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CASE REPORT

Carcinosarcoma of the tongue

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Summary Carcinosarcoma, which consists of both carcinomatous and sarcomatous tissues, is extremely rare in the oral cavity. An 80-year-old woman was referred to us, complaining of a painful, polypoidal, and rapidly growing mass at the lateral border of her tongue. We performed an excision with adequate margin around the stalk of the polyp. The mass was histologically and immunohistochemically diagnosed as a carcinosarcoma composed of both squamous cell carcinoma and rhabdomyosarcoma which is, to our knowledge, the first such case in the tongue.
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Introduction

Carcinosarcoma composed of both carcinomatous and specific differentiated sarcomatous elements with typical features of malignancy is extremely rare in the oral cavity,[1,2] it has been reported in the tongue, lower gingiva, and floor of the mouth. To our knowledge, there have been only five previously reported cases of carcinosarcoma of the tongue.[3–7] Of these five cases, the sarcomatous component was identified as chondrosarcoma or spindle-cell carcinoma, but there was no case of rhabdomyosarcoma. We present the first case of carcinosarcoma of the tongue, that consists of a squamous cell carcinoma component and a sarcomatous component that was diagnosed as rhabdomyosarcoma.

Report of a case

An 80-year-old woman was referred to the Division of Oral and Maxillofacial Surgery, University of Tsukuba Hospital, complaining of a tongue mass with spontaneous and contact pain. Clinical examination revealed a polypoid mass at the right lateral border of the tongue, measuring $30 \times 25 \times 15$ mm, with a smooth surface (Fig. 1A). There was no cervical lymphadenopathy. The patient's past medical history revealed congestive heart failure and insulin-dependent diabetes mellitus. She was being treated with warfarin, digoxin, and

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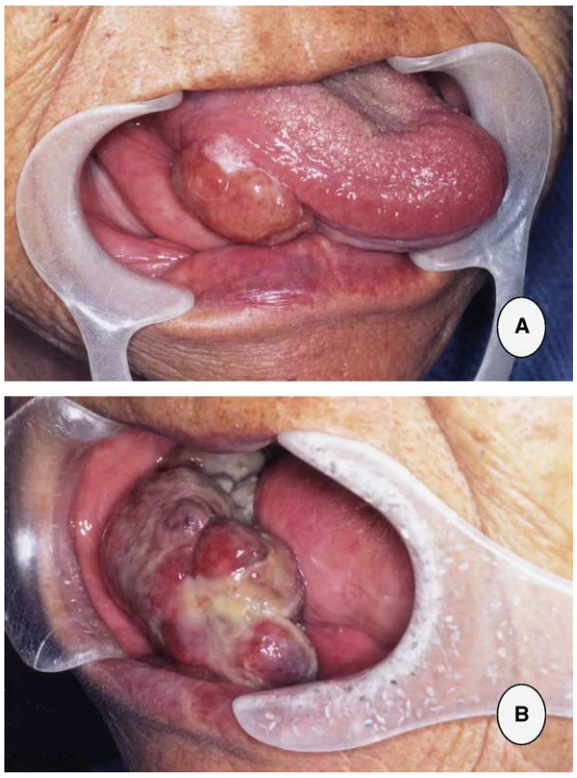

Figure 1 (A) Clinical appearance showing polypoid at the right lateral border of the tongue at the initial presentation (A) and 3 weeks later (B).

furosemide. Laboratory examination results were within normal range, except for the white blood cell count of 19.6×10^3/mm and the C-reactive protein level of 9.31 mg/dl. Three weeks after it first appeared, the tumor rapidly grew to approximately $50 \times 45 \times 35$ mm in size (Fig. 1B). Computed tomography (CT) scans with medium contrast and Magnetic Resonance Imaging (MRI) with a T2-weighted sequence depicted the lesion as a heterogeneous and multilobulated mass. A chest X-ray film showed no evidence of pulmonary or metastatic spread. The tumor was excised with an adequate margin around the stalk under general anesthesia. The patient's postoperative course was uneventful, and there was no evidence of recurrence at four years after the surgery.

Microscopic examination of the resected specimen showed that the tumor consisted of malignant epithelial and mesenchymal components. The two components were distinctly demarcated without a transition zone (Fig. 2). The epithelial element was a well-differentiated squamous cell carcinoma at the stalk of the tumor. The mesenchymal element was a rhabdomyosarcoma occupying most of the tumor. These tumors were characterized by the proliferation of oval or polyhedral cells with

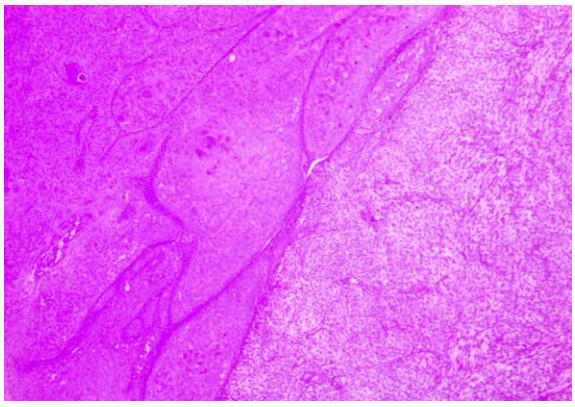

Figure 2 Photomicrograph showing the distinct demarcation between carcinomatous and sarcomatous components (H & E, ×200).

hyperchromatic nuclei and abundant acidophilic cytoplasm. Immunohistochemical studies showed that the sarcomatous cells were positive for myoglobin and vimentin and negative for desmin

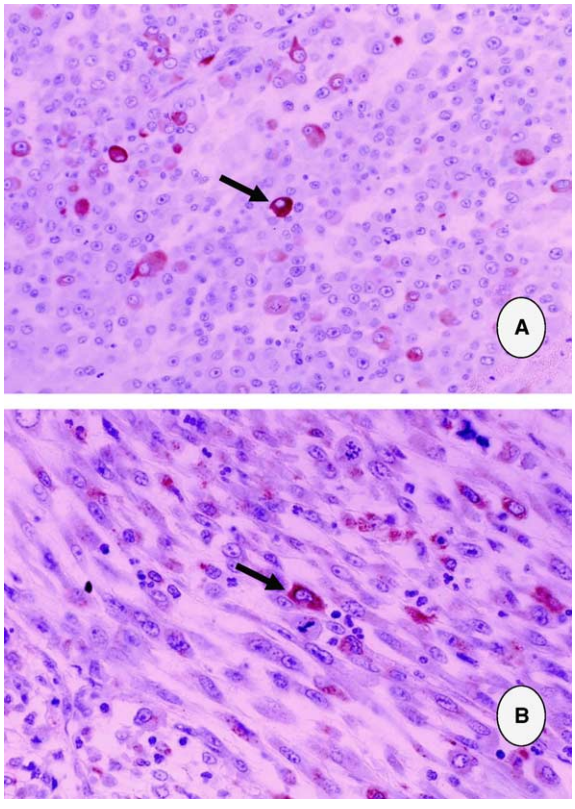

Figure 3 (A) Photomicrograph showing cytokeratin-positive cells (arrow) scattered in the in the mesenchymal component (immunostaining for myoglobin, ×400). (B) Photomicrograph showing positive staining for myoglobin (arrow) in the mesenchymal component (immunostaining for cytokeratin, ×400).

and s-100 protein, and that the carcinomatous cells were strongly positive for cytokeratin. A few cytokeratin-positive cells were scattered in the sarcoma (Fig. 3A and B). Consequently, a diagnosis of carcinosarcoma was made.

Discussion

Two antithetical hypotheses have been advanced to explain the histogenesis of carcinosarcomas:one proposes a multiclonal origin arising from two or more stem cells; the other a monoclonal origin from a single totipotential stem cell that differentiates in separate epithelial and mesenchymal directions.[8] Recent immunohistochemical and chromosomal analyses appear to have settled this argument in favor of the monoclonal hypothesis.[8,9] The current case, however, showed a sharp demarcation between the carcinomatous and sarcomatous elements without a transition zone, and the different immunohistochemical staining pattern of the two components for mesenchymal and epithelial markers, suggests a different origin for each tumor cell type. However possibility remains that the demarcation may not signify a different origin, since carcinoma components are demarcated in some sarcomatoid carcinomas in which the sarcomatous component derives from metaplasia of the epithelial cells.[10] The positive immunohistochemical staining for vimentin in the sarcomatous elements of our case is not diagnostic of mesenchymal cells, as several types of carcinomas, including squamous cell carcinomas, show coexpression of vimentin and cytokeratin.[4,11] Additionally, we found a few cytokeratin-positive cells scattered in the sarcoma. Although we cannot resolve the histogenesis in the present case with certainty, the findings suggest that the rhabdomyosarcoma cells arose from the carcinoma cells.

The primary treatment modality for sarcomatoid carcinoma should be the same as for squamous cell carcinoma, and surgical excision is the preferred treatment.[12,13] In the present case, the tumor was excised with an adequate margin and a suspicion of malignancy because of its rapid development. Poor prognosis has been reported in patients treated with radiotherapy, which is considered to be ineffective,[12–14] although adjuvant irradiation may be beneficial in patients who have positive surgical margins or who have nodal metastasis at the time of diagnosis.[12] The role of chemotherapy has not been established, but it may decrease the incidence of recurrence or metastasis of primarily sarcomatous tissue.

The lethality for oral sarcomatoid carcinoma has been reported to be 60%.[15] Of 59 patients with oral cavity sarcomatoid carcinoma studied by Ellis and Coriob,[16] 25 died from their disease, 12 suffered a recurrence, and 16 had metastases. The mean survival time of the patients who died of the cancer was 1.9 years. The gross morphology and size of the tumor, the differentiation of the carcinoma component, the depth of invasion, and prior treatment with radiation were reported as possible, although controversial, factors that might influence the prognosis.[12,17]

Excision with adequate margin around the stalk of the polypoid mass is effective not only for the diagnosis, but also for the treatment of a rapidly-growing tumor. The present case showed no evidence of recurrence or metastasis four years after surgery, but careful follow-up is necessary because of the poor prognosis in many cases.

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