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Unilateral Dacryoadenitis as the First and Predominant Manifestation in a Patient with Idiopathic Orbital Inflammation Mimicking Thyroid-Associated Orbitopathy

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Abstract **Case Report**

A 52-year-old female presented with left eyelid swelling and redness. An orbital magnetic resonance imaging (MRI) showed a swollen left lacrimal gland. On laboratory examination, she was positive for thyroid autoantibodies. A diagnosis of idiopathic orbital inflammation with thyroid autoantibodies was made based on clinical symptoms, laboratory findings, and MRI findings. She was treated with oral prednisolone. On orbital MRI, lacrimal gland swelling improved to normal appearance after steroid treatment. Coexistent idiopathic orbital inflammation and thyroid-associated orbitopathy is extremely rare. This case highlights the importance for clinicians to be aware of a concomitant diagnosis of idiopathic orbital inflammation and thyroid-associated orbitopathy.
Keywords: idiopathic orbital inflammation, dacryoadenitis, thyroid-associated orbitopathy.

INTRODUCTION

Idiopathic orbital inflammation (IOI) is defined as a benign, noninfective clinical syndrome characterized by features of nonspecific inflammatory conditions of the orbit without identifiable local or systemic causes. The clinical features of IOI are highly variable ranging from a diffuse to very focal process targeting specific orbital tissues, such as the lacrimal gland, extraocular muscles, and orbital fat [1, 2]. Thyroid-associated orbitopathy (TAO) is often mentioned as a disease to exclude in the diagnosis of IOI [1, 2].

Herein, we discuss a case of IOI associated with unilateral dacryoadenitis mimicking TAO.

CASE REPORT

A 52-year-old female presented with painless left eyelid swelling and redness, which developed two months before. She had no history of smoking, and a non-significant familial history. Her thyroid gland examination was unremarkable. Serum levels of TSH (thyrotropin), FT3 (triiodothyronine), and FT4 (thyroxine) were 0.123 μIU/mL (normal: 0.500-5.000), 4.03 pg/mL (normal: 2.30-4.00) and 1.30 ng/dL (normal: 0.90-1.70), respectively; TRAb (TSH receptor antibody (third generation)) level was 4.4 IU/mL (normal; < 2.0 IU/mL), TSAb (thyroid stimulating antibody) was 314 % (normal; < 180 %), TgAb (anti-thyroglobulin antibody) < 10 IU/mL (normal; < 28 IU/mL), and TPOAb (antithyroid peroxidase antibody) 9.0 U/mL (normal; < 16.0 U/mL), respectively. A complete blood count, serum electrolyte, renal function and liver function tests were normal. The level of serum C-reactive protein (CRP) was 0.06 mg/dL (normal; < 0.30 mg/dL). The serum levels of IgG and IgG4 were 1,100 mg/dL (normal: 870-1,700 mg/dL) and 24.1 mg/dL (normal; 4-105 mg/dL), respectively. Test for antinuclear antibodies was negative. Based on these laboratory findings, a diagnosis of TAO was considered. Orbital magnetic resonance imaging (MRI) showed markedly swollen left lacrimal gland extending to the upper eyelid (Figure 1a, b). The extraocular muscles were not enlarged (Figure 1c), and exophthalmos was not observed.

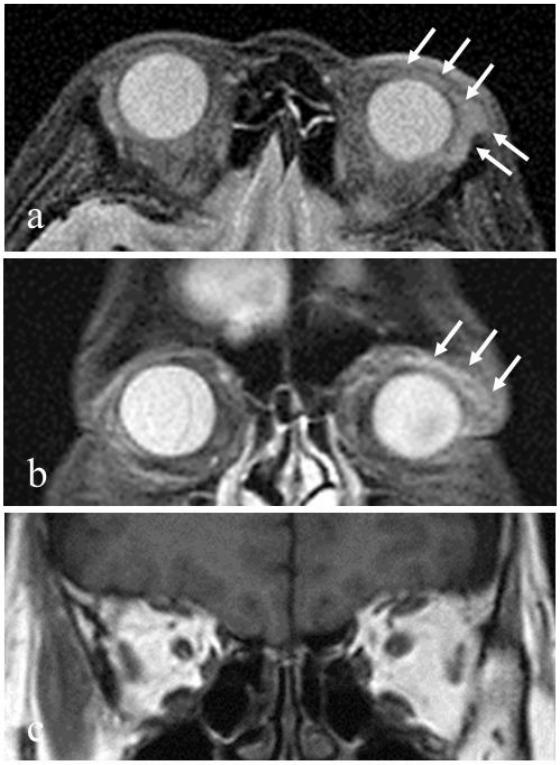

Fig-1: Axial (a) and coronal (b) T2-weighted MRI showed swollen left lacrimal gland extending to the upper eyelid (arrows). The extraocular muscles were not enlarged (c: coronal T1-weighted MRI)

She was referred to an endocrinologist, who excluded TAO. She was referred to our hospital for further evaluation. Ocular examination revealed left upper eyelid retraction. No reductions in visual acuity were observed. Eye movements were not restricted. No diplopia was observed. A diagnosis of IOI with thyroid autoantibodies was made based on clinical symptoms, laboratory findings, and previous MRI findings. She was treated with oral prednisolone (25 mg/day). One month following steroid therapy, the symptoms showed gradual amelioration. The steroid dosage was gradually reduced over several months and tapered off. On orbital MRI, left lacrimal gland swelling improved to normal appearance after steroid treatment (Figure 2a). In addition, the serum levels of TSH, FT3, and FT4 improved to 0.500 μIU/mL, 3.19 pg/mL, and 1.23 ng/dL, respectively. The patient was doing well and there were no signs indicative of recurrence.

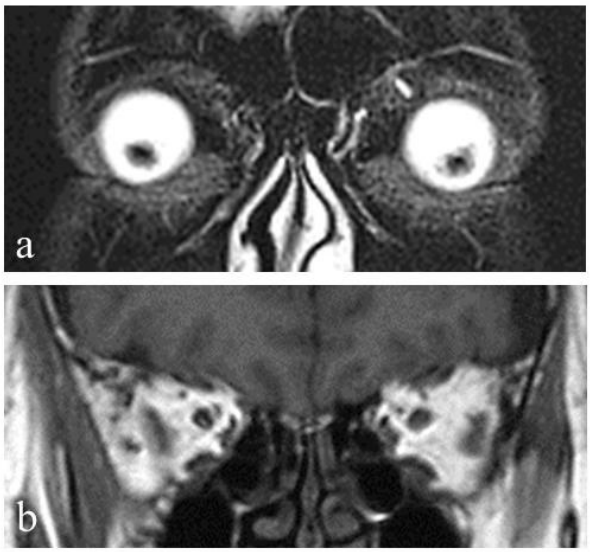

Fig-2: Coronal (a) T2-weighted MRI showed left lacrimal gland swelling improved to normal appearance after steroid treatment. The extraocular muscles were not enlarged (b: coronal T1-weighted MRI)

Discussion

We present a case of IOI where unilateral lacrimal gland enlargement was the predominant early clinical sign. In this patient, we initially suspected TAO based on endocrinological investigations. However, in brief, serum FT4 level was within the normal range, TgAb and TPOAb were negative, and MRI showed no extraocular muscle enlargement. In addition, lacrimal gland enlargement as an early clinical or radiological sign in TAO is extremely rare [3]. Therefore, we considered that these findings were compatible with IOI rather than TAO.

By definition, IOI excludes lesions with identifiable local or systemic etiology [1, 2]. Thyroid-associated orbitopathy is an important differential diagnosis in patients with IOI; TAO has characteristic manifestations such as eyelid retraction, eyelid lag, proptosis, restrictive extraocular myopathy, and optic neuropathy [1]. Radiological findings include enlargement of extraocular muscles and an increase in orbital fat volume [1, 4]. These findings may occur in a euthyroid setting in the absence of any objective thyroid dysfunction as the initial presentation of the disease process or after adequate control. Generally, a number of clinical and radiological features distinguish IOI from TAO [1, 4]. Abrupt onset of pain and inflammatory signs, such as periorbital erythema and swelling, are typical early manifestations of IOI. In contrast, TAO has a slower, more insidious course, and extraocular motility dysfunction and visual disabilities tend to occur later in the disease process. Radiological findings for IOI are typically unilateral and may involve any of the orbital structures, including the extraocular muscles, tendons, lacrimal gland, orbital fat, perineural connective tissues, Tenon capsule, and sclera [1, 4]. In contrast, TAO has findings that are typically bilateral, and the primary focus is on the enlargement of extraocular muscles and increased orbital fat volume.

Although definitively distinguishing between IOI and TAO is difficult in this case, we suggest a common etiology as an autoimmune-like disease among IOI and TAO. The coexistence of IOI and TAO is extremely rare [5, 6]. Bijlsma *et al*. [5] described 4 patients with both IOI and TAO *separated* in time of onset and localization in the orbit. Shen *et al.* [6] reported 3 patients with concomitant, *simultaneous* IOI and TAO. The finding of both IOI and TAO in the same patient could be explained by the tendency of autoimmune diseases to occur together. Therefore, the theory that TAO automatically rules out IOI is not necessarily true.

Conclusions

Although our current findings were based on a single case, long-term follow-up and additional cases will need to be examined so that these rare and unusual associations between IOI and TAO can be definitively characterized. This case highlights the importance for clinicians to be aware of a diagnosis of concomitant IOI and TAO.

Disclosure: The author declares no conflict of interest.

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