Co-Infection of the Human Cornea With Stenotrophomonas Maltophilia and Aspergillus Fumigatus

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Purpose. To report a case of corneal co-infection of Stenotrophomonas maltophilia and Aspergillus fumigatus.

Methods. We describe a culture and biopsy proven infectious keratitis with a large, brown, round anterior chamber mass attached to the endothelium.

Results. Stenotrophomonas maltophilia was cultured from external scrapings of a corneal ulcer and septate hyphae were stained with Gomori’s methenamine silver stain along the wall of the excised intracameral mass. Aspergillus fumigatus was cultured from the mass and pus pockets developed along the corneoscleral incision for removal of the mass.

Conclusion. Co-infection of cornea with Stenotrophomonas maltophilia and Aspergillus fumigatus with existence of a large, brown, smooth-surfaced mass in the anterior chamber makes this case unique and interesting.

Key Words: Aspergillus—Infectious keratitis—Stenotrophomonas maltophilia.

Stenotrophomonas maltophilia that is closely related to the Pseudomonas species is emerging as an important opportunistic and a nosocomial pathogen with the increased use of broad-spectrum antimicrobials.1 Patients compromised by debilitating illness, surgical procedures, indwelling catheters, immunocompromise, long term antibiotic therapy, and malignant neoplasm are most prone to S. maltophilia infections.2,3

Infectious keratitis caused by S. maltophilia is rare with only 10 cases reported worldwide.4–6 None of these cases, however, has been reported in association with Aspergillus fumigatus. We report an infectious keratitis where S. maltophilia and Aspergillus fumigatus were cultured simultaneously. The former was cultured from the corneal ulcer and the latter from a large, round, brown-pigmented intracameral mass that was attached to the endothelium.

CASE REPORT

A 36-year-old man was referred for corneal ulcer and secondary uveitis in his right eye. Foreign body sensation had started 20 days earlier after riding a motorcycle. He had been diagnosed to have infectious keratitis without any corneal foreign body in another hospital where the culture of corneal lesion grew Stenotrophomonas maltophilia. Despite the use of 1.4% tobramycin and 0.3% ciprofloxacin every 30 minutes, his corneal lesion enlarged and intraocular pressure (IOP) increased due to severe uveitis.

At our initial examination, he appeared in good health, except for his eye problem and best-corrected visual acuity in the right eye was 20/100. IOP was 30 mmHg and 90% of the cornea was deep epithelialized with 3.5 x 1.8 mm well-demarcated, milky-white, and dense infiltrative lesion observed at 7 o’clock peripheral cornea, where a brown-pigmented nodular mass was attached intracamerally, and associated with 4+ cellular reaction (Figs. 1, 2). After scrapings were collected from the lesion for culture and smear, concentrated antibiotics were restarted with oral corticosteroid to alleviate the secondary uveitis.

The culture confirmed the growth of Stenotrophomonas maltophilia, after which we started using 4.0 mg/mL ticarcillin-clavulanic acid eye drops hourly in addition to 50mg/ml amikacin and ceftazidime respectively, guided by in vitro drug sensitivity test. Oral prednisolone was also prescribed at the initial dosage of 1 mg/kg with tapering.

Despite the maintenance of the above medications over 20 days, the corneal lesion and uveitis continued to deteriorate with the appearance of a ring infiltrate, spreading of ulcerative lesion, and persistence of intracameral nodular mass (Fig. 3). We did the...
second smear and culture of external scrapings from the ulcer area without recovery of other organism except S. maltophilia. Anterior chamber irrigation with antibiotic solution and excisional biopsy of the intracameral mass were performed through superior corneoscleral incision for pathologic and microbiologic examination. The mass was a grossly brown-pigmented cystic one enclosing thick, pus-like material. Two days after removal of the mass, multiple pus pockets developed along the superior corneoscleral incision, from which the culture was performed (Fig. 4).

Pathology of the mass showed multiple brown pigment particles superficially along the wall of the mass, presumably originated from iris pigment epithelium (Fig. 5). Gomori’s methenamine silver (GMS) stain showed acutely angled septate hyphae along the wall of the mass (Fig. 6). Fungal infection was further confirmed with the identification of Aspergillus fumigatus from the culture of the intracameral mass and pus pockets developed along the superior corneoscleral incision, from which the culture was performed (Fig. 4).

The pus pockets along the superior corneoscleral incision completely disappeared in 7 days, followed by slow amelioration of anterior chamber reaction. The deep epithelialized area of the cornea also rapidly contracted (Fig. 7) and was completely epithelialized over the next 30 days. New vessels grew into the ulcer area where white infiltration slowly cleared out to residual scar. It has been 6 months since the discontinuation of the antifungal and antibiotic use and there have been no recurrences. At his last examination, opaque corneal scar, irregular astigmatism, and development of a secondary cataract added to his decreased vision of hand motion (HM) with findings of iris stromal atrophy and depigmented iris epithelium (Fig. 8). The patient refused further intervention and was lost to follow-up.

DISCUSSION

Stenotrophomonas maltophilia that is closely related to Pseudomonas species is an aerobic, nonfermentative, and gram-negative bacillus. It can be isolated from soil, water, various animals, and plants. Although community-acquired infections have been reported, S. maltophilia is primarily considered a nosocomial and an opportunistic pathogen. The pathogenicity of S. maltophilia has been documented in cases of septicemia, endocarditis, pneumonia, postoperative wound infections, and urinary tract infections.
Unlike systemic S. maltophilia infections, most ocular infections were community-acquired ones predisposed by compromised ocular surface such as trauma or contact lens use. In addition to suppurative keratitis, other ocular infections associated with S. maltophilia included conjunctivitis, infected scleral buckles, endophthalmitis, dacryocystitis, and preseptal cellulitis. This organism also has been recovered as a contaminant from contact lens or lens care systems without being implicated in infection or in acanthamoeba keratitis cases.

Although S. maltophilia recovered much less often and has lower virulence than Pseudomonas aeruginosa, it is becoming clinically important because of resistance to many drugs such as aminoglycosides, quinolones, and most beta-lactam antibiotics. In a recent study, the combination of ticarcillin and clavulanic acid was one of the most active compounds against S. maltophilia in vitro.

In the initial treatment of this case, the explanation for poor response to the combined use of sensitive antibiotics and ticarcillin-clavulanic acid might have been either the emergence of new resistance to antibiotics used or the failure to use antifungal medication. However, drug sensitivity test showed S. maltophilia to be sensitive to aminoglycosides, such as amikacin, used in this case.

In addition, the fact that the corneal lesion rapidly improved after the discontinuation of corticosteroids and implementation of antifungal medication favors the latter possibility over the first one. The development of pus pockets along the superior corneoscleral incision after the removal of intracameral mass and the recovery of Aspergillus fumigatus from them might be because of the inoculation of the organisms during the removal procedure.

The common clinical features of infectious keratitis caused by filamentous fungi such as Aspergillus and Fusarium are known to be fine or coarse granular infiltrates within the epithelium and stroma with feathery edges, satellite lesions, corneal rings, and endothelial plaques. The absence of the above clinical manifestations and the large, brown-pigmented, smooth-surfaced intracameral mass dissimilar to known fungus balls, and the growth of rare S. maltophilia from the surface of corneal ulcer make this case unprecedented to the best of our knowledge.

It is difficult to explain the correct pathogenic process of this unusual corneal infection with certainty at present. However, based on the fact that S. maltophilia was cultured initially and the large intracameral fungal mass would have taken a relatively long time to form, we are speculating that a corneal foreign body inflicted from motorcycle riding, which was spontaneously dislodged later, might have inoculated the two causative organisms. The large intracameral mass might be an exaggerated form of deep fungal keratitis such as fungal plaque or ball, resulting from the failure to use an appropriate antifungal medication early.

From our experience of this case, we suggest an early excisional biopsy and microbiologic testing of an anterior chamber mass associated with trauma. At the same time, it would be helpful to use an antifungal medication such as amphotericin B intracameral after microbiologic confirmation of fungal organism as recommended by Kaushik et al.

REFERENCES


3. Laing FPY, Ramotar K, Read RR, et al. Molecular epidemiology of


