CASE REPORT

Corneal infection by *Pseudomonas stutzeri* following excision of trigeminal nerve schwannoma

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SUMMARY

A 25-year-old woman underwent intracranial surgery for trigeminal nerve schwannoma (TGNS) with persistent left-sided facial hypoesthesia. Two months later, she developed a central corneal ulceration. Scraping of the corneal lesion revealed Gram-negative bacilli. Genus level identification was achieved using standard techniques and species level identification, revealing *Pseudomonas stutzeri*, was aided by a VITEK 2 compact system. Broad-spectrum fortified antibiotics were initially started followed by species-sensitive fortified antibiotics. Ocular surface toxicity developed a week later; this was managed with a non-fortified antibiotic. The epithelial defect healed in 3 weeks with subsequent corneal scar formation. Visual rehabilitation was achieved with deep anterior lamellar keratoplasty. Six months following surgery, the patient had a visual acuity of 20/40 with -1.25 170° -0.5 refractive correction and a clear graft. This case report, for the first time, highlights *P. stutzeri*, an aetiological agent of corneal ulcer following excision of TGNS and its successful management.

BACKGROUND

Pseudomonas spp are known to cause devastating ocular infections including bacterial keratitis. Among the various species of *Pseudomonas*, *P. aeruginosa* has been mainly implicated in the causation of bacterial keratitis. *Pseudomonas stutzeri*,¹ an aetiological agent for keratitis has been sparingly described in the literature and, to the best of our knowledge, this report is the first of its kind to describe corneal infection by *P. stutzeri* in a post-operative setting in a patient who underwent excision of trigeminal schwannoma.

CASE PRESENTATION

A 25-year-old woman, non-diabetic and non-hypertensive, reported with pain, redness, photophobia and diminution of vision of almost 2 months duration. She underwent intracranial surgery for left trigeminal nerve schwannoma about 2 months prior following which hypoesthesia persisted on the left side of her face including her eye. Initial ocular examination revealed distant visual acuity as 20/20 in the right eye and hand movements close to the face in the left eye. Right eye examination was essentially within normal limits. Left eye examination showed oedema of upper and lower eyelids, circumciliary congestion, corneal epithelial defect with underlying multifocal infiltrates of varying size and 360° peripheral superficial vascularisation involving mainly the upper two quadrants (figure 1). Anterior chamber showed

Figure 1 Slit lamp photograph showing ocular surface infection on initial evaluation.

grade 1 flare and cells. The pupil was central, circular and sluggishly reacting to light. The lens was clear and fundus was unremarkable. Corneal and conjunctival anaesthesia was assessed with a cotton wisp and found to be reduced as compared to the normal eye. Schirmer's test with anaesthesia was 15 mm in the right eye and 5 mm in the left. Moreover, the Schirmer's test without anaesthesia showed a higher than 25 mm reading, as expected, due to reflex secretion in the affected eye. Intraocular pressure as measured by applanation tonometer was 14 mm Hg in the right eye and digitally normal in the left eye.

INVESTIGATIONS

Scraping of the corneal lesion revealed Gram-negative bacilli on staining. Colonies on blood and chocolate agars exhibited irregular margins, were oxidase positive and attained a wrinkled appearance after incubation of 48 h. Non-lactose fermenting dry colonies were obtained on McConkey agar. Oxidative reaction on Hugh and Leifson's media, motility, utilisation of glucose and reduction of nitrate were seen. Growth was seen at both 37°C and 42°C. Genus level identification was achieved by standard techniques and species level identification of *P. stutzeri* was aided by a Gram-negative identification (GNID) card of the VITEK 2 compact automated microbiology system (bioMérieux, France) with 99% probability.

TREATMENT

Broad-spectrum topical antimicrobial therapy in the form of fortified cefazolin 5% every hour and fortified amikacin 2.5% every hour was initially started. With the availability of an antibiotic

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Figure 2 Slit lamp photographs showing ocular surface toxicity (A) and gradual resolution (B).

sensitivity report through the VITEK 2 compact automated microbiology system, fortified amikacin was continued, however, fortified cefazolin was stopped. Although the initial response was positive, unfortunately, a week later, the patient developed features suggestive of ocular surface toxicity in the form of a non-healing ulcer with punched out epithelial defect, angry looking 360° peripheral superficial vascularisation and diffuse conjunctival congestion involving bulbar and palpebral conjunctiva (figure 2A). Hence, the therapy was modified. The fortified antibiotic was stopped and replaced with a non-fortified antibiotic, that is, 0.3% ciprofloxacin (every 2 hours), based on a susceptibility profile obtained by an antibiotic susceptibility testing (AST) N-280 card of the VITEK 2 compact automated microbiology system. The isolate was found susceptible to all antimicrobials tested by the AST N-280 card, which included β -lactams, third-generation cephalosporins, aztreonam, carbapenems, aminoglycosides, quinolones, minocycline, tigecycline, cotrimoxazole and colistin. It exhibited intermediate susceptibility to levofloxacin with a minimum inhibitory concentration of 4 $\mu\text{g/dL}$. Repeat scraping confirmed Gram-negative bacilli on direct smear and *P. stutzeri* on culture and identification by GNID card.

OUTCOME AND FOLLOW-UP

The patient responded to the management described above. The epithelial defect started healing with reduction in conjunctival congestion (figure 2B). Over a period of 3 weeks, the patient showed marked improvement with healed epithelial defect and absence of corneal infiltrate with subsequent corneal scar formation (figure 3A). At this stage, her visual acuity was 20/400. One

month later, the corneal scar was managed with layer by layer lamellar dissection technique of deep anterior lamellar keratoplasty. Six months following surgery, the patient had a visual acuity of 20/40 with -1.25 170° -0.5 refractive correction and a clear graft (figure 3B).

DISCUSSION

Trigeminal schwannoma, a rare entity, makes up 0.2% of all intracranial tumours.² Complete tumour removal is considered to be the treatment of choice.³ However, in spite of the best facilities and the most experienced hands, it is often associated with surgically induced trigeminal nerve dysfunction. Ramina *et al*,³ in a series of 17 patients, observed postoperative trigeminal anaesthesia in 20% of cases after complete excision of trigeminal schwannoma. Trigeminal anaesthesia often results in persistent epitheliopathy secondary to reduction in number of limbal stem cells,⁴ reduced mitotic and metabolic activities of corneal epithelium, and lower activities of acetylcholine and cholineacetyl transferase at the corneal nerve endings.^{5 6} This chronic epithelial defect may lead to the formation of corneal ulcer with secondary bacterial infection, as seen in our case. Besides neurotrophic keratitis, the affected eye may have dryness due to reduced corneal reflex.³ Lambiase and associates,⁷ in an observational case series, reported additional corneal changes following neurosurgical trigeminal damage in the form of reduced endothelial cells and an increase in the number of hyper-reflective keratocytes. Presence of corneal anaesthesia and dry eye may lead to a decline in local immunity, thereby rendering the cornea more prone, and leading to infection by *P. stutzeri*, as seen in our case.

Figure 3 Slit lamp photographs showing healed corneal lesion (A) and subsequent management by deep anterior lamellar keratoplasty (B).

P. stutzeri is an aerobic, ubiquitous Gram-negative bacterium generally considered a contaminant.⁸ It becomes an opportunistic pathogen with decline in local or generalised immunity and has been reported to cause iatrogenic infections and pseudo-outbreaks. *P. stutzeri* has been isolated from surgical wounds, blood, respiratory samples, urine and various other samples, though it is rare in the eyes.⁹ A MEDLINE search reveals only a few reports of its ocular involvement in the form of conjunctivitis,¹⁰ delayed onset endophthalmitis¹¹ and late onset bleb-related panophthalmitis.¹² The role of *P. stutzeri* in causing corneal ulcers is extremely rare, as highlighted by the presence of only one report existing in the literature.¹

In our case, the presence of Gram-negative bacilli in corneal scraping, pure growth on solid agar and response to treatment, establishes its pathogenicity. Species identification and antimicrobial susceptibility were achieved using a VITEK 2 compact automated system.¹³ Identification by manual techniques is demanding in time and effort. Automated systems such as the VITEK 2 compact, utilise growth-based technology and colourimetric cards to identify organisms. Automated systems provide accuracy and rapidity in species identification and detection of antimicrobial resistance, facilitating early diagnosis and treatment that can help prevent complications such as posterior perforation of corneal ulcers leading to endophthalmitis. These systems facilitate standardisation, quality control and reproducibility of results, and are suited for medium-sized laboratories. Nakasone *et al*,¹³ in a laboratory-based evaluation, found 95.5% accuracy in species identification and 97.7% accuracy in detection of antimicrobial resistance. Molecular methods employing 16s ribosomal RNA and *rpoD* gene sequencing can also aid species level identification, however, they are resource intensive.¹⁴

Early species identification and antimicrobial susceptibility enables early institution of treatment curtailing therapy with broad-spectrum antimicrobials. Fortified antibiotics in combination have traditionally been administered in many eye centres, including ours, as broad-spectrum antibiotics. However, they are often associated with ocular surface toxicity, including damage to ocular surface epithelium.^{15 16} We observed a similar phenomenon in our case, and hence we stopped fortified antibiotics and switched over to a non-fortified broad-spectrum antibiotic, namely, ciprofloxacin. The literature has suggested that ciprofloxacin monotherapy is as efficacious as combined fortified

antibiotics and offers an advantage in terms of reduction of ocular surface inflammation.¹⁷ After termination of the fortified antibiotics, we found a dramatic improvement in ocular surface inflammation in our case, with subsequent early healing and corneal scar formation.

This case report is unique as we are reporting, for the first time, *P. stutzeri* as a causative agent of corneal ulcer in a patient with trigeminal nerve schwannoma. Moreover, it highlights the diagnostic utility of the VITEK 2 compact automated microbiology system and also the need for an early diagnosis of ocular surface toxicity for better management of corneal ulcers.

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Learning points

- Hypoaesthesia often persists following excision of trigeminal nerve schwannoma. As a result, ocular surface immunity is often compromised.
- Reduced localised immunity makes the surface conducive to infection by opportunistic organism, as seen in our case.
- In the postoperative setting following excision of trigeminal schwannoma, regular ophthalmic reviews are recommended so as to detect ocular infection at an early stage.

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