

Squamous Cell Carcinoma of the Tongue Dorsum: Incidence and Treatment Considerations

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Abstract Squamous cell carcinoma (SCC) of the tongue is the most common oral cancer. Most cases occur on the lateral border of the tongue and only very rarely on the dorsum. We retrospectively investigated the incidence of SCC of the tongue dorsum in our department between April 2006 and March 2015. Of the 368 patients with tongue cancer, only 3 had SCC of the tongue dorsum (incidence, 0.8 %). All patients with advanced cancer of the tongue dorsum underwent superselective intra-arterial chemoradiotherapy for organ preservation. We discuss the present findings in relation to past case reports and series in the English language literature as well as discuss treatments for SCC of the tongue dorsum.

Keywords Squamous cell carcinoma · Tongue dorsum · Incidence · Treatment · Chemoradiotherapy

Introduction

Squamous cell carcinoma (SCC) of the tongue is one of the most common oral cancers, with most cases occurring on the lateral border of the tongue. SCC of the tongue dorsum is very rare, especially at the midline [1–15]. Lesion of the dorsum might be overlooked or misdiagnosed as a more common benign lesion or oral manifestation of systemic disease [6, 16]. The purpose of this study was to investigate

the incidence of SCC of the tongue dorsum. We discuss the present findings in relation to case reports and series reported in the English language literature, as well as discuss the treatments available for SCC of the tongue dorsum.

Materials and Methods

We retrospectively investigated the incidence of SCC of the tongue dorsum in our department between April 2006 and March 2015. Cases of SCC of the tongue dorsum were pathologically re-evaluated to reconfirm the initial diagnosis and we reviewed the case series for the following factors: age, sex, TNM stage, lesion on the tongue dorsum before development of SCC, treatment, treatment response, and prognosis.

Results

Of the 368 patients with tongue cancer referred to our department, only 3 (1 man and 2 women; mean age, 61.7 years; range, 46–80 years) had SCC of the tongue dorsum (Figs. 1, 2, 3, Table 1), for an incidence was 0.8 %. These 3 patients, all with advanced cancer (T3 or T4a), underwent retrograde superselective intra-arterial chemoradiotherapy [17] for organ preservation. A complete response was observed in 2 patients, who had no dysfunction, such as swallowing or speech, and no recurrence or metastasis at 64–78 months after chemoradiotherapy. The remaining patient underwent salvage reconstructive surgery for a partial response, but died due to distant metastasis 3 months after the surgery.

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Fig. 1 Case 1. **a** Intraoral photograph before treatment. **b** Intraoral photograph after treatment

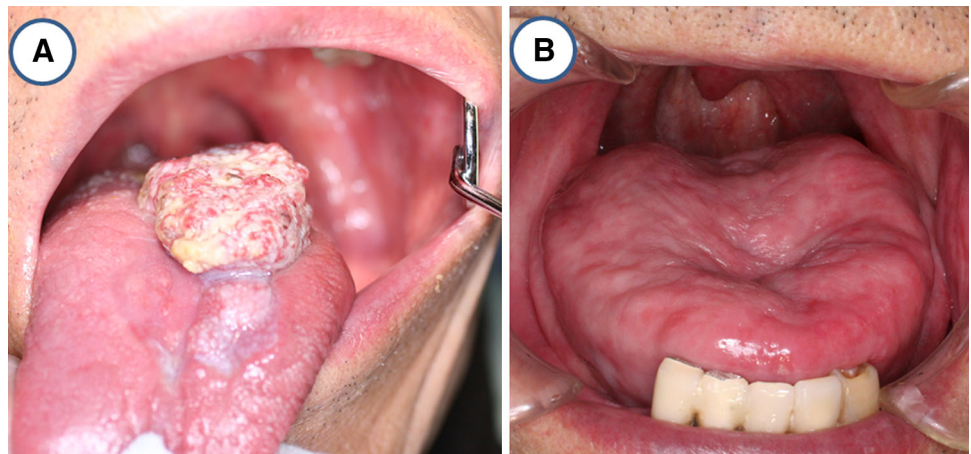


Fig. 2 Case 2. **a** Intraoral photograph before treatment. **b** Intraoral photograph after treatment

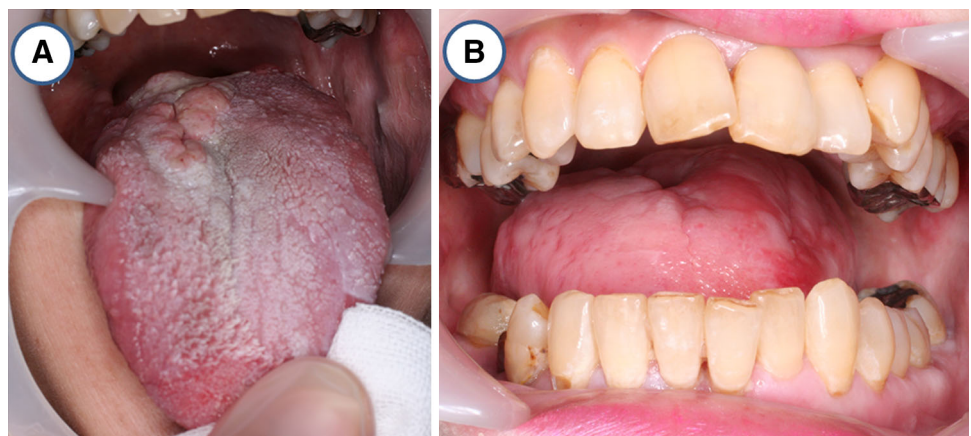


Fig. 3 Case 3. Intraoral photograph before treatment

Discussion

Most tongue cancers occur on the lateral border, making cancer of the dorsum very rare, with an incidence of 2.9–5.0 % (Table 2) [1–6]. SCC of the dorsal midline is even rarer and accounts for less than 1 % of tongue carcinomas [6, 10]. In the present study, pathological re-examination of 368 patients with tongue cancer revealed only

3 with SCC of the dorsum, for an incidence of 0.8 %. To our knowledge, only 17 cases, including our 3 cases, with SCC of the tongue dorsum have been well documented in the English language literature [6–15]. Among these 17 cases, the ratio of men to women was 1:1.43 (7 men and 10 women) and mean age was 60.8 years (range, 35–80 years). Seven patients were smokers and 10 were non-smokers. Lesions arose from the midline of the tongue in 7 patients, the right side in 3, and the left side in 2, and was not described in 5. SCC of the tongue dorsum is typically a single lesion, but Pastore et al. [15] reported a patient with 3 lesions as a rare case.

Clinically, SCC of the tongue dorsum may be difficult to suspect because it can resemble other lesions such as median rhomboid glossitis, granular cell myoblastoma, amyloidosis, and oral lichen planus [6, 8, 16]. In 1976, Ogus et al. [16] investigated the original records of 302 cases of SCC of the tongue and performed pathological re-examination. Although the original diagnosis revealed 7 cases (2.3 %) with SCC of the tongue dorsum in which surgery and/or radiotherapy was performed, pathological re-examination revealed that all cases were not in fact SCC

Table 1 Summary of patients with squamous cell carcinoma of the dorsum of the tongue in our series

Case	Age/Sex	TNM	Lesion on the tongue dorsum before SCC arising	Treatment	Treatment response of primary site	Outcome
1	80/M	T3N0M0	Bald tongue	SIACRT	CR	No recurrence or metastasis 78 months after CRT
2	46/F	T4aN1M0	Lichen planus	SIACRT and ND after induction CT	CR	No recurrence or metastasis 64 months after CRT
3	59/F	T3N2cM0	Geographic and fissured tongue with chronic glossitis	SIACRT and salvage surgery	PR	Died 3 months after salvage surgery for distant metastasis

M Male; *F* Female; *CRT* Chemoradiotherapy; *SIACRT* Superselective intra-arterial chemoradiotherapy; *CT* Chemotherapy; *ND* Neck dissection; *CR* Complete response; *PR* Partial response

Table 2 Incidence of squamous cell carcinoma of the tongue dorsum

Author	Reference	Year	Country	Incidence
Frazell and Lucas	[1]	1962	USA	4.1 % (63/1554)
Flamant et al.	[2]	1964	France	4.1 % (37/904)
Saxena	[3]	1970	Canada	5.0 % (22/440)
Mendelson et al.	[4]	1976	USA	4.1 % (14/340)
Kari et al.	[5]	1997	Finland	2.9 % (3/105)
Goldenberg et al.	[6]	2000	Israel	5.1 % (5/99)
Present study	–	2016	Japan	0.8 % (3/368)

but benign lesions, namely medial rhomboid glossitis, granular cell myoblastoma, amyloidosis, or ulcerated hemangioma. SCC may not occur on the dorsum of the tongue as frequently as has been suggested in earlier English language reports, because many reported cases may indeed have been misdiagnosed [16]. To avoid misdiagnosis, surgeons should obtain specimens of sufficient size as atraumatically as possible for differential diagnosis [16].

Several authors reported that SCC of the dorsum of the tongue arose from median rhomboid glossitis [12–14] or oral lichen planus [6–11, 15]. On the contrary, no lesions prior to developing SCC arose on the tongue dorsum in any patients [6]. There seems to be no relationship between median rhomboid glossitis and carcinoma [12], although a biopsy should be performed for lesions with an irregular border, increased size, induration, or accompanying tenderness or pain [13, 14]. In our series, lesions on the tongue dorsum prior to SCC arising were geographic and fissured tongue with chronic glossitis, and lichen planus. However, we consider lesions excluding lichen planus not to be precancerous.

According the World Health Organization's definition, oral lichen planus is considered a precancerous condition. Whether or not it is precancerous remains controversial, but several authors have reported cases of long-standing

lichen planus on the dorsum of the tongue that was diagnosed as SCC [6–11, 15]. Case 2 in the present study provides further histopathologic evidence of the malignant potential of oral lichen planus. The erosive and/or atrophic forms of oral lichen planus undergo malignant transformation more readily than other variants of lichen planus [7, 10, 11]. However, several authors have reported SCC arising from keratotic forms [8, 15], as well as from erosive and/or atrophic forms [7, 10, 11] of lichen planus of the dorsum of the tongue. Case 2 in the present study had a keratotic form of lichen planus of the dorsum as a precancerous condition, and Coombes [8] reported a case of lichen planus of the tongue dorsum that showed malignant transformation (SCC) in a patient who did not attend regular follow-up. Accordingly, long-term regular follow-up of patients with oral lichen planus is advisable [8, 10, 15]. Specifically, oral and maxillofacial surgeons should follow cancer patients regularly every 6–12 months for oral cancer surveillance [9]. Furthermore, when changes in the appearance of the lesion are observed, a biopsy should be performed for the possibility of a malignant transformation [8].

Frazell and Lucas [1] reported 63 patients with SCC of the tongue dorsum. Of 58 patients in which the size of the primary lesion was estimated, at least 44 (75.9 %) had early cancer of the tongue dorsum [1]. Of the 59 patients

who were treated, 39 (66.1 %) underwent surgery and 20 (33.9 %) underwent radiotherapy [1]. Cervical lymph node metastasis was noted in 9 of the 63 patients (14.3 %), with bilateral cervical lymph node metastasis evident in 3 patients [1]. According to Mendelson et al. [4], the incidence of cervical lymph node metastasis is 21.4 % (3 of 14 cancer patients). Our review of 17 well-documented cases of SCC of the tongue dorsum, including our 3 cases, showed that 9 (64.3 %) were T1 or T2 SCC of the tongue dorsum and 5 of 14 (35.7 %) were T3 or T4a SCC. Furthermore, 4 of 14 cases (28.6 %) involved at least one metastatic lesion in the cervical lymph nodes, with bilateral cervical lymph node metastasis observed in only 1 case. Initial treatment of the primary site in the 17 cases involved surgery in 9 cases, radiotherapy in 5, and chemoradiotherapy in our 3 cases. Of the 5 patients with T3 or T4a SCC of the tongue dorsum, 1 patient (T4aN0) underwent palliative radiotherapy but died 7 months after diagnosis. Although 1 patient (T4aN2b) underwent partial glossectomy and neck dissection followed by radiotherapy of the neck, the patient died 2 months after chemoradiotherapy due to local recurrence that was noted 2 months after the surgery. Our 3 patients underwent superselective intra-arterial chemoradiotherapy; 2 achieved a complete response with no recurrence or metastasis 64–78 months after chemoradiotherapy, and the remaining patient underwent salvage reconstructive surgery for a partial response, but died 3 months after the surgery due to distant metastasis.

Early-stage tongue cancer is generally treated with surgery (partial glossectomy) or radiotherapy (brachytherapy), whereas advanced tongue cancer requires wide excision and reconstructive surgery. In patients with locally advanced tongue cancer, laryngeal preservation in total glossectomy may also preserve swallowing function and maintain airway protection over the long term, compared with concurrent total laryngectomy. According to a systematic review by Dziegielewski et al. [18], 24 % of patients had used a gastrostomy tube at 1 year post-surgery. Rihani et al. [19] reported that of 94 patients with T3 or T4 tongue cancer who underwent total glossectomy with laryngeal preservation and free or pedicled flap reconstruction, the rates of tracheostomy decannulation and gastrostomy tube removal 1 year after surgery were 84.1 and 28.7 %, respectively. Although these rates in patients treated with primary surgery and postoperative radiation were 94.5 and 55.6 %, respectively, the corresponding rates in patients treated with primary radiation and salvage surgery were 77.6 and 12.1 %. On the contrary, chemoradiotherapy is one of the treatment options aimed at organ preservation in patients with advanced tongue cancer. McDowell et al. [20] investigated disease control and functional outcomes in patients with T4 SCC of the tongue

without mandibular involvement who had undergone surgery or definitive chemoradiotherapy. Disease outcomes 5 years after surgery or systemic chemoradiotherapy were not significantly different (61 vs. 70 % for the local control rate, 56 vs. 55 % for the progression-free rate, and 27 Vs. 40 % for the overall survival rate, respectively). McDowell et al. [20] concluded that chemoradiotherapy may be a reasonable alternative to surgery in patients with T4 tongue cancer, while Stenson et al. [21] reported that systemic chemoradiotherapy in such patients may negate the need for total glossectomy. Pederson et al. [22] reported that the rates of 5-year locoregional progression-free survival, overall survival, and disease-free survival in 127 patients with stage III or IV base of tongue cancer treated with systemic chemoradiotherapy was 87, 58.2, and 46 %, respectively. Concurrent chemoradiotherapy offers promising locoregional control for base of the tongue cancer [22]. Another option for advanced tongue cancer is intra-arterial chemoradiotherapy, especially superselective intra-arterial infusion [17, 23, 24]. Fuwa et al. [23] reported that the rates of 3-year local control and survival in 88 advanced tongue cancer patients were 72 and 57 %, respectively. In their study, 22 of 88 patients underwent not superselective but conventional intra-arterial chemoradiotherapy. Therefore, therapeutic results can improve further by increase of superselective catheterization to tumor-feeding artery. In our department, superselective intra-arterial chemoradiotherapy is performed for organ preservation in patients with advanced oral cancer [17]. Mitsudo et al. [17] reported 5-year local control and survival rates of 73 and 70.2 %, respectively. Superselective intra-arterial chemoradiotherapy can preserve organs and minimize functional disturbance, and as described above, the therapeutic results of chemoradiotherapy are not inferior to those of surgery.

Because total or subtotal glossectomy and microsurgical reconstruction is commonly performed for advanced SCC of the tongue including that involving the dorsum, post-operative function of the tongue may be poor. In the present study, superselective intra-arterial chemoradiotherapy achieved a favorable outcome (complete response) and preserved oral function in 2 of the 3 patients with advanced SCC of the tongue dorsum. However, salvage surgery was required in the third patient who had a poor response to superselective intra-arterial chemoradiotherapy and died 3 months after the surgery without tracheostomy decannulation or gastrostomy tube removal. According to Rihani et al. [19], most patients with advanced tongue cancer treated with primary surgery and postoperative radiation can achieve gastric tube independence and maintain adequate per oral intake, compared with patients treated with salvage surgery. Therefore, treatment for each patient should be selected carefully.

In conclusion, SCC of the tongue dorsum may be mimicked by a wide variety of benign and premalignant lesions, but SCC should be suspected when diagnosing lesions of the tongue dorsum, and surgeons should perform biopsy to avoid delayed diagnosis and enable early treatment. We would like to stress that careful treatment selection is essential in patients with advanced cancer of the tongue dorsum, with consideration given to the possibility of tumor tissue remaining after chemoradiotherapy, as well as poor function and prognosis after salvage surgery compared with primary surgery. Nevertheless, chemoradiotherapy, especially superselective intra-arterial chemoradiotherapy, can be considered as a treatment option.

Compliance with Ethical Standards

Conflict of interest All authors declare no conflicts of interest.

Ethical Approval All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

Informed Consent Informed consent was obtained from all individual participants included in the study.

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