithelia, haze at the base, and corneal edema (Fig. 1). Fluorescein pooling and staining were observed in the affected area. The ophthalmic examination of the right fundus and the left eye were normal. In vivo CCM (Heidelberg Retina Tomograph III, Rostock Cornea Module, Heidelberg Engineering GmbH, Heidelberg, Germany) revealed hyper-reflective cysts of 20-25 µm in diameter surrounded by a hyporeflective halo (Fig. 2a). The presence of unexpected fusiform cells with bright cytoplasm in the wing cell layer, which, aside from the shape, resembled epithelial cell shedding, prompted the suspicion of trophozoites (Fig. 2a). CCM also revealed activated keratocytes in a honeycomb pattern, hyper-reflectivity in the basement membrane and the anterior stromal level (Fig. 2 b-d). CCM images of

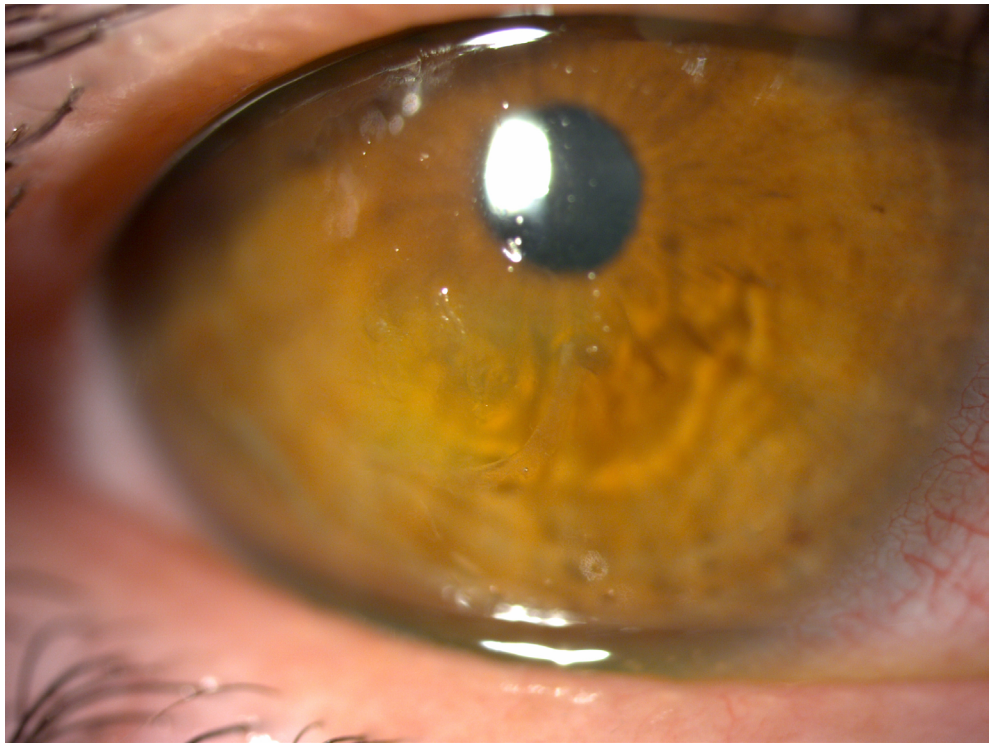

Figure 1. Corneal photo of the right eye.

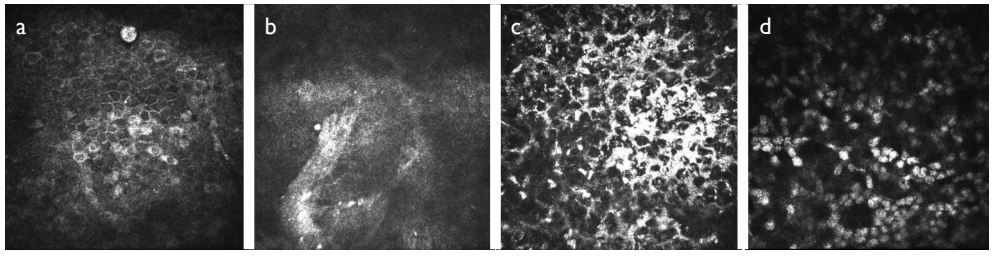

Figure 2. (a) Corneal hyperreflective cyst surrounded by a hyporeflective halo. Fusiform cells with hyperreflective cytosol that were similar to shedding epithelial cells aside from non-hexagonal shape can be seen among the wing cells; **(b)** Hyperreflective tissue resembling the rolled basement membrane seen in basement membrane dystrophy; **(c)** Increased reflectivity in the anterior stromal level; **(d)** Activated anterior stromal keratocytes forming a honeycomb pattern.

the left eye were normal. Mechanical debridement of the loose epithelium was performed with a microsponge. The tissue and the CL of the patient were sent to the laboratory for direct staining and bacteria, fungi, and *Acanthamoeba* cultures. Treatment with systemic analgesia for pain, oral itraconazole 2x100 mg/day, topical moxifloxacin 0.5% 4x1 drop/day, topical cyclopentolate 1% 3x1/day, and neomycin containing ophthalmic pomade at bedtime was initiated. Topical propamidine isethionate 0.1% treatment was planned, however, it was not initiated since the symptoms subsided on the third day, the epithelium had completely healed on the 10th day, and a microbiological study was negative. Only artificial tears were used thereafter (Fig. 3a, 3b). Informed consent was obtained from the patient for all treatment strategies and publication of the case details.

Discussion

The etiological factors for recurrent epithelial erosion are primarily mechanical trauma, corneal dystrophy, or diabetes mellitus (6). Since the left eye was normal and the patient was free of any systemic disease, the causative factor for recurrent epithelial erosion was thought to be the trauma reported by the patient.

The disproportionately severe pain described by the patient and the CCM findings revealing the presence of unexpected cysts in the epithelial layer made us question the diagnosis of a typical case of recurrent epithelial erosion. The absence of a symptomatic response to the use of a therapeutic CL was another point of suspicion about the diagnosis.

CL use, trauma history, basement membrane dystrophy, herpetic keratitis, and diabetic epitheliopathy are predisposing factors for AK and 2 of these were present in this patient (3, 4). A diagnosis of AK in the early phases can be difficult as a result of non-specific and atypical findings (2). In this case, the significant pain and the cysts observed with CCM suggested a diagnosis of AK, which when diagnosed and treated appropriately at a stage restricted to the epithelium, can save the vision of the patient (7).

CCM is a valuable technique to diagnose AK as well as other corneal diseases (8). However, false-positive and negative results must be considered (9). The typical hexagon-shaped, double-walled cysts are usually considered pathognomonic of AK (5). However, there are reports of various shapes and sizes of cysts and trophozoites detected in AK patients confirmed by a positive culture or polymerase chain reaction (PCR) test, which may be attributed to the pathogenicity of different subtypes (10). The cysts detected in our case were only in the epithelial layer, not in the stroma, which can also be seen in the early phases of AK. In recurrent epithelial erosion syndrome, cysts are located in the epithelial layer due to abnormal epithelial turnover during wound healing. They tend to be of various sizes and accumulated in a single area (6, 11). In our patient, the cysts were 20-25 μm in diameter with a uniform pattern. Normal shedding surface epithelial cells are hexagonal and have a bright cytoplasm. The presence of unexpected fusiform cells with bright cytoplasm in the wing cell layer evoked the suspicion of trophozoites. The hyper-reflective tissue at the Bowman layer resembled the basement membrane fold in epithelial basement membrane dystrophy, though the unilaterality of our case excluded this diagnosis (11).

Mechanical debridement of the epithelium was performed and the loose epithelium was removed in order to treat the recurrent corneal erosion, obtain tissue for microbiologic evaluation, and to debulk the infected tissue. Epithelial debridement has been shown to be curative in the early phase of AK (12).

The stain and culture results of our patient were negative, however, false-negative results have been reported at rates as high as 50% in the literature (13). We were not able to study the epithelial biopsies using a complementary PCR test (14). However, the clinical course of the patient was

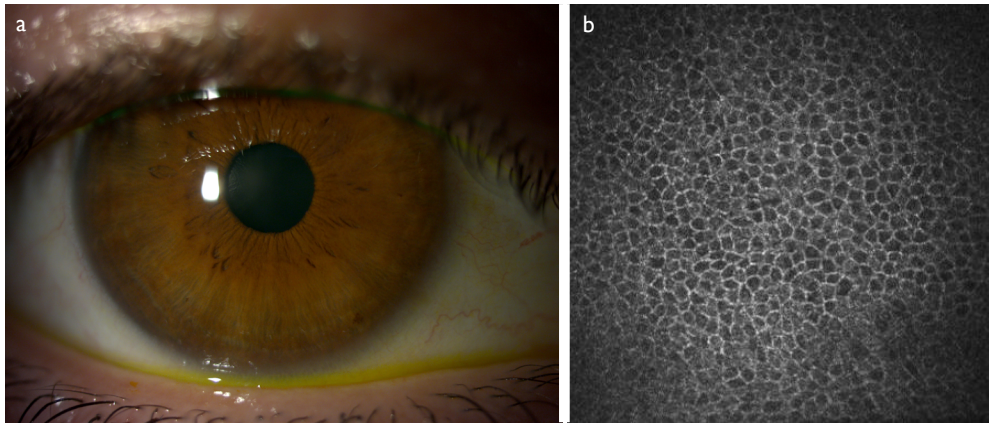

Figure 3. (a) Corneal photo of the patient on the 10th day; **(b)** Corneal confocal microscopy image at the wing cell level demonstrating a normal, healthy appearance.

straightforward without anti-Acanthamoebea treatment, which led us to the conclusion of a diagnosis of recurrent epithelial erosion.

Despite the presence of trauma, CL use, pain, corneal edema, and cysts in confocal images in our case, the final diagnosis was recurrent corneal epithelial erosion syndrome. Symptoms at presentation may resemble AK, and since AK can potentially have devastating sequelae, samples should be obtained for microbiological study and patients should be followed closely to observe the clinical course until a final diagnosis can be achieved.

Disclosures

Informed consent: Written informed consent was obtained from the patient for the publication of the case report and the accompanying images.

Peer-review: Externally peer-reviewed.

Conflict of Interest: None declared.

Authorship Contributions: Involved in design and conduct of the study (ASD); preparation and review of the study (ASD, CG, EUK, NK); data collection (ASD, CG, EUK, NK, OC).

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