Journal of Acute Disease

Case Report

doi: 10.4103/2221-6189.307392 jadweb.org

Infective crystalline keratopathy due to *Citrobacter koseri*

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ABSTRACT

Rationale: Infectious keratopathy is an ocular emergency with the potential to cause irreversible blindness.

Patient's concerns: A 63-year-old diabetic man presented with a 3-day history of painful red right eye. He had a history of multiple ocular surgeries in the affected eye without recent ocular trauma.

Diagnosis: Infective crystalline keratopathy secondary to *Citrobacter koseri*.

Interventions: Topical corticosteroids were discontinued, and dual topical antibiotic therapy of moxifloxacin 5% and gentamycin 0.3% were applied.

Outcomes: Twelve hours after the start of treatment, the crystalline nature of the infiltrate disappeared, with enlargement of the epithelial defect. The antibiotic regime was continued and the lesion healed within a week of presentation with residual scarring.

Lessons: Infective crystalline keratopathy classically presents with intrastromal branching fern-like opacities and minimal anterior segment inflammation in an immunosuppressed eye. The mainstay of management is corticosteroid discontinuation and the administration of empirical antibiotics until the results of the culture and sensitivity are available. In Citrobacter-related cases, treatment may result in a suppurative appearance before healing of the lesion.

KEYWORDS: Infectious crystalline keratopathy; *Citrobacter koseri*; Gram-negative bacteria

1. Introduction

Infectious crystalline keratopathy (ICK) is infective keratitis which classically presents with intra-stromal needle-like branching opacities and lack of inflammatory signs in the anterior segment[1,2]. It is usually seen in an immunosuppressed eye, such as post-penetrating keratoplasty, glaucoma filtering surgeries, and other ocular diseases requiring prolonged corticosteroid therapy[1,3-5]. Gram-positive cocci are the most common aetiology, since other organisms have rarely been reported[1].

Citrobacter koseri is an anaerobic Gram-negative bacillus in the family Enterobacteriaceae. As part of the normal intestinal flora, it is commonly found in soil, water, and sewage[6]. The most common site of human infection is the urinary tract, followed by respiratory tract[6,7]. To the best of our knowledge, only one case of ICK caused by *Citrobacter koseri* has been reported[8]. We report a case of ICK caused by *Citrobacter koseri* and discuss the pathogenesis, clinical features, and management of this condition.

2. Case report

Informed consent was obtained from the patient. A 63-year-old man with underlying diabetes presented with a 3-day history of a painful, red right eye. He was unable to recall any recent ocular trauma. Right extracapsular cataract extraction with posterior chamber intraocular lens implantation was done 26 years ago. However, the posterior chamber intraocular lens was dislocated 4 years ago, requiring the explantation of the lens. The situation was more complicated due to secondary glaucoma which required multiple glaucoma filtering surgeries; the last surgery was done three years ago. Since then, he had been on topical corticosteroid eye drops.

The visual acuity of the affected eye was limited to finger-

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How to cite this article: Thiam-Hou L, Tai ELM, Khairy SST, Mohtar I. Infective crystalline keratopathy due to *Citrobacter koseri*. J Acute Dis 2021; 10(1): 36-38.

Article history: Received 30 March 2020; Revision 19 November 2020; Accepted 23 November 2020; Available online 25 January 2021

counting at close to face distance. His premorbid vision a month before the onset was already poor (finger-counting at 1 meter), due to corneal decompensation secondary to multiple ocular surgeries and glaucoma. The right eye examination revealed eyelid swelling, injected conjunctiva, an oedematous and hazy cornea, and multiple punctate epithelial erosions. A stromal infiltrate in a branching fern-like pattern with an overlying epithelial defect was seen at the mid-peripheral cornea (Figure 1). A glaucoma drainage device tube was visualized in the anterior chamber, with no endothelial touch. There was no hypopyon. The cornea oedema precluded examination of the posterior segment, but the B scan was normal. The left eye examination was unremarkable.

A diagnostic corneal scraping for microscopy revealed Gram-negative bacilli. The topical corticosteroids were withheld, and topical antibiotics (G. moxifloxacin 5% and G. gentamycin 0.3%) initiated were administered. The crystalline nature of the infiltrate disappeared 12 h after initiation of the therapy, with enlargement of the overlying epithelial defect and blurred appearance of edges of the lesion (Figure 2). The culture from the corneal scraping grew *Citrobacter koseri*, sensitive to the antibiotics given. At the end of a week, the lesion had healed with faint scarring and the patient was discharged. Penetrating keratoplasty for corneal decompensation was performed 4 months later. However, the vision post-operation showed no improvement, due to underlying glaucomatous optic neuropathy.

3. Discussion

ICK was first described by Gorovoy *et al.* in 1983 in a post-penetrating keratoplasty patient[1]. It occurs as a secondary disease in immunosuppressed corneas, most commonly in patients on prolonged topical corticosteroids or those who have undergone surgery like penetrating keratoplasty, trabeculectomy, or laser in situ keratomileusis[1,3,5]. Other causes such as topical anaesthetic abuse and diabetes have also been associated with an increased risk of ICK[1,4,9].

Most ICK is caused by streptococci[1-3]. *Citrobacter koseri*, previously known as Citrobacter diversus, is a rare cause of eye infections. It has uncommonly been reported in association with microbial keratitis and endophthalmitis[6,7]. To the best of our knowledge, only one case of ICK secondary to *Citrobacter koseri* has been documented, which resulted in perforation[9].

ICK is believed to occur due to impaired ocular surface defenses, thus allowing bacteria to enter the stroma *via* a break in Bowman membrane. The crystalline pattern has been attributed to the

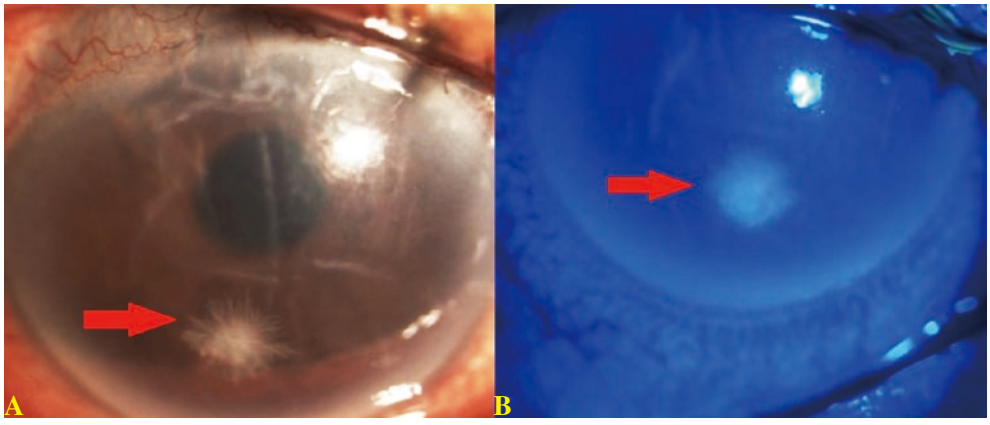

Figure 1. Photographs of the anterior segment of the right eye upon presentation. A: A branching snowflake pattern of stromal infiltrate in the mid-peripheral cornea (red arrow); B: Fluorescein staining demonstrates the epithelial defect overlying the stromal infiltrate (red arrow).

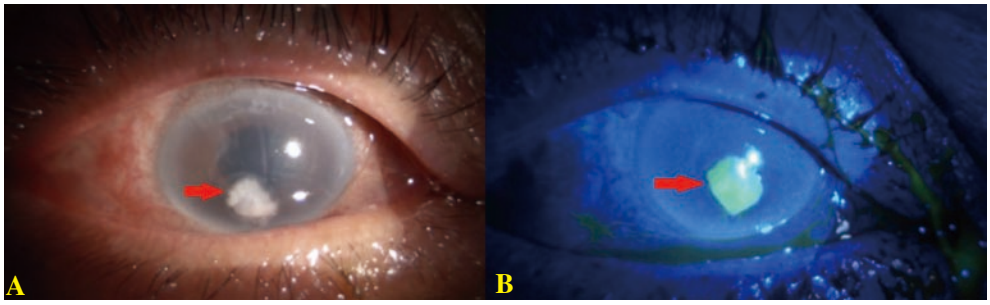

Figure 2. Photographs of the anterior segment of the right eye after 12 hours of treatment. A: Loss of the characteristic snowflake pattern of infectious crystalline keratopathy, with the confluence of edges of the lesion (red arrow). B: Fluorescein staining shows an enlargement of the overlying epithelial defect (red arrow).

deposition of biofilm from insidious growth of intrastromal bacteria or precipitation of immune complexes as part of the host cornea's defense[4,10]. In our case, the patient's risk factors were diabetes and prolonged topical corticosteroid use.

ICK may be asymptomatic or present with similar symptoms as microbial keratitis, such as blurred vision, photophobia, and pain. Ocular inflammation in ICK is less marked than in typical microbial keratitis because of the indolent nature of the common causative organisms and the underlying corneal immunocompromise[1,2]. The treatment of ICK involves early and aggressive topical antibiotics, with the cessation of topical corticosteroids[1,4,5]. Antimicrobial therapy is empirical while awaiting the results of culture and sensitivity[2]. Double antimicrobial therapy may be required as ICK can occur in the presence of mixed microbial keratitis[3]. Other adjunctive therapies are Nd: YAG laser, intrastromal antibiotic injection, and corneal biopsy to disrupt the microbial biofilm[1-3]. In resistant cases, therapeutic penetrating keratoplasty or amniotic membrane transplantation should be performed[3].

In the only previously reported case of Citrobacter-related ICK, the characteristic crystalline features of the keratopathy disappeared 24 h after discontinuation of corticosteroids and initiation of antibiotics[8]. We observed the same phenomenon in our patient, in which the lesion appeared more poorly defined and 'wet' during the initial phase of treatment. Although this might mislead clinicians to believe that the condition is worsening, this suppurative appearance is believed to reflect less production of biofilm, thus allowing better antibiotic penetration[1]. Clinicians should persevere with the empirical antibiotics until the results of the culture and sensitivity are available.

A particular clinical challenge is partially treated ICK, which may mimic a fungal ulcer. In these patients, a thorough history involving risk factors such as corticosteroid use is essential. As corticosteroids are a risk factor for both ICK and fungal infection, empirical treatment with antifungals may be indicated when the clinical picture is indistinguishable. Unfortunately, the final visual outcome may be limited, as in our case, due to concomitant ocular pathology.

Conflict of interest statement

The authors report no conflict of interest.

Authors' contributions

Writing, revision and figures: L.T.H., E.L.M.T.; Review: L.T.H., E.L.M.T., S.S.T.K., I.M.

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