**Eosinophilic Sialodochitis: A Rare Comorbidity of Severe Asthma**

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Diseases of the parotid gland vary widely, the most frequent being obstructive sialadenitis, which can be caused by mucus plugs, salivary stones, or anatomic anomalies, although in some cases the cause remains unclear [1-2].

The relationship between recurrent swelling of the parotid gland and mucus plugs containing leukocytes and Charcot–Leyden crystals was first described in 1876 by Kussmaul [3]. Since then, similar cases, also involving eosinophilia or high serum IgE levels, have been diagnosed as “allergic parotitis” or “eosinophilic sialodochitis” [4-6,8].

We report the case of an 80-year-old woman with severe persistent eosinophilic asthma, chronic rhinosinusitis with nasal polyps with good tolerance to nonsteroidal anti-inflammatory drugs, bronchiectasis, allergic bronchopulmonary aspergillosis, and eosinophilic esophagitis. She was initially treated with omalizumab for 2 periods (2009-2013 and 2016-2021).

In the last 25 years, she has experienced countless episodes of bilateral parotid gland swelling. These were initially mild and self-limiting but became progressively worse in intensity and frequency over the years, affecting her quality of life. The episodes were not related to a food or drug. She noticed that the oral corticosteroids (OCS) she took for asthma exacerbations not only improved the exacerbations, but also reduced the number of parotiditis episodes. Therefore, she started taking short courses of OCS solely to treat parotiditis, as her asthma control improved significantly while receiving omalizumab.

She has been evaluated by many specialists. Below, we summarize the investigations performed.

Computed tomography of the parotid glands showed no sialolithiasis, lymphadenopathy, or other organ involvement. The cervical lymph nodes were not enlarged.

Given the recurrent and alternating additional symptoms in response to OCS and xerostomia, the patient was referred to the systemic autoimmune diseases department with an initial suspected diagnosis of Sjögren syndrome. Results for ANA, Anti-Ro/La, Ro52, and ANCA were all negative.

Eye tests and parotid scintigraphy ruled out keratoconjunctivitis sicca, and scintigraphy revealed moderate-to-severe dysfunction (grade 3), with a marked decrease in salivary gland uptake and delayed excretion.

Given the clinical and analytical discrepancy, the study was expanded with a parotid magnetic resonance scan, which showed moderate hypertrophy, fatty infiltration on both parotid glands, and diffuse minimal enhancement with dilatation of the parotid ducts, all compatible with “chronic parotitis”.

Minor salivary gland biopsy revealed non-specific chronic sialadenitis. There was no histological evidence of Sjögren syndrome, amyloid deposit, or sarcoid granulomas. Immunohistochemistry for IgG4 was negative. The periductal infiltration was lymphocytic, and eosinophils were not observed, probably owing to the ongoing therapy with OCS.

Throughout the process, periodic analyses showed elevated total IgE levels ranging between 368 and 3017 kU/L, peripheral eosinophilia between 130 and 1320/mm³, and normal IgG, IgA, IgM, and IgG4. Testing for the fusion gene FIP1L1-PDGFRα was negative, as was the ANCA titer. This, together with the absence of other manifestations suggestive of vasculitis, also ruled out the diagnosis of eosinophilic granulomatosis with polyangiitis.

With the suspicion that the inflammation responsible for recurrent parotiditis was due to eosinophilia, a cytological analysis of the parotid secretion was performed. Fine needle aspiration cytology of the salivary glands revealed normal ductal epithelium cells with mild inflammation, macrophages, and eosinophilic granulocytes. As eosinophils were the only inflammatory cell present, an intraductal eosinophilic inflammatory process was suspected. Following these analyses, the patient was diagnosed with eosinophilic sialodochitis.

The patient initially responded to omalizumab. OCS for asthma exacerbations were suspended, thus triggering multiple episodes of parotid gland involvement and forcing her to resort to OCS at least twice per month. Given the good clinical control of asthma and the need to take recurrent OCS cycles for episodes of parotid swelling, it was decided to suspend omalizumab in 2013; the asthma was controlled for 2 years with an inhaled corticosteroid, a long-acting β-agonist, and a long-acting muscarinic antagonist. However, after the progressive loss of asthma control led to the need for continued OCS therapy in 2016, omalizumab was restarted. The patient's...
We present a case of eosinophilic sialodochitis that was successfully treated with benralizumab. Eosinophilic sialodochitis is an infrequent disease that has classically been associated with allergy and/or eosinophilia, which we now refer to as type 2 inflammation. It should be considered in patients with type 2 inflammation who show suggestive symptoms. Benralizumab is a therapeutic option in these cases owing to its absolute antieosinophilic action. It can be prescribed off-label in the absence of severe eosinophilic asthma, its currently approved indication.

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Conflicts of Interest

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