CASE REPORT

Kuttner tumor involving minor salivary glands in a patient undergoing radiotherapy in the head and neck

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Summary Kuttner tumor (KT) was first described in 1896 by Kuttner. It occurs mainly in the submandibular gland and usually presents as a firm and painful swelling. An important aspect of KT is its clinical resemblance to a salivary gland neoplasm. Histologically, the disease is characterized by progressive periductal sclerosis, acinar atrophy, and gland infiltration by lymphocytes. However, for a reliable diagnosis of KT an immunohistochemical analysis of lymphocyte subtypes is required. We report a case of KT that involved minor salivary glands of a patient that was undergoing radiotherapy in the head and neck region.

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Introduction

Kuttner tumor (KT) was first described in 1896 in Germany by Kuttner.1 It occurs mainly in the submandibular gland of middle-aged adults,2 usually as a firm and relatively painful swelling.3 Indeed, it is classified as a tumor-like lesion of the salivary glands due to its clinical similarity to a neoplasm.4 Histologically, KT is characterized by progressive periductal sclerosis, acinar atrophy, and gland infiltration by lymphocytes.3 Recent research has shown that in KT a T-lymphocyte immune reaction predominates.5

The aim of this paper is to report a case of KT that involved minor salivary glands of a patient that was undergoing radiotherapy in the head and neck region. This is the first described case associated to radiotherapy in the head and neck region.

Case report

A 33-year-old Caucasian male was referred to the Oral Oncology Service of the School of Dentistry at the Federal
University of Minas Gerais (UFMG) for dental treatment. The patient had been surgically treated for a squamous cell carcinoma of the palpebra. He would be further submitted to 31 sessions of radiotherapy. At the 24th radiotherapy session, a submucosal swelling in the right buccal mucosa was noticed during clinical examination. Provisional clinical diagnosis was of an inflammatory fibrous hyperplasia. The patient underwent seven more sessions of radiotherapy and when he returned there was now a tri-nodular, asymptomatic, normal-colored, 3.0 cm swelling in the right buccal mucosa (Fig. 1(A)). Clinical diagnosis was of a mesenchymal neoplasm and an incisional biopsy was performed. Histological examination revealed oral mucosa fragments with minor salivary gland tissue and monocytic infiltrate (Fig. 1(B) and (C)). The salivary gland tissue showed periductal monocytic cells infiltration (lymphocytes and plasma cell), slight increase in duct diameter and atrophy of acini (Fig. 1(C) and (D)). Paraffin-embedded tissues were immunostained for CD20 (Dako Corporation, Carpinteria, CA, USA; clone L26; dilution 1:50, antigen retrieval with 0.01 M citric acid, 95 °C, 30 min), VS38c (Dako Corporation, Carpinteria, CA, USA; clone M7077; dilution 1:50, antigen retrieval with 0.01 M citric acid, 95 °C, 30 min), CD3 (Dako Corporation, Carpinteria, CA, USA; Clone M0835; dilution 1:50, antigen

Figure 1  (A) Clinical aspect showing tri-nodular, asymptomatic, normal-colored, 3.0 cm swelling in the right buccal mucosa. (B) Oral mucosa with minor salivary gland tissue in the deep lamina propria (hematoxylin and eosin stain; 100× magnification). (C) Minor salivary gland tissue associated with monocytic cells (hematoxylin and eosin stain; 200× magnification). (D) Lymphocytes and plasma cell (*), duct diameter increased (arrow head) and atrophy of acini (arrow) are observed in the minor salivary gland tissue (hematoxylin and eosin stain; 400× magnification). (E) Scarce CD20-positive lymphocytes between the monocytic cells (streptavidine–biotine, 400× magnification). (F) Abundant CD45RO-positive lymphocytes between the monocytic cells (streptavidine–biotine, 400× magnification). (G) Abundant CD8-positive lymphocytes subsets between the monocytic cells (streptavidine–biotine, 400× magnification). (H) Eighteen months follow-up demonstrated no signs of clinical recurrence.
neously affecting parotid and submandibular glands, and less, unusual cases affecting the parotid gland, simultaneously affecting both parotids, both submandibular sialoliths, the effects of infectious agents, secretory dysfunction, duct abnormalities and immune processes have also been considered. In our case, it is very likely that the alteration of the salivary flow induced by radiotherapy lead to development of the lesion. Moreover, direct damage to the salivary gland may have played a role as a possible etiological factor. In either case, this would be the first report of a KT consequent of radiation treatment, and hence the lesion may be included in the scope of radiotherapy-induced oral lesions in the head and neck region.

We presented a case of a KT that exclusively affected minor salivary glands. To our knowledge, this has not yet been reported. Furthermore, it may be the first radiotherapy-induced described case in the literature.

Discussion

The clinical, histological and immunohistochemical features of the presented case qualifies it as a case of KT. It has recently been stressed that this lesion has been under-recognized and awareness of this process has been emphasized.

The clinical presentation of KT varies from an asymptomatic swelling to a recurrent pain, and some authors state that KT only affects the submandibular gland. Nevertheless, unusual cases affecting the parotid gland, simultaneously affecting parotid and submandibular glands, and simultaneously affecting both parotids, both submandibular and minor salivary glands, were recently reported. A case that bilaterally affected the submandibular and lacrimal glands was also described. This may be the first report of KT limited to minor salivary glands.

Histological and immunohistochemical examination is required for definitive diagnosis of KT. The disease was staged by Seifert and Donath into four stages. Our case was in stage 1, i.e., focal periductal lymphocytic infiltration, slight increase in duct diameter and acini atrophy. In order to obtain definitive diagnosis we performed immunophenotyping (CD45RO, CD3 and CD8) were more pronounced (Fig. 1(E)–(G)). Histological diagnosis of chronic sclerosing sialadenitis, stage 1, was obtained. The remaining portion of the lesion resolved spontaneously within ten days. The patient has been followed-up for 18 months with no signs of recurrence (Fig. 1(H)).

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References