Plasma cell gingivitis, an underestimated condition in patients with Sjögren’s syndrome?

Sir,

We would like to share with the medical and scientific community this case that describes a plasma cell gingivitis in a woman with primary Sjögren’s syndrome (pSS).

A 61-year-old woman was referred to hospital dental department for comprehensive care. Her medical history revealed a IgG kappa multiple myeloma (MM) in complete remission after intensive chemotherapy followed by autologous stem cell transplantation 8 years before, a hypothyroidism treated by L-thyroxine and several depressive episodes. She complains from dry mouth, dry eyes, and sore and bleeding gums when brushing for the past 2.5 years. A severe carious disease and moderate generalized periodontitis (stade II grade B) associated with hyposalivation were diagnosed (Fig. 1A). A diagnosis workup to search for pSS revealed objective ocular dryness without keratitis, positive AAN antibodies (high titers=1:2560, with no specificity), and minor salivary gland biopsy revealed a focal sialadenitis (Focus score =1.4) confirming the diagnosis of pSS according to the classification of the 2016 ACR/EULAR (1). The patient did not present other immunological abnormalities (normal gammaglobulin and complement level, no cryoglobulin), or systemic manifestations.

The clean-up treatment with tooth extraction, caries treatment and a periodontal treatment (oral hygiene instruction and scaling and root planning), lead to clinical improvement. However, gums remained locally red and swollen at 6 weeks and the patient still complained about soreness and bleeding on brushing (Fig. 1B). As oral mucosal lesions in patients with pSS had been described in the literature (2), a gingival biopsy was performed. It revealed a highly vascularised inflammatory cellular infiltrate made of polytypic plasma cells both kappa and lambda. IgG4 staining revealed no evidence of significant IgG4 plasma cell infiltration. Together, these features were consistent with a diagnosis of plasma cells gingivitis (3). Due to the patient medical history, a relapse of MM or flare of autoimmune disease was investigated. MM was still in complete remission and the polytypic nature of the plasma cell infiltrate excluded a possible link between MM and this gingivitis. Moreover, no systemic flare of pSS was observed. We investigated allergenic factors in the diet (red pepper, chewing-gum) and cosmetic (herbal toothpaste) but we didn’t find any. Nevertheless, we decided to change the patient’s toothpaste to one that did not contain sodium lauryl sulphate, a very common and highly allergenic component, and prescribed a topical corticoid treatment (40 mg of prednisolone mouthwash twice a day during three months) (6). No improvement was observed reinforcing the idea of a non-allergic etiology (Fig. 1C). Plasma cells gingivitis is a rare disease that could be linked to type IV hypersensitivities with allergenic (4) or idiopathic (5) origin. Interestingly, it has also been described in patients with auto-immune disease, i.e., secongenous rheumatoid arthritis (6) and coeliac disease (7). pSS is a rare auto-immune disease that is characterized by lymphocytic infiltration of the exocrine glands leading to their destruction and dry syndrome. Gingiva inflammation is current in patients with pSS because of the absence of humidification, lubrication, clearance, buffering capacity, and local immunity (8). Herein, the local role of a lack of saliva could not be excluded. On the other hand, the gum could also be a place of inflammation of the pSS autoimmune process. Interestingly, Likar-Manookin et al., reported in a multi-center retrospective cohort study on 155 pSS patients that 12% had oral mucosa lesions of auto-immune etiology, mainly lichen planus and aphthous stomatitis, and rarely chronic ulcerative stomatitis, but no case of plasma cells gingivitis (2). Therefore, in case of refractory gingivitis in a patient with pSS, plasma cell gingivitis might be search since the treatment of this entity differ from that of classical pSS involvement.

References