Case Report: Inflammatory Pseudotumor of the Orbit and Buccal Space

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ABSTRACT

Educational Objective: Participants should have an understanding of inflammatory pseudotumor (IPT), its imaging features, histological features, and management.

A 31 year old female presented to the emergency department with a three month history of orbital pain and visual changes. On examination, she was noted to have a 2 cm right buccal mass as well as right eye proptosis and diplopia affecting right lateral and vertical gaze. CT and MRI imaging revealed a 4 x 3 x 1 cm lesion within the right orbit with buccal extension and displacement of orbital contents medially. Biopsies from the buccal space and the orbit were histologically consistent with inflammatory pseudotumor. The patient responded to a one month steroid taper with near resolution of her extraocular movement limitations and diplopia. She was subsequently lost to follow-up.

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ABSTRACT

Educational Objective: To present a rare case of inflammatory pseudotumor of the orbit with buccal space extension and discuss appropriate evaluation and treatment options.

Case report and literature review.

METHODS

Case report and literature review.

RESULTS

A 31 year old female presented with a one month history of right eye pain and visual changes. On examination, she was noted to have a 2 cm right buccal mass as well as right eye proptosis and diplopia affecting right lateral and vertical gaze. CT and MRI imaging revealed a 4 x 3 x 1 cm lesion within the right orbit with buccal extension and displacement of orbital contents medially. Biopsies from the buccal space and the orbit were histologically consistent with inflammatory pseudotumor. The patient responded to a one month steroid taper with near resolution of her extraocular movement limitations and diplopia. She was subsequently lost to follow-up.

INTRODUCTION

Inflammatory pseudotumors are idiopathic, space occupying lesions, most commonly affecting the orbit. They are frequently mistaken for neoplastic processes. Non-orbital inflammatory pseudotumors of the head and neck have been reported in the paranasal sinuses, salivary glands, nasopharynx, larynx, and trachea.1 They present an unusual case of an inflammatory pseudotumor presenting with simultaneous masses in the orbit and the buccal space.

DISCUSSION

In 1905, Britch-Hirshfield described the first reported case of inflammatory pseudotumor (IPT), located in the orbit.1 Multiple terms have been used to describe this process in the literature, including inflammatory myofibroblastic tumor, inflammatory myofibrohistiocytic proliferation, inflammatory thymus tumor, sarcomaous pseudotumor, postinflammatory pseudotumor, plaque cell malignant xanthoma, and inflammatory xanthoma.2,3 IPT is most commonly found in the orbit. However, non-orbital IPT has been reported in the maxillary sinus, parotid, sinonasal, lacrimal, dural, parotid, and cranial nerves.4-6 Non-orbital IPT has no predilection for gender or age. It usually presents as a progressively enlarging, space occupying lesion which can cause symptoms related to mass effect. Presentation includes a vague sense of fullness, headache, and epiphora.

Although the etiology of IPT is unknown, its responsiveness to steroids suggests an immune-mediated process. IPT typically responds to high-dose corticosteroids within 2-3 weeks, which differentiates these lesions from neoplastic lesions. Acute IPTs have a rapid presentation, and histologically have a mostly lymphocytic component. Chronic IPTs are more fibrous. As a result, acute lesions have a greater reduction in size in response to steroids. While neoplasms and other inflammatory processes may also decrease in size in response to steroids, they do so at a much slower rate. A less dramatic response to steroids has been noted in adults with chronic inflammation. For those patients with a contraindication to steroid use, low dose radiation therapy can be considered.

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