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Case Report

Simultaneous triple primary malignant melanomas occurring in the buccal mucosa, upper gingiva, and tongue: A case report

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ABSTRACT

Malignant melanoma of the oral cavity is a rare disease. Here, we present an extremely rare case of simultaneous triple primary malignant melanomas occurring in the buccal mucosa, upper gingiva, and tongue.

A 97-year-old male was referred because of tumors of the right buccal mucosa and left upper gingiva. Macroscopically, neither the tumors nor the adjacent mucosae exhibited melanin pigmentation. Biopsies of the two lesions showed that both were malignant melanomas. The patient underwent resection of the two tumors and right neck dissection. A histological examination of the surgical specimens demonstrated primary malignant melanomas and a metastatic lymph node. Most of the tumor cells were amelanotic, but displayed positive reactions for S100 or HMB45.

One month after surgery, a small amelanotic nodule appeared on the dorsum of the patient's tongue. An excisional biopsy demonstrated that this was also a primary malignant melanoma. The patient is well at 6 months after the initial surgical procedure.

1. Introduction

Malignant melanoma of the oral cavity accounts for a relatively high proportion of all cutaneous melanomas among the Japanese population [1]. However, the optimal methods for classifying and treating oral melanoma have not been established. Malignant melanoma of the oral cavity frequently affects the hard palate and upper gingiva, but rarely develops in the buccal mucosa or tongue. Most oral melanomas are considered to originate from pre-existing precancerous lesions, which is similar to the radial growth phase exhibited by acral lentiginous melanoma (ALM) of the skin [2,3]. Here, we present an extremely rare case of simultaneous triple primary malignant melanomas occurring in the buccal mucosa, upper gingiva, and tongue.

2. Case report

A 97-year-old male was referred to our hospital because of painless, pinkish-brown tumorous lesions in his right buccal mucosa and left upper gingiva, which he had first noticed a month ago (Fig. 1). There were no pigmented areas in the mucosae surrounding the tumors. Under a clinical diagnosis of malignant tumors, incisional biopsies were performed, and both lesions were pathologically diagnosed as malignant melanomas. A computed tomography examination demonstrated that the right submandibular lymph node was enlarged. Although the patient had a history of hypertension, chronic heart dysfunction, dementia, and pneumoconiosis, his performance status was 2, and it was considered that he would be able to tolerate radical surgery.

Under general anesthesia, he underwent right modified radical neck dissection (levels I–V), resection of the tumor in the right buccal mucosa, and left partial maxillectomy. Full thickness skin was grafted onto the raw surface after the buccal tumor had been resected. In a histological examination, a submandibular lymph node was found to contain metastatic melanoma cells. The operative time was 3 h, and 170 g of blood were lost during the procedure. The patient's postoperative course was good, and he was discharged 20 days after surgery.

☆ Asian AOMS: Asian Association of Oral and Maxillofacial Surgeons; ASOMP: Asian Society of Oral and Maxillofacial Pathology; JSOP: Japanese Society of Oral Pathology; JSOMS: Japanese Society of Oral and Maxillofacial Surgeons; JSOM: Japanese Society of Oral Medicine; JAMI: Japanese Academy of Maxillofacial Implants.

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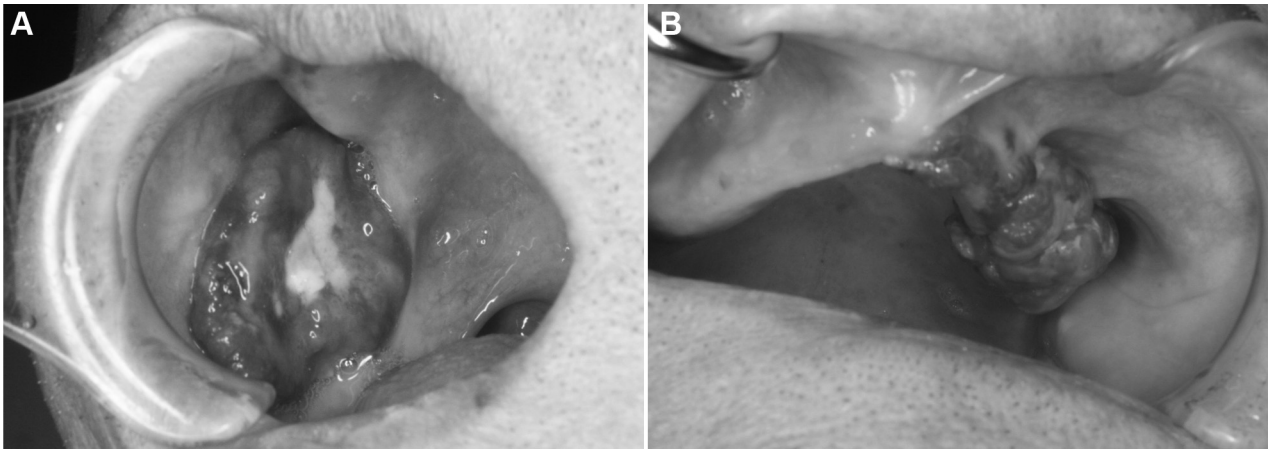

Fig. 1. A macroscopic examination showed painless, pinkish-brown tumors in the right buccal mucosa (A) and left upper gingiva (B). There were no pigmented areas in the adjacent mucosae.

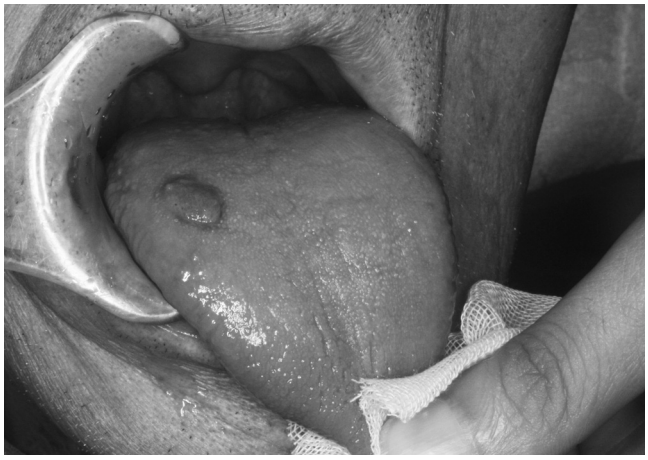

Fig. 2. An amelanotic nodule appeared in the dorsum of the tongue 4 weeks after the initial surgical procedure.

One month after the initial surgical procedure, a 10 mm × 10 mm amelanotic nodule appeared in the dorsum of his tongue. The nodule was located away from the scar caused by the first surgical procedure (Fig. 2). An excisional biopsy was performed under local anesthesia, which resulted in a histological diagnosis of malignant melanoma. The patient is well at 6 months after the initial surgical procedure.

Histological examinations of the surgical specimens obtained from the buccal mucosa, upper gingiva, and tongue detected the dense proliferation of atypical polymorphic tumor cells. In addition, there were focal areas of melanin pigmentation, although most of the tumor cells were amelanotic. The tumor cells exhibited positive reactions for S100 and HMB45 (DAKO Japan, Co. Ltd., Tokyo, Japan), and all three lesions were diagnosed as malignant melanomas (Fig. 3). The tumors in the buccal mucosa, upper gingiva, and tongue were 10, 7, and 2 mm thick, respectively. The intraepithelial spread of atypical tumor cells in the adjacent epithelia was observed in all three lesions. In these epithelia, individual proliferating atypical melanocytes were observed along the basal cell layer, and tumor cell nests in the lower epithelium and pagetoid upward migration were sometimes seen (Fig. 4). These findings resembled those of the radial growth phase of ALM of the skin or premalignant melanocytic dysplasia of the oral mucosa [3]. Furthermore, the above histological findings indicated that the three tumors were primary, not metastatic, malignant melanomas.

3. Discussion

Malignant melanoma of the oral cavity is a rare but potentially fatal disease. Among Japanese people, mucosal melanoma involving the oral cavity accounts for a relatively high percentage of all malignant melanomas [1]. Keratinizing mucosae, such as the hard palate or gingiva, are the most common sites for melanomas of the oral cavity, while melanoma occurring in the tongue is extremely rare. To the best of our knowledge, only seven cases of primary malignant melanoma of the tongue have been reported in the English literature [4–10].

Cutaneous melanomas are classified into four major types: lentigo maligna melanoma (LMM), superficial spreading melanoma (SSM), nodular melanoma, and ALM. However, the optimal methods for classifying and treating oral melanoma are disputed. Prior to the introduction of the ALM concept, many authors who attempted to classify oral melanoma pointed out the similarities between the histological features of oral melanoma and LMM of the skin [11–13], but the biological behaviors of these two lesions are quite different. After palmar, plantar, and subungual melanomas involving a radial growth phase were recognized as a separate entity from SSM and classified as ALM in 1976 [14], some investigators suggested that oral melanoma should be classified as ALM rather than LMM, in consideration of the poor clinical outcomes of oral melanoma patients [15–18]. On the other hand, many investigators have indicated that oral melanoma should be classified separately from cutaneous melanoma because of its extremely poor prognosis, although the histological features of oral melanomas involving a radial growth phase are similar to those of ALM of the skin [19–23]. Umeda et al. reported that most oral melanomas exhibit a radial growth phase that resembles that seen in ALM of the skin. In addition, they also found that the prognosis of oral melanoma was not worse than that of cutaneous melanoma when adequate therapy was performed [24] and advocated that ALM and oral melanomas involving a radial growth phase should be recognized as a single clinical, histological, and epidemiological category [1–3].

Although the optimal method for classifying oral melanoma is disputed, it is generally agreed that most oral melanomas exhibit a radial growth phase; i.e., a premalignant stage. There are few reports about precursors to oral melanoma. Takagi et al. reported that many cases of oral melanoma involved pre-existing melanosis [12]. Umeda et al. also described three cases of oral premalignant melanocytic dysplasia that displayed similar features to precursors of ALM of the skin [3]. It is known that multiple squamous cell carcinomas can occur simultaneously or metachronously in the oral cavity, pharynx, or esophagus. Such cases tend to involve