Sarcomatoid salivary duct carcinoma of the palate: a rare case report

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Salivary duct carcinoma (SDC) is an uncommon neoplasm that most commonly occurs in major salivary glands, mainly the parotid gland. SDC is rarely found in the minor salivary glands of the oral cavity. This report presents an extremely rare case of sarcomatoid SDC originating in a minor salivary gland of the palate. The tumor was histologically characterized by the presence of both carcinomatous and sarcomatoid components. The patient presented with a painless mass in the right palate, which slowly increased in size over 20 years. The clinical course of the present case suggests that the tumor most probably developed as a result of malignant transformation of a preexisting benign tumor of the palatal salivary gland. This report describes the clinical and histologic features of this extremely rare case of sarcomatoid SDC with reference to the relevant literature. (Oral Surg Oral Med Oral Pathol Oral Radiol 2015;119:e27-e32)

Salivary duct carcinoma (SDC) is an uncommon, highly aggressive, malignant salivary gland tumor that predominantly involves the major salivary glands.[1,2] It was first described by Kleinsasser et al. in 1968[3] and was then classified as a distinct entity of salivary gland tumors by the World Health Organization in 1991.[4] Histopathologically, SDC consists of invasive and intraductal components that morphologically resemble high-grade ductal carcinoma of the breast. The intraductal component shows a cribriform, papillary, or solid growth pattern. The cribriform growth pattern often shows comedo-like central necrosis. There are several variants of SDC, and sarcomatoid, invasive micropapillary, and mucin-rich forms have been described.[5-7] The sarcomatoid form of SDC, characterized by the presence of both sarcomatoid and carcinomatous components, was initially described by Henley et al. in 2000.[5] They reported 3 cases of this new type of SDC arising in the parotid gland as a sarcomatoid variant of conventional SDC. Although several cases of sarcomatoid SDC arising in the parotid gland have been reported since then,[6] reports of sarcomatoid SDC arising in the minor salivary glands are extremely rare. To the best of the authors' knowledge, only 2 cases have been reported in the English literature to date.[8,9] This report describes an additional case of sarcomatoid SDC in a minor salivary gland of the palate. The tumor was present for 2 decades and slowly increased in size. Based on this clinical course, the sarcomatoid SDC probably developed as a result of malignant transformation of a preexisting pleomorphic adenoma.

CASE REPORT

A 74-year-old woman was referred to the authors' hospital in 2012 with a complaint of a painless mass in the right palate. The patient was aware of the swelling in her right palate for approximately 20 years. However, recently, the swelling rapidly increased in size, for which the patient initially consulted a general dental practitioner. She was referred to the Department of Oral and Maxillofacial Surgery at a local hospital, where a diagnosis of neoplasm of the palate was suggested. The patient was then referred to the authors' department. Her medical history included well-controlled hypertension, and her family history was unremarkable. On initial assessment, no systemic symptoms were evident. Several enlarged lymph nodes were palpable bilaterally at levels I, II, and III. Intraoral examination found a dome-shaped mass measuring 4.6 cm × 3.4 cm on the right side of the hard palate, extending anteriorly to the distal second premolar area, posteriorly to the soft palate, and medially crossing the midline to the left side of the palate. The mucosal surface of the mass was smooth, but its anterior aspect was partially necrotic and ulcerated (Figure 1, A). On palpation, the mass was elastic and hard. Panoramic radiography found opacity of the right maxillary sinus. Computed tomography (CT) found a large, nonhomogeneous mass extending from the right palate to the right maxillary sinus and right nasal fossa with concomitant destruction of the right posterior hard palate and the medial right maxillary sinus walls (Figure 1, B). CT of the neck found multiple homogeneously enhancing lymph nodes in bilateral levels I, II, and III. CT of the abdominal and thoracic regions was also performed, but no other specific findings were observed. The tumor in the right palate was clinically malignant and was staged as T4 aN2 cM0. Biopsy found features of carcinoma including both carcinomatous and sarcomatoid components. The patient, therefore, underwent excision with a subtotal right maxillectomy and ipsilateral classical radical neck dissection along with

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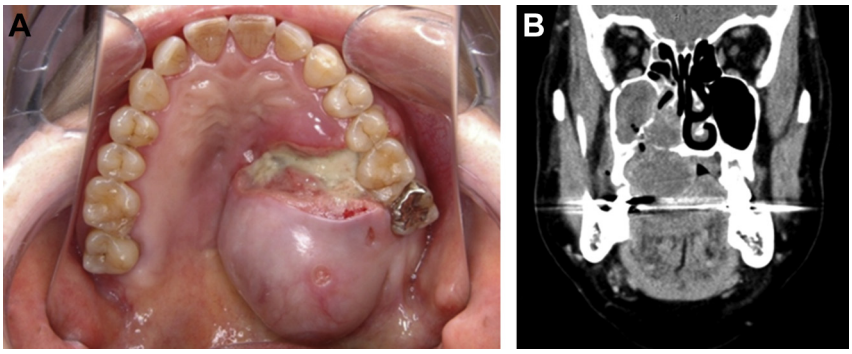

Fig. 1. **A**, An intraoral photograph showing an ulcerated mass on the right side of the hard palate. **B**, Computed tomography scan showing a homogeneous mass in the right palate with bone destruction in the maxillary sinus and nasal fossa.

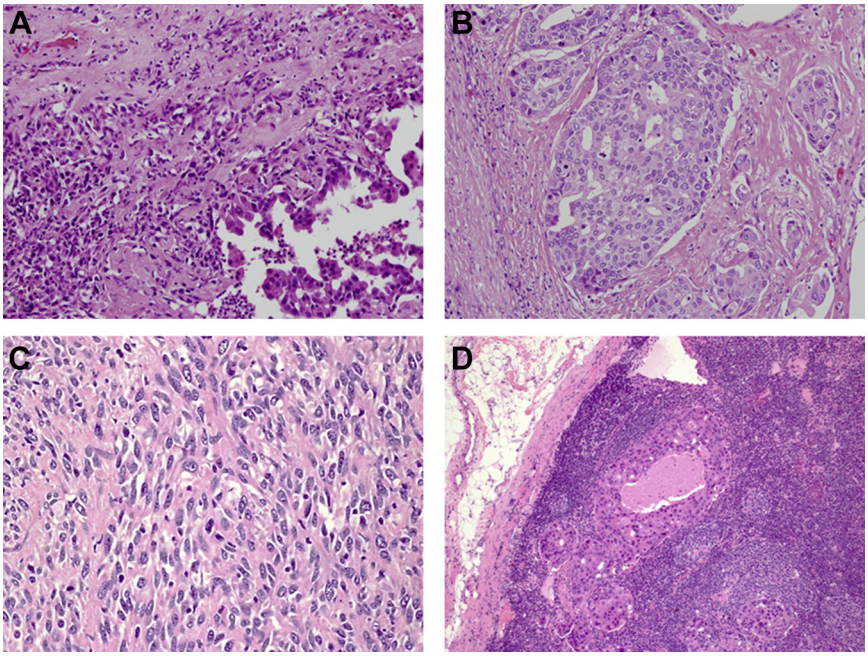

Fig. 2. Histologic appearance of the tumor. **A**, A biphasic tumor with both carcinomatous and sarcomatoid components. The transitional zone between carcinomatous and sarcomatoid components is seen (hematoxylin-eosin, original magnification ×200). **B**, The carcinoma component contained atypical epithelial cells with a cribriform growth pattern (hematoxylin-eosin, original magnification ×200). **C**, The sarcomatoid component contained spindle-shaped cells with atypia and mitosis (hematoxylin-eosin, original magnification ×400). **D**, Central comedo necrosis was observed in the metastatic lymph node (hematoxylin-eosin, original magnification ×100). A high-resolution version of this slide is available as eSlide: VM00332.

subsequent contralateral modified radical neck dissection. The surgical defect was rehabilitated with an obturator. The patient also underwent postoperative adjuvant chemoradiotherapy. Macroscopically, the surgical specimen measured 60 mm × 45 mm, and the tumor was found to be located in the hard palate and soft palate and had destroyed the palatal bone. It consisted of mostly dark gray necrotic areas and white-gray solid areas in the deeper part of the cut surface. The overlying mucosal surface was mostly normal, but the anterior part was partially necrotic and ulcerated. Microscopically, the tumor mainly comprised areas of necrosis. In the deeper parts of the tumor, 2 different morphologic patterns were observed: 10% carcinomatous and 90% sarcomatoid components (Figure 2, A). The carcinomatous component contained atypical epithelium with a cribriform growth pattern (Figure 2, B). The epithelium was primarily composed of polygonal cells with abundant eosinophilic cytoplasm and large nuclei. The sarcomatoid component contained atypical spindle-shaped cells with high mitotic activity (Figure 2, C). Both perineural infiltration and lymphovascular invasion were observed. There was no evidence of a preexisting pleomorphic adenoma. Moreover, metastases were observed in several cervical lymph nodes bilaterally. The metastatic carcinoma exhibited ductal structures with a cribriform growth pattern and comedo-like central necrosis (Figure 2, D). Supplemental Figure S1 gives additional views (available at www.oooojournal.net). The immunohistochemical findings are