Journal of Oral and Maxillofacial Surgery, Medicine, and Pathology 26 (2014) 488–491

Contents lists available at ScienceDirect

Journal of Oral and Maxillofacial Surgery, Medicine, and Pathology

journal homepage: www.elsevier.com/locate/jomsmp

Case report

Acantholytic squamous cell carcinoma of the maxillary gingiva: Case report and literature review

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ARTICLE INFO

Article history:
Received 24 November 2012
Received in revised form 12 April 2013
Accepted 17 April 2013
Available online 25 May 2013

Keywords:
Acantholytic squamous cell carcinoma
Maxillary gingiva
Oral cavity
Prognosis

ABSTRACT

Acantholytic squamous cell carcinoma (ASCC) is an uncommon, histologically distinctive variant of squamous cell carcinoma. It occurs rarely in the oral cavity, and is especially rare at the maxillary gingiva. We report here the case of a 64-year-old female who requested examination of a diffuse swelling of the anterior maxilla. The histological diagnosis was ASCC. Maxillectomy involving the full-thickness of the upper lip, bilateral selective neck dissection, and immediate reconstruction with a double-folded, free radial forearm flap was conducted. The postoperative course was quite good. Neither recurrence nor metastases was found 30 months postoperatively. Only 15 cases of ASCC in the oral cavity have previously been reported, and our present case was the sixteenth. To improve understanding of the treatment and prognosis of ASCC, all published intraoral cases were reviewed.

1. Introduction

Acantholytic squamous cell carcinoma (ASCC) was first described by Lever [1] in 1947. ASCC is one of the variants of squamous cell carcinoma, such as spindle squamous cell carcinoma, basaloid squamous cell carcinoma, adenosquamous cell carcinoma, and papillary squamous cell carcinoma. ASCC is found more frequently on skin exposed to the sun, and is extremely rare in the oral mucosa [2]. Only 15 case reports of ASCC on the oral mucosa have been published in the English literature [2–12]. ASCC on the skin has been prognostically found to be poor with a higher risk of recurrence and metastasis compared with squamous cell carcinoma of the skin [12]. However, the prognosis of intraoral ASCC is controversial.

We report here the sixteenth case of ASCC of the maxillary gingiva that was managed with a good outcome. We also reviewed published cases of ASCC in the oral cavity with respect to the prognosis and treatment method using Kaplan–Meier analyses.

2. Case report

A 64-year-old female was referred to our facility because of diffuse swelling of the anterior maxilla (Fig. 1). On initial clinical examination, a 6-cm-diameter tumour of the maxillary gingiva was recognized. The clinical diagnosis was a malignant tumour of the maxillary gingiva. Histological examination of the incisional biopsy showed ASCC. MRI revealed a 5.0 cm × 3.8 cm × 2.7 cm tumour of the anterior maxilla, which invaded just beneath the skin of the upper lip (Fig. 2). Preoperative imaging (CT and PET/CT) showed no other tumours in the body. Maxillectomy involving the full thickness of the upper lip, bilateral selective neck dissection, and immediate reconstruction with a double-folded, free radial forearm flap was conducted (Fig. 3A and B). The surgical margins were free of tumour, and metastatic lymph nodes were not observed with the pathological diagnosis. The postoperative course was uneventful. No recurrence and no metastases were found 30 months after surgery. Chewing, swallowing, speech functions and facial aesthetics were satisfactory.

3. Pathological findings

Grossly, the resected tumour, which was 5.0 cm × 4.0 cm × 3.5 cm in size, was an exophytic mass in the upper gingiva that was invading maxillary bone and the upper

☆ AsianAOMS: 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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2212-5558/$ – see front matter
http://dx.doi.org/10.1016/j.ajoms.2013.04.008

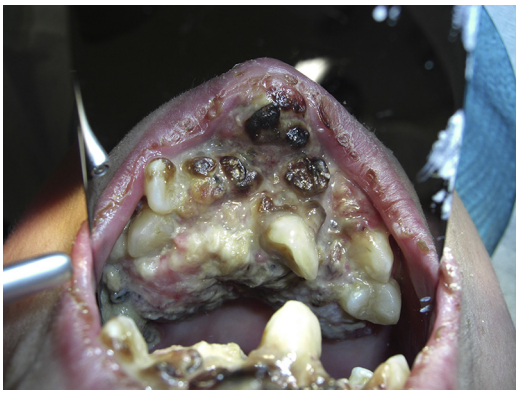

Fig. 1. Preoperative view of the anterior maxilla (mirror image). The tumour is seen to infiltrate just beneath the skin of the upper lip.

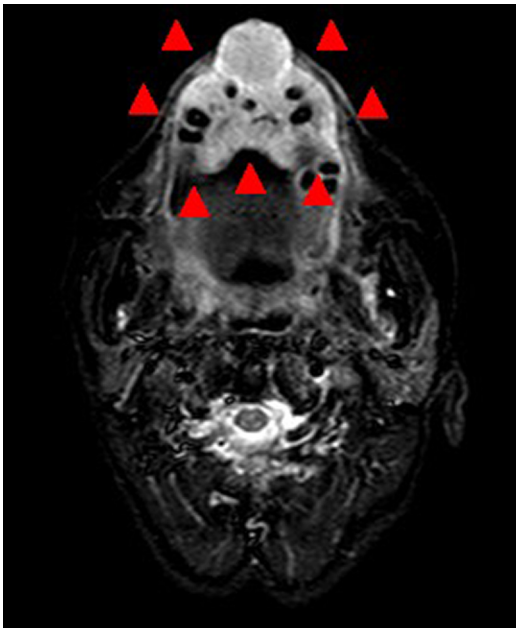

Fig. 2. Preoperative MRI (STIR) shows a lesion in the anterior maxilla (arrows).

lip. Microscopically, the tumour was composed of atypical stratified squamous epithelium with a sheet-like arrangement. In the periphery of the tumour growth, tumour cells were separated from one another, leading to cleft formation lined by a single layer of "tombstone-like" adenoid cells (Fig. 4). Desquamated dyskeratotic cells ("acantholytic cells"), like those of cutaneous pemphigus

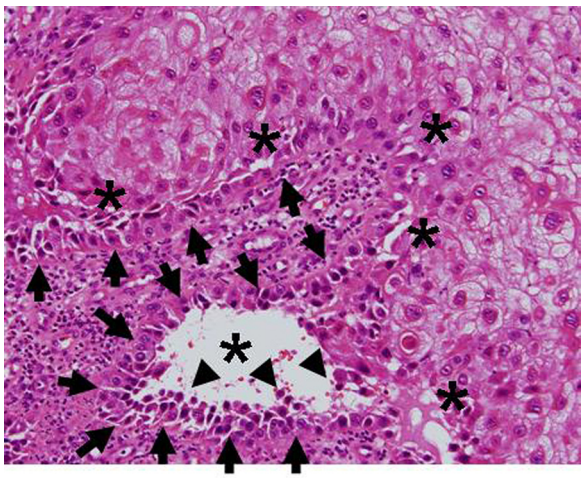

Fig. 4. High-power magnification (original magnification, ×400) shows loss of intercellular cohesion (*) at the periphery of tumour growth. Tombstone-like adenoid cells are seen (arrow). An acantholytic cell is shown by the arrowhead.

vulgaris, were observed. Adenoid cells were negative for periodic acid-Schiff and alcian blue stains. The present case showed the typical characteristics of an acantholytic (adenoid) squamous cell carcinoma. The underlying mucosa was not dysplastic. Vascular invasion and perineural invasion were not observed.

4. Discussion

ASCC of intraoral sites was first reported in 1977 [3], and 16 cases (including the present case) have been reported until now. Table 1 summarizes the clinical features and prognosis of all published cases of ASCC in the oral mucosa [2–12] (including the present case). The sites of ASCC in the oral mucosa were: tongue, 5; mouth floor, 4; maxillary gingiva, 3; mandibular gingiva, 2; and buccal mucosa, 2. Overall, there were 9 males and 7 females, and the ratio was 1.3:1.0. The average age was 61.6 (range 38–86) years. No cases with ASCC in their twenties or forties were seen, as is usual with SCC. Kaplan–Meier analyses with the pooled 16 cases revealed that survival at 46 months of intraoral ASCC was 0% (Fig. 5A), and disease-free survival was only 20.4% at 30 months (Fig. 5B). Furthermore, none of the cases with recurrence could be salvaged. ASCC in the oral cavity carries an extremely poor prognosis. More aggressive biological behaviour and a poorer prognosis are more common in ASCC of the skin than in typical SCC in the skin, the same applies to ASCC in the oral cavity.

Treatment modalities were described in 13 reported cases and the present case. Surgery alone was undertaken in 7 cases (tumour

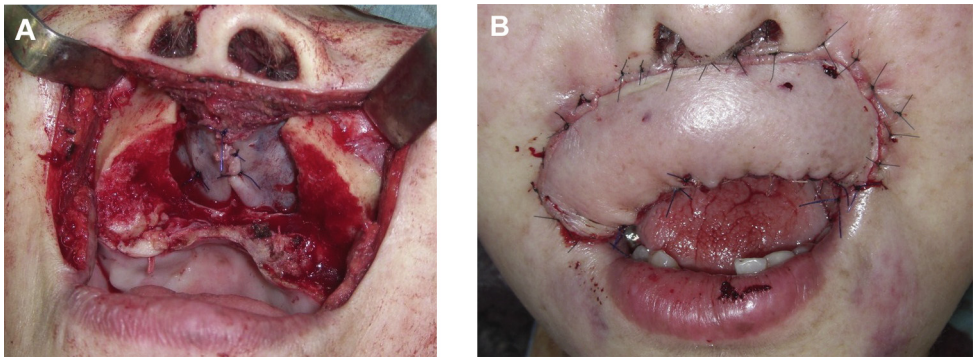

Fig. 3. (A) Defect after maxillectomy, including the full-thickness of the upper lip, and bilateral selective neck dissection. (B) Reconstruction with a double-folded, free radial forearm flap.