

Case Report

Verrucous Carcinoma of the Tongue: Report of two cases

Michiro KAWAKAMI¹, Katsuhiko YOSHIMURA¹, Ibuki HAYASHI¹,
Kanako ITO², and Sawako HYO²

1 Department of Otolaryngology, Osaka National Hospital, Hoenzaka, Osaka, Japan

2 Department of Otolaryngology, Osaka Medical College, Takatsuki, Osaka, Japan

Key Words : verrucous carcinoma, tongue, laser surgery

ABSTRACT

Two cases of verrucous carcinoma (VC) in the tongue are reported. VC is a variant of well differentiated squamous cell carcinoma. VC grows gradually, has a tendency of local invasion and seldom metastasizes. The patients were treated by laser partial glossectomy. There was neither local recurrence nor distant metastasis observed. Surgical resection with sufficient safety margin is recommended.

INTRODUCTION

Verrucous carcinoma (VC) is a rare variant of well differentiated squamous cell carcinoma that has some unique characteristics. We encountered two cases of VC in the tongue. This paper documents the management of VC in the tongue and reviews the diagnosis and therapy of VC.

Case Report

Case 1

A 49-year-old man complained of the swelling and slight pain of the tongue for about three years. He was an ex-smoker and his Brinkman index was 600. There was a papillomatous lesion on the right lateral tongue with no induration (Fig.1). The punch biopsy was performed and the biopsy specimen was reported as VC. Cervical lymph node metastasis was not detected by ultrasonography. Preoperative TNM classification was T1N0M0.

Partial glossectomy by a laser knife was carried out. Safety margins were 5 - 10mm and no cancer cells were detected pathologically by frozen sections.

Pathological findings of the operation specimen (Fig.2):

Papillary proliferation and severe thickening of squamous epithelium are observed. Swelling of nuclei and mitoses are found occasionally. The invasion into the basement membrane is not detected. Pathologically, it was diagnosed as VC.

He made a satisfactory recovery. There was



Fig. 1 Macroscopic findings of case 1

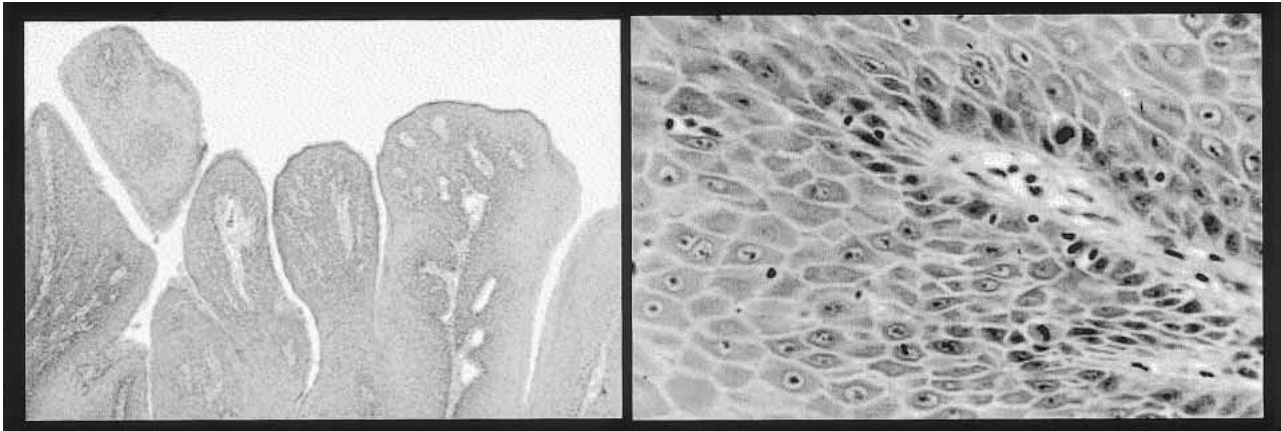


Fig. 2 Microscopic findings of operation specimen in case 1 (Hematoxylin and eosin stain) Left: original magnification x40, Right: original magnification x400

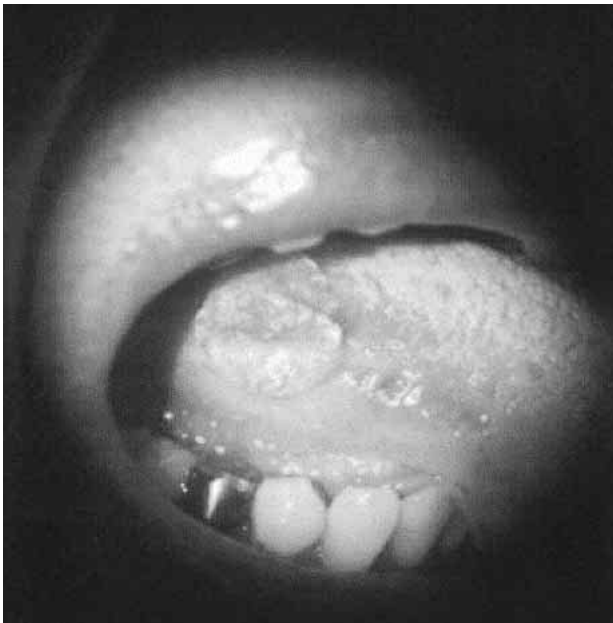


Fig. 3 Macroscopic findings of case 2

neither local recurrence nor distant metastasis observed for about two years.

Case 2

A 57-year-old woman complained of the swelling and pain of the tongue for about two years. She was a smoker and her Brinkman index was 1200. She also had low dental hygiene. There was a papillomatous lesion on the right lateral tongue with no induration (Fig.3). The punch biopsy was performed and following report was obtained. Pathological findings of the biopsy specimen (Fig.4, upper):

Papillary proliferation and thickening of squamous epithelium are observed. Cellular atypia is

not seen. The biopsy specimen was reported as papilloma.

Cervical lymph node metastasis was not detected by ultrasonography. Partial glossectomy by a laser knife was carried out. Safety margins were 5 - 10mm.

Pathological findings of the operation specimen (Fig.4, lower):

Papillary proliferation and severe thickening of squamous epithelium are also observed. Single cell keratinization and incomplete pearl formation are found occasionally. The basement membrane is partially disappeared. The pathological diagnosis was VC. Pathological TNM classification was T1N0M0.

She made an uneventful recovery. There was neither local recurrence nor distant metastasis observed for one year.

DISCUSSION

Verrucous carcinoma (VC) is a variant of well-differentiated squamous cell carcinoma (ACKERMAN, 1948). The oral cavity is one of the predilection sites for VC. This carcinoma has also been reported in the nasal cavity, larynx and esophagus. In the oral cavity, the buccal mucosa and lower gingiva is the common site (IMAI et al, 1995). VC of the tongue is clinically rare. Most patients are elderly males with smoking history (ACKERMAN, 1948). Our two cases also had histories of smoking. Regional lymph node metastases are exceedingly rare, and distant metastases have not been reported.

The pathological diagnosis of typical VC is not so difficult. But in some cases, superficial biopsies will show only hyperkeratosis, acanthosis and

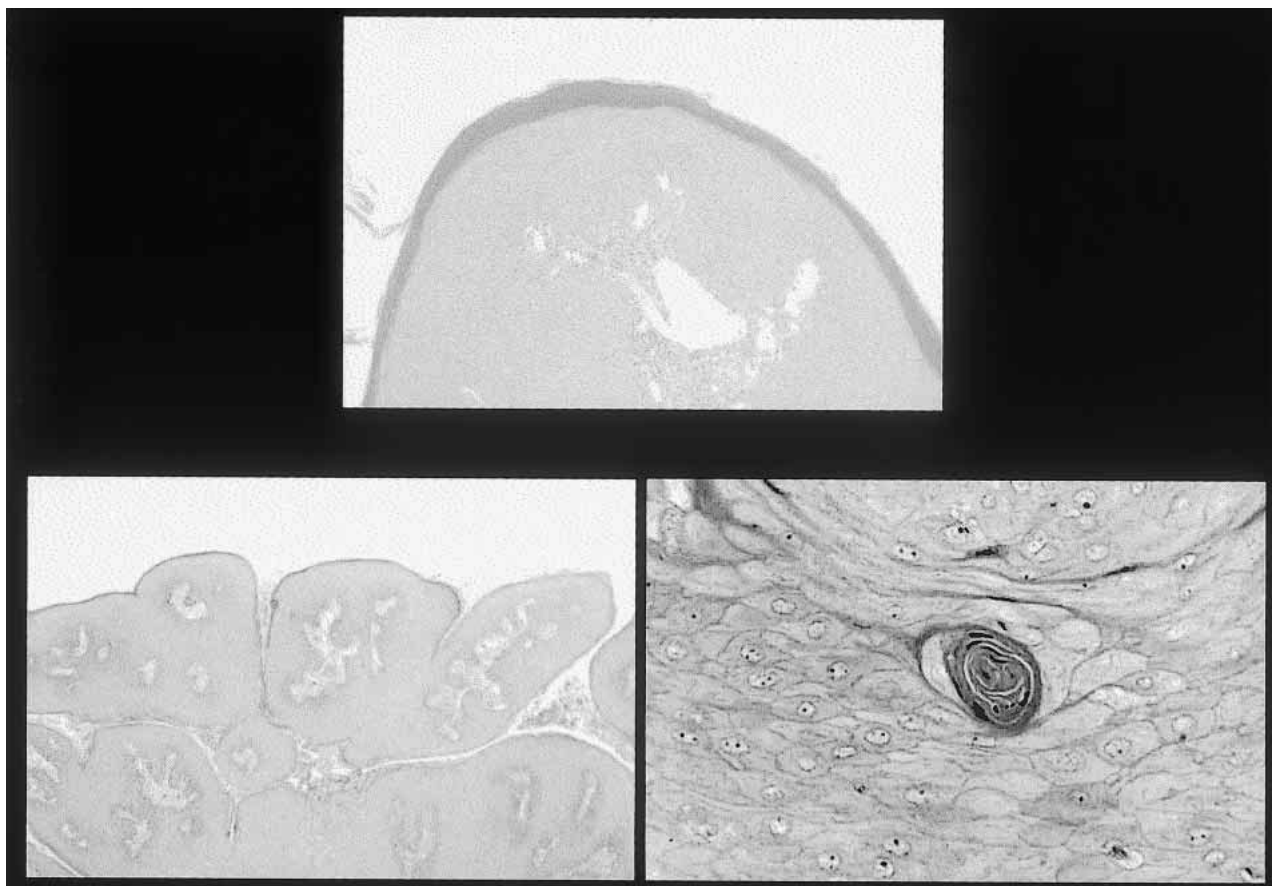


Fig. 4 Microscopic findings of in case 2 (Hematoxylin and eosin stain)
 Upper: biopsy specimen; original magnification x40
 Lower: operation specimen
 Left: original magnification x40, Right: original magnification x400

benign papillomatosis. Deeper adequate biopsies must be needed. The most important pathological difference with squamous cell carcinoma is the good cytological differentiation throughout the tumor. VC can also be mistaken as a benign lesion histologically. The close discussion between the clinicians and pathologists is necessary. Especially, identification of VC in papilloma is difficult. In our case 2, first biopsy specimen was diagnosed as papilloma. For exact diagnosis, sufficient volume is necessary. But in head and neck region, it is often difficult technically to obtain enough amount of biopsy specimen. There is also a problem of so called "hybrid" tumor: VC and papilloma, VC and usual squamous cell carcinoma. From such a point of view, we must take enough surgical margins when we resect papilloma of the tongue.

A link between VC and human papillomavirus (HPV) is a concern. Several studies implied a role for HPV in the development of VC. It is thought that the canceration mechanism of human papillo-

mavirus is the modulation of cell cycle control. In the oral cavity, several HPV subtypes were detected: 16 and 18 (JORDAN, 1995; ORVIDAS et al, 1999). However HPV's role in the etiology of VC has not been confirmed. In the field of gynecology, trial of a HPV type 16 vaccine had been applied to eradicate the cervical cancer (KOUTSKY et al, 2002). Tombs R, et al also suggest the relevance between CV of the tongue and lichen planus (TOMB et al, 2003). It is regrettable for us not to have checked HPVs in our two cases. Further studies must be needed to explicate the etiology of VC.

Complete resection of the tumor is the best treatment for VC. At the case of ordinary squamous cell carcinoma of the tongue in stage 1 and 2, we usually perform supraomohyoid neck dissection (MEDINA et al, 1989) prophylactically regardless of the existence of lymph node metastasis or not. In almost all cases of VC, neck dissection is not necessary because lymph node metastases are

extremely rare (KATO et al, 1991). In cases of tongue cancer, sentinel lymph nodes are thought to exist in the upper internal jugular chain and submandibular region. Generally, ultrasonography is very effective to detect lymph node metastases. But, especially in submandibular region, inflammatory lymph node swellings are fluent, so, it is difficult to distinguish lymph node metastases from inflammatory lymph node swellings. At the case of cervical lymph node swellings detected, an ultrasound-guided fine needle aspiration biopsy is absolutely recommended to avoid redundant neck dissection of the VC patient.

In our two cases, we used a laser knife as a cutting device. In the head and neck region, to keep safety margins sufficiently is very difficult. Using a laser knife, the cutting edges were heat-degenerated and heat-degeneration might contribute the improvement of the local control of the carcinomas. We can also operate easily with less bleeding and no muscle constrictions.

Radiation therapy is not usually applied since it may change the nature of the tumor to a poorly differentiated squamous cell carcinoma. But some authors (HAMAMOTO et al, 2000; THARP et al, 1995) reported that anaplastic transformation of VC after irradiation is not common and the radiosensitivity of VC is usually good, therefore radiotherapy can become a radical treatment for VC. In our cases, fortunately, tumors were not so invasive and curable resection with sufficient safety margin could be performed and we did not need to consider the irradiation. In all cases of VC, surgery should be used if the procedure has acceptable morbidity. Irradiation might be the second choice for VC treatment when sufficient operation could not be done. On chemotherapy, Tanaka, et al reported the effectiveness of preoperative chemotherapy for advanced VC of the tongue (TANAKA et al, 1992). But the effect of chemotherapy on VC has not been thoroughly estimated at this moment in time. In most cases, operations are the first choice for the treatment of VC and chemotherapy has only a complementary role to surgical procedure.

REFERENCES

- ACKERMAN LV. Verrucous carcinoma of oral cavity. *Surgery* 23:670-678, 1948
- HAMAMOTO Y, NIINO K, YOSHINO M, ITAGAKI T, YAMAGUCHI K, YOSHIZAWA N, et al. Radiation therapy for oral verrucous carcinoma. *J Jpn Soc Ther Radiol Oncol* 12:307-313, 2000.
- IMAI H, YOSHIHARA T. Verrucous Carcinoma of the Tongue; Report of a Case. *Otolaryngology-Head and neck Surgery (Tokyo)* 67: 1165-1169, 1995.
- JORDAN RC. Verrucous carcinoma of the mouth. *J Can Dent Assoc* 61:797-801, 1995.
- KATO A, TAKAHASHI Y, YANOHARA K. Verrucous Carcinoma of the Tongue; A Case Report. *Practica Oto-Rhino-Laryngologica* 84:775-780, 1991.
- KOUTSKY LA, AULT KA, WHEELER CM, BROWN ER, BARR E, ALVAREZ FB, et al. A controlled trial of a human papillomavirus type 16 vaccine. *N Engl J Med* 347:1645-1651, 2002.
- MEDINA JE, BYERS RM. Supraomohyoid neck dissection; Rationale, indications and surgical technique. *Head & Neck* 2: 111-122, 1989.
- ORVIDAS LJ, LEWIS JE, OLSEN KD, WEINER JS. Intranasal verrucous carcinoma: Relationship to inverting papilloma and human papillomavirus. *Laryngoscope* 109:371-375, 1999.
- TANAKA J, YOSHIDA K, TAKAHASHI M, SUZUKI M. A case of verrucous carcinoma of the tongue, effectively treated with preoperative chemotherapy (UFT, CDDP, PEP) and irradiation. *Gan To Kagaku Ryoho*. 19:525-7, 1992.
- THARP MET, SHIDNIA H. Radiotherapy in the treatment of verrucous carcinoma of the head and neck. *Laryngoscope* 105:391-396, 1995.
- TOMB R, EL-HAJJ H, NEHME E, HADDAD A. Verrucous carcinoma of the tongue occurring on lesions of lichen planus. *Ann Dermatol Venereol* 130:55, 2003.

Received August 19, 2004

Accepted October 17, 2004