

her knees and loose-jointedness. Her vision was 6/4 bilaterally, without any angioid streaks. His son had stiff joints except his knees, extensible skin over the neck and face and a normal ocular examination.

It was felt that our case was a PXE heterozygote and that angioid streaks were part of this clinical phenotype. In the absence of cutaneous changes homozygosity for the PXE gene is unlikely, although it cannot be totally excluded. DNA has been extracted and banked to test for the PXE gene in the future. A formal skin biopsy from the side of the neck showed minimal elastotic degeneration and elastic fibres were not Von Kossa positive. There were no particular features suggestive of PXE. The recent normal colonoscopy rules out Familial Adenomatous Polyposis (FAP).

### Comment

Congenital hypertrophy of the retinal pigment epithelium (CHRPE) is a well-known association of Familial Adenomatous Polyposis (FAP). On reviewing literature there is one case report of a middle-aged Caucasian male with angioid streaks but no CHRPE who had extensive FAP.<sup>4</sup>

The relationship between CHRPE and angioid streaks is probably coincidental and has not been reported before. The previously reported case had extensive FAP necessitating a total colectomy, though his fundi did not show any features of CHRPE. Both these patients were Caucasian males, of similar age and with radiological evidence of degenerative changes in their lumbar vertebrae. The striking similarity between these cases and the fact that CHRPE and FAP often are part of the same syndrome suggests that their relationship with angioid streaks could be part of the same syndrome. In the present case the cutaneous manifestations of PXE were minimal, while in the previously reported case FAP was not associated with CHRPE, possibly indicating an incomplete expression.

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Sir,

### Gonococcal keratoconjunctivitis in adults

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Gonococcal ocular infection can be divided into two distinct forms, one affecting neonates and the other affecting sexually active adults. Most cases occur in neonates or sexually active adults and are transmitted by contact with infected urine or genital secretions. Recently, the incidence of adult gonococcal conjunctivitis has shown a tendency toward increasing with time, especially penicillinase-producing *N. gonorrhoeae* (PPNG).<sup>1,2</sup>

As gonococcal eye infection in adults is relatively rare, the clinical diagnosis may be delayed. However, it is very important that a prompt confirmatory culture for isolation of gonococcal organisms and earlier parenteral antibiotic treatment is required, because the outcome of gonococcal conjunctivitis is related to the severity of disease at the start of adequate therapy.<sup>3</sup>

We present a rare case of bilateral gonococcal conjunctivitis with keratitis, gram-negative intracellular diplococci, penicillin-resistant *Neisseria gonorrhoeae*, associated with a sexual history of relations with a prostitute.

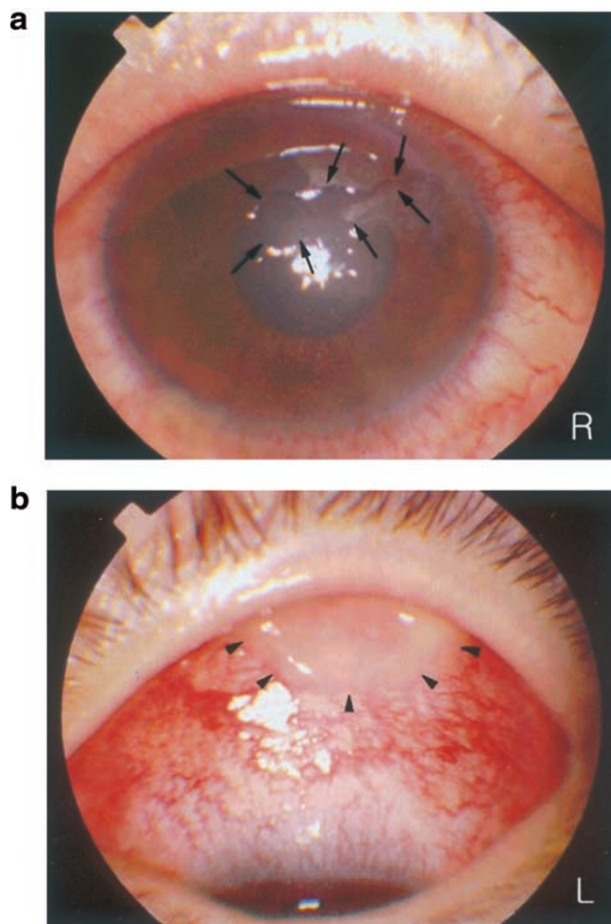
### Case report

A 29-year old man presented redness and ocular discharge from both eyes for 13 days before admission to Pusan National University Hospital in Pusan city. At first, the left conjunctiva was markedly inflamed and there was intense dilatation of the conjunctival vessels without small petechial hemorrhages with purulent exudates. The right eye was completely normal. The other eye became involved on the 7th day after left conjunctivitis developed. Eventually, he failed to respond, so was referred to our clinic for proper management of mucopurulent conjunctivitis.

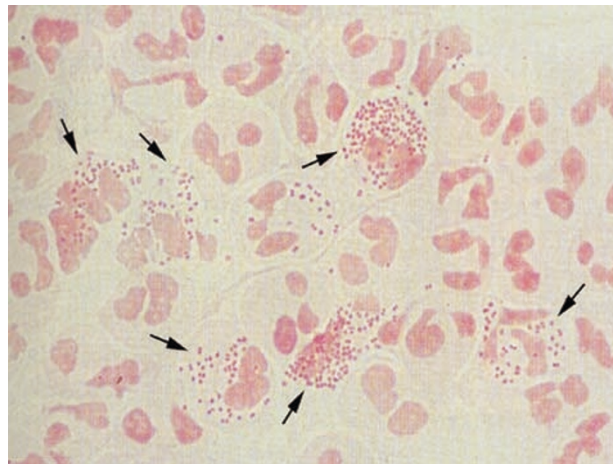
He had a distinctive sexual contact history with a prostitute 17 days previously, but there was no evidence of genitourinary symptoms. On the day of

admission, visual acuity was 20/40 in the right eye and 20/20 in the left. The right stromal cornea was mildly diffusely hazy with an epithelial defect just in the central portion, and the conjunctival signs were markedly inflamed with a purulent discharge in the left eye (Figure 1).

Initial laboratory examination consisted of conjunctival scrapings for Gram stain, and conjunctival cultures on chocolate, blood, phenylethyl alcohol with 5% sheep blood, and MacConkey media, all incubated at 37°C. Chocolate medium was incubated in a 5–10% carbon dioxide-enriched environment. This patient, with gram-negative intracellular diplococci bilaterally on gram stain, subsequently had positive conjunctival cultures for *N. gonorrhoeae* (Figure 2). Sensitivity studies showed the organism to be resistant to penicillin but sensitive to 3rd cepharosporin. Further, we performed the diagnostic test of sexually transmitted diseases such as direct fluorescent antibody



**Figure 1** At first visit, a keratitis with corneal epithelial defect in the right eye and injected conjunctivitis with purulent discharge in the left eye were present.



**Figure 2** There were many intracellular diplococci (*Neisseria gonorrhoea*) with gram-negative staining in culture media.

testing for chlamydia and a serum rapid plasma reagin test for syphilis, but did not find any positive signs.

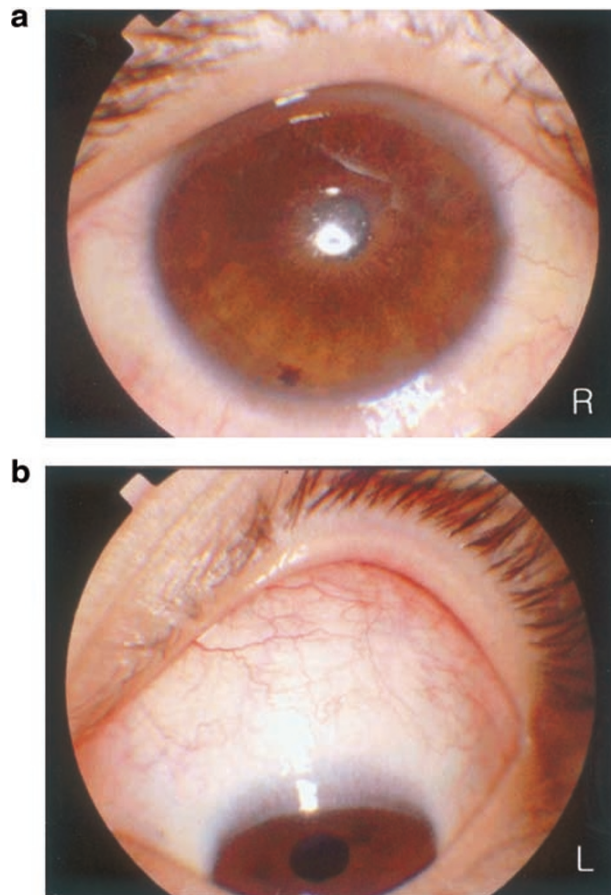
Single dose treatment was begun with 1 g of intramuscular ceftriaxone (Rocephin<sup>®</sup>) for 5 days and topical cefmenoxime 0.5% (Bestron<sup>®</sup>) every 30 min for 2 days. Within 24 h, this patient responded clinically with marked reduction of ocular discharge and inflamed conjunctiva. Repeated conjunctival culture for 12 h after topical and systemic therapy was negative for *N. gonorrhoeae*. On the 3rd hospital day, cefmenoxime 0.5% (Bestron<sup>®</sup>) was tapered every 2 h for 2 days, and every 6 h for 10 days. A 7-day course of doxycycline 100 mg was administered to prevent activity of collagenase released from destructive corneal lesions.

This patient was discharged after 5 days of intramuscular ceftriaxone (Rocephin<sup>®</sup>). Three weeks after discharge, this patient had a visual acuity of 20/20 in both eyes without any complications of the conjunctiva and cornea (Figure 3). Currently, there is no evidence of serious complications of the cornea and conjunctiva.

#### Comment

*N. gonorrhoeae* in adults causes an extremely profuse, hyperacute purulent discharge accompanied by severe conjunctival chemosis and intense dilatation of the conjunctival vessels without small petechial hemorrhages, eyelid swelling, and epithelial or stromal keratitis.<sup>1</sup> The degree of corneal involvement is highly variable, but common types are marginal corneal melt, subepithelial or stromal infiltrates, and a discrete edema of the entire surface of the cornea.<sup>3</sup>

The incubation period of gonococcal ocular infection generally ranges from 3–19 days, and the urethral



**Figure 3** After the antibiotics were initiated for 3 weeks, the cornea was normal without corneal epithelial defect and corneal opacity in the right eye and the conjunctiva was normal in the left eye.

symptoms precede the ocular symptoms from one to several weeks.<sup>1</sup> Compared with typical gonococcal keratoconjunctivitis, this case has some atypical findings such as localized stromal infiltrates with epithelial defect in the central cornea, no dysuria or urethral discharge, and a relatively long incubation period.

If inadequately treated, gonococcal conjunctivitis can progress in an extremely rapid and fulminant fashion, leading to corneal perforation within 24 h. If aggressive parenteral antibiotic therapy is instituted before severe corneal destruction occurs, it appears that the risk of serious sequelae and visual loss is greatly reduced. Therefore, the Centers for Disease Control issued the recommendation that all patients with adult gonococcal conjunctivitis be treated with hospitalization and a 5-day course of high doses of parenterally administered antibiotics like penicillin or cephalosporin therapy.<sup>4-6</sup>

The local incidence of PPNG currently represents less than 1% of all gonococcal infections.<sup>7</sup> To initiate adequate antibiotics therapy is clinically important,

because the outcome of gonococcal conjunctivitis is related to the severity of disease at the start of adequate therapy.

For the treatment of PPNG conjunctivitis, the Centers for Disease Control recommend the administration of 1.0 g of cefoxitin or 500 mg of cefotaxime intravenously four times daily, or 1.0 g of ceftriaxone intramuscularly daily, for 5 days.<sup>4,5,8</sup> The World Health Organization recommends cefotaxime 1.0 g intravenously four times daily for 5 days or intramuscularly spectinomycin 2.0 g for 3 days.<sup>9</sup> Recently, oral norfloxacin 1200 mg for 3 consecutive days may be a useful alternative for the treatment of adult gonococcal keratoconjunctivitis, especially for penicillin-resistant strains.<sup>1</sup> We started with 1.0 g of ceftriaxone intramuscularly daily for 5 days, because this PPNG organism was sensitive to 3rd cephalosporin.

Frequently ocular saline lavage and topical antibiotics have been recommended as ancillary therapy in the treatment of gonococcal ophthalmia, but are not essential for successful treatment of *N. gonorrhoeae* conjunctivitis in adults.<sup>10</sup> This patient was administered a topical 0.5% Bestron<sup>®</sup>, because we feel strongly that aggressive topical antibiotics should be used to cure ocular lesions. Also we started with doxycycline 100 mg orally once daily for 1 week, to prevent activity of collagenase released from destructive corneal lesions,<sup>11</sup> or to treat possible coexistent chlamydial infection with a false negative response by laboratory test.<sup>12</sup>

The rising incidence of PPNG strains and the increasing number of isolates with a high-level resistance to penicillin should lead us to suggest that a prompt confirmatory culture for isolation of PPNG organisms and a sensitivity test to antibiotics are elementary.<sup>1,2</sup> When a case with sexual contact history is suspected of being a hyperacute purulent conjunctivitis or bacterial conjunctivitis refractory to primary antibiotic eye drops, a prompt confirmatory culture for isolation of gonococcal organisms and earlier parenteral antibiotic treatment are mandatory.

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Sir,

**Topical and intralesional interferon therapy for recurrent lacrimal papilloma**

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**Introduction**

Papillomas of the lacrimal sac and canaliculus are an uncommon cause of epiphora. Surgical excision with adjunctive cryotherapy is the standard treatment, but results are poor, with multiple recurrences.<sup>1</sup> We present a case of recurrent lacrimal sac papilloma, treated successfully with topical and intralesional interferon alfa-2b.

**Case report**

A 10-year-old boy was referred with a 2-year history of right epiphora. The left lacrimal outflow system was freely patent, but the right was highly resistant to syringing. A Dacryocystogram (DCG) was requested and he was listed for a Canaliculo-Dacryocystorhinostomy (CDCR) with intubation. A week later, he presented with bloody tears, and epistaxis. Clinical examination and nasoendoscopy were normal. The right nasolacrimal duct was narrow, but patent, with no demonstrable filling defect on the DCG, and the CT scan was normal.

At the time of surgery, multiple papillomata were found within the sac and common canaliculus. Complete excision was performed and the rhinostomy completed. Histology confirmed lacrimal sac papillomas and no evidence of dysplasia. DNA typing confirmed Human Papilloma Virus (HPV) subtype 11. Four months later, he had a recurrence, growing out of the upper canaliculus. This was excised after performing a canaliculotomy, and cryotherapy applied to the base. Histological examination again revealed a lacrimal system papilloma, but with moderate epithelial dysplasia (Figure 1). Two months later there was a second recurrence, which was again excised (Figure 2), but the papilloma reappeared after 3 months.

The patient refused further surgery, and requested alternative therapeutic options. After obtaining informed consent, topical interferon alfa-2b 1 million units/ml four times a day was prescribed. The papilloma dramatically reduced in size within 3 weeks. To augment the effect of topical treatment, a single injection of 3 million-units per 1 ml of interferon alpha-2b was given into the canaliculus. Within one week of the injection, complete clinical resolution was seen (Figure 2). Topical treatment was given for a total duration of 2 months. The only adverse effect seen was a transient, moderate follicular conjunctivitis (Figure 1), during treatment. At 12 months follow-up, the patient remains symptom-free and patent to syringing.

**Comment**

Many therapeutic options are available for lacrimal papillomas. However, the results have been disappointing.<sup>1</sup> Complete surgical excision with adjunctive cryotherapy is currently the standard treatment. Adjunctive cryotherapy has not been proven to be effective in preventing recurrences, and may lead to stenosis of the lacrimal passages. Interferons are part of the human body's natural defence mechanism against tumours and viruses. Interferons exert their