CASE REPORT

Rare case of bilateral perforated corneal ulcer due to gonococcal infection, managed with temporary periosteal graft

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SUMMARY

A 17-year-old female patient was referred to Kirana Ophthalmology Unit, Cipto Mangunkusumo Hospital, with a 10-day history of redness and swelling of the eyes, and inability to open them. Other symptoms included pain, blurred vision and excessive yellowish-white discharge from both eyes. There was a history of multiple sexual partners. The patient was assessed with bilateral perforated corneal ulcer due to gonococcal infection, based on the findings of intracellular and extracellular Gram-negative diplococci found on the Gram staining examination. The cornea in both eyes showed perforation with iris prolapse inferiorly. The perforations were treated with temporary periosteal grafts. The grafts remained in place after the surgery. Final uncorrected visual acuity was 6/20 in the right eye and 6/24 in the left eye, a few months after surgery.

BACKGROUND

Neisseria gonorrhoeae ocular infection can turn into a vision-threatening condition, especially in cases where corneal involvement has resulted in scarring and possibly perforation.^{1–3} Ocular infection is transmitted through autoinoculation of the bacteria from infected urine and genital secretion by hands in sexually active adults.^{4–5} *N. gonorrhoeae* is one of the few organisms that can penetrate intact corneal epithelium, leading to stromal and epithelial keratitis, rapidly progressing to ulcerative keratitis, and delayed treatment may progress to corneal perforation.^{2 3 5 6} Gonococcal ocular infection is usually treated with antibiotics, either topical or parenteral, according to the culture result.³ When corneal perforation occurs, it should be managed by a combination of intensive medical and surgical intervention.³ Therapeutic keratoplasty is the most effective treatment in such cases.³ In this case report, we present a rare case of perforated bilateral gonococcal corneal ulcer, associated with a history of multiple sexual partners and irrational traditional medication using the patient's own urine. In cases such as this, immediate surgical intervention should be performed combined with intensive medical therapy. In this case, where keratoplasty as the definitive surgical management was not possible to perform immediately, temporary periosteal graft as an alternative surgical procedure was successful.

CASE PRESENTATION

A 17-year-old female patient presented with redness and swelling of the eyes for the past 10 days, with the inability to open them. She also had symptoms of pain, blurred vision and excessive yellowish-white discharge from both eyes. There was a history of multiple sexual partners within the past 3 months, and the patient was unmarried. Vaginal discharge was present during the last 1 week.

Visual acuity of both eyes was light perception with wrong projection. The eyelids showed severe oedema and spasm, were hyperaemic and excreted profuse purulent discharge. The tarsal conjunctiva showed giant papillae and membrane, while the bulbar conjunctiva was chemotic. The cornea of the right eye revealed infiltrate with thinning at inferior site, shallow anterior chamber and iris shadowing. The cornea of the left eye also revealed infiltrate and perforation at inferior site, with a 2.5×5 mm iris prolapse. The rest of the areas could not be evaluated (figure 1). General physical examination revealed that the right and left neck lymph nodes were palpably soft, painless and mobile, and measuring 10 mm×10 mm, with positive vaginal discharge. Gram staining revealed extracellular and intracellular Gram-negative diplococcus among polymorphonuclear neutrophils (figure 2).

The patient was diagnosed with impending perforated corneal ulcer of the right eye and perforated corneal ulcer of the left eye due to *N. gonorrhoeae* infection. She was given intravenous injection of 1 g ceftriaxone two times daily. Topical treatment consisted of Levofloxacin eye drops hourly, Dibekacin eye drops hourly, Protagent A eye drops hourly, Timol eye drops two times a day, Tropin eye drops three times a day, Gentamycin eye

Figure 1 (A) Right eye: profuse purulent secretion with severe conjunctival chemosis and thinning at inferior site; (B) left eye: severe conjunctival chemosis with perforation at inferior site with iris prolapse.

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Figure 2 (A) The intracellular diplococci (*Neisseria gonorrhoeae*) with Gram-negative staining. (B) The arrow showing intracellular diplococci and the arrowhead showing extracellular diplococci among neutrophils.

ointment three times a day and normal saline irrigation three times a day. Additional oral therapies were 250 mg acetazolamide tablets three times a day and 300 mg potassium tablets two times a day. HIV screening was carried out and was negative, and Gram-staining examination was scheduled to be repeated 3 days after therapy. The eye discharge was also sent to microbiology for culture and resistance examination. Stuart agar was used as a media transport for culture. A dermatovenereologist consult was ordered for the patient's vaginal discharge, with an assessment of non-specified genital infection, which was treated with an oral single dose of azithromycin 1000 mg. The vaginal discharge later resolved. In anticipation of endophthalmitis, an ultrasound examination was planned to be repeated every 3 days; this showed anterior vitreous opacities and retinal oedema (figure 3). The patient was planned to undergo bilateral keratoplasty but, unfortunately, a corneal donor was not available at the eye bank at the time. Hence, temporary periosteal grafts were planned instead of keratoplasty, to close the corneal perforations.

Two days after treatment, the patient showed clinical improvement. There was decrease of discharge and the eyelid oedema gave minimal pain. The visual acuity was hand movement with good projection. However, the right eye became perforated with a 1.5×4 mm iris prolapse (figure 4). The Gram staining showed extracellular Gram-negative diplococcus. The culture result came back 5 days after the specimen was submitted. It grew *Staphylococcus saprophyticus* and did not reveal any *N. gonorrhoeae* growth. The sensitivity test showed the culture to be sensitive to ceftriaxone, among other antibiotics. Nine days after admission, the Gram staining showed absence of diplococcus.

The periosteal graft procedure was carried out 10 days after admission. Periosteum was taken by making an incision along the anterior tibial crest and dissecting the periosteal tissue. The periosteum was then given a 2×2 mm incision and lifted free of its underlying attachments with a periosteal elevator. The

Figure 3 Ultrasound examination of the left eye (A) and right eye (B) showing anterior vitreous opacities and retinal oedema.

Figure 4 Eye condition 2 days after therapy. (A) A 1.5×4 mm perforated corneal ulcer inferiorly; (B) infiltrate (arrow) at superior part with perforated corneal ulcer inferiorly.

autologous graft was then harvested at the perforated cornea with vicryl 6.0. The surgery covered only the inferior part of the cornea where the perforation existed (figure 5). After surgery, all previous therapies were continued except acetazolamide and potassium tablets. Intravenous ceftriaxone was changed to oral cefixime. Hourly instillation of ceftazidime and chloramphenicol eye drops was added to the therapy. The patient was finally discharged from the hospital 4 days after the surgery.

Visual acuity gradually increased from light perception and wrong projection in both eyes before surgery to 3/60 in 3 weeks in the right eye and 1/60 in the left eye. One month postsurgery, the visual acuity of both eyes was even better. The uncorrected visual acuity (UCVA) of the right eye was 6/20 and of the left eye it was 6/24. The corneas in both eyes were relatively clear at the superior with signs of scarring at the mid-central to superior areas affected up to the stromal layer, and the grafts were attached steadily at the half inferior part of the cornea (figure 6). The patient was still planned to undergo corneal transplantation but, unfortunately, this was postponed indefinitely due to financial constraint.

TREATMENT

The patient was given ceftriaxone 2×1 g intravenously as a treatment recommendation for her gonococcal ocular infections. She was also given artificial tears, antibiotics and antiglaucoma eye drops. Oral antiglaucoma medication was also given to the patient. Since there was no corneal donor available, periosteal grafts were chosen as the best alternative procedure in managing the corneal perforations in this case.

OUTCOME AND FOLLOW-UP

Final UCVA was 6/20 in the right eye and 6/24 in the left eye 1 month postsurgical and medical therapy. The corneas in both eyes were relatively clear at the superior with sign of scarring at the mid-central to superior areas affected up to the stromal

Figure 5 (A and B) Right and left eye preoperatively. (C and D) Right and left eye immediate postoperatively.

layer, and the grafts were attached at the half inferior part of the cornea. The patient was still planned to undergo corneal transplantation but, unfortunately, this was postponed indefinitely due to financial constraint.

DISCUSSION

N. gonorrhoeae is the most common bacterial pathogen associated with hyperacute bacterial conjunctivitis, as shown in this patient. The simplest and quickest way to verify the diagnosis of gonorrhoea in a routine setting is through direct microscopy with Gram staining showing intracellular Gram-negative diplococci within the neutrophils.⁵ In this patient, conjunctival smears of the exudate showed numerous intracellular and extracellular Gram-negative diplococci, which was highly suggestive of *N. gonorrhoeae*, making this a straightforward case with a clear diagnosis.

Treatment principles consist of definitive antibiotic and supportive therapy, and surgical management. Antibiotics were given intravenously and topically to maintain high concentration in the plasma and cornea. The treatment recommendations of

gonococcal ocular infections are independent of the severity of the ocular infection. Patients with corneal ulceration, as in our case, should be admitted to the hospital and treated with intravenous ceftriaxone (1000 mg intravenous every 12 h) for 3 consecutive days.⁵ In our case, the intravenous ceftriaxone was given starting on the first day of admission and continued until 3 days postoperatively (13 days), considering the severity of the corneal infection and the bilateral corneal involvement. Fransen *et al*³ reported an increased cure rate with the addition of topical gentamycin ointment. The benefit of using topical therapy is that it delivers a large concentration of antibiotic locally without great risk of systemic side effects.^{2 3} That explained this patient also receiving topical antibiotic therapy with Gentamycin eye ointment, Levofloxacin and Dibekacin eye drops.

In this patient, Stuart agar was used as a transport system for culture since it was the only medium available. The culture grew *Staphylococcus saprophyticus*. This was different from what we expected as the culture result. There are two possibilities concerning this culture result. First, *N. gonorrhoea* is known to be

Figure 6 (A and B) Eye condition 1 month after the surgery showed that the periosteal grafts were attaching to both eyes and sign of scarring at superior parts of cornea (arrow).

very fragile and even delayed transportation without obligatory anaerobic reservoir will cause death of the vital organism.³ Second, the possibilities of co-infection were present, since the patient used urine as an ocular irrigation agent. The culture sensitivity showed that the organism was resistant to penicillin but sensitive to third-generation cefalosporin, ceftriaxone, an intravenous antibiotic that we gave to this patient. Culture also showed sensitivity to ceftazidime and chloramphenicol, so they were added to the topical therapy after the periosteal grafts were performed. Frequent ocular saline lavage has been recommended as an adjunctive irrigation with approximately 50 cc of saline every hour as needed to remove the possibly toxic purulent conjunctival discharge.^{2 3} Three times daily saline lavage was enough to remove the purulent discharge in this patient.

Learning points

- ▶ A bilateral case of gonococcal keratoconjunctivitis is a rare finding, yet it carries a double-fold threat to blindness and, when associated with corneal perforation, the situation becomes an emergency that requires prompt surgical management, besides adequate medical management.
- ▶ The diagnosis of gonococcal infection of the eye should be made as early as possible since any delay may lead to irreversible consequences.
- ▶ Gram stain and confirmatory culture are mandatory because of the systemic and therapeutic implications.
- ▶ When definitive surgical management is unfeasible, resulting from corneal donor absence, temporary periosteal graft can be a reasonable alternative. This procedure offers a good result in terms of avoiding secondary endophthalmitis, controlling the infection and re-establishing the structural integrity of the globe.

Therapeutic keratoplasty was planned initially when the patient came to the hospital. Since there was no corneal donor available, the autogenous periosteum was chosen as substitute because of its ease in harvesting and ready vascularisation. The rich vascularisation of periosteum also gave benefit in fighting against the microorganism, thus eliminating it. The graft was harvested using vicryl 6.0 since it is strong enough to stitch the rough periosteal tissue to cover perforation and the fact that it will be absorbed spontaneously so it will not require additional time for suture removal. This was also supported by a descriptive study conducted by Mulyawarman and Edwar⁷ at Kirana Ophthalmology Department, Cipto Mangunkusumo Hospital, in 2012, which showed periosteal graft as being a possible procedure that can be chosen, besides penetrating keratoplasty, to preserve the integrity of the globe, especially where a corneal donor is not available.¹ In this case, periosteal graft was used as a temporary measure.

Competing interests None declared.

Patient consent Obtained.

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