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Case report of *Stenotrophomonas maltophilia* in corneal ulcer: why is it difficult to treat?

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ABSTRACT

Introduction: *Stenotrophomonas maltophilia* is an opportunistic bacterium and has rarely been reported as a cause of infection in the cornea. Ocular infection due to *Stenotrophomonas maltophilia* is an opportunistic infection followed by instability of the ocular surface. Casalita et al. (2020), in their research at Cipto Mangunkusumo Hospital, Jakarta, Indonesia found two cases of infected corneal ulcers due to *Stenotrophomonas maltophilia*.

Case description: A 57-year-old man came to the eye clinic with pain in the left eye and blurred vision. The patient was diagnosed with severe corneal ulcer and regularly came for the treatment. The patient felt that his eye suddenly got worse with visual acuity only to his hand movement. Corneal ulcer swab procedure is performed when the patient firstly visit the clinic. Samples are sent to the microbiology laboratory for culture examination. Culture results obtained were coagulase negative *Staphylococcus* and *Stenotrophomonas maltophilia*.

Conclusion: *Stenotrophomonas maltophilia* has several virulence factors, including resistance to various antibiotics, making it difficult to treat.

Keywords: Cornea ulcer, Corneal swab, Resistance, *Stenotrophomonas maltophilia*, Virulence factors.

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INTRODUCTION

Eyes are one of the most important senses in humans. If there is disease in the eye, it can be at risk of impaired vision or blindness. Based on the World Health Organization (WHO), 5.1% of blindness is caused by diseases of the cornea. One of the eye diseases (especially the cornea) is a corneal ulcer. Corneal ulcers can be caused by both non-infectious and infectious (bacteria, fungi, viruses and protozoa). Data from the Asia Cornea Society (multicentre study) in 2014-2015 found corneal ulcers caused by bacteria amounting to 38% of all occurrences of corneal ulcers. Casalita et al. (2020), in their research at Cipto Mangunkusumo Hospital, Jakarta, Indonesia found two cases of infected corneal ulcers due to *Stenotrophomonas maltophilia*.¹⁻⁵

Stenotrophomonas maltophilia is a gram-negative bacterium that is often found in the environment (water, soil, plants), animals (especially aquatic animals), and healthcare settings. *Stenotrophomonas maltophilia* is an opportunistic bacterium

that causes nosocomial infections and is an uncommon cause of infection in the cornea. *Stenotrophomonas maltophilia* is difficult to treat because of its resistance to several antibiotics.⁶⁻¹⁰ Reports of *Stenotrophomonas maltophilia* causing eye infections (especially corneal ulcers) in Indonesia are still very rare and this case is not research so there has been no previous preliminary study. Therefore, the author decided to write this article and hopes that based on this report further studies can be carried out.

CASE DESCRIPTION

A 57-year-old man came to the eye clinic at RSUP Dr. Sardjito with pain on the left eye and blurred vision. The patient felt that complaint has suddenly became heavier, in which the patient's left eye cannot see, feels lumpy, and tears come out constantly. The patient has a history of diabetes mellitus (blood sugar test results at 220 mg/dl). The results of the vital sign examination obtained blood pressure 190/105 mmHg, heart rate 111x/

minute, respiration rate 20x/minute, and temperature 36.2°C. Examination results of the right eye were within normal limits with visual acuity 6/12. The results of the examination of the left eye showed visual acuity hand movement, palpebral spasm grade 2, hyperemic conjunctiva, cloudy cornea and hypopyon in anterior chamber (Figure 1). The patient was diagnosed with left eye severe corneal ulcer clinically bacterial infection and treated with empirical treatment. The patient received topical antibiotics, pain medication, and was planned to undergo corneal collagen cross-linking and amniotic membrane transplant procedures. The patient was advised to have regular check-ups once a week. In the next control, the results of the vital sign examination were within normal limits. There was no change in the results of the eye examination. At the third week of control, the patient complained persisted. The results of the examination of the left eye showed poor light perception while good color perception, palpebral spasm grade 3, conjunctival injection [+] and paracorneal injection [+] on

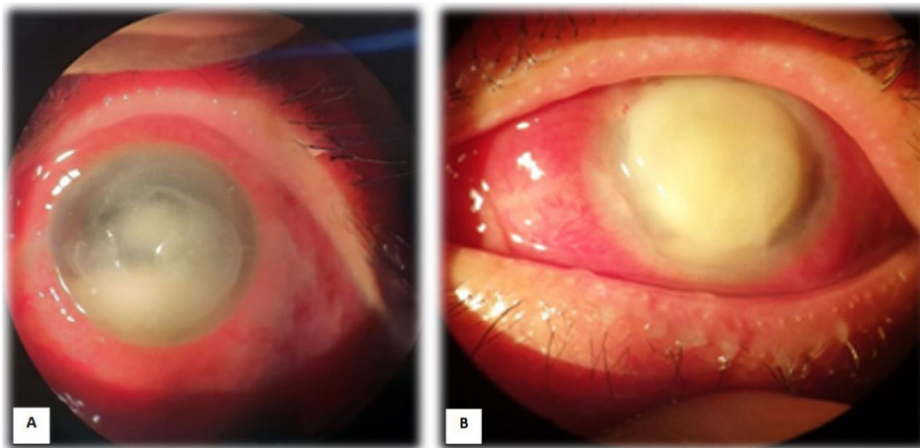


Figure 1. Results of eye examination [A] when the patient first came to the eye clinic; [B] at the time of control to the 28th day.

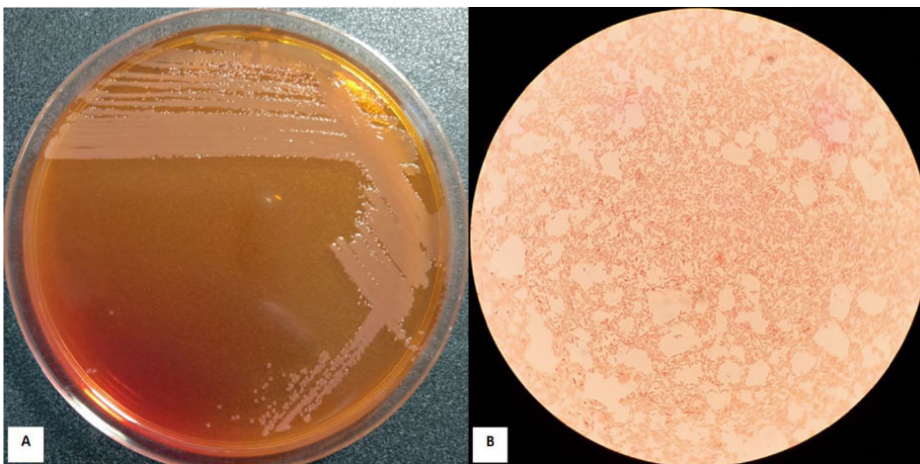


Figure 2. [A] Colony appearance of *Stenotrophomonas maltophilia* on MacConkey's medium. colonies appear round, yellow and smooth; [B] Microscopic view of *Stenotrophomonas maltophilia*. The results of the Gram staining of the bacteria are rod-shaped and red in color (gram-negative bacteria).

Table 1. The result of the antibiotic sensitivity tests in this case report

Antibiotic	<i>Stenotrophomonas maltophilia</i>
Ceftazidime	R
Levofloxacin	S
Cotrimoxazole	S

the conjunctiva, cloudy cornea and the anterior chamber was difficult to assess. At the fourth week of control, the patient still complained of a lump in his eye and tears kept coming out. Examination results of the left eye obtained visual acuity showing poor light and color perception, palpebral and calm conjunctiva, cloudy cornea and the anterior chamber was difficult to assess.

When he first came to the eye clinic, the patient received an action in the form of a corneal ulcer swab. The sample was

sent to the microbiology laboratory for culture examination. The culture results showed coagulase-negative *Staphylococcus* and *Stenotrophomonas maltophilia* (Figure 2). *Stenotrophomonas maltophilia* was identified by API 20NE identification system by Biomérieux (99.7% identification percentage). Test antibiotics for *Stenotrophomonas maltophilia* were carried out using the Kirby-Bauer method and interpretation based on CLSI 2021 (Table 1). The patient gives permission for the use and publication of the data.

DISCUSSION

Eyes are one of the most important senses in humans. If there is disease in the eye, it can be at risk of impaired vision or blindness. Indonesia is one of the top five countries with a large number of visually impaired people. The World Health Organization (WHO) reports that 5.1% of blindness is caused by diseases of the cornea. One of the diseases of the eye is a corneal ulcer. Corneal ulcers can be divided into two based on the cause, namely non-infectious corneal ulcers and infected corneal ulcers. Infectious corneal ulcers can be caused by bacteria, fungi, viruses or protozoa.¹⁻⁴ Corneal ulcers caused by bacteria have symptoms in the form of pain, redness, discharge, lacrimation, foreign body sensation, photophobia, conjunctival injection, and vision loss to varying degrees. There is an infiltrate accompanied by inflammation, stromal oedema, epithelial defects, and anterior chamber reaction +/- on examination with a slit lamp.^{2,4}

Corneal ulcers caused by viruses have similar symptoms to bacterial ones, but on slit lamp examination there are dendritic lesions. Fungal corneal ulcers are slow and there is no sensation of conjunctival injection as occurs with bacterial infections. On slit lamp examination, the lesion appears grey-white with irregular edges. Corneal ulcers caused by protozoa (*Acanthamoeba*) have symptoms in the form of pain that is disproportionate to physical examination findings and severe photophobia. Slit lamp examination shows that this may show a dendritic-like lesion and a ring-shaped infiltrate.⁴

To find out the cause and to guide treatment for corneal ulcer, culture is necessary. Corneal specimens are taken for culture by an ophthalmologist by scraping the cornea. Corneal scraping begins by providing an explanation of the procedure to the patient, then the patient is positioned sitting comfortably in front of the slit lamp and asked to open his eyes and reduce blinking during the procedure. Local anaesthesia is administered to the eye to reduce discomfort. The ophthalmologist uses a 21-gauge needle/kimura scalpel to take a corneal specimen. The specimen is then streaked onto a glass object and inoculated into solid media

such as blood agar, chocolate agar, Mac Conkey, sabouraud dextrose agar, non-nutrient agar and liquid media such as brain heart infusion, thioglycolate broth, cooked meat broth. The samples are then sent to the microbiology laboratory.^{4,11-13}

In this case, the cornea is scraped using a cotton swab which is then inserted into the brain heart infusion and then sent to the microbiology laboratory. At the analysis stage carried out in the microbiology laboratory, it is started with microscopic examination (Gram staining/KOH) and process specimens that have been inoculated into media starting by inserting the media into an incubator, reading culture results, carrying out biochemical tests, identifying pathogens, and carrying out tests. antimicrobial sensitivity. The final stage (post-analysis) is reporting the results of pathogen identification and antimicrobial susceptibility testing to the ophthalmologist.^{14,15} In this case, coagulase negative *Staphylococcus* and *Stenotrophomonas maltophilia* was identified with the results of the antibiotic sensitivity test for *Stenotrophomonas maltophilia* can be seen in Table 1.

Data from the Asia Cornea Society (multicentre study) in 2014-2015 found that corneal ulcers caused by bacteria amounted to 38% of all corneal ulcers. The National Eye Center of Cicendo Eye Hospital reported that the most common cause of bacterial corneal ulcers was infection with gram-positive cocci bacteria (56.6%). The most common gram-positive bacteria found are *Staphylococcus aureus* and coagulase-negative staphylococcus (CoNS). While gram-negative bacteria that mostly often cause corneal ulcers are *Pseudomonas* sp.^{3,16} *Stenotrophomonas maltophilia* is a rare cause of corneal ulcers. Casalita et al. (2020), in their research at Cipto Mangunkusumo Hospital, Jakarta, Indonesia found two cases of infected corneal ulcers due to *Stenotrophomonas maltophilia*.⁵

Stenotrophomonas maltophilia is the only member of the genus *Stenotrophomonas*. It is often found in the environment (water, soil, plants), animals (especially aquatic animals), and healthcare settings. *Stenotrophomonas maltophilia* is a gram-negative rod, obligate aerobic, motile, non-fermentative

and oxidase-negative.^{6,7} Little is known about the virulence factor of *Stenotrophomonas maltophilia*. Adherence ability is known to be possessed by the *Stenotrophomonas maltophilia* strain both from the environment and clinically. They adhere firmly to plastics (including intravenous cannulas), glass and teflon. In addition, firstly, *Stenotrophomonas maltophilia* has cell wall proteins that can bind IgG via their Fc region. These cell wall surface proteins have a possible role in pathogenicity in humans and animals. Though, this requires further research.^{7,17} Secondly, *Stenotrophomonas maltophilia* can form biofilms and thirdly they are intrinsically resistant to some broad-spectrum antibiotic agents (mostly beta-lactam antibiotics).⁶

Stenotrophomonas maltophilia is an opportunistic bacterium that causes nosocomial infections, especially in immunocompromised individuals such as pneumonia, bacteremia, meningitis, endocarditis, upper respiratory tract infections, urinary tract infections, and skin/soft tissue infections.^{8,9} *Stenotrophomonas maltophilia* infection of the eye is rare. However, it seems that the incidence is increasing as reports circulate. Conjunctivitis, keratitis, corneal ulcers, endophthalmitis, infantile dacryocystitis, and preseptal cellulitis are ophthalmological syndromes caused by these bacteria. Corneal ulcers can occur when the eye's defense system, such as the corneal epithelium, is disturbed so that pathogens can enter and cause infection. Predisposing factors for eye infection due to *Stenotrophomonas maltophilia* include long-term use of corticosteroids, use of contact lenses, eye trauma/surgery, disease on the surface of the eye and immunodeficiency.^{2,8-10,18} The percentage of the risk of bacterial corneal ulcers in the use of contact lenses is 46%. The incidence of keratitis due to contact lens use is 34.6%; this result is similar to the results of a study conducted by Wu et al. that is equal to 42.9%. The high cases of infection due to the use of contact lenses is possible because *Stenotrophomonas maltophilia* can form biofilm on contact lenses and their resistance to lens care systems/disinfectant solutions.^{2,8,9} Immunodeficiency/systemic factors such as old age, AIDS, Steven-Johnson syndrome, Sjogren's syndrome,

vitamin A deficiency and diabetes mellitus can also affect the occurrence of corneal ulcers. The percentage of the risk of bacterial corneal ulcers in people with diabetes mellitus is 52.1%.²

The correct diagnosis determines the right therapy. Misdiagnosis of cases of *Stenotrophomonas maltophilia* is often caused by other suspected causes causing severe complications. Treatment of *Stenotrophomonas maltophilia* infection is complicated because this bacterium shows resistance to many drugs, both clinical and environmental strains show similar tendencies.¹⁹ Although *Stenotrophomonas maltophilia* has increased resistance to beta-lactam antibiotics (most of all), aminoglycosides, and quinolones; however, based on the results of research by Wu et al., fluoroquinolones are the drug of choice for treating infections caused by these bacteria.⁸⁻¹⁰ In this study, the patient underwent therapeutic penetrating keratoplasty to manage the disease.

In this case, good coordination between the ophthalmologist and the clinical microbiologist to help find the cause so that appropriate treatment can be provided is one of the strengths of this case. Apart from that, a complete explanation of *Stenotrophomonas maltophilia*, including the clinical history that appears in cases of corneal ulcers, is expected to help understand this bacterium.

There are several disadvantages such as 1) Not carrying out direct gram staining or inoculating the specimen directly into the media; 2) Repeated specimen collection for evaluation in this patient was also not carried out and 3) Studies have not been carried out on other cases of corneal ulcers caused by *Stenotrophomonas maltophilia*.

CONCLUSIONS

In this case, the corneal ulcer was caused by coagulase negative *Staphylococcus* and *Stenotrophomonas maltophilia*. *Stenotrophomonas maltophilia* itself has several virulence factors and a tendency to be resistant to many antibiotics. In addition, the patient had a history of diabetes mellitus which could be one of the factors that aggravated the clinical condition of the corneal ulcer. Of course, all of these things complicate the treatment.

DISCLOSURES

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Ethical Approval

The Medical and Health Research Ethics Committee of the Faculty of Medicine, Public Health and Nursing, Universitas Gadjah Mada/Dr. Sardjito Hospital approved this study (KE/FK/1645/EC/2022).

Conflict of Interest

There is no conflict of interest in this study.

Author Contributions

Conceptualization, methodology and writing original draft preparation: Kusumadewi YP; Formal analysis: Kusumadewi YP; Data curation: Kusumadewi YP, Karina SM, Setyowati R, Suhardjo, Rizki LP; Validation: Setyowati R, Rizki LP; Writing, review and editing: Kusumadewi YP; Approval of final manuscript: all authors.

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