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Basal cell adenocarcinoma of the soft palate with multiple bone metastases

KEYWORDS

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Basal cell adenocarcinoma (BCAC) is a rare salivary gland malignant tumor with variable basal and ductal neoplastic cells forming nests and glandular structures, accounting for <2% of all salivary gland neoplasms.^{1,2} It has no sex predilection. The average age of BCAC patients is 60 years, and only adults have been affected.³ Approximately 90% of these tumors occur in the parotid gland,³ followed by the submandibular gland, and BCAC of the minor salivary gland is extremely rare.^{4,5} Although BCAC is locally destructive, distant metastasis is exceedingly rare.⁴ Here, we reported a rare case of basal cell adenocarcinoma of the soft palate with multiple bone metastases.

A 69-year-old woman with an ulcer on the left soft palate was referred to our department. Intraoral examination revealed a 2 × 2 cm elastic hard mass with a 5 × 5 mm ulcer (Fig. 1A). Magnetic resonance imaging revealed a 2 × 2 cm well-circumscribed mass in the left soft palate had an intermediate signal intensity on T2-weighted images (Fig. 1B and C). A biopsy was performed, and the pathological diagnosis was BCAC. The patient underwent tumor resection under general anesthesia, with a safety margin of 10 mm. Histopathologically, the tumor was composed of small dark cells showing glandular and solid proliferation,

and had duct-like structures in which secretory product was seen in some areas. Each tumor nest consisted of basaloid epithelial cells with nuclear palisading at the periphery of the tumor nests (Fig. 1. D). Immunohistochemically, some tumor cells were positive for p53 protein, but most tumor cells were negative for vimentin, S-100 protein, and carcinoembryonic antigen. The postoperative course was uneventful, and the patient was followed up. However, the patient had lower back pain 12 months after surgery. Bone scintigraphy showed multiple bone metastases (Fig. 1. E). Therefore, radiotherapy (total of 33 Gy in doses of 3 Gy/day), as palliative therapy, was administered to the lumbar region. Although the pain was controlled by supportive care, the patient died 14 months after surgery.

The primary treatment of BCAC arising in the minor salivary gland is surgery. Because the incidence of cervical lymph node metastasis is approximately 10%, neck dissection is unnecessary unless there is cervical lymph node metastasis on clinical or radiologic examination.^{4,5} In patients with distant metastases, watchful waiting, radiation therapy, palliative surgery, and systemic chemotherapy would be treatment options.⁴ To our knowledge, there is only one report of distant metastases of BCAC arising from the minor salivary gland.⁴ Chen et al.⁴ reported liver metastases 14 months after primary surgery of BCAC of the buccal mucosa. The patient underwent a partial hepatectomy with adjuvant chemotherapy for liver metastasis and achieved long-term survival for 20 months after the hepatectomy. As this case, surgery is possible depending on the metastatic site. However, in the present case with multiple bone metastases, palliative radiotherapy was performed for pain control because surgery could not be applied. These two cases with distant metastases had no local recurrence and occult cervical lymph node metastasis.

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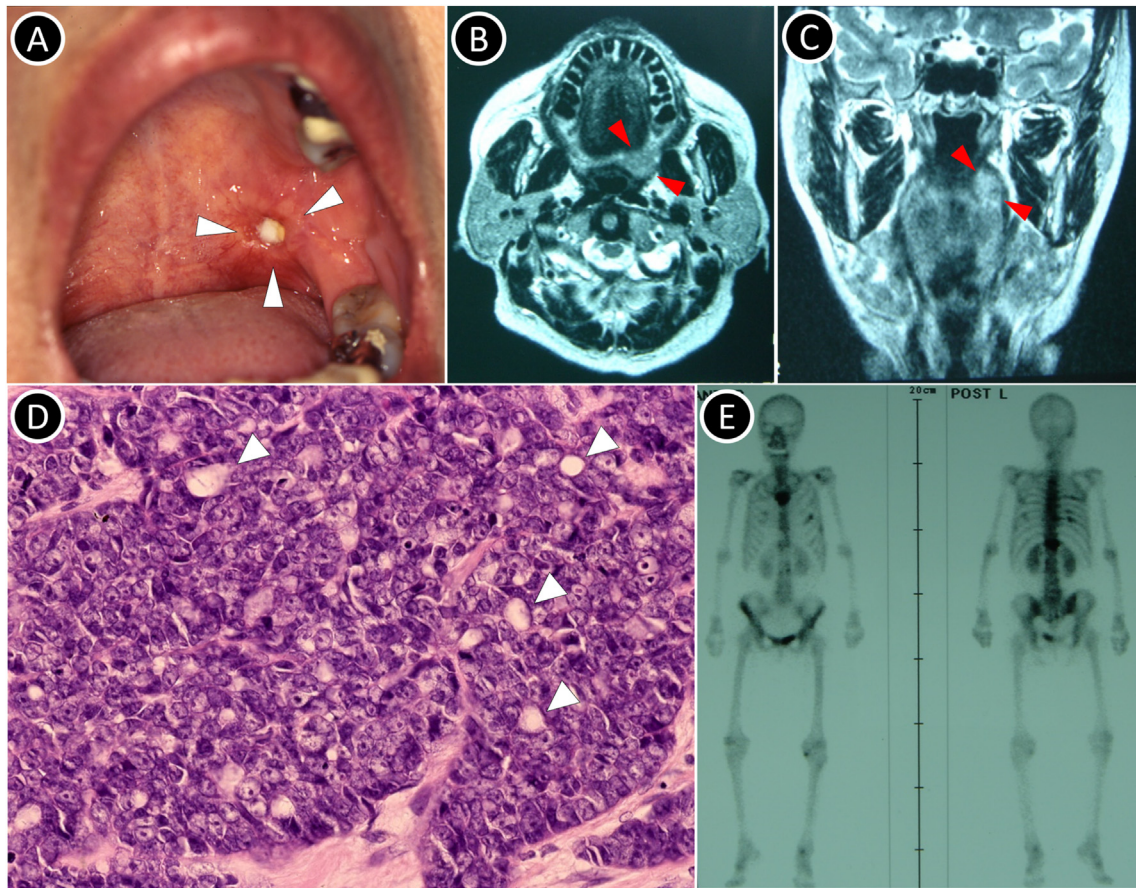


Figure 1 Intraoral photograph, radiological images, and histopathological photomicrographs (A) The Intraoral view showed an elastic hard mass (arrowheads) with an ulcer on the left soft palate (B and C) Magnetic resonance imaging (MRI) showed a well-circumscribed mass (arrowheads) in the left soft palate on T2-weighted axial and coronal images (D) Histopathologic examination showed each tumor nest consisted of basaloid epithelial cells with nuclear palisading and duct-like structures (arrowheads) (hematoxylin and eosin stain, $\times 200$) (E) Bone scintigraphy showed multiple bone metastases.

Declaration of competing interest

The authors have no conflicts of interest relevant to this article.

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Yuichiro Hayashi
Department of Oral and Maxillofacial Surgery, Shonan
Kamakura General Hospital, Kamakura, Kanagawa, Japan

Toshinori Iwai*
Satomi Sugiyama
Kenji Mitsudo
Department of Oral and Maxillofacial Surgery/
Orthodontics, Yokohama City University Hospital,
Yokohama, Kanagawa, Japan

*Corresponding author. Department of Oral and Maxillofacial Surgery/Orthodontics, Yokohama City University Hospital, 3-9 Fukuura, Kanazawa-ku, Yokohama, Kanagawa 236-0004, Japan.
E-mail address: iwai104@yokohama-cu.ac.jp (T. Iwai)

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