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

Sirs,

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 generalised 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, the 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 gingiva 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 were 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 did not 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 aetiology (Fig. 1C).

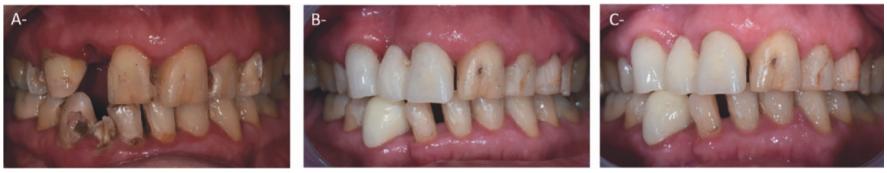

Fig. 1. A: Initial clinical view of the gum. The patient suffered from a serious carious disease and a moderate generalised periodontitis with redness and oedema of the gum, and generalised dryness (mouth, eyes). The clinical characteristics of the gum are coherent with the importance of the dental lesions, the bad control of plaque and oral dryness. **B**: Clinical view after treatment of carious disease and periodontitis. After tooth extraction, treatment of dental caries, temporary oral rehabilitation and periodontal treatment, the gums remain red and swollen at the level of the marginal gum and the papillae. Spontaneous bleeding is visible around the right maxillary canine. The aspect of the gums is not coherent with a controlled oral hygiene and a stabilised periodontal disease. In parallel, a diagnosis of pSS was made. **C**: Clinical view after local treatment of the gingiva with a topical corticosteroid for 3 months to treat plasma cell gingivitis. Note that clinical signs of inflammation of the gums persist, indicating a specific form of gingiva in a patient with pSS.

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.* seronegative rheumatoid arthritis (6) and coeliac disease (7). pSS is a rare auto-immune disease that is characterised by lymphocytic infiltration of the exocrine glands leading to their destruction and dry syndrome. Gingiva inflammation is current inpatients 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-centre retrospective cohort study on 155 pSS patients that 12% had oral mucosa lesions of auto-immune aetiology, mainly lichen planus and aphtous 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.

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Competing interests: none declared.

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