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Case report

A case of rheumatoid nodules in the buccal subcutaneous : A novel case report of review and the literature

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ABSTRACT

Methotrexate (MTX) has been reported to be effective for the treatment of rheumatoid arthritis (RA) when a weekly low dose is administered to patients. We carried out microscopic examination of a biopsied specimen suspected of being Wegener's granulomatosis, IgG4-related disease, or malignant lymphoma. Finally we diagnosed a rheumatoid nodule because his medical history included treatment with MTX. Then we immediately discontinued the MTX and his rheumatoid nodule disappeared after 2 months. Rheumatoid nodules are granulomatous nodules that appear subcutaneously and in tendons in the active phase of RA and commonly occur on the exterior part of the elbow joint and the back of the head where subcutaneous tissue is subject to recurrent pressure against bone. The development of an intraoral rheumatoid nodule in the maxillofacial region of a patient with RA who received MTX has not been few reported. To date, this patient has been followed up for 6 years without recurrence.

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1. Introduction

Rheumatoid nodules are granulomatous nodules of the tendon sheath or subcutaneous tissue that appear in the active phase of rheumatoid arthritis (RA), and generally are one of the painless extra-articular symptoms of RA. They occur in about 25% of RA patients, and are a diagnostic sign of the disease [1,2]. Accelerated nodulosis with multiple rheumatoid nodules is induced by MTX administration, though it ameliorates the symptoms of arthritis [3]. In the process of pharmacological action, MTX migrates to the bone and synovial membrane, achieving a high concentration resulting in anti-inflammatory action. In other words, the concentration in synovial fluid reaches about 10 times that in blood, whereas it is relatively low outside the joints [4]. Although the anti-inflammatory pharmacological action of long-term administration of MTX is effective for arthritis, the suppressive effect of extra-articular symptoms such as rheumatoid nodules and vasculitis is weak, which causes these symptoms. It has been reported that rheumatoid nodules occur in sites subjected to pressure, such as the

back of the head and the elbow extension side [5], and also in the thyroid gland [6]. However, cases in the maxillofacial area are very rare [7].

On the other hand, the use of methotrexate (MTX) for RA can cause lymphoma during treatment. The primary site of the MTX-induced lymphoma can be various tissues such as those of the lung [8,9], axillary lymph nodes [10], ovary [11], and lower lip [12].

We suspected Wegener's granulomatosis, IgG4-related diseases, or malignant lymphoma from his biopsy and initial pathological findings. We finally diagnosed of rheumatoid nodules from a re-biopsy and his history of MTX treatment prior to examination.

2. Case report

In November 2011 a 60-year-old man with RA who was treated for 4 years with MTX was referred to our department by his dentist because of the formation of a 36 mm x 15 mm mass with diffuse swelling of his right buccal region. We noted diffuse swelling with erythema in that region and swelling of elastic lymph nodes with tenderness in the submandibular region. We suspected a malignant tumor due to the intra- and extraoral findings (Fig. 1A, B) and mass formation in the right buccal region on computed tomography (CT) (Fig. 1C). Fluorodeoxyglucose (FDG)-positron emission tomog-

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K. Ogi et al. / Journal of Oral and Maxillofacial Surgery, Medicine, and Pathology xxx (2018) xxx-xxx



Fig. 1. His intraoral findings show swelling with redness of the buccal mucosa (A) and his extraoral findings also show swelling of the right cheek (B). Axial CT shows a mass spreading in the buccal region (C).

raphy (PET)-CT showed accumulation of FDG in the right buccal region (SUV max; 11.5) and in the right submandibular lymph nodes (SUV max; 4.6). Then we performed biopsy of the right upper canine gingiva in January 2012, which revealed a hard elastic mass.

Histopathological findings revealed that lymphoid cells invaded the stroma with fibrosis and coagulation necrosis (Fig. 2A). Staining findings led us to suspect granulomatous vasculitis in the elastic fibers. However, we could not make a definitive diagnosis from the

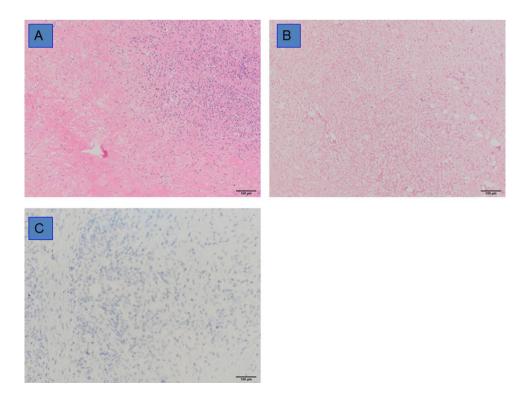


Fig. 2. Hematoxylin and eosin (H-E) staining shows fibrinoid necrosis surrounded by palisaded histocytes and lymphocytes (A). Immunohistochemistry was negative for EBER-1 (B) and CD56 (C). (Magnification: X10).

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