A Rare Case of Mucoepidermoid Carcinoma of Hard Palate with Thinning of the Palatine Bone

TWMUJ 4: 90-93, 2020

Case Report

A Rare Case of Mucoepidermoid Carcinoma of Hard Palate with Thinning of the Palatine Bone

Munenaga Nakamizo,[1] **Kazuhiko Yokoshima,**[2] **Taisei Kato,**[2]
Shunta Inai,[1] **Atsuko Sakanushi,**[3] and **Ryuji Ohashi**[4]

[1]Department of Oto-Rhino-Laryngology, Tokyo Women's Medical University, Tokyo, Japan
[2]Department of Head & Neck Surgery, Tochigi Cancer Center, Tochigi, Japan
[3]Department of Oto-Rhino-Laryngology Head & Neck Surgery, Nippon Medical School, Tokyo, Japan
[4]Department of Pathology, Nippon Medical School, Tokyo, Japan

(Accepted July 31, 2020)
(Advance Publication by J-STAGE September 29, 2020)

We reported a rare case of low-grade mucoepidermoid carcinoma of the hard palate with bone changes. Preoperative coronal computed tomography was useful to reveal thinning of the palatine bone. We performed the total resection of the tumor including palatine bone with a safety margin of 5 mm or more in monobloc fashion and reconstructed with a palatal mucoperiosteal flap. There is no recurrence and complications for five years after surgery.

Key Words: mucoepidermoid carcinoma, hard palate, bone thinning, resection

Introduction

Mucoepidermoid carcinoma of the hard palate is a rare disease, and there are cases in which its pathological diversity and anatomical complexity make it difficult to select a treatment approach. Since it is impossible for a single facility to handle a large number of cases,[1-3] it is important that each case should be reported as a detailed record. Here, we describe a case in which diagnostic imaging revealed changes in the palatine bone and discuss the problems in treating this disease.

Case Presentation

A 46-year-old male had become aware of an intraoral mass 5 years earlier and visited the hospital after noticing that the mass had increased in size during recent months. The initial examination found a 7-mm-long submucosal mass on the left side of the hard palate (**Figure 1**). There was no spontaneous pain or bleeding. MRI of the neck revealed a mass with an abnormal signal and a major axis of 7 mm on the left side of the hard palate (**Figure 2A**). Because of his history of bronchial asthma, contrast examination was not possible. A plain cervical computed tomography (CT) scan revealed a tumor at the same site and thinning of the palatine bone (**Figure 2B**). Fine nee-

Corresponding Author: Munenaga Nakamizo, Department of Oto-Rhino-Laryngology, Tokyo Women's Medical University, 8-1 Kawada-cho, Shinjuku-ku, Tokyo 162-8666, Japan. nakamizo.munenaga@twmu.ac.jp
doi: 10.24488/twmuj.2020008

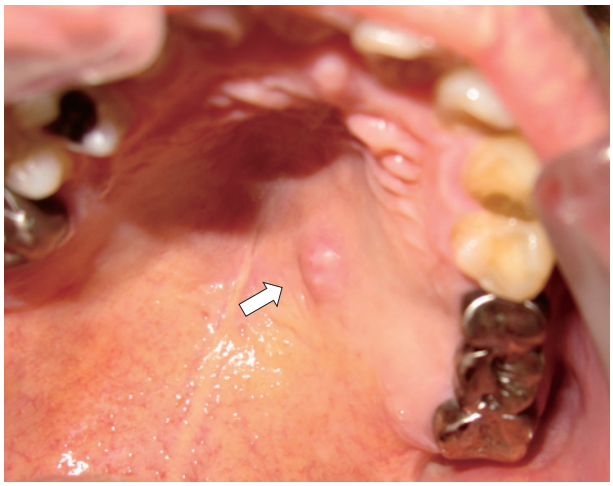

Figure 1 Oral findings at first visit: A submucosal tumor (arrow) was found on the hard palate.

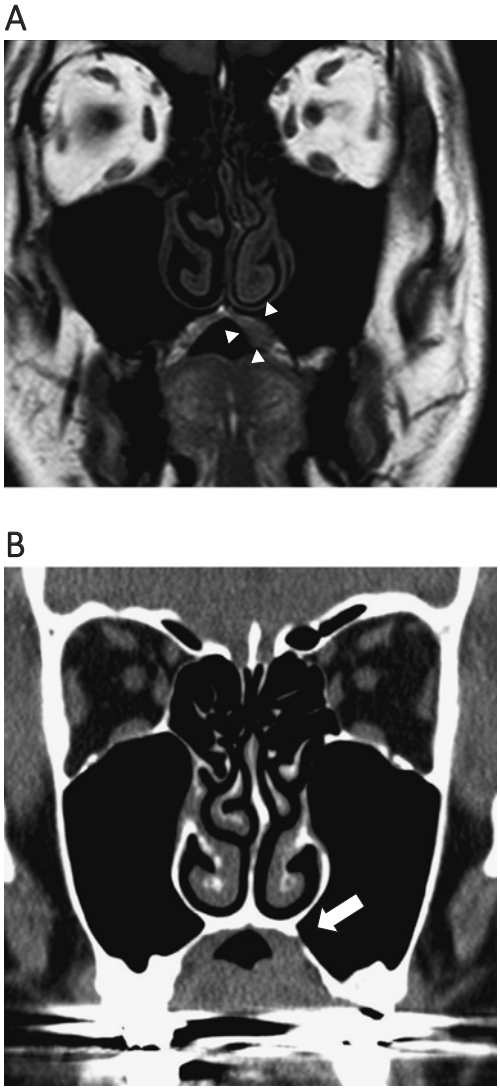

Figure 2 The plain coronal MRI T1-weighted image shows an elliptical mass (arrowheads) with a slightly lower signal than the muscles and no hyperintense area with a somewhat irregular margin at the side of the maxillary floor (**A**). Coronal CT shows thinning (arrow) in part of the maxillary sinus floor (**B**).

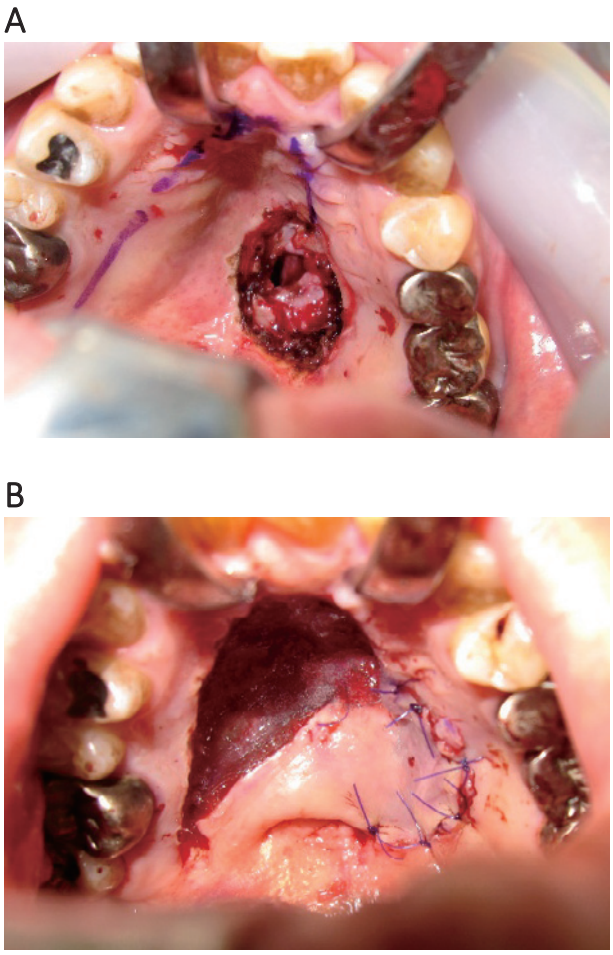

Figure 3 The communication (**A**) created with the left nasal cavity and maxillary sinus by the resection was reconstructed using a right palatine mucoperiosteal flap (**B**).

dle aspiration cytology found cells having a round nucleus with small, clear nucleoli, and cell images showed a mixture of squamous epithelial-like and intermediate-like cells containing mucus. A mucoepidermoid carcinoma was considered. Although there was no finding suggesting a high-grade malignancy, the pathological grade could not be precisely established.

Based on the above findings, surgery was performed under a diagnosis of a mucoepidermoid carcinoma of the hard palate, T1N0. The palatal mucosa and submucosa were incised with a safety margin of 5 mm or more around the tumor. Partial palatine osteotomy was performed down to the left nasal cavity and the maxillary sinus floor in the vertical direction, and the palate was opened to remove the tumor and palatine bone with the left nasal and maxillary sinus mucosa in monobloc fashion (**Figure 3**). Consequently, the left nasal and maxillary sinus mucosa without tumor invasion were the depth

margin of resected specimen. We did not perform intraoperative rapid diagnosis, since there was no finding suggesting a high-grade malignancy. The hard palate defect was reconstructed by rotating a right mucoperiosteal flap. The surface of the right hard palate where the mucoperiosteal flap was raised was filled with a polyglycolic acid sheet as a wound dressing, which was fixed with fibrin glue.

Pathologically, mucus-producing cells were observed, with a large cystic portion. The solid portion contained mucus-producing cells, intermediate cells and squamous cells. This was a typical low-grade mucoepidermoid carcinoma. There was sparse connective tissue between the carcinoma and the maxilla, and the bone margin was irregular, but no bone infiltration was observed. The surgical margin was negative, and no lymphatic invasion was seen (**Figure 4**). Immunostaining was consistent with a mucoepidermoid carcinoma. Oral intake was started on the 5th postoperative day, and the patient was discharged on the 9th day. Epithelialization of the donor site of the mucoperiosteal flap was good, and there was no evidence of recurrence as of 5 years after the surgery.

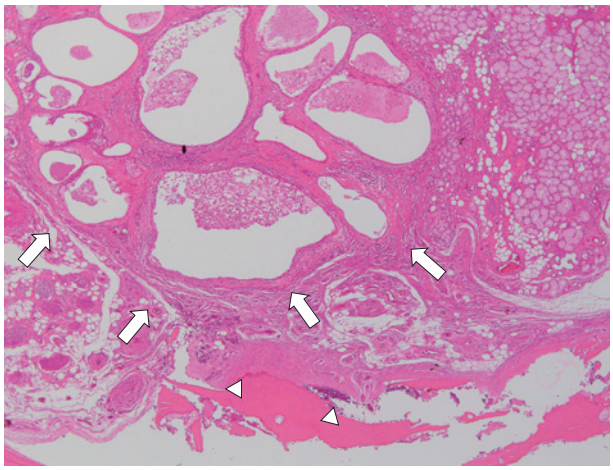

Figure 4 The histopathological diagnosis was a low-grade mucoepidermoid carcinoma, and the surgical margin was negative. The tumor component had spread to the palatine bone at a point where only a small amount of connective tissue remained (arrows). The bone margin was irregular, and fibrosis was spreading (arrowheads). (×20)

Discussion

Minor salivary gland tumors are relatively rare, but almost half of them are carcinomas.[4,5] According to the Japanese Head and Neck Cancer Registry from 2011 to 2016, 2 to 3% of malignant oral tumors were minor salivary gland cancers, most of which were adenoid cystic carcinomas and mucoepidermoid carcinomas, with the latter occurring most commonly in the hard palate.[6,7] There have been multiyear compilations[1-3] of minor salivary gland carcinomas or hard palate mucoepidermoid carcinomas. However, since they are rare diseases and the medical technique changes with time, reporting of each case should provide useful information for future treatment selection.

Mucoepidermoid carcinomas are usually classified into three pathological categories: low-, intermediate- and high-grade. Mucus-producing cells predominate in low-grade cancers, whereas intermediate- to high-grade cancers contain few mucus cells but many nonkeratinized squamous epithelial cells.[8] Since the pathological features of these lesions are sometimes reflected in cytology, we performed cytological analysis of this case. Although we inferred that the tumor was a mucoepidermoid carcinoma based on typing of the collected cells, we were unable to confirm that since histological information is essential[9,10] for determining the grade of malignancy. On the other hand, we did not perform a biopsy because of the possibility that it would influence the margin of mucosal resection. Rather, the problem was setting the vertical margin because CT showed thinning of the palatine bone in spite of the small mass.

In the case of minor salivary gland carcinomas, the histology and resected margin have been reported to affect recurrence-free survival and disease-specific survival.[1,2] Therefore, it is said that making the resected margin negative is important for obtaining a good prognosis.[11] In fact, Hay et al. reported that a 30-year summary of minor salivary gland cancers at the Sloan-Kettering Cancer Center showed a 56% increase in the risk of death when the resected margin was positive,[3] although there was no statistically significant difference in the prognosis. Ord et al.[1] reported that only 2 cases were found to have bone changes by preoperative imaging out of 18 low-grade mucoepidermoid carcinoma cases over a 15-year period, and recommended palatal bone resection for them because 1 of them showed pathological bone infiltration.

In our present case, we estimated that the possibility of

high-grade carcinoma was low based on our preoperative examination, but we thought that a sufficient margin should be taken on the maxillary side since CT showed bone thinning. As a result, there was no pathological bone infiltration, and the image of bone thinning in this patient can be thought to have been caused by chronic compression by the tumor. Depending on the pathological findings, bone preservation may be possible, but in that case as well, verification of the resection margin may be difficult.

Then there is the problem of communication between the oral cavity and the nasal cavity as well as the maxillary sinus when combined resection of the palate is performed. For follow-up observation after surgery, the resected stump is clearly visible and easy to observe. In addition, by wearing a denture for a palatal defect, it is possible to prevent rhinolalia aperta and inflow of food into the nasal cavity. However, in our present case, we were able to observe the surgical stump from the nasal cavity by using a nasopharyngeal fiberscope. In addition, the patient's dental condition was good. And by palatal mucoperiosteal flap reconstruction, we were able to avoid the need to make a prosthesis and perform maintenance, thus reducing the costs.

The reliability and safety have been established for the palatine mucoperiosteal flap as an axial pattern flap including the greater palatine artery and vein.[12] The maximum area that can be reconstructed with the flap is 16 cm^2, while larger palatal defects are reconstructed with buccal myomucosal flaps or free forearm flaps.[13-15] The hard palate defect in our patient was sufficiently closed with a mucoperiosteal flap. Five years after the operation, the color and texture of the donor site and transplanted hard palate mucosa were good, and there was no recurrence. Moreover, the quality of life was the same as before the operation.

We obtained written informed consent for a case report from the patient.

Conflicts of Interest: We have no conflict of interests about this case report.

References

1. Ord RA, Salama AR: Is it necessary to reset bone for low-grade mucoepidermoid carcinoma of the palate? Br J Oral Maxillofac Surg 50: 712−714, 2012
2. Li Q, Zhang XR, Liu XK et al: Long-term treatment outcome of minor salivary gland carcinoma of the hard palate. Oral Oncol 48: 456−462, 2012
3. Hay AJ, Migliacci J, Zanoni DK et al: Minor salivary gland tumors of the head and neck—Memorial Sloan Kettering experience: Incidence and outcomes by site and histological type. Cancer 125: 3354−3366, 2019
4. Pires FR, Pringle GA, de Almeida OP et al: Intra-oral minor salivary gland tumors: a clinicopathological study of 546 cases. Oral Oncol 43: 463−470, 2007
5. Yih WY, Kratochvil FJ, Stewart JCB: Intraoral minor salivary gland neoplasms: review of 213 cases. J Oral Maxillofac Surg 63: 805−810, 2005
6. Japan Society of Head and Neck Cancer, Cancer registry committee: Report of Head and Neck Cancer Registry of Japan. Clinical Statistics of Registered Patient, 2011-2016. http://www.jshnc.umin.ne.jp/pdf/2011syourei_houkoku.pdf - http://www.jshnc.umin.ne.jp/pdf/2016syourei_houkoku.pdf (Accessed June 7, 2020)
7. Takahashi H, Fujita S, Tsuda N et al: Intraoral minor salivary gland tumors: A demographic and histologic study of 200 cases. Tohoku J Exp Med 161: 111−128, 1990
8. WHO Classification of Head and Neck Tumours, 4th ed (El-Naggar AK, Chan JKC, Grandis JR et al eds), IARC Press, Lyon (2017)
9. Auclair PL, Goode RK, Ellis GL: Mucoepidermoid carcinoma of intraoral salivary glands. Evaluation and application of grading criteria in 143 cases. Cancer 69: 2021−2030, 1992
10. Goode RK, Auclair PL, Ellis GL: Mucoepidermoid carcinoma of the major salivary glands: clinical and histopathologic analysis of 234 cases with evaluation of grading criteria. Cancer 82: 1217−1224, 1998
11. Iyer NG, Kim L, Nixon IJ et al: Factors predicting outcome in malignant minor salivary gland tumors of the oropharynx. Arch Otolaryngol Head Neck Surg 136: 1240−1247, 2010
12. Jamali JA: Palatal flap. Oral Maxillofac Surg Clin North Am 26: 305−311, 2014
13. Kimia R, Shakir S, Jackson OA et al: Buccal myomucosal flap reconstruction after resection of palatal mucoepidermoid carcinoma. Ann Plast Surg 83: 538−541, 2019
14. Nakamizo M, Yokoshima K, Yagi T: Use of free flaps for reconstruction in head and neck surgery: A retrospective study of 182 cases. Auris Nasus Larynx 31: 269−273, 2004
15. Duflo S, Lief F, Paris J et al: Microvascular radial forearm fasciocutaneous free flap in hard palate reconstruction. Eur J Surg Oncol 31: 784−791, 2005