Chronic bilateral lacrimal gland pseudotumor in an adult

Dear Editor,

A 38-year-old male presented to us with a history of bilateral painless upper eyelid swelling over the lateral region increasing slowly in size over a period of seven years. On examination there was fullness at the lateral aspect of the upper eyelid in either eye giving rise to S-shaped ptosis. There was associated mild injection of the palpebral portion of the lacrimal gland and the adjacent conjunctiva. Best corrected visual acuity was 20/20 in both eyes. Other ocular and systemic examinations were normal.

Ultrasound of the lesions showed a heterogeneous mass in the lacrimal gland area. Computed tomography scan revealed heterogeneous lesion in both lacrimal glands extending into preseptal tissue with no evidence of bony erosion [Fig. 1]. Fine needle aspiration cytology (FNAC) from the right lacrimal gland was suggestive of pleomorphic adenoma, however, incisional biopsy was advised for confirmation. A week later, incisional biopsy was undertaken, which showed lymphocytes, plasma cells and other features suggestive of pseudotumor.
He was started on oral prednisolone 1.5 mg/kg/day, with which he showed dramatic response. The swelling disappeared by second week of treatment. Steroids were tapered over the next two months. However, he presented again with similar swellings bilaterally six months later [Fig. 2]. The same treatment was instituted but this time steroids were tapered very slowly over a period of six months. He did not have any further recurrence in the last one year.

Idiopathic orbital inflammation, also known as orbital pseudotumor (OP), is a term describing a spectrum of idiopathic, non-neoplastic, non-infectious, space-occupying orbital lesions without identifiable local or systemic cause. It may present as dacroadenitis, sclerotenovitis or as a diffuse anterior soft tissue inflammation. Bilateral OP may be seen in children, however, unilateral disease is the rule in adults. Isolated bilateral lacrimal gland pseudotumor in adults is exceedingly rare. Lacrimal gland pseudotumors tend to be recurrent and refractory to treatment.

Pseudotumor, solely limited to the lacrimal gland may harbor malignancies, especially if chronic enlargement of gland is present alike our case. The FNAC diagnosis and differentiation of benign from malignant primary orbital lymphoid lesions is often difficult. When the results are equivocal open biopsies are recommended for confirmation as was required in our case.

This case underscores the need of prolonged systemic steroid therapy in cases of bilateral lacrimal gland pseudotumor and the limited utility of FNAC in establishing the diagnosis of lacrimal gland diseases.

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References