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Case report

A case of myeloid sarcoma in the soft palate

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ABSTRACT

Myeloid sarcoma occurs commonly in various parts of the body, however it is rare in the oral cavity, especially, in the soft palate. The purposes of this paper were to report a rare case of 68-year-old Japanese man who was diagnosed with acute myeloid leukemia due to the onset of a myeloid sarcoma in the palate and discuss the treatment or prognosis of this lesion based on a review of the literature.

1. Introduction

Myeloid sarcoma is also called granulocytic sarcoma or chloroma and is defined as a myelocyte-derived mass-forming tumor based on the WHO classification of tumors of haematopoietic and lymphoid tissues [1,2]. This tumor is closely related to acute myeloid leukemia (AML) and chronic myelogenous leukemia (CML). It occurs commonly in various parts of the body, such as the lymph nodes, bone, spinal cord, soft tissue, and skin, but it is rare in the oral cavity [1–4]. We report here the case of a patient who was diagnosed with AML due to the onset of a myeloid sarcoma in the palate, and we include a review of literature.

2. Case report

The patient was a 68-year-old Japanese man who visited the Department of Oral and Maxillofacial Surgery, Mie University Graduate School of Medicine for a chief complaint of soreness of the palatal mucosa. He had no remarkable family or medical history. The patient suffered pain in the area of the palate to the pharynx for 1 month before the initial examination by a local physician. A mass was found in the soft palate, and the patient was referred to our department in the hospital for further examination and treatment.

No systemic abnormalities and no abnormalities were observed in the face, but a 13 mm × 16 mm sharply circumscribed tumor spread from the soft palate to hard palate on the left side of the oral cavity (Fig. 1).

A panoramic X-ray image at the initial examination revealed no particular problems, and in a MRI, T1-weighted images show that the lesion from the oral cavity side to the pharynx side of the soft palate was similar signal to surrounding tissues. In addition, there was a protrusion in the left lateral wall of the oropharynx. T2-weighted images showed that the lesion had high intensity but low intensity was observed inside the above-mentioned protrusion in the pharynx. Contrast T1-weighted images showed a slightly higher contrast effect in the lesion, but the enhancement was observed only on the surface of the protrusion in the pharynx (Fig. 2). FDG-PET indicated a strong accumulation, SUV9.8, from the soft palate to the glottis.

A blood test showed that the white blood cell count and CRP were 13,800/μl and 10.53, respectively. Based on differential leukocyte count, the neutrophil, lymphocyte, basophil, eosinophil, and monocyte counts were 76.1%, 19.5%, 0.1%, 0.3%, and 4.0%, respectively. The findings at the initial examiantion suggested that the lesion might be a maxillary malignant tumor, and a biopsy was performed on the anterior side of the lesion. Histopathological examination revealed the presence of lymphocytic infiltration under the fibrous tissues, but there were no malignant findings. A microbial examination was performed at that time, but bacteria were not detected. A second biposy performed in the central part of the lesion showed similar findings. Therefore, a third biopsy from the pharynx was performed by the department of otorhinolaryngology. Histopathological examination showed tumor cells with

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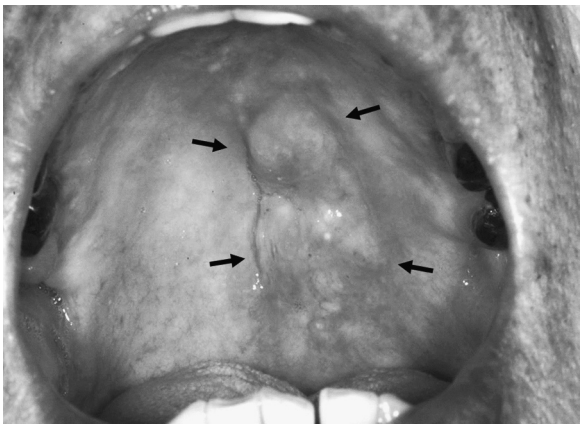

Fig. 1. Intraoral tumor finding at the initial examination. A 13 mm × 16 mm tumor was observed in the soft palate.

eosinophilic granular cytoplasm beneath the epithelium and large atypical nuclei, and revealed the presence of prominent diffuse growth and cell division (Fig. 3A). Immunohistochemical staining showed the lesion was positive both to myeloperoxidase (MPO) (Fig. 3B) and c-kit while negative to TdT antigen. The patient was diagnosed as having myeloid sarcoma based on the above-mentioned findings.

The blood test included with the biopsy showed that the white blood cell count was 15,300/μl and the differential blood test results were neutrophil count 39.0%, lymphocyte count 23.5%, monocyte count 10.5%, basophil count 1.01%, and blast cell count 25.5%. Based on the above-mentioned results, leukemia was suspected and the patient next consulted with a hematology specialist. A bone marrow aspiration biopsy by the hematology department was performed for a definitive diagnosis and revealed a significant growth of monocytoid cells with atypical nuclei. (Fig. 4). The atypical cells were positive for MPO, a nonspecific esterase, and for CD13 and CD33. Based on the results, the patient was diagnosed as having acute monocytic leukemia (AMoL) of the French-American-British Classification.

A combination of daunorubicin and cytarabine was given to the patient in accordance with the AML201 protocol after he was admitted to the hematology department, followed by three courses of consolidation chemotherapy. Three months after the chemotherapy, the tumor in the palate disappeared (Fig. 5). MRI also confirmed the tumor size had decreased. The patient was discharged due to symptom remission. However, the patient had AML relapse 1 month after the discharge and was rehospitalized. The same chemotherapy was performed again but was not effective. The patient died 9 months after the initial onset due to respiratory insufficiency.

3. Discussion

The tumor cells of myeloid sarcoma originate from immature bone marrow cells, and in myeloid sarcoma myeloblasts or undifferentiated myeloid cells form tumors in bones and/or in extramedullarysites [1,2,5,6]. Furthermore, it has been reported that this tumor is related to AML, CML, and myeloproliferative disorder, and that the coincidence rate with AML, all CML, and acute leukemic transformation was 3–8% [7,8], 3.9%, and 4.2%, respectively [6].

According to the classification of myeloid sarcoma by timing of onset, there are 3 types as follows: (i) patients who do not develop leukemia at the onset of myeloid sarcoma (preceding leukemia), (ii) those who have already developed leukemia at the onset (secondary to leukemia), and (iii) those with myeloproliferative disorder or myelodysplastic syndrome as an underlying disease. Neiman et al. [6] reported that the percentage of patients with the above-mentioned types was 50%, 30%, and 20%, respectively, and that the number of patients with sarcoma preceding leukemia type and secondary type was 15 and 11 of 61 patients, respectively. The patient was diagnosed with leukemia by bone marrow biopsy that was performed at the same time as the biopsy from the side of the pharynx diagnosed with myeloid sarcoma. Therefore, we cannot exclude the possibility of secondary type. However, since there were no abnormal findings in blood tests performed at the first biopsy, we diagnosed the case as a preceding

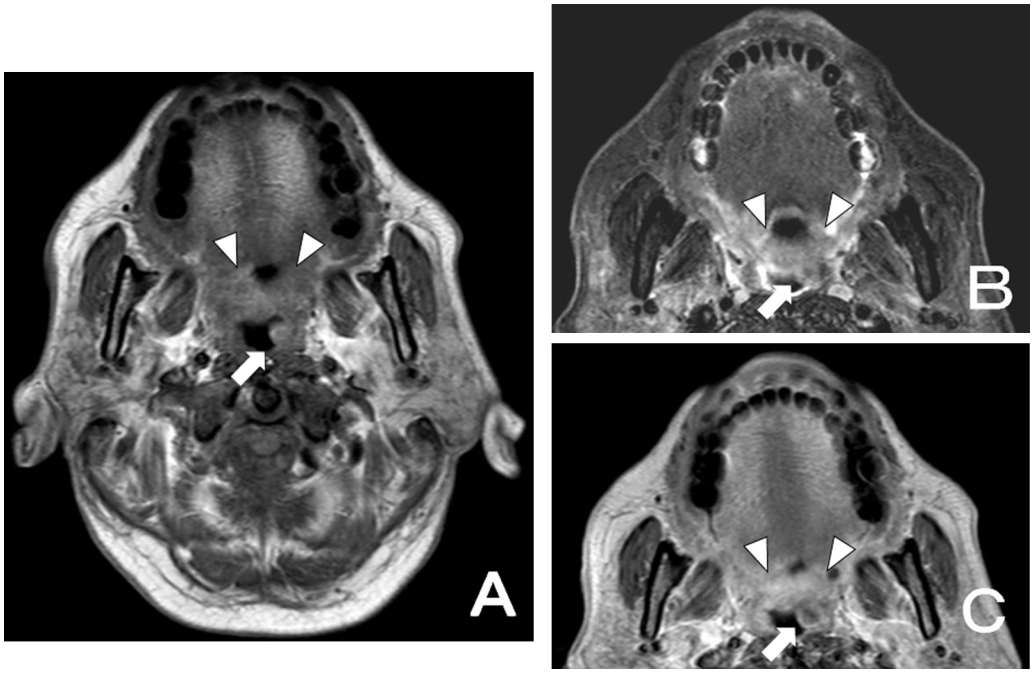

Fig. 2. MRI images. The lesion spread from the soft palate to the pharynx, had similar signal intensity to the surrounding tissues (arrowhead), and a protorusion was observed in the left lateral wall of the oropharynx (arrow) (A: T1, B: T2, C: Contrast T1).