

CLINICAL INVESTIGATIONS

Chronic Dacryoadenitis Misdiagnosed as Eyelid Edema and Allergic Conjunctivitis

Kaan Gündüz, İlhan Günalp and Rabia Gürses Ozden

Ocular Oncology Service, Eye Clinic, Faculty of Medicine, Ankara University, Ankara, Turkey

Purpose: To report the case of a 53-year-old woman with a 2-year history of episodic upper eyelid swelling and nonspecific complaints, who was diagnosed as having allergic conjunctivitis.

Methods: A complete ocular examination, orbital computerized tomographic (CT) scans followed by complete physical and systemic examinations.

Results: The results of physical and systemic examinations were unremarkable for systemic lymphoma and a primary focus of cancer. The results of the ocular examination were normal. CT scans demonstrated well-defined lesions bilaterally with a homogeneous internal structure in the lacrimal gland fossa, which suggested a diagnosis of chronic dacryoadenitis. The differential diagnosis included lymphoma and orbital metastases. The patient refused a biopsy and was started on a tapering dose of 60 mg oral prednisolone daily. The follow-up CT scans 1 month after cessation of 6-week oral corticosteroid treatment showed near complete resolution of the orbital lesions.

Conclusion: This case demonstrates that orbital inflammation can be misdiagnosed as refractory allergic conjunctivitis. **Jpn J Ophthalmol 1999;43:109–112** © Japanese Ophthalmalogical Society

Key Words: Allergic conjunctivitis, corticosteroids, dacryoadenitis, eyelid swelling, idiopathic orbital inflammation.

Introduction

Orbital pseudotumor is a term referring to a spectrum of idiopathic orbital inflammations but excludes neoplastic, infectious, and systemic inflammatory or immunologic etiologies.¹ This term generally has been replaced by idiopathic orbital inflammation. Especially excluded from this generic term are the various forms of lymphomas including benign reactional lymphoid hyperplasia and atypical lymphoid hyperplasia. Kennerdall and Dresner² divided idiopathic orbital inflammations into diffuse and localized types, and further subclassified the localized type into four categories: myositis, dacryoadenitis, periscleritis, and perineuritis.³

In this report, we present a case of presumed chronic dacryoadenitis. The symptoms of eyelid edema and nonspecific ocular complaints led to a misdiagnosis of allergic conjunctivitis.

Case Report

A 53-year-old woman was referred to us with a 2-year history of eyelid swelling, ocular itching, burning, and foreign body sensation. She was diagnosed as having allergic conjunctivitis and was treated with various antiallergic eye drops including fluoromethalone 0.1%, iodoxamide 0.1%, sodium cromoglycate 2% drops, dexamethasone acetate 0.1% ointment and oral antihistamines. The symp-

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Address correspondence and reprint requests to: Kaan GÜNDÜZ, MD, G.M.K. Bulvari 1163, Maltepe 06570, Ankara, Turkey.



Figure 1. Facial view of patient showing upper eyelid edema.

toms waxed and waned over several months, but did not completely resolve.

At referral, the ocular examination showed upper eyelid edema (Figure 1), moderate conjunctival injection, and mild upper tarsal papillae. Visual acuity was 20/20 in both eyes. The results of ocular examinations were otherwise within normal limits. There was no sign of dry eye and the Schirmer test showed normal results. There was no proptosis and an orbital mass was not palpable. However, there was some puffiness overlying the lacrimal gland fossa bilaterally.

Orbital computerized tomographic (CT) scans demonstrated well-defined masses with a homogeneous internal structure in the lacrimal gland fossa bilaterally (Figure 2). Retroocular fat planes and extraocular muscles were normal. There was no bone erosion. The tomographic appearance, together with the clinical findings, suggested a diagnosis of dacryoadenitis. The differential diagnosis included orbital lymphoma and metastatic tumors.

The patient refused an orbital biopsy. To rule out orbital involvement from systemic lymphoma and orbital metastasis, complete physical and systemic examinations were performed. Results of the physical examination were normal. The patient did not have a history of allergic rhinitis, asthma, or salivary gland enlargement. The findings of laboratory studies were unremarkable for hemoglobin, leukocyte count, platelet count, erythrocyte sedimentation rate, serum chemistries including calcium, serum proteins, eosinophils, IgE levels, and serum angiotensin-converting enzyme. Sinus and chest radiographs, electrocardiogram, and urinanalysis data were unremarkable. In addition, results of serum protein electrophoresis, and patient data on C3 and C4 complement levels, Cl esterase inhibitor, antinuclear antibody, single and double-stranded DNA, antinuclear cytoplasmic antibody, purified protein derivative, Kveim-Siltzbach tests, and bone marrow biopsy were also within normal limits. Computerized tomography of the bones, abdomen, and brain showed nothing abnormal.

Although the systemic examinations showed negative results, the possibility of a primary orbital lymphoma could not be ruled out. The situation was discussed with the patient. She elected to be treated with oral corticosteroids and 60 mg oral prednisolone daily was prescribed. The medication was tapered over 6 weeks. At the follow-up examination 1 month later, the eyelid edema had decreased considerably (Figure 3). The follow-up CT scan showed near complete resolution of the orbital lesions (Fig-



Figure 2. Initial computerized tomographic scan showing well-defined homogeneous masses in lacrimal gland fossa bilaterally with normal retro-ocular fat planes, extraocular muscles, and bone structures.



Figure 3. Facial view of patient after systemic corticosteroid treatment showing resolution of upper eyelid edema.



Figure 4. Computerized tomographic scan after systemic corticosteroid treatment demonstrating near complete resolution of lesions in lacrimal gland area bilaterally.

ure 4). The patient has been followed for 2 years with no evidence of recurrence.

Discussion

The clinical course of idiopathic orbital inflammation can be acute or chronic.⁴ Acute idiopathic orbital inflammation develops over the course of days and is characterized by the abrupt onset of pain, lid swelling, erythema, conjunctival injection, extraocular muscle dysfunction, impaired visual acuity, proptosis, and a palpable mass. The chronic form of idiopathic orbital inflammation develops over the course of weeks to months. Chronic idiopathic orbital inflammation may be characterized by proptosis, orbital mass, pain and visual acuity decrease, or it may present with nonspecific signs and symptoms such as eyelid swelling or conjunctival injection that may resemble allergic conjunctivitis, as occurred in this case.⁵

Significant advances have been made in the diagnosis of idiopathic orbital inflammation syndromes through CT scanning and magnetic resonance imaging.^{2,6–8} Important features revealed by imaging are retrobulbar fat infiltration, proptosis, extraocular muscle enlargement, thickening of the optic nerve and sclera, edema of Tenon's capsule, and an enlarged lacrimal gland conforming to the shape of the globe. Our patient had bilateral, well-defined, homogeneous orbital masses in the lacrimal gland area which suggested a diagnosis of dacryoadenitis although the possibility of orbital lymphoma and metastasis could not be ruled out. Chronic dacryoadenitis is the most common lesion of the lacrimal gland fossa. It accounted for 54% of all biopsies of the lacrimal fossa lesions and for 7% of all orbital biopsies in the series of Shields and associates.⁹ In many cases of chronic dacryoadenitis, the etiology of the inflammatory process cannot be precisely determined, whereas in others, sarcoidosis, tuberculosis, leprosy, and parasitic infestations, can be identified as the underlying factor.⁴ In our patient, there was no underlying etiologic factor.

There is controversy as to whether patients with presumed idiopathic orbital inflammation should undergo biopsy or not. It is generally accepted that biopsy may not be necessary in all patients with idiopathic orbital inflammation and should generally be reserved for cases with suspected neoplasms, patients refractory to corticosteroids and other immunosuppressives, and those with uncertain diagnosis. On the other hand, Moseley and Wright¹⁰ stated that biopsy must be performed on most patients with idiopathic orbital inflammation because a number of infective, inflammatory, and especially neoplastic conditions may also respond to corticosteroids, leading to an incorrect diagnosis of orbital pseudotumor.

Other authors have pointed out that the problem is more complicated because even if a histopathologic diagnosis was obtained, there was still the possibility that reactive lymphoid hyperplasia might be confused with idiopathic orbital inflammation.¹¹ Furthermore, lymphoma has been reported to arise in histologically benign pseudotumor cases.¹² As for the timing of the biopsy procedure, it is generally accepted that biopsy should be performed in the stable period of the disease and not during the acute stage.¹³

We did not perform a biopsy in our patient because she refused the procedure initially and the orbital lesions bilaterally responded completely to systemic corticosteroids. Therefore, although the diagnosis of chronic dacryoadenitis remains presumptive, the diagnostic investigations failed to disclose systemic lymphoma or a primary focus of cancer. We recognize that orbital lymphoma can occur as a primary disease without systemic involvement. Although orbital lymphoma or, more rarely, metastatic foci may respond to oral corticosteroid treatment, near complete resolution without recurrence, as in our patient, is very unusual.

Several authors have reported good success rates in idiopathic orbital inflammation after treatment with 60–100 mg oral prednisone.^{14,15} Patients with focal encapsulated lesions and dacryoadenitis responded less favorably compared to those with diffuse lesions and acute myositis.¹ We have managed 11 dacryoadenitis cases during 1971 to 1994 whose 1-year follow-up examination results after treatment were available. Six of 11 cases were resolved with corticosteroids; 1 of these 6 cases still had recurrent disease.¹³ It has been suggested that the chronic forms of orbital pseudotumor often respond less well because of significant fibrosis and collagen deposition into the orbital tissues.⁵ This reported case with chronic dacryoadenitis improved readily with 60 mg oral corticosteroid treatment.

In conclusion, this case demonstrates that chronic orbital inflammation may present with signs and symptoms leading to an erroneous diagnosis of refractory allergic conjunctivitis. Such patients with suspected orbital inflammation should be evaluated by CT or magnetic resonance imaging to reveal the orbital lesions.

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