Case Report

Ophthalmoplegia without severe painful eyelid swelling in acute dacryoadenitis: a case report

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Abstract: Here, we present a case of right eyelid drooping in a 79-year-old man. Neurological examination revealed ptosis of the right eye without severe painful eyelid swelling and redness. An ocular motility examination of the right eye revealed upward limitation and downward overshoot. The results of routine blood examinations were within normal limits, and no autoantibodies were detected. Orbital magnetic resonance images revealed mild right eyelid swelling and lacrimal gland enlargement, indicating orbital inflammation. The ocular discharge was positive for *Staphylococcus hominis* by culture and the patient was diagnosed as having acute dacryoadenitis. Treatment with topical and systemic administration of antibiotics rapidly improved symptoms. Ocular infection is not usually suspected in the absence of local severe painful swelling and redness, and painless acute dacryoadenitis presenting as ophthalmoplegia and ptosis may be misdiagnosed. Orbital inflammation may rapidly progress to orbital cellulitis with treatment delay, which may also lead to aggravation of ophthalmic prognosis. Therefore, neurologists should be aware of the possibility of acute dacryoadenitis occurring without the local severe inflammatory findings mimicking neurological diseases, and acute dacryoadenitis should be considered in patients with ophthalmoplegia even in the absence of severe painful eyelid swelling and redness.

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Key words： acute dacryoadenitis, ptosis, ophthalmoplegia

Introduction

Dacryoadenitis is a relatively rare disorder that results from a lacrimal duct obstruction anywhere between the lacrimal gland and the conjunctiva[1)]. Acute dacryoadenitis generally presents as inflammation and enlargement of the lacrimal gland that leads to severe swelling of the lateral eyelid, tenderness over the lacrimal gland fossa, and injection over the lacrimal gland palpebral lobe[2)]. The condition also is symptomatically characterized by downward globe displacement and diplopia[3)]. We observed a rare case of acute dacryoadenitis that presented with ptosis with oculomotor disturbance in the absence of severe eyelid redness, swelling, and tenderness.

Case report

A 79-year-old man visited our hospital with drooping of the right eyelid that began upon wakening on the consultation day. General physical examination revealed redness and swelling of the right bulbar conjunctiva and slight eyelid redness without tenderness. Neurological examinations showed right ptosis despite a normal ocular primary position. Ocular motility evaluation revealed an upward limitation and a downward overshoot of ocular movement in the right eye. The patient complained of diplopia during the downward gaze (Fig. 1). Other examinations yielded normal results. Routine blood and urine examinations were within normal limits, and the patient did not show autoantibodies associated with Sjögren's syndrome and

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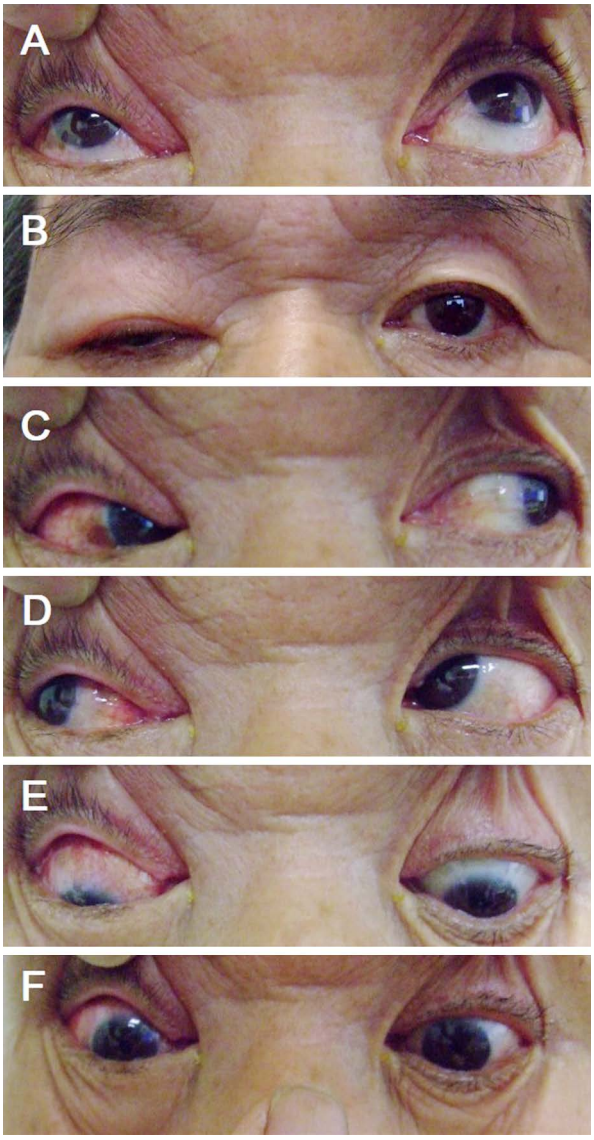

Fig. 1　Initial ocular motility evaluation.
Photographs were taken while the patient attempted an upward gaze (eyelids retracted by examiner, A), was in the primary position (B), attempted a leftward gaze (eyelids retracted by examiner, C), was in the right gaze (eyelids retracted by examiner, D), attempted a downward gaze (eyelids retracted by examiner, E), and during accommodation (F). The eye showed normal position in the primary gaze, but displayed remarkable ptosis without severe eyelid swelling (B). During vertical movement, a monocular upward limitation (A) was associated with downward overshoot (E) in the right eye. The right eye also had bulbar conjunctival redness (C–E). Horizontal movement was normal (C, D) and convergence was possible (F).

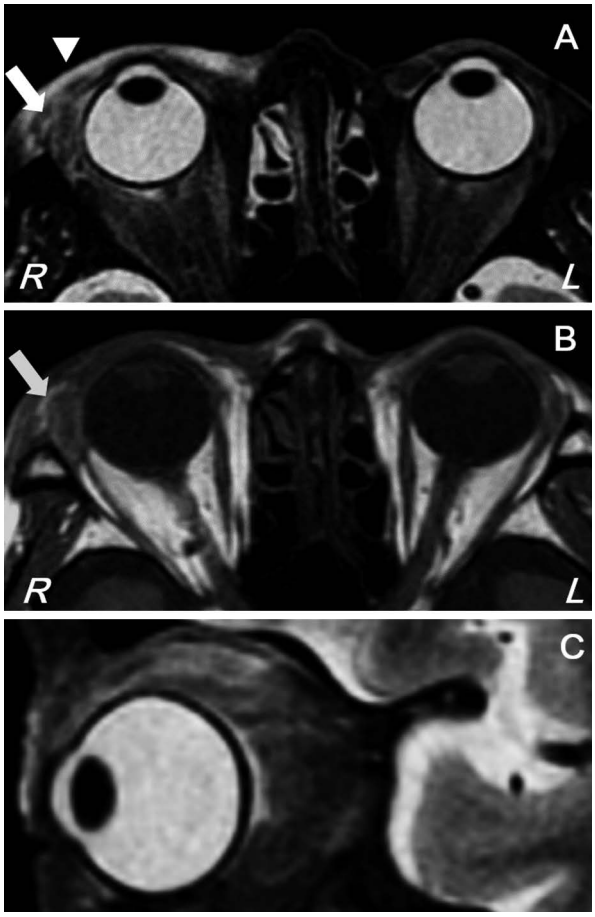

Fig. 2　Initial orbital magnetic resonance imaging.
(A) An axial short inversion TI recovery (STIR) image (TR: 4,740 ms, TE: 41.82 ms) shows mild right eyelid swelling (arrowhead) and lacrimal gland enlargement (arrow). (B) An axial T_1-weighted image (TR: 540 ms, TE: 12 ms) shows lacrimal gland enlargement. (C) An orbital tissue hyperintensity is present on the sagittal STIR image (TR: 4,700 ms, TE: 42.11 ms), indicating orbital inflammation in the right eye.

myasthenia gravis. Ocular discharge culture was positive for *Staphylococcus hominis*. Blood culture was not performed.

Brain magnetic resonance imaging and angiography (MRI/MRA) showed normal configuration. Contrast-enhanced MRI was not performed because the patient was allergic to contrast agent. Orbital MRI revealed mild right eyelid swelling, lacrimal gland enlargement, and orbital fat hyperintensity on an axial short TI inversion recovery image, which was suggestive of orbital inflammation (Fig. 2). Based on these results, a diagnosis of acute dacryoadenitis was made.

The patient was treated with oral sultamicillin tosilate (1,125 mg/day) for 10 days, along with ocular instillation of moxifloxacin and fluometholone, which rapidly improved his symptoms. Follow-up oculomotor evaluation performed 17 days after treatment revealed resolution of ptosis and normal ocular motility on the right side (Fig. 3). Orbital MRI at this time showed improvement of right eyelid swelling, lacrimal gland enlargement, and orbital inflammation (Fig. 4).

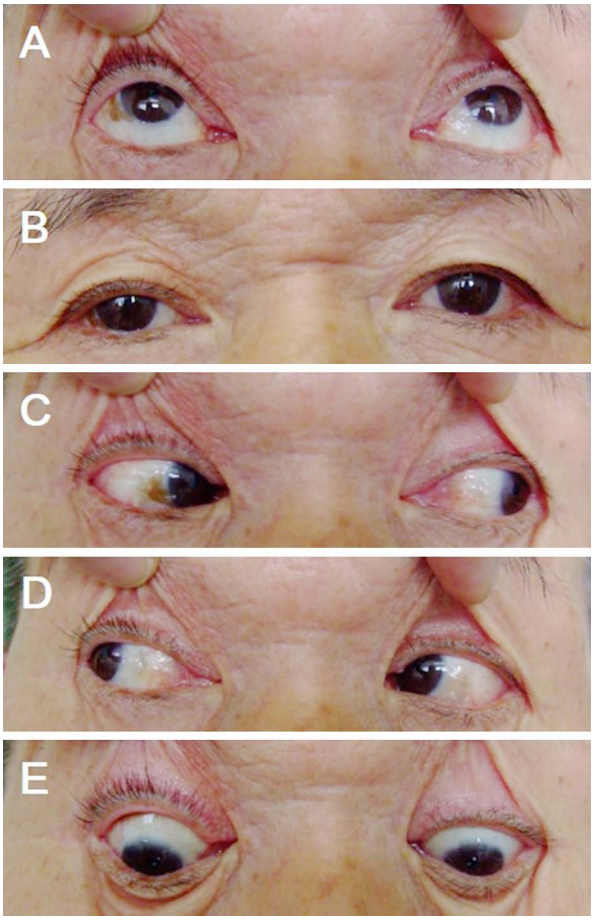

Fig. 3 Ocular motility examination after treatment.
Photographs taken 17 days after treatment in an attempted upward gaze (eyelids retracted by examiner, A), the primary position (B), an attempted leftward gaze (eyelids retracted by examiner, C), a right gaze (eyelids retracted by examiner, D), an attempted downward gaze (eyelids retracted by examiner, E). Ptosis and ophthalmoplegia in the right eye had improved.

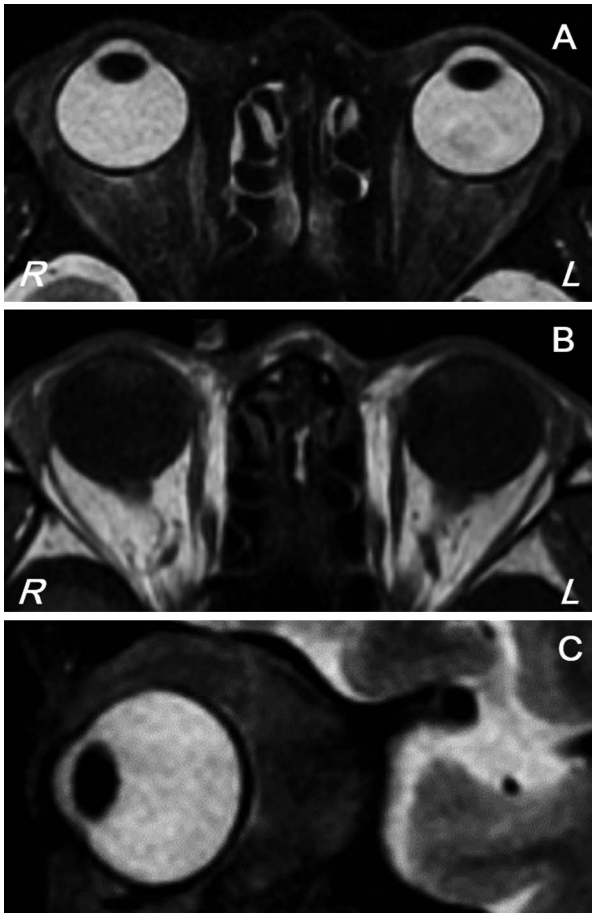

Fig. 4 Orbital magnetic resonance images after treatment.
(A) An axial short TI inversion recovery (STIR) image (TR: 4,700 ms, TE: 41.98 ms) shows resolution of mild right eyelid swelling and lacrimal gland enlargement. (B) An axial T_1-weighted image (TR: 540 ms, TE: 12 ms) shows a normal lacrimal gland and orbit. (C) A sagittal STIR image (TR: 4,760 ms, TE: 42.66 ms) reveals improvement of orbital inflammation in the right eye.

Discussion

Acute dacryoadenitis occurs in approximately 1 in 10,000 ophthalmic outpatients[4]; severe eyelid redness and tenderness commonly occur with the disorder and limited ocular movement is suggestive of orbital cellulitis[1]. The disorder generally results from a causative viral (e.g., mumps, measles, influenza, infectious mononucleosis and herpes virus[5)6)]) or bacterial (e.g., *Staphylococcus aureus*, *Streptococcus pneumonia*, *Streptococcus pyogenes*, *Haemophilus influenza*, and *Neisseria gonorrhea*) infection[5)~8)]. Common routes of such orbital infections include sinusitis, insect bites, and ocular trauma[9]. However, these causes were excluded in our patient. The causative pathogen in our patient was thought to be *Staphylococcus hominis*, based on ocular discharge cultures, although the bacterium belongs to normal bacterial flora, and the efficacy of antibiotic treatment.

This patient had a unique presentation of acute dacryoadenitis, in which noticeable symptoms were ophthalmoplegia and ptosis compared to local infectious signs. This is surprising because severe painful eyelid swelling is the predominant clinical feature of the disorder. Although this case showed conjunctival hyperemia and slight eyelid redness, these findings were not enough to strongly suspect involvement of orbital inflammation. However, these minor finding may provide an important clue to the diagnosis because these symptoms suggest ophthalmic disorders. The advanced age of the patient might explain why ptosis and ophthalmoplegia occurred without severe painful eyelid swelling, because the connective tissue of the eyelid can become loose with age. The orbital inflammation might have extended to the external ocular muscles, resulting in the ophthalmoplegia,

because the lacrimal gland is closest to the upper eyelid elevator muscle and the superior rectus muscle. Additionally, the superior branch of oculomotor nerve might be involved in the symptom. Downward overshoot upon right ocular movement may have resulted from eyeball compression by the enlarged lacrimal gland and inflamed orbital fat.

Painless acute dacryoadenitis presenting as ophthalmoplegia and ptosis may be misdiagnosed for other disorders, including cerebral aneurysm, myasthenia gravis, diabetic ophthalmoplegia, and Fisher's syndrome. A misdiagnosis could lead to treatment delays, and orbital inflammation may rapidly progress to severe orbital cellulitis, which may lead to worsening of the ophthalmic prognosis. Therefore, neurologists should be aware of acute dacryoadenitis without the local severe inflammatory findings mimicking neurological diseases.

In conclusion, acute dacryoadenitis should be considered in the differential diagnosis in elderly patients with ptosis and ophthalmoplegia, even in the absence of severe painful eyelid swelling and redness.

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※ The authors declare there is no conflict of interest relevant to this article.

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