Rare disease

Sequential right then left acute dacryoadenitis in Crohn's disease

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Summary

A 41-year-old woman presented with painful swelling of the lateral aspect of the right upper lid along with ipsilateral conjunctival injection. Her right eye had limited abduction and pain on attempted lateral gaze. Funduscopy was normal. Medical history included Crohn's disease treated with sulphasalazine and azathioprine. White blood cell count, erythrocyte sedimentation rate and C reactive protein were raised. CT of orbits revealed right-sided lacrimal gland enlargement, with signs of abscess formation within it. She was treated initially with intravenous antibiotics for 3 days and then switched to orals but it took 2 weeks for full resolution of symptoms. Eight months later, she presented again with signs of acute left-sided dacryoadenitis, not responding at all to oral antibiotics after 3 days. She was subsequently started on oral steroids, and her symptoms resolved within 48 h. Steroids were tapered without any recurrence after a period of 6 months.

BACKGROUND

- ▶ Acute dacryoadenitis is a rare ophthalmic manifestation of Crohn's disease. There are only a few case reports in the literature, but a late recurrence of dacryoadenitis has not been documented so far.
- ▶ Although the first episode in our patient did resolve after intravenous antibiotics, recovery and resolution of pain was much faster with systemic steroids.

CASE PRESENTATION

A 41-year-old Caucasian woman presented with a 1-week history of a swollen, painful and red right eye. She had already had a 5-day course of oral flucloxacillin 500 mg four times a day from her general physician (GP) but without any improvement in her symptoms.

Medical history included Crohn's disease (proctocolitis) diagnosed 3 years earlier treated with sulphasalazine 1 g and azathioprine 150 mg daily.

On examination, corrected visual acuity was 6/12 in the right eye and 6/6 in the left eye. There was swelling mainly on the lateral aspect of the right upper lid (figure 1), which was tender on palpation, along with right bulbar conjunctival injection and chemosis. On assessment of extraocular movements, she had limited right eye abduction, with pain, worse on lateral gaze. No proptosis was noted. Pupils and fundi were normal. Her temperature was 37.5°C and she was otherwise systemically well.

INVESTIGATIONS

White blood cell count was 11.2, with neutrophils 8.67. Erythrocyte sedimentation rate (ESR) measured 88 mm/h and C reactive protein (CRP) 244 mg/l. Thyroid function tests were normal. Extractable nuclear antigens, antinuclear antibodies (ANAs) and antidouble-stranded DNA antibodies (anti-dsDNA) were negative.

Contrast-enhanced CT of the orbits showed an enlarged right lacrimal gland, with signs of an early abscess formation within the centre of the gland (figure 2). There was no evidence of cavernous sinus thrombosis, bone involvement or subperiosteal abscess.

Figure 1 Presenting right eye condition, with signs of acute right dacryoadenitis. There is swelling mostly of lateral aspect of the right upper lid. Right chemotic conjunctiva can be also noted.

Figure 2 Axial CT of orbits, which shows right enlarged lacrimal gland, pushing right eye globe and lateral rectus muscle. There are some areas of low attenuation within the centre of the gland, suggesting developing abscess formation. No bony involvement.

DIFFERENTIAL DIAGNOSIS

1. Acute dacryoadenitis (more commonly idiopathic, bacterial or viral)
2. Orbital cellulitis
3. Orbital myositis
4. Cavernous sinus thrombosis
5. Arteriovenous fistula
6. Thyroid eye disease
7. Ruptured dermoid cyst
8. Orbital tumour
9. Lacrimal gland tumour

TREATMENT

Because of the diagnosis of right acute dacryoadenitis, she was admitted for intravenous broad spectrum antibiotics, ceftriaxone 1 g twice daily and oral metronidazole 500 mg thrice daily, for the first 3 days. Then, she was discharged on oral ciprofloxacin 750 mg twice daily and metronidazole 400 mg thrice daily, for the next 10 days.

OUTCOME AND FOLLOW-UP

Within 2 weeks, her symptoms had entirely settled and the dacryoadenitis had resolved with no sequelae off antibiotic treatment.

However, 8 months later, she presented urgently with symptoms of painful left acute dacryoadenitis. Vision, eye movements and intraocular examination were all normal.

She was treated initially with oral co-amoxiclav 625 mg and metronidazole 400 mg, both thrice daily. Three days later, there was no clinical improvement (figure 3) and her ESR had increased from 71 to 100 mm/h with a CRP of 105 mg/l. White blood cell count had increased from 7.5 to 12.3, with neutrophil count of 9.9. Conjunctival swabs showed no microorganism growth.

In the light of probable Crohn's-related dacryoadenitis, oral prednisolone 40 mg daily was started and dramatic improvement followed over the next 48 h. Steroids were tapered by 10 mg/month and she made a full recovery.

No recurrence has been noted after a period of 6 months.

Figure 3 This photo shows the recurrent episode of acute dacryoadenitis on the left side this time. Left upper lid appears ptotic, swollen and inflamed.

DISCUSSION

The incidence of ocular manifestations in Crohn's disease can be up to 10%, with episcleritis, scleritis and uveitis occurring more frequently than retinal, orbital or neuro-ophthalmic complications.^{1 2}

Although the occurrence of orbital inflammation in Crohn's disease is recognised, dacryoadenitis is rare and extremely unusual to happen twice in one person. In the literature, there are only four cases of lacrimal gland involvement associated with Crohn's disease and two of those were bilateral.²⁻⁵ Three of them responded well to a short course of oral steroids while the fourth proved self-limiting, and immunosuppression was not necessary. Relapse of ipsilateral lid swelling within a month happened in one case.

This is the first reported case of a relatively late contralateral recurrence of acute dacryoadenitis occurring as an extraintestinal manifestation of Crohn's colitis.

Steroid treatment resulted in a dramatic resolution of the disease process within the first 48 h.

Learning points

- Dacryoadenitis can be associated with Crohn's disease.
- Resolution may be possible with antibiotic treatment alone.
- Steroid treatment results in much faster resolution of symptoms.

Competing interests None.

Patient consent Obtained.

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