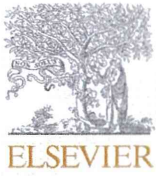


lines discourage tooth extraction in patients at high risk of BRONJ.¹⁸ However, tooth decay with periapical or periodontal disease causes infection and is a major risk factor for BRONJ.¹⁹ Therefore extraction of infected teeth may be preferable.^{20,21} We continued to irrigate the periodontal pockets of the second and third molars even after the teeth fell out, and invasive surgical tooth extraction was avoided.

For suppurative arthritis of the TMJ caused by BRONJ, the diagnosis should be determined by the clinical presentation, imaging findings, joint fluid analysis, and laboratory findings. Adequate antibiotic therapy and joint immobilization can be recommended in the acute stage by examination of the suppurative material. Continuous local irrigation at the TMJ from the preauricular fistula could be necessary, even after the symptoms of inflammation have disappeared.

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Oral Pathology/Case report

A case of malignant melanoma discovered as a result of metastatic disease of the temporomandibular joint[☆]

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ABSTRACT

We report the case of a patient with malignant melanoma discovered upon her experiencing pain when opening her mouth. In 2009, a 42-year-old female patient was referred to the Sakai Municipal Hospital because of preauricular pain when opening her mouth. We suspected a malignant tumor because a simple X-ray showed a radiolucent lesion in the mandibular condyle, and this was confirmed after computed tomographic (CT) and magnetic resonance imaging (MRI) scans. We also detected fluorine-18-deoxyglucose accumulation in the left mandibular condyle, right inferior lobe of lung, and lymph nodes throughout the body during a positron emission tomography PET-CT scan. We performed a biopsy on the bronchoscopic tissue and malignant melanoma was confirmed by histopathology. The patient was subsequently treated at another hospital with systemic chemotherapy along with radiotherapy applied to the left mandibular condyle, but the lymph node tumor enlarged. Accordingly, palliative therapy was given, and the patient died from the tumor 10 months after her first consultation. We could find the malignant melanoma by identifying the radiolucent lesion in the TMJ on panoramic X-ray. Hence, it is crucial not to overlook images indicating bone destruction in the mandibular condyle if there is a malignant tumor in the TMJ.

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1. Introduction

Malignant melanoma occurs in 1.5% of all cancers [1]. It is comparatively rare in the oral mucosa and mucus membrane of the maxillary sinus [2], and a few reports have indicated that it can metastasize into the mouth. Seven cases of metastatic melanoma have reported that occurred in the posterior mandible [3]. On the other hand, metastases to the oral cavity only account for 1–8% of all oral malignancies [4–6]. Because of the sparse blood flow in the mandibular condyle, it is rare for a malignant tumor to metastasize here. We have identified 52 cases similar to the current one [7]. However, most of these cases involved adenocarcinomas and only 2 involved metastatic malignant melanoma [8,9]. This report details the case of a patient with malignant melanoma that was found as a

result of investigations made because the patient experienced pain upon opening her mouth.

2. Case report

The patient was a 49-year-old female. She was referred to the Department of the Oral and Maxillofacial surgery at Sakai Municipal Hospital, for pain in the left temporomandibular joint (TMJ) upon opening of the mouth in 2009. Since the beginning of June 2009, the patient felt pain radiating to the top of her head when chewing food. She also had difficulty in opening her mouth immediately after waking. She did not feel any pain when she was at rest.

Medical history included undifferentiated spondylarthritis which was diagnosed and treated in 2004. She smoked 10 cigarettes per day and drank alcohol occasionally. She was medium build, and had good nutritional status. Other than that, there was no special findings systematically.

The woman's face was symmetrical, and we did not observe any mandibular deviation when her mouth was open. As a symptom outside the oral region, we found slight oppressive pain in the left TMJ when the distance between the incisors was 25 mm (maximum opening width, 42 mm), but no swelling or lumps were observed.

[☆] Asian AOMS: Asian Association of Oral and Maxillofacial Surgeons; ASOMP: Asian Society of Oral and Maxillofacial Pathology; JSOP: Japanese Society of Oral Pathology; JSOMS: Japanese Society of Oral and Maxillofacial Surgeons; JSOM: Japanese Society of Oral Medicine; JAMI: Japanese Academy of Maxillofacial Implants.

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Fig. 1. Extra oral findings: no trismus and no aneurysm in the preauricular region.

The skin over the TMJ was normal, and there was no blistering or rubefaction of her earlobe and external auditory canal (Fig. 1).

We observed slight oppressive pain in the transition area between the maxilla molar part and buccal gingiva as symptoms within the oral region, but the mucous membrane in this area was normal. The patient was able to bite with her molars and no distortion was observed during closed and open biting. The movement of the mandibular condyle appeared to be normal upon palpation.

Panoramic and frontal head X-ray detected a round radiolucent lesion in the area of the left mandibular condyle. No indication of osteolysis in the articular eminence and glenoid cavity of the temporal bone was found (Fig. 2a).

The computed tomography (CT) scans showed bone resorption imaging, which confirmed destruction of the cortical bone at the outside of the left front mandibular condyle (Fig. 2b).

The hemogram was normal in her blood test. C-reactive protein and rheumatoid factor tests were also negative.

Therefore our clinical diagnosis was a malignant tumor on the left mandibular condyle. We prescribed an anti-inflammatory drug and a magnetic resonance imaging (MRI) examination, which revealed the destruction of cortical bone of the front mandibular condyle and infiltration of the lateral pterygoid muscle, and as a result, a malignant tumor was suspected (Fig. 2c).

Accordingly, a positron emission tomography (PET)-CT scan was undertaken because there was a possibility of metastasized malignant melanoma. We detected fluorine-18-deoxyglucose accumulation in the left mandibular condyle, right inferior lobe of lung, bronchial lymph node, liver, right common iliac artery lymph node, and around the hip joint (Fig. 3). Therefore, we consulted with the department of medical oncology about the possibility of lung cancer in the inferior lobe.

Blood tests were also performed, which showed that carcinoembryonic antigen, carbohydrate antigen 19-9, and cytokeratin fragments were within the normal range, and levels of Neuron-specific enolase were high.

We subsequently performed bronchoscopic and histopathologic examination in July (Fig. 4a–c). Microscopically, outgrowth of uniform tumor cells under the mucous membrane of the bronchiole was confirmed. These cells were atypical cell with small densely stained nuclei and pigment-producing cells with brown granules. Epidermal markers were negative while S100 protein and HMB-45 were positive. HMB-45 was stained Premelanosome, showing a positive marker in 80% of malignant melanoma. HMB-45 was positive for the parenchyma of the tumor. Melan-A antigen is specific for the melanocyte lineage, found in normal skin, the retina, and melanocytes, but not in other normal tissues. It is thus useful as a marker for melanocytic tumors (melanomas) with the caveat that it is normally found in benign nevi as well. Melan-A was weak positive. NKI/C3 initially was reported as a marker for a formalin-resistant, melanoma-associated antigen. NKI-C3 was positive. We diagnosed the condyle tumor was metastatic malignant melanoma of the lung.

After the examination, the patient went to the department of dermatology in a cancer center. She was given 2 cycles of chemotherapy with dacarbazine, nimustine, and cisplatin along with 28 Gy irradiation of the tumor on the left TMJ at the cancer center. Although the diameter of the tumor on the mandibular condyle was observed to have reduced on a CT scan performed after radiotherapy, treatment was switched to best supportive care because pathological changes in the lung had progressed despite chemotherapy. The patient was then referred to the affiliated hospital of Kinki University, Faculty of Medicine for palliative care.

Pleural drainage was performed for pleural effusion, and malignant melanoma cells were confirmed in the pleural cell block. Subsequently, we administered picibanil and transferred the patient to palliative care.

A CT scan performed in March 2010 indicated that the left mandibular condyle had been absorbed and had disappeared, (Fig. 5) and preauricular swelling had enhanced. The patient died from her disease 10 months after her first consultation.

3. Discussion

It is very likely that preauricular pain upon opening of the mouth and during mastication will be diagnosed as temporomandibular joint disorder (TMD) because these symptoms are the most common complaints relating to TMD in daily dental and oral surgery practice. However, other possible causes should also be considered as several reports have indicated that patients suffering the exact same symptoms as TMD were diagnosed with malignant melanoma [3,7,10,11].

Malignant tumors in the TMJ are roughly classified into those originating in the TMJ, those caused by a neighboring organ invading the TMJ, and metastasized tumors that have spread to the TMJ [12]. Although the occurrence of a tumor originating in the TMJ is extremely rare, osteosarcoma, chondrosarcoma, and giant cell malignant tumors have been reported. The parotid gland is the most likely neighboring organ from which a malignant tumor can be derived, but oral, maxillary, and pharyngeal tumors are also possible. A metastasized tumor is the most common cause of a malignant tumor detected in the TMJ. On the other hand, malignancy in the TMJ is extremely rare with only 52 other cases identified between 1954 and 2010 [7]. A total of 73% of these cases were adenocarcinomas, which are frequent in bone metastasis from the prostate, mammary glands, lungs, and thyroid glands; there were only 2 cases of malignant melanoma. Hence we can say that this case is very unusual.

Malignant melanoma is a malignant tumor originating in melanocytes although the reason for its occurrence remains

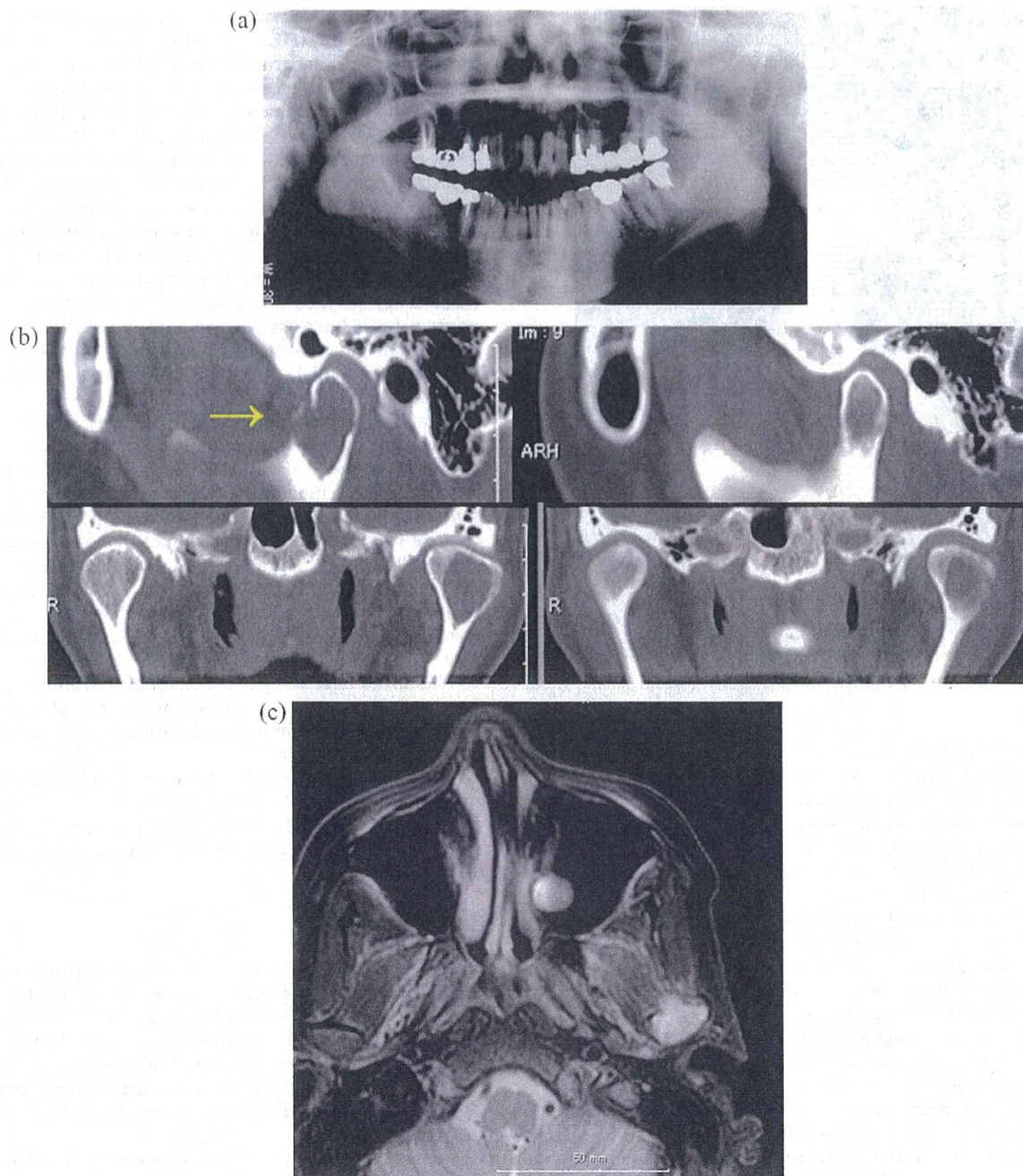


Fig. 2. (a) Panoramic radiographs of the patient at first examination showing a radiolucent lesion in the mandibular condyle. (b) Coronal and sagittal CT (bone window setting) revealed osteolysis of the left mandibular condyle. (c) Axial and sagittal MRI (short time inversion recovery) revealed destruction of the left mandibular condyle and invasion of the lateral pterygoid muscle. (Hydrocele revealed in the left maxillary sinus.)

unknown. Malignant melanoma affects approximately 1500–2000 patients annually in Japan and this number is increasing; 52.4% of patients are female and 47.6% are male, and the incidence of this disease is greatest in those aged 40 years or older.

Malignant melanoma commonly originates in the foot (sole) (27.0%), trunk (13.8%), face and neck (13.8%), upper limbs, nails, and lower limbs [13]. Malignant melanoma has also been reported to occur at the mucosa. Ariyoshi et al. [2] reported the incidence of malignant melanoma as 0.7% of malignant tumors in the oral and maxillofacial region, and a few reports have indicated that it can metastasize into the mouth. For 7 examples of which have reported that occurred in the posterior mandible [3].

Because we found fluorine-18-deoxyglucose accumulation in the bronchi and regional lymph nodes during a PET scan, as for the primary lesion of the malignant melanoma, there was no sign of malignant melanoma including the black lesion which disappeared in the skin. Moreover, because PET-CT scan showed uptakes in the temporomandibular joint, lungs, iliac bone and inguinal lymph node, it was unlikely to be the malignant melanoma that was originated in bone. Also, there are no reports of the cases of malignant melanoma originated in bone to date as much as I can collect, so that we suspected the malignant melanoma that was originated in lungs which I could find a large number of case reports. Metastasis to the TMJ in this case appears to have originated from a tumor

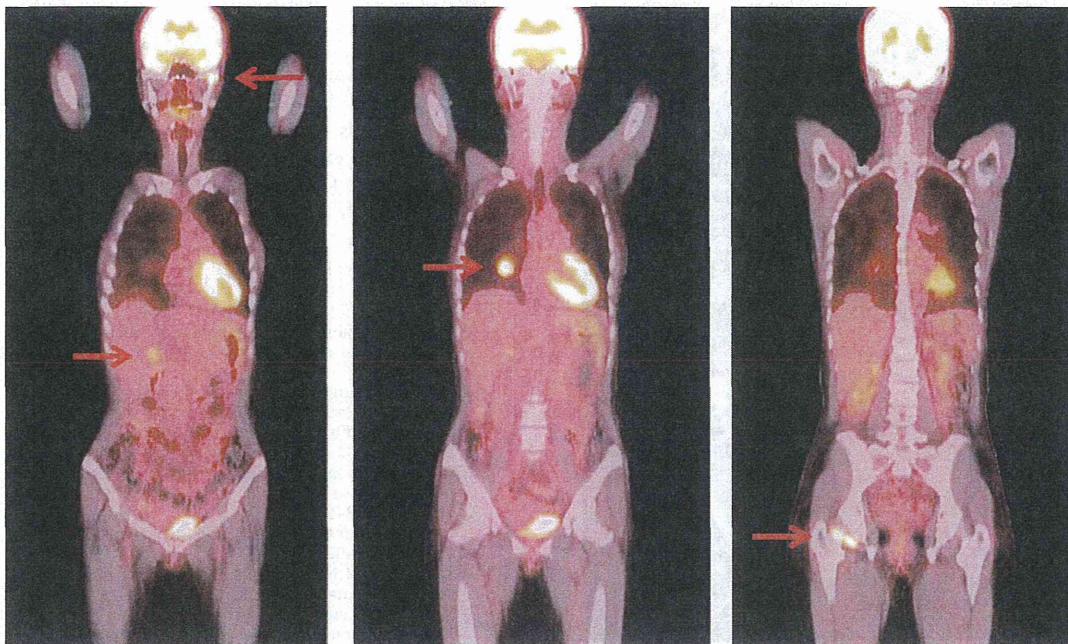


Fig. 3. (Right) Whole body fluorine-18-deoxyglucose PET-CT revealed a density anomaly mass in the liver and left mandibular condyle with increased glucose metabolism. (Center) Increased glucose metabolism in the lower left lobe, (left) increased glucose metabolism in the right common iliac artery lymph node.

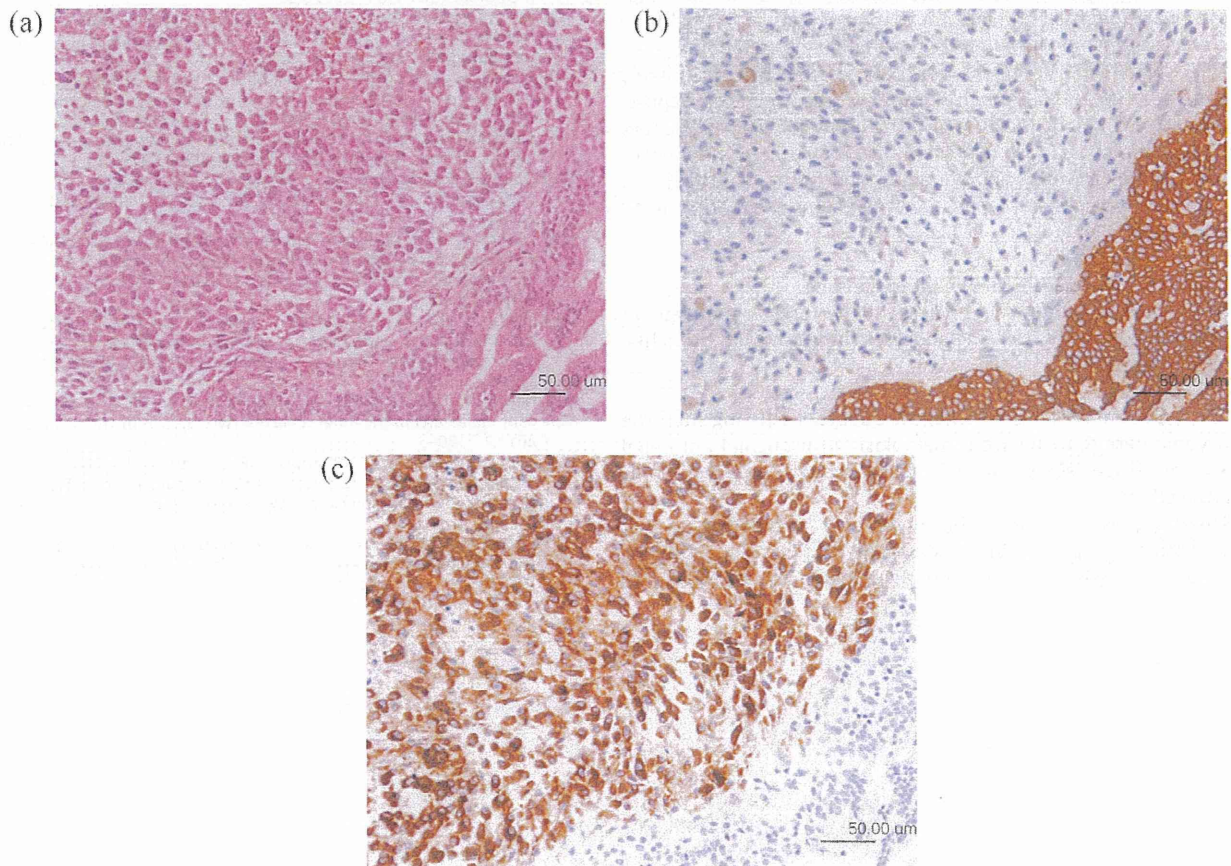


Fig. 4. Histopathological features compatible with those of malignant melanoma. (a) Hematoxylin and eosin stain confirmed outgrowth of uniform tumor cells under the mucous membrane of the bronchiole. Atypical cells with small densely stained nuclei and pigment-producing cells with brown granules were identified (original magnification $\times 100$). (b) Immunohistochemical staining for MNF116 was negative (original magnification $\times 100$). (c) Immunohistochemical staining for HMB 45 was positive (original magnification $\times 100$).

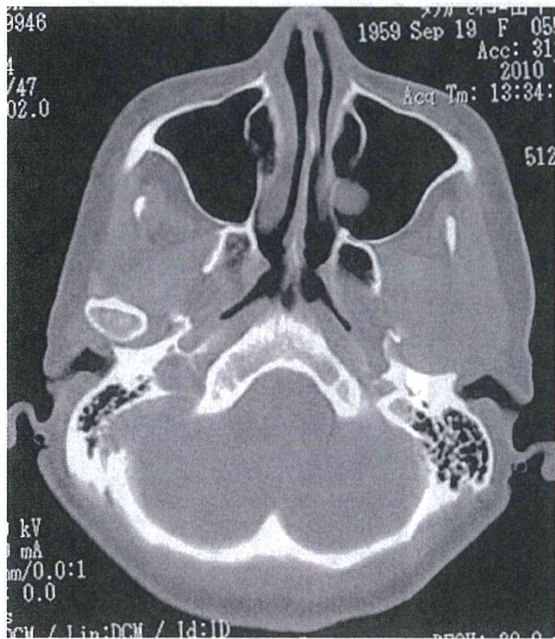


Fig. 5. CT (bone window setting) demonstrated an expansive soft tissue mass and resorption of the left mandibular condyle.

in the inferior lobe of lung because 72% of tumors that metastasize to the mandibular condyle are adenocarcinomas [7,14]. There was no clear sign of the origin of the malignant melanoma, such as a clear black mole or appearance of skin lesions. The most likely candidate is the lung although there have been few case reports of primary malignant melanoma of the lung [15,16]. A primary malignant tumor is a rare neoplasm that accounts for only 0.01% of primary lung tumors [17]. We did not perform an autopsy, but we arrived at a diagnosis of malignant melanoma originating in the lung because:

- (a) there was no existing malignant melanoma;
- (b) malignant melanoma originating in the lung is a malignant transformation of the melanocytes beneath the bronchial epithelial cells.

The argument remains whether melanocytes, existing since the embryonic period, result from metaplasia in bronchial epithelial cells or are a specialized part of intraneural secretory cells (Kulchitsky cell).

The prognosis of malignant melanoma originating in the lung is generally poor [16]. This patient died despite undergoing chemotherapy. There was no clear lower jaw distortion or

limitation to opening the mouth initially, and her symptoms were identical to those of TMD.

We were able to find the malignant melanoma because of the radiolucent lesion in the TMJ identified on panoramic X-ray. In other words, it is crucial not to overlook images indicating bone destruction in the lower mandibular condyle that appear on simple X-rays if there is evidence of a malignant tumor in the TMJ.

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モーニングセミナーから

下顎骨に発生した良性歯原性腫瘍および 悪性腫瘍の鑑別

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抄 録

歯原性腫瘍は歯を形成する組織由来の腫瘍で、一般に顎骨内に発生する。大部分が良性腫瘍であり、無痛性の膨隆をきたし発見されたり、レントゲン撮影にて偶然発見される場合も多い。歯原性腫瘍は良性腫瘍が多いものの、局所再発傾向の強い腫瘍があることや、頻度は低いものの悪性腫瘍も存在するため、正確な診断と適切な処置が必要とされる。今回、当科で経験した良性歯原性腫瘍と顎骨内に発生した悪性腫瘍を提示し、その鑑別診断について検討する。

緒 言

歯原性腫瘍とは歯を形成する組織、すなわち外胚葉由来のエナメル質を作る細胞、中胚葉由来の象牙質、歯髄またはセメント質を形成する細胞を発生母地とする腫瘍で、その多くが良性腫瘍である。良性腫瘍が大部分を占めるが、その代表的な腫瘍であるエナメル上皮腫は良性腫瘍に分類されているものの、局所再発傾向の強い腫瘍として知られている。一方、顎骨に発生する腫瘍には頻度は高くはないものの悪性腫瘍が存在する。言うまでもなく、悪性腫瘍の場合は迅速な診断、処置を要する。今回、当科にて経験したエナメル上皮腫と顎骨中心性の扁平上皮癌症例の初診時の症状、パノラマX線所見より鑑別診断について検討し、その診断・治療について考察する。

症 例 1

患者：61歳、男性

主訴：左側下顎部の腫脹

現病歴：平成20年12月頃より左側下顎部の腫脹を自覚するも放置していた。平成21年3月近畿大学歯科医院を受診し、パノラマX線にて左側下顎部の透過像を指摘され、精査目的で当科へ紹介となった。

現症：左側下顎部に腫脹を認めるものの、疼痛等の著明な症状は認めなかった(図1A)。

画像所見：パノラマX線所見では下顎左側第1大白

歯遠心から筋突起前縁にかけて境界明瞭な透過像を認めた(図1B)。CT所見(冠状断)では下顎左側第1大白歯遠心から下顎枝にかけて頬舌的に膨隆した透過像を認めた(図2A)。CT所見(軸位断)頬舌側の骨は非薄化しており、下顎管は下方に圧平さ

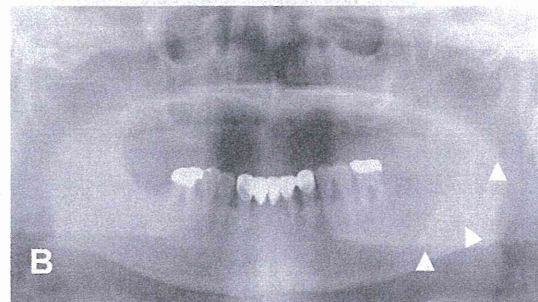
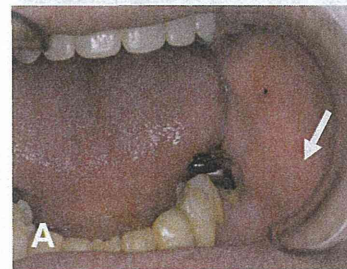


図1 A：症例1口腔内写真、左側下顎臼歯部に腫脹が認められた(白矢印)。 B：症例1パノラマX線：下顎左側第1大白歯遠心から筋突起前縁にかけて境界明瞭な透過像を認めた(白矢頭)。

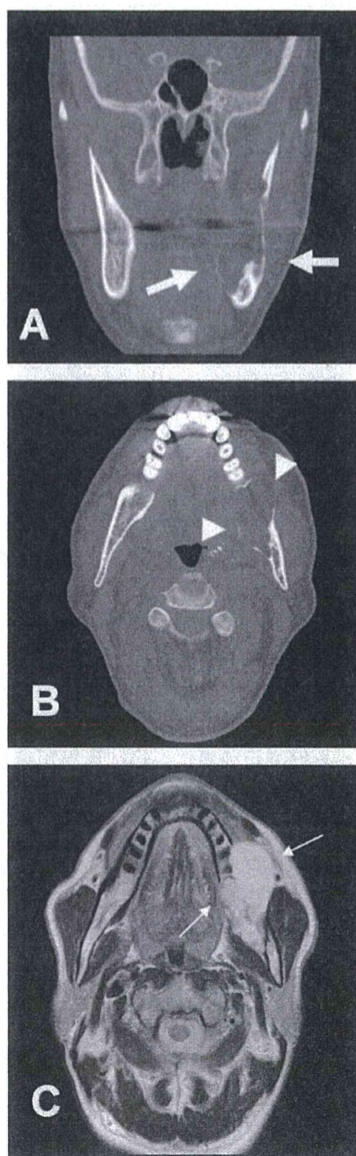


図2 A：症例1 CT 冠状断，頬舌側の骨は非薄化しており，下顎管は下方に圧平されていた（白矢印）．B：症例1 CT 軸位断，下顎左側第1大臼歯遠心から下顎枝にかけて頬舌的に膨隆した透過像を認めた（白矢頭）．C：症例1 MRI 軸位断，T2 強調像にて下顎左側第1大臼歯遠心に多房性の高信号を認め，病変は左側咬筋とは境界明瞭であった（細白矢印）．

れていた（図2 B）．MRI 所見においては T2 強調画像にて下顎左側6番遠心に多房性の高信号を認め，病変は左側咬筋とは境界明瞭であった（図2 C）．
臨床診断：左側下顎骨歯原性腫瘍
生検：外来にて局所麻酔下で生検術を施行した．
病理組織所見：腫瘍実質は歯胚のエナメル器に類似する島状の胞巣よりなる（弱拡大，図3 A）．腫瘍細胞は内腔面では類円形核を持っており，嚢胞内腔に

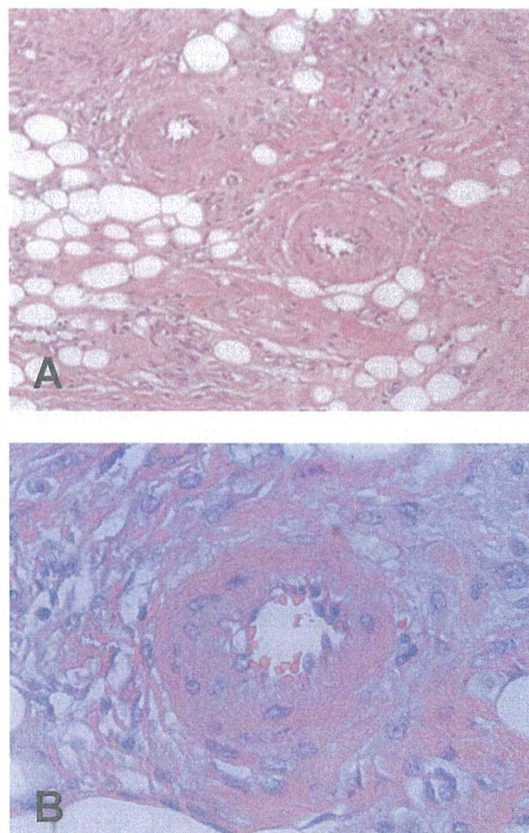


図3 A：症例1 病理組織像（弱拡大）腫瘍実質は歯胚のエナメル器に類似する島状の胞巣よりなる．B：（強拡大）腫瘍細胞は内腔面では類円形核を持っており，嚢胞内腔に微小な乳頭状構造をとっている．

微小な乳頭状構造をとっている（強拡大，図3 B）．
病理組織診断：乳頭状角化エナメル上皮腫（papilliferous keratoameloblastoma）
処置および経過：平成21年5月，全身麻酔下にて腫瘍摘出開窓術を施行した．手術から2年経過しているが，経過良好である．

症例 2

患者：68歳，男性
主訴：左側下唇麻痺
現病歴：平成22年7月頃より左側下唇の麻痺を認め，近耳鼻科を経て，本院耳鼻科を受診した．画像検査にて左側下顎部の透過像を認め，顎骨腫瘍疑いにて当科を紹介された．
既往歴：特記事項なし．
画像所見：パノラマX線所見では左側下顎第2大臼歯遠心に，下顎枝上方に至る智歯を含む辺縁不整な透過像を認めた（図4 A）．また，MRIT2強調像にて左下顎角部に充実性信号を示した（図4 B）．

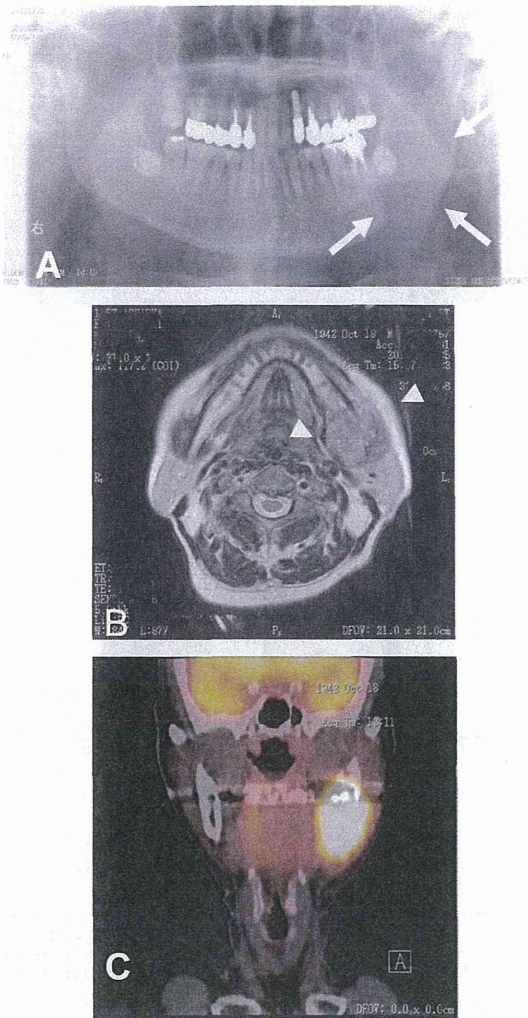


図4 A：症例2パノラマX線：左側下顎第2大臼歯遠心に、下顎枝上方に至る智歯を含む辺縁不整な透過像を認めた。B：症例2 MRI 軸位断 T2 強調像にて左下顎角部に充実性信号を示した。C：症例2 PET-CT 所見にて左側下顎骨および左頸部リンパ節に集積を認め、左側頸部リンパ節転移を疑った。

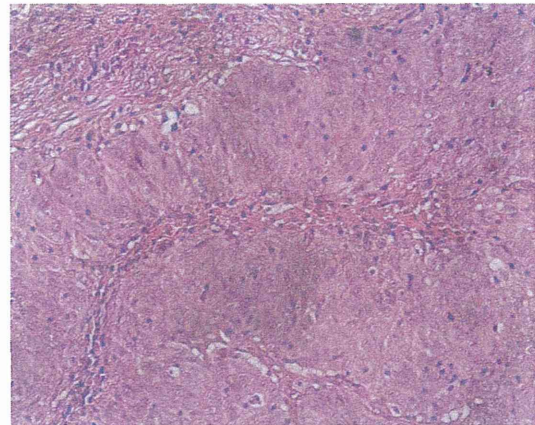


図5 症例2病理組織像(弱拡大)未分化な扁平上皮細胞の増殖を認めた。

ート再建術を施行した(図6 A, B, C)。手術より1年経過しているが、再発もなく経過良好である(図7 A, B)。

考 察

歯原性腫瘍とは歯胚を形成する組織、すなわち外胚葉由来のエナメル質を作る細胞、中胚葉由来の象牙質、歯髄またはセメント質を形成する細胞を発生母地とする腫瘍で、病理学的に複雑な組織像を示す¹⁾。歯原性腫瘍はその多くが良性腫瘍であり、その特徴として、初期に臨床症状が乏しく、無痛性に進行し顎骨の膨隆、変形を訴えて来院することが多く、パノラマX線撮影によって偶然発見されることも多い¹⁾。

歯原性腫瘍は良性腫瘍が大多数を占めるものの、局所浸潤性が高く、再発傾向が高いとされる。エナメル上皮腫や角化嚢胞性歯原性腫瘍の発生頻度が高い^{1,2)}。大阪大学歯学部附属病院検査部で歯原性腫瘍821例を検索した報告によると、角化嚢胞性歯原性腫瘍が293例(35.7%)、エナメル上皮腫が219例(26.7%)であった^{3,4)}。

エナメル上皮腫は歯原性腫瘍でありながら、局所浸潤・再発傾向の高い腫瘍とされている。一方、角化嚢胞性歯原性腫瘍は2005年のWHO 歯原性腫瘍の分類変更に伴い、腫瘍に再編された腫瘍で、以前は嚢胞に分類されながら、娘嚢胞が存在し、再発傾向の高い疾患である^{2,4,5)}。

原発性下顎骨中心性扁平上皮癌はWHOにより、顎骨内に生じ、初期には口腔粘膜と連続性がなく、歯原性上皮遺残から発生したと推定され、かつ他臓器からの転移でない扁平上皮癌と定義されている²⁾。その診断は初期では困難であり、発見時はすでに下顎骨内で広範囲に進展していることが多く、一

現症：左側下顎部に知覚異常を認めるものの、その他の著明な症状は認めなかった。

生検：平成22年8月、外来にて局所麻酔下に生検を施行した。

病理組織検査：未分化な扁平上皮細胞の増殖を認めた(弱拡大、図5)。

診断：顎骨中心性扁平上皮癌

PET-CT 所見：左側下顎骨および左頸部リンパ節に集積を認め、左側頸部リンパ節転移を疑った(図4 C)。

処置および経過：平成22年9月、全身麻酔下に下顎骨区域切除、左側頸部郭清術、ならびに下顎骨プレ

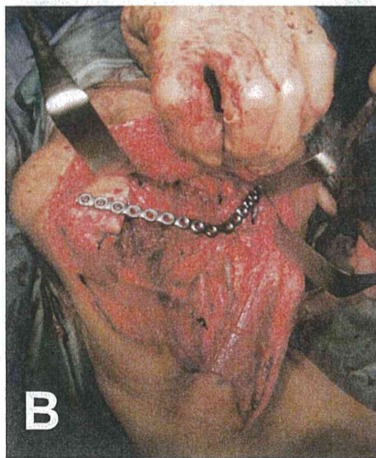


図6 A：症例2手術直前 B：左側頸部郭清および顎骨腫瘍切除後，チタンプレートにて再建。
C：左側頸部リンパ節組織と顎骨腫瘍を一塊に切除。

般的に予後不良とされている^{1,2,6,7}。

今回、顎骨に発生した良性の歯源性腫瘍であるエナメル上皮腫と顎骨中心性扁平上皮癌のX線所見、

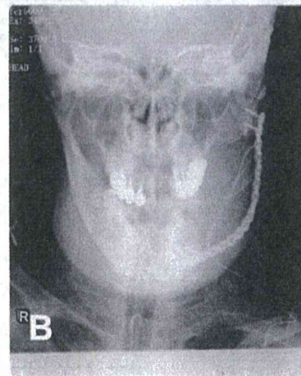
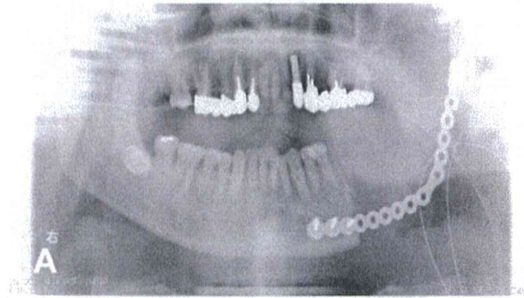


図7 A：症例2術後パノラマX線，B：症例2術後頭部正面像，顎骨腫瘍切除後チタンプレートにて再建されている。

表1 症例1，症例2のパノラマX線所見，臨床所見，病理組織診断，治療法の比較

	症例1	症例2
X-P 所見	顎骨の透過像	顎骨の透過像
多胞，単胞	多房性	多房性
歯の存在	なし	埋伏歯
辺縁	比較的明瞭	不明瞭
臨床症状	無痛性膨隆	下唇に麻痺
病理組織診断	エナメル上皮腫	扁平上皮癌
治療法	腫瘍摘出開窓術	顎骨区域切除 +プレート再建

初診時の症状，診断，治療法を比較し，表1に示した。

症例1，2を比較すると，顎骨内に腫瘍が存在することから両者ともパノラマ写真にてX線透過像が認められた。パノラマX線写真は歯科で高頻度に撮影され，顎骨病変の描写には極めて有効である。透過像と顎骨との境界に関して，良性のものが比較的明瞭であったのに対し，悪性のものは境界不明瞭であった。境界不明瞭な像を呈するのは，浸潤性の強い悪性腫瘍の特徴であるといえる。臨床症状も鑑別の指標となり，良性のものが無症状であることが多いのに対し，悪性のものは知覚異常などの神経症状