Invasive micropapillary salivary duct carcinoma mixed with mucin-rich salivary duct carcinoma in minor salivary gland: a rare case report

Kei Tomihara, DDS, PhD,[a] Shigeharu Miwa, MD, PhD,[b] Takeshi Takazakura, DDS,[c] Yuichi Kamisaki, MD, PhD,[d] and Makoto Noguchi, DDS, PhD[a]
University of Toyama and Kurobe City Hospital, Toyama, Japan

Background. Invasive micropapillary salivary duct carcinoma (SDC) is a rare variant of SDC. Although several cases involving major salivary glands have been reported, no cases arising de novo in minor salivary glands have been reported to date. Here we report the first case of invasive micropapillary SDC that arose in a minor salivary gland of the parapharyngeal space.
Methods. A 72-year-old male patient presented with an enlarging mass in the left parapharyngeal region along with trismus and swollen lymph nodes. Clinical examinations and biopsy findings were suggestive of a salivary gland malignant tumor with regional lymph node metastases. The tumor, therefore, was excised with partial mandibulectomy with unilateral radical neck dissection.
Results. Histologically, the tumor consisted of an invasive micropapillary growth pattern of SDC and mixed with mucinous component of SDC. Local recurrence and lung metastasis developed, and the patient died of disease 13 months after the initial treatment.
Conclusions. We describe the clinical and histologic features of this extremely rare case of minor salivary gland SDC that was histologically characterized by the presence of both invasive micropapillary growth pattern and mucinous component. (Oral Surg Oral Med Oral Pathol Oral Radiol 2016;121:e162-e167)

Salivary duct carcinoma (SDC) is rare malignant tumor that was first described by Kleinsasser et al. in 1968[1] and classified by the World Health Organization in 1991 as a highly aggressive, malignant salivary gland tumor that predominantly involves the major salivary glands, especially the parotid.[2] Histopathologically, SDC consists of invasive and intraductal components that are characterized by their morphologic resemblance to high-grade ductal carcinoma of the breast.[3] Described morphologic variant forms include papillary, invasive micropapillary, sarcomatoid, mucin-rich, and oncocytic.[4-7]

Invasive micropapillary growth pattern of carcinoma was originally described as a highly aggressive malignancy in breast cancer, and subsequently it has been described in various other organs, including urinary bladder, lung, ovary, stomach, colorectum, and salivary gland.[5,8-13]

Invasive micropapillary growth pattern of SDC was initially described by Nagao et al. in 2004 in the report of 14 cases of invasive micropapillary growth pattern of SDC arising in the major salivary glands (12 involving the parotid gland and 2 the submandibular gland) as an invasive micropapillary variant of conventional SDC,[5] and subsequently an additional new case of an invasive micropapillary variant of SDC in the parotid gland was described by Yamamoto et al.[14] More recently, carcinoma ex pleomorphic adenoma of the palate with an invasive micropapillary SDC component was reported by Sedassari et al.[15] However, no case of invasive micropapillary variant of SDC arising de novo in the minor salivary glands has been reported so far.

We report a first case of invasive micropapillary SDC arising de novo in the minor salivary gland of the parapharyngeal space. The present case is also highly unusual because the tumor was histologically mixed with mucin-rich SDC. Mucin-rich SDC in minor salivary glands is exceedingly rare, and only 6 cases have been reported to date. To the best of authors' knowledge, no case of invasive micropapillary SDC accompanied with mucin-rich component of SDC has been reported in either the major and minor salivary glands.

CASE REPORT

A 72-year-old male patient was referred to our hospital in 2013 with a complaint of a painless enlarging, asymptomatic parapharyngeal mass. The patient had been aware of the

[a]Department of Oral and Maxillofacial Surgery, Graduate School of Medicine and Pharmaceutical Sciences for Research, University of Toyama, Toyama, Japan.
[b]Department of Diagnostic Pathology, Graduate School of Medicine and Pharmaceutical Sciences for Research, University of Toyama, Toyama, Japan.
[c]Department of Oral and Maxillofacial Surgery, Kurobe City Hospital, Toyama, Japan.
[d]Department of Radiology, Graduate School of Medicine and Pharmaceutical Sciences for Research, University of Toyama, Toyama, Japan.
Received for publication Jun 23, 2015; returned for revision Sep 8, 2015; accepted for publication Oct 11, 2015.

2212-4403/$ - see front matter
http://dx.doi.org/10.1016/j.oooo.2015.10.012

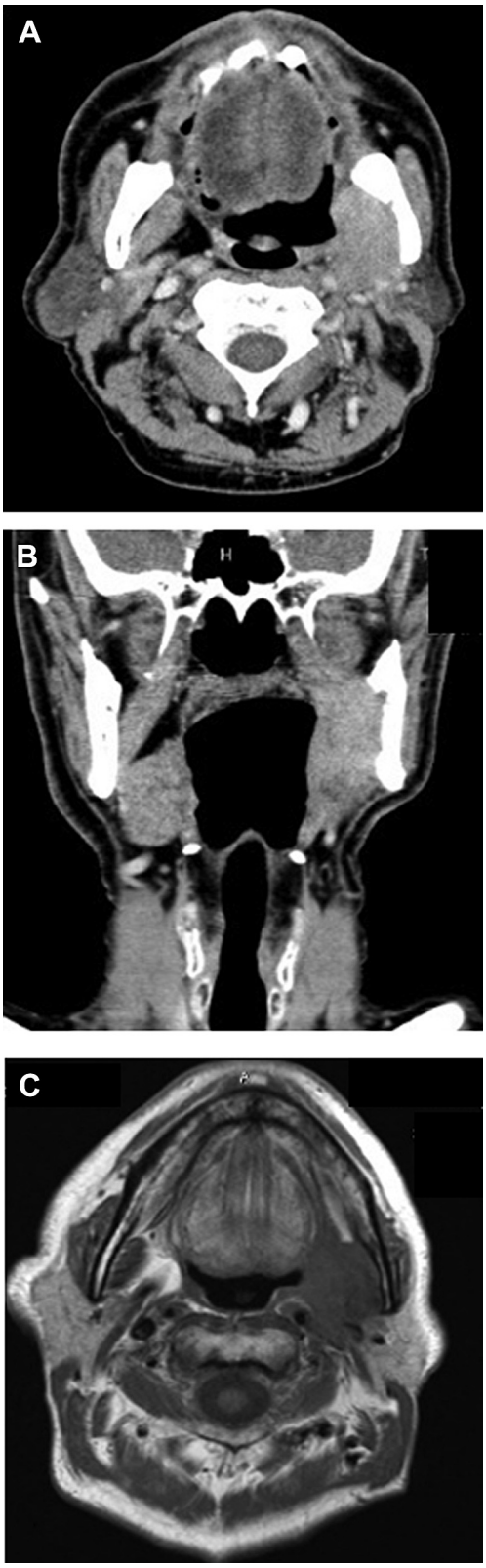

Fig. 1. (**A**) Computed tomography (CT) scan showing a homogeneous enhancing lesion in the left parapharyngeal space extending to the medial pterygoid muscle. (**B**) Irregular osteolytic cortical bone resorption adjacent to the tumor was observed. (**C**) Non−contrast enhanced T1-weight magnetic resonance image of the left parapharyngeal mass without involving deep lobe of the parotid gland.

swelling in his left parapharyngeal region for a couple of months. However, the swelling had recently increased in size along with severe trismus, and the patient initially had been seen by the Department of Oral and Maxillofacial Surgery at a local hospital, where a diagnosis of neoplasm of the parapharyngeal region was suggested. The patient was then referred to our department. His medical history revealed deep vein thrombosis and hypercolesterolemia. His family history was unremarkable. On initial assessment, no systemic symptoms were evident. Several enlarged lymph nodes were palpable unilaterally. Physical examination revealed a submucosal swelling in the lateral pharyngeal wall with extension to the medial pterygoid muscle causing severe trismus. Computed tomography (CT) and magnetic resonance imaging (MRI) indicated ill-demarcated homogeneously enhancing of the mass in the left parapharyngeal space extending to the medial pterygoid muscle (Figure 1). The mass appeared discrete from the deep lobe of the left parotid gland in both CT and MR imaging, suggesting the tumor was arising from the paraphayngeal space (Figures 1A and C). CT of the neck indicated multiple homogeneously enhancing lymph nodes in unilateral levels II, III, and IV, which suggested lymph node metastases. No other specific findings were observed by CT or positron emission tomography (PET) of the abdominal and thoracic regions. The tumor in the left parapharyngeal space was clinically malignant and was staged as T4 aN2 bM0. An intraoral biopsy of the left parapharyngeal mass was performed under general anesthesia, and histopathologic findings were suggestive of malignant salivary gland neoplasm. The tumor was excised by a transcervical-transmandibular approach with segmental mandibulectomy, and unilateral classical radical neck dissection was performed. The surgical defect was reconstructed with a latissimus dorsi myocutaneous flap.

On gross examination, macroscopically the tumor measured 3.0 × 2.2 cm and appeared grayish-white and solid in the cut surface. Microscopically the lesion was composed of 2 different morphologic patterns: 80% invasive micropapillary component and 20% mucinous component (Figure 2A). The invasive micropapillary component was characterized by small tight clusters of neoplastic cells floating in clear spaces (Figures 2B and C). The tumor cells were mostly polygonal with large nuclei and eosinophilic cytoplasm. On the other hand, the mucinous component had the typical morphologic appearance of colloid carcinoma, which contained clusters of neoplastic cells floated in mucin lakes (Figures 2D and E). Direct invasion into mandibular bone was also observed (Figure 2F). Both lymphovascular and perineural invasion were observed (Figure 2G). Lymph node metastasis was evident in 15 of 29 dissected lymph nodes; 7 in level IIB, 5 in level III, and 3 in level IV. Metastatic lymph nodes had similar histopathologic appearance in the invasive micropapillary component to the primary site (Figure 2H).