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(CASE REPORT)

Infectious Keratitis following acute corneal hydrops in Keratoconus patient: A rare case report

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Abstract

Purpose: To report a rare case of infectious keratitis complicating acute corneal hydrops (ACH) in a patient with Down syndrome and to discuss risk factors, clinical features, and management strategies.

Observation: A 20-year-old female with Down syndrome, atopic dermatitis, and a history of eye rubbing presented with acute corneal hydrops. Anterior segment OCT confirmed the rupture of Descemet's membrane. Initial conservative treatment was initiated. Three days later, the patient presented with severe infectious keratitis. Corneal cultures grew *Staphylococcus aureus*, sensitive to topical tobramycin and ciprofloxacin. Gradual resolution of the infiltrate and the corneal edema was observed, leading to formation of a central corneal opacity after one month of treatment.

Conclusion: Infectious keratitis is an uncommon but rapidly progressive complication of ACH. The reported risk factors included atopic dermatitis, eye rubbing, the use of topical corticosteroids and contact lens wear. Early detection through careful clinical assessment and imaging is critical to initiate antibiotic treatment. Despite infection control, visual prognosis remains poor due to residual corneal scarring and neovascularization. Prophylactic antibiotics may be considered in high-risk cases to prevent superinfection.

Keywords: Keratoconus; Acute Corneal Hydrops; Infectious Keratitis; Atopic Dermatitis; Prophylactic Antibiotics

1. Introduction

Acute corneal hydrops (ACH) is a complication of ectatic corneal disorders including advanced keratoconus. It is characterized by stromal corneal swelling secondary to aqueous humor leakage through a rupture of Descemet's membrane. It is a rare sight-threatening condition, with an incidence varying between 2.4% and 3% in keratoconus patients [1,2].

The natural course of the disease may lead to several complications - acute, such as perforation, and chronic, including neovascularization, intrastromal clefts or pseudocysts, and corneal opacity [3]. Exceptionally, an episode of infectious keratitis may occur. Only a few cases have been reported in the literature, with an incidence less than 2% [4,5,6].

We present a case of infectious keratitis complicating an acute hydrops in a patient with Down syndrome. Through this observation, we aim to illustrate the clinical aspects, warning signs, risk factors and management of this unusual complication.

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2. Case report

We report the case of a 20-year-old female patient with Down syndrome, a past medical history of atopic dermatitis and eye rubbing, who presented to the emergency department with a three-day history of a painful red right eye, tearing and photophobia.

The right eye visual acuity was severely reduced to hand motion perception. Slit-lamp biomicroscopy examination showed a clear mucoid discharge, multiple eyelid papillae, a central and paracentral stromal corneal edema with subepithelial bullae (Figure 1A, B). The rest of the cornea was ectatic. The fluorescein test was negative. There were no signs of infection or anterior chamber inflammation.

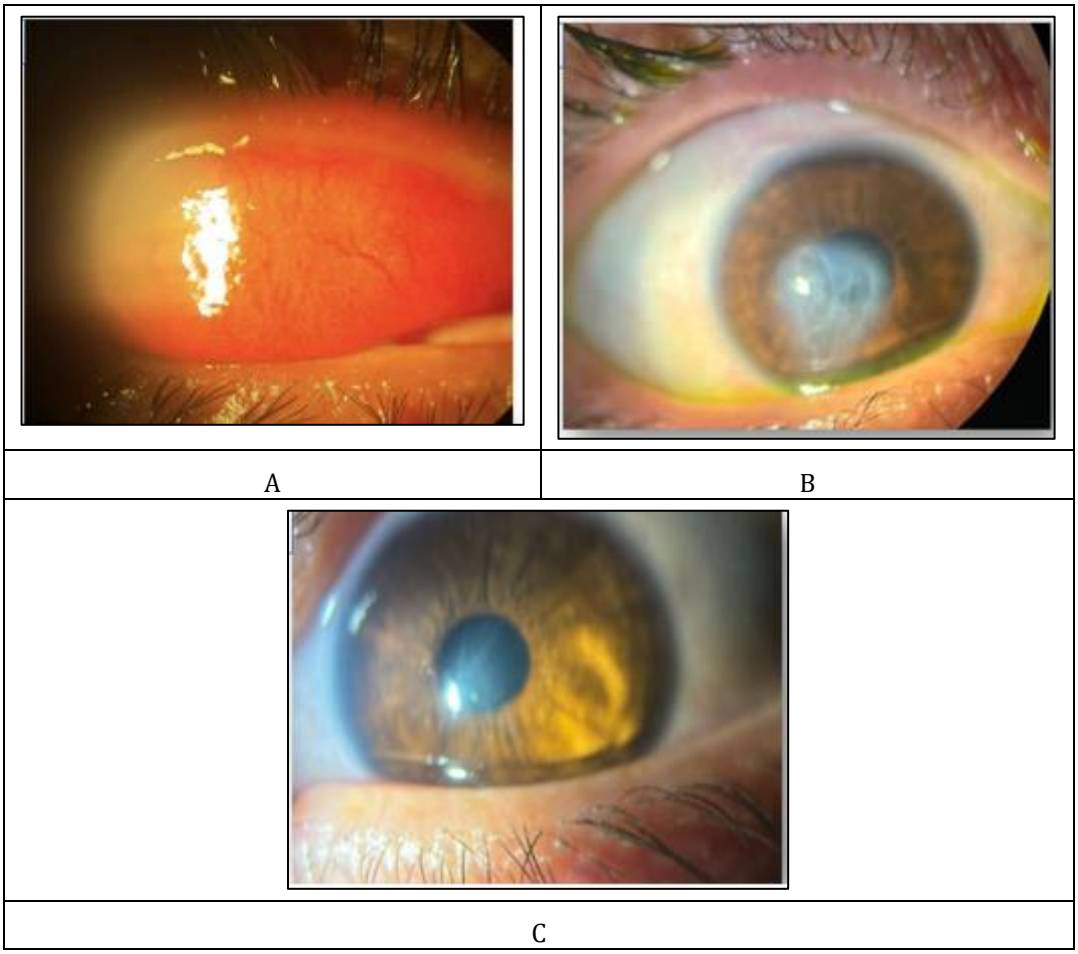

Figure 1 Slit lamp examination of the right eye showing multiple eyelid papillae (A), a central and paracentral corneal stromal edema (B) and multiple corneal scarrings in the fellow eye (C)

Examination of the fellow eye showed signs of advanced keratoconus stage: multiple eyelid papillae, an ectatic cornea with a positive Munson's sign, and multiple linear corneal scars (Figure 1C).

General examination revealed features of atopic dermatitis including erythema, xerosis and lichenification of the face, eyelids and hands (Figure 2A, B).

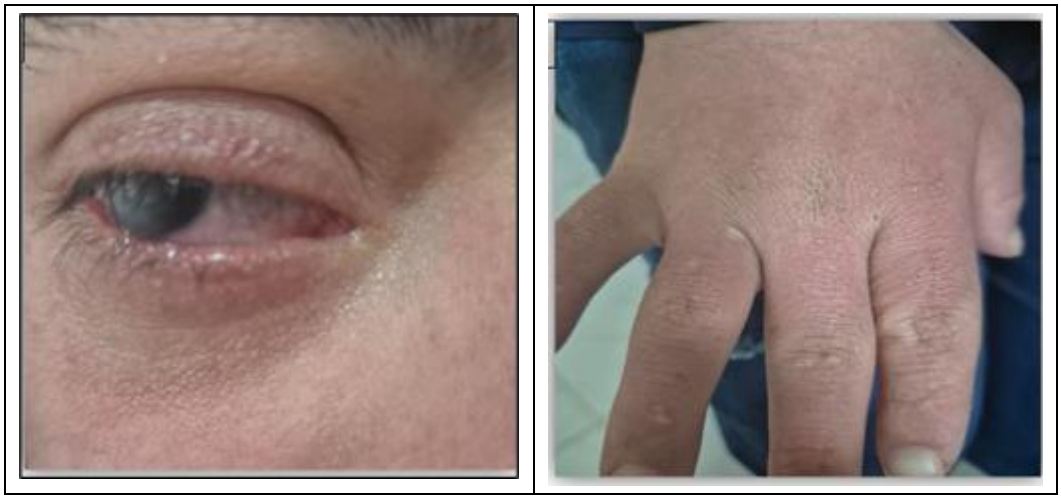

Figure 2 Signs of atopic dermatitis including erythema and xerosis of the eyelid (A) and hands (B)

The evaluation of the corneal structure by anterior segment OCT showed subepithelial bullae, stromal swelling, intra-stromal hypo-reflective cystic spaces and a rupture of Descemet's membrane (Figure 3A, B). These signs were consistent with the diagnosis of an acute hydrops. The contralateral eye AS-OCT image showed significant corneal steepening and thinning to 270 µm.

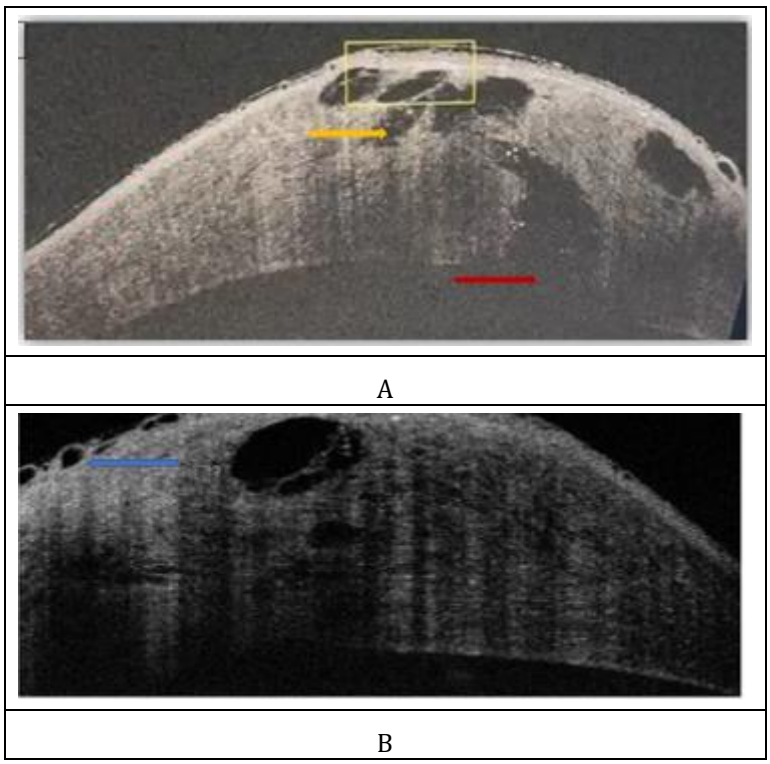

Figure 3 A, B Anterior segment OCT of the right eye showing stromal swelling, subepithelial bullae (blue arrow), intra-stromal hypo-reflective cystic spaces (yellow arrow), and a rupture of Descemet's membrane (red arrow)

Conservative medical treatment of ACH was initiated, including lubricants and topical dexamethasone 0.1%. Antiglaucoma drugs were also prescribed in order to reduce the fluid ingress into the corneal stroma by decreasing the intraocular pressure.

Three days later, the patient returned with increasing ocular pain. Examination revealed purulent discharge, a central corneal ulcer with an infiltrate overlying the area of edema, and a hypopyon (Figure 4). Anterior segment OCT showed hyperreflective corneal infiltrates along with persistent corneal edema, and Keratic precipitates (Figure 5).

Figure 4 Corneal infiltrates overlying the area of stromal edema (A), fluorescein test reveling a corneal ulcer (B)

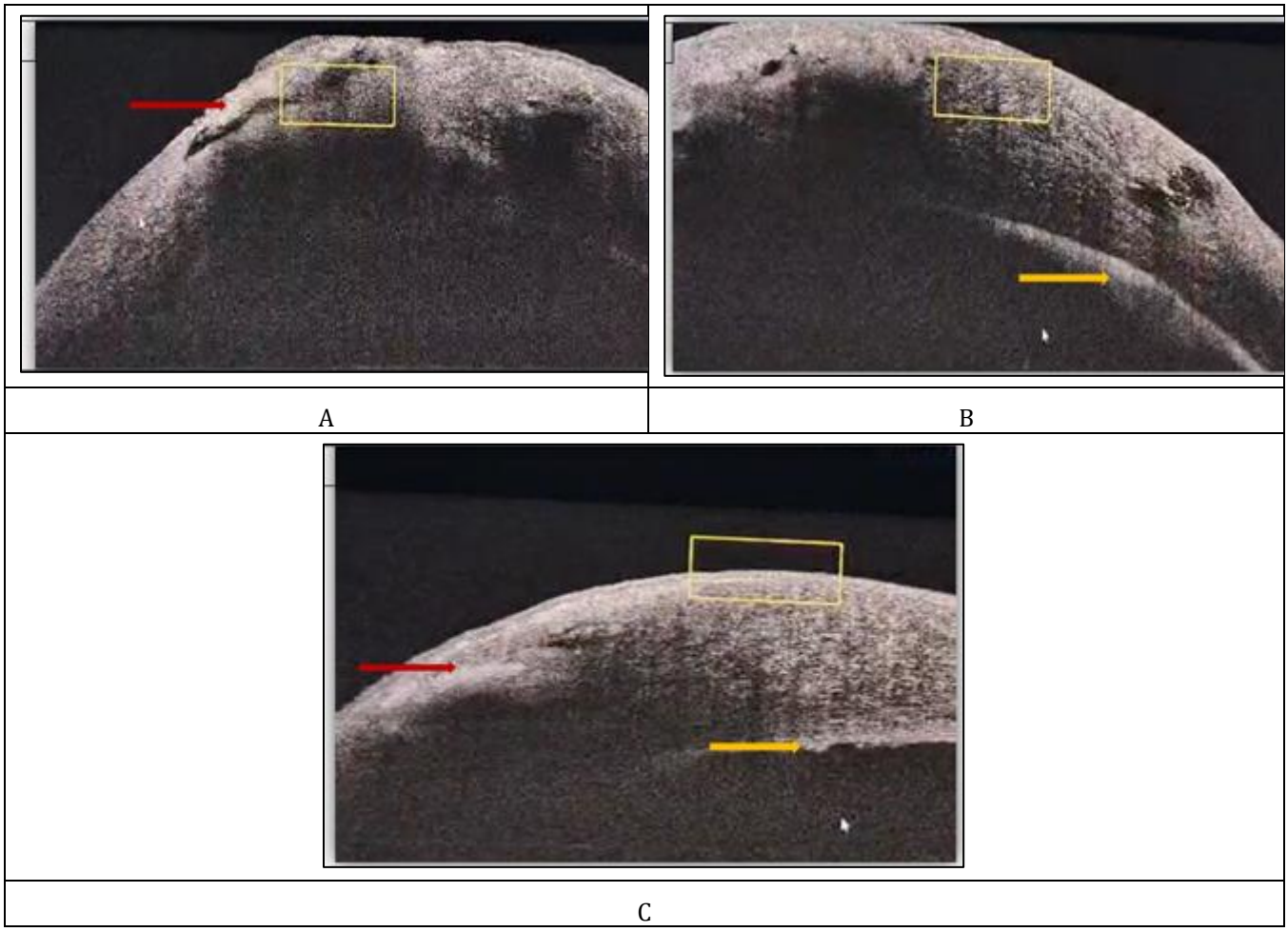

Figure 5 Anterior segment OCT revealing hyper-reflective corneal infiltrates (red arrows) with Keratic precipitates (yellow arrow)

After hospital admission, corneal scrapings were performed, and an empiric antibiotic therapy based on topical tobramycin 0.3% and ciprofloxacin 0.3% hourly was initiated. Cultures grew Staphylococcus aureus, sensitive to the antibiotics previously prescribed. Oral ciprofloxacin 500 mg was also administered twice daily. We observed a gradual

resolution of the infiltrate, healing of the corneal ulcer, regression of corneal edema, and the formation of a central corneal opacity after one month of treatment (Figure 6). The patient is awaiting a keratoplasty.

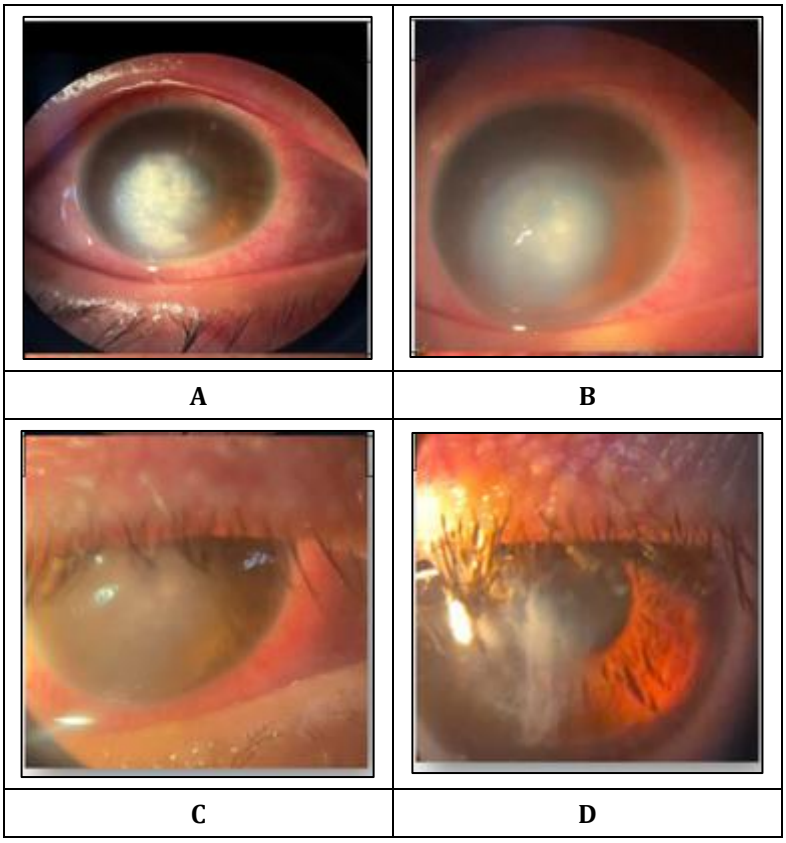

Figure 6 Panoramic images showing the evolution of the corneal keratitis: initial clinical presentation (A), clinical aspect at day 5 of treatment (B), further improvement at day 10 (C), and ulcer healing with the development of a stromal linear scar one month after admission (D)

3. Discussion

Infectious keratitis is a rare complication of acute corneal hydrops. Only a few cases have been reported in the literature [4,5,6,7]. The estimated incidence is 2% among patients with ACH. According to Meyer et al. [4], the epithelial bullae and the corneal edema formed during hydrops may lead to corneal ulceration, thereby increasing the risk of secondary infection.

Other risk factors have been described, including atopic dermatitis which alters the microbiota and favors colonization by antibiotic-resistant bacteria; eye rubbing which helps spreading of microorganisms from the lids or hands to the ocular surface and promotes rupture of epithelial bullae; the use of topical corticosteroid, and contact lens wear [4,5].

In the current case, untreated atopic dermatitis and eye rubbing were likely the risk factors of superinfection. Similarly, Koji et al. [5] reported three cases in which atopic dermatitis was the common risk factor.

The diagnosis could be challenging: corneal edema due to hydrops may mask the early signs of superinfection [5]. The worsening of symptoms should raise suspicion. OCT imaging can help in diagnosis by detecting hyperreflective corneal infiltrates.

Infection often progresses rapidly as the cornea is particularly vulnerable due to structural disruption during hydrops. In the present case, only three days elapsed between the onset of hydrops and the appearance of infectious keratitis. The three cases reported by Koji et al. showed active corneal infiltration at the initial consultation, as did the case reported earlier by Jhanji et al. [6].

While infection control is usually achieved with targeted antibiotic therapy, the visual prognosis remains poor due to residual corneal opacity and neovascularization that limits the success of subsequent corneal transplantation. Therefore, it is crucial to monitor this complication, especially when the previously described risk factors are present. Meyer et al. recommended the use of prophylactic topical antibiotics in the early course of ACH to prevent superinfection, particularly if an epithelial defect or large bullae is present.

The reduced number of affected patients reported in the literature limits our current understanding of its characteristics and management. Further investigations and analyses based on larger case series are needed.

4. Conclusion

Infectious keratitis occurring in the course of acute corneal hydrops is a rare but rapidly progressive and a potentially severe complication. Careful clinical assessment and monitoring is required in patients with ACH in order to detect and treat infection at the slightest suggestive sign.

Compliance with ethical standards

Acknowledgments

The authors have no acknowledgements to declare.

Disclosure of conflict of interest

No conflict of interest to be disclosed

Statement of informed consent

Informed consent was obtained from all individual participants included in the study.

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