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[Clinical Report]

Postoperative Follow Up with Radionuclide Angiography and Bone Scintigraphy in Malignant Hemangioendothelioma

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Abstract

A case of malignant hemangioendothelioma is presented in which the postoperative observation was carried out by means of radionuclide angiography and bone scintigraphy with ^{99m}Tc labeled methylene diphosphonate. In addition, these two modalities were compared with conventional dental radiography. The angiographic images obtained as a serial scintigrams from the first pass revealed an extremely decreased bone blood flow in the incisor region of the mandible, in which the radiographic images could not be contrasted without contrast media. The static scan bone images showed a well defined bone defect corresponding to the angiographic images and radiographic images. All findings obtained using these modalities showed that radionuclide angiography and bone scintigraphy were useful in following up the postoperative progress of malignant hemangioendothelioma.

Key word : Radionuclide angiography, bone scintigraphy, malignant hemangioendothelioma, malignant vascular tumor

Introduction

Malignant hemangioendothelioma is a rare malignant vascular tumor which is seldom observed in the oral regions. A limited number of case reports regarding this tumor are available in literature.¹⁻⁴⁾ Most of them, however, were examined or followed up primarily only with conventional radiography. Radionuclide angiography and bone scintigraphy were seldom used either in the diagnosis or in the follow up of malignant vascular tumors,^{5,6)} although these modalities were safe and sufficiently sensitive compared with conventional radiography using contrast media.⁷⁻¹⁰⁾

This report presents a case of malignant hemangioendothelioma and shows the usefulness of radionuclide procedures in diagnosing and in following up the postoperative progress of angiomatic malignant tumors which appeared in the oral regions.

Case Report

A 27-year-old man was admitted to the Department of Dental Radiology, Higashi-Nippon-Gakuen University Dental Hospital, with an easily bleeding painless swelling in the incisor region of the mandible. No noteworthy events were seen in the family history or in the patient's past history. The conventional dental radiograms and panoramic dental radiogram taken on the first admission (Fig. 1 and Fig. 2) showed a destructively advanced tumor lesion which was invading the alveolar bone with a moth-eaten appearance. Histological examination of the specimen from the lesion revealed malignant hemangioendothelioma. Then, surgical resection of the tumor lesion was carried out at the Department of Oral Surgery.

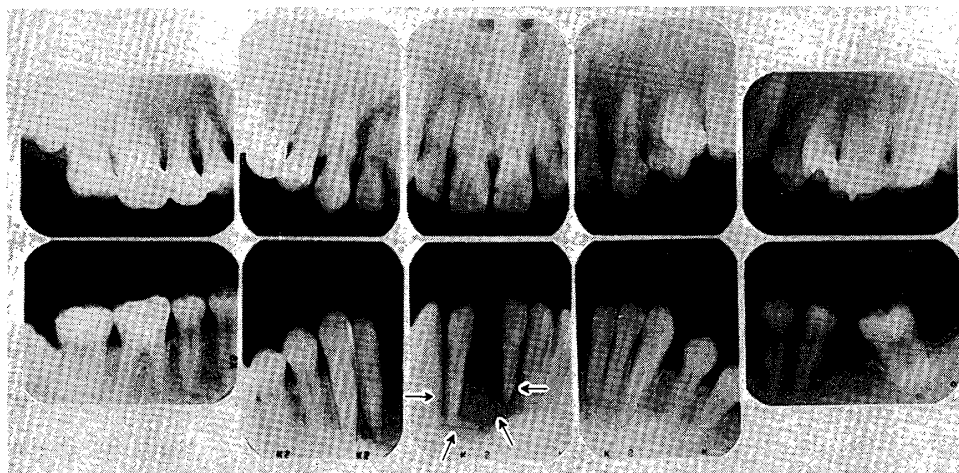


Fig. 1 Full mouth dental radiograms obtained on the first admission, which reveals a destructively advanced tumor lesion and two floating teeth.

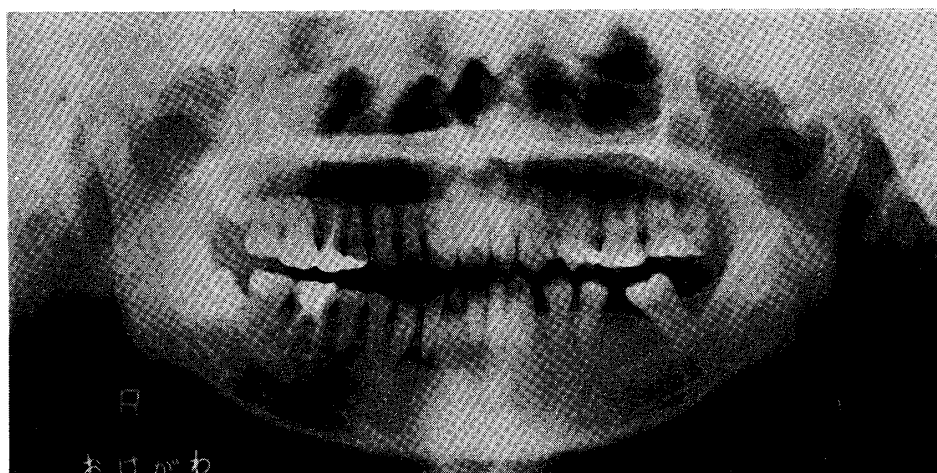


Fig. 2 Panoramic dental radiogram obtained on the first admission, which shows as the same findings as Fig. 1.

In order to examine the postoperative progress of the resected region of the mandible, radio-nuclide angiography was carried out, in addition to static scan bone scintigraphy and conventional dental radiography.

$7.4 \times 10^8 \text{ Bq}$ (20 mCi) of $^{99\text{m}}\text{Tc}$ labeled methylene diphosphonate was administered intravenously. Dynamic scintigraphy was conducted at the same time of the injection, using a gamma camera with a parallel collimeter. Three hours after the injection, static scan bone images were taken.

The angiographic images obtained from the first pass of the blood flow into the resected region (Fig. 3) showed an exceedingly decreased bone blood flow in the incisor region of the mandible, which extended between the left lateral incisor region and the right lateral incisor region. The static scan bone images (Fig. 4) demonstrated a well defined bone defect corresponding to the angiographic images and radiographic images (Fig. 5). The whole body scan images obtained to detect metastases (Fig. 6) demonstrated no abnormalities in the whole body skeleton except for a well defined negative accumulation in the incisor region of the mandible.

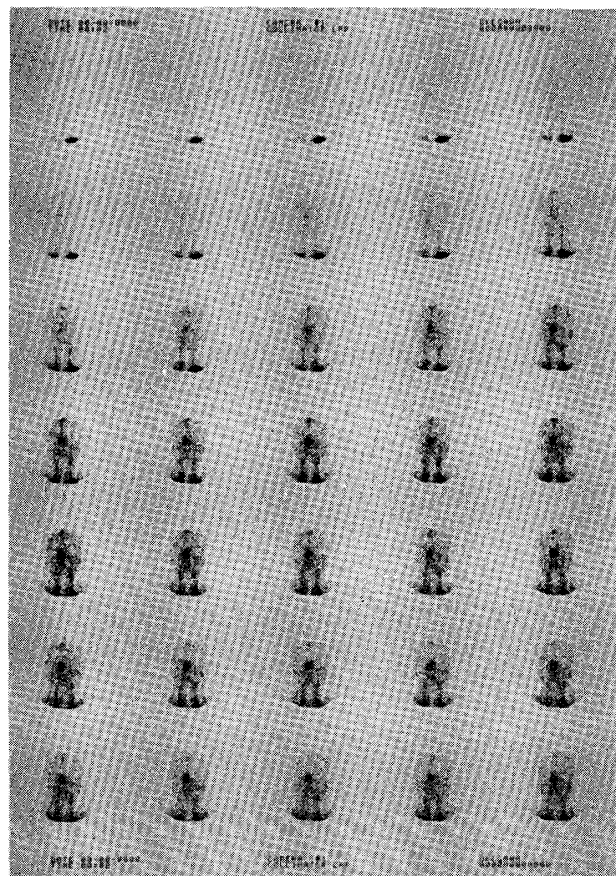


Fig. 3 Serial scintigrams obtained in the postoperative follow up, which demonstrates a decreased bone blood flow in the incisor region of the mandible.

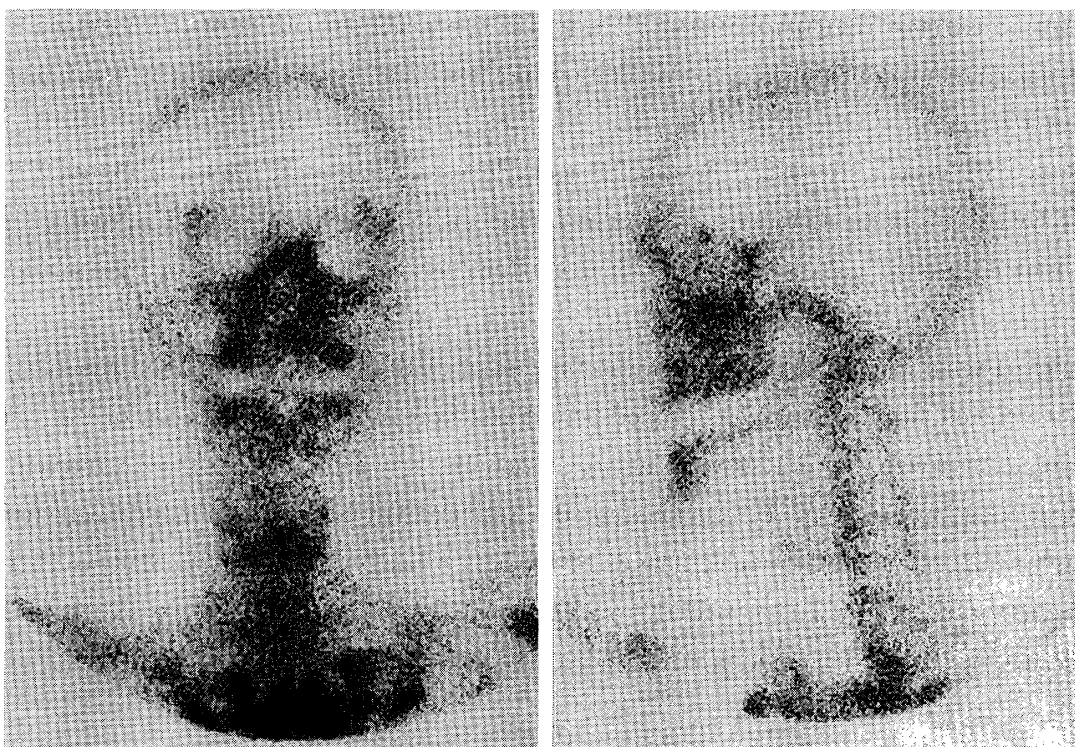


Fig. 4 Static scan bone images obtained in the postoperative follow up, which shows a well defined bone defect in the incisor region of the mandible.

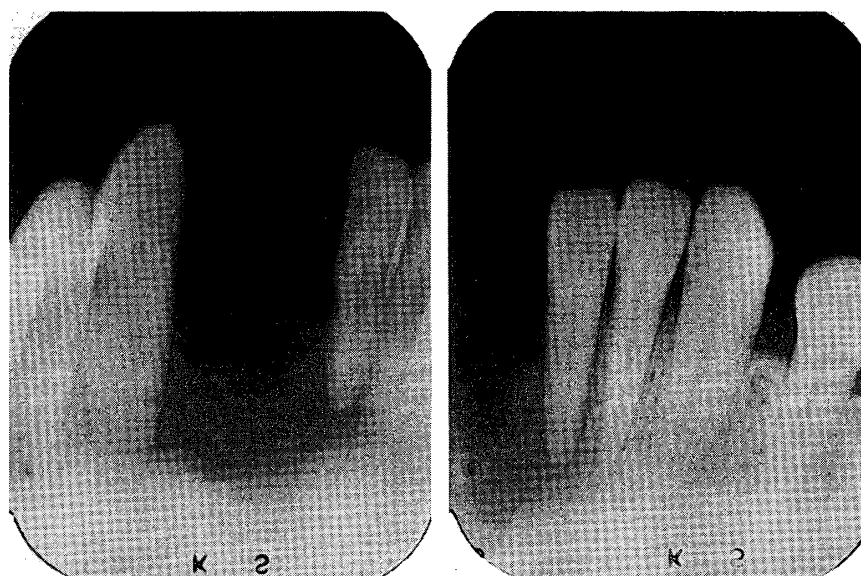


Fig. 5 Dental radiograms obtained in the postoperative follow up.

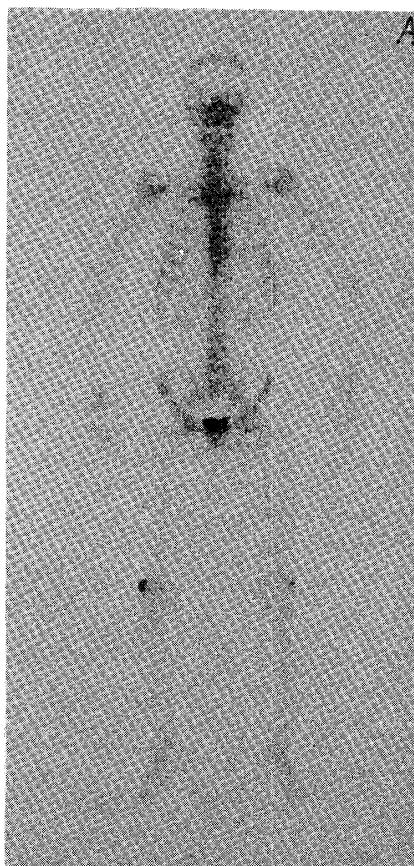


Fig. 6 Whole body scan bone images obtained in the postoperative follow up, which shows no abnormalities in the whole body skeleton, except for the negative accumulation in the mandible.

Discussion

Malignant hemangioendothelioma, although its etiology is still uncertain, is a very rare malignant tumor of vascular origin in the oral regions. This tumor has acquired many other names such as hemangioendothelioma, hemangiopericytoma, hemangiosarcoma, hemangioendothelial sarcoma, angiosarcoma and angiofibrosarcoma. The term hemangioendothelioma was first used by Borrmann (1899)¹¹⁾ when he introduced a complete histological classification of blood vessel tumors. Staut (1943)¹²⁾ further defined the hemangioendothelioma and presented two main criteria for its classification. Lichtenstein (1959)¹³⁾ first proposed that the term malignant hemangioendothelioma should be used for all genuine malignant tumors of vascular origin except for metastasizing hemangiopericytomas. Jaffe (1968)¹⁴⁾ defined malignant vascular tumors as sarcomas which demonstrated a vasoformative capacity as part of their intrinsic natures. Ishikawa and Akiyoshi (1973)¹⁵⁾ proposed that the malignant vascular tumors were classified into two types. One of them is malignant hemangioendothelioma, and the other is malignant hemangiopericytoma. They described that the malignant hemangioendothelioma was characterized by a remarkable proliferation of endothelial cells with vasoformative natures.

In any events, most of these malignant tumors described above have their origins in the various vascular tissues. According to the etiology or characteristics of malignant vascular tumors, radionuclide angiography and bone scintigraphy appears to be helpful to facilitate the diagnosis and to define the extent of the lesions of these malignant tumors. However, only a few reports were available in literature which described these malignant vascular tumors examined or followed up with radionuclide procedures.^{4,5)} Most cases were examined and followed up only with conventional radiography,^{1~4)} although these modalities were proved safe, sensitive and useful in diagnosing the early stage of malignant tumors. Conventional radiography without angiographic procedures failed to reveal the soft tissue lesions. Radionuclide angiography showed that it could detect the tumor blood flow without using contrast media.^{5~9)} Due to the remarkable development of nuclear medicine, radionuclide angiography has become utilized to visualize the peripheral blood flow.

Bone scintigraphy also has been used in examining the dental diseases as frequently as in the medical ailments.^{6,16~19)} But, the utilization ratio in dentistry is lower than that in medicine, because the target areas are limited to the maxillofacial regions. In the dental examinations bone scintigraphy has been used primarily in detecting comparatively rare diseases such as malignant tumors. In most cases, they are limited to the static scan bone images.

In our case, the angiographic images obtained with radionuclide procedure showed clearly an extremely decreased bone blood flow in the incisor region of the mandible, and, the bone scan images could revealed a well defined bone defect in the same area corresponding to the radiographic images and angiographic images. Furthermore, the whole body scan bone images showed no abnormalities in the whole body skeleton. Thus, it was shown that bone scintigraphy is more sensitive than conventional radiography which can only demonstrate the bony lesions when the calcium content in the bone tissues becomes lower or higher over 30%. Only a small change of calcium content in the bony lesions could be available on the bone scan images.

As to the postoperative follow up of malignant hemangioendothelioma reported above, radionuclide angiography and bone scintigraphy were more useful than conventional dental radiography.

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RIアンギオグラフィーと骨シンチグラフィーで 術後観察を行った悪性血管内皮腫の1例

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(主任：金子昌幸 教授)

抄 録

口腔領域の悪性血管内皮腫は、ほとんどの場合、X線検査のみによって、診断や術後観察が行われてきたといえる。われわれは、本腫瘍の術後観察を、RIアンギオグラフィーと骨シンチグラフィーで行い、X線所見と比較検討した。得られたRIアンギオグラムは、下顎骨正中部における血流量の減少を示し、骨シンチグラムは同部の骨欠損像を示した。

これらの所見から、本腫瘍の診断や術後観察に、RIアンギオグラフィーや骨シンチグラフィーは、極めて有用であろうと考えられた。