

Metastasis to the Lingual Lymph Node in Patients with Squamous Cell Carcinoma of the Floor of the Mouth: A Report of Two Cases

MASAHIRO UMEDA*, TSUTOMU MINAMIKAWA,
TAKASHI SHIGETA, AKIKO OGUNI, TOMOKO KATAOKA,
HIDENORI TAKAHASHI, YASUYUKI SHIBUYA,
and TAKAHIDE KOMORI

Department of Oral and Maxillofacial Surgery, Kobe University Graduate School of Medicine, 7-5-1Kusunoki-cho, Chuo-ku, Kobe, 650-0017, Japan.

Received 30 June 2009/ Accepted 7 August 2009

Key Words: Carcinoma of the floor of the mouth, lingual lymph node, neck dissection, recurrence

Cancer of the tongue or the floor of the mouth sometimes metastasizes to the lingual lymph node. We present two patients with squamous cell carcinoma of the floor of the mouth who developed metastases to the lateral lingual lymph nodes.

Case 1, a 62-year old male, had squamous cell carcinoma of the floor of the mouth (T3N2cM0). He underwent tumor resection and bilateral neck dissection, and histological examination revealed five metastatic nodes including the lateral lingual node near the hyoid bone. No recurrent tumors were evident, but he died of pneumonia 10 months after the surgery.

Case 2, a 62-year old male, had squamous cell carcinoma of the floor of the mouth (T2N2cM0). He underwent tumor resection and bilateral neck dissection, and histological examination revealed three metastatic nodes including the lateral lingual node near the sublingual gland. No recurrence was found in the oral and neck regions, but he died of liver metastasis 18 months after the surgery.

Metastasis to the lingual lymph node may cause a recurrence of oral cancer in the neck, since conventional neck dissection cannot remove this node even in the case of en bloc resection of the primary tumor and the neck. When CT, MRI, or intra-operative palpation findings lead to a suspicion of metastasis to the lingual lymph node, the area of neck dissection should be extended to include this node.

Squamous cell carcinoma of the oral cavity often metastasizes to the cervical lymph nodes, and the prognosis depends upon the control of the metastatic rather than the primary tumor³⁾. Neck dissection, including functional, modified, and radical dissection, has been used for patients with oral cancer accompanied by lymph node metastasis, but in some cases it resulted in neck failure. It has been reported that tumor recurrence after neck dissection often occurs in patients with multiple positive nodes and extra-nodal invasion²⁾. However, recurrence near the hyoid bone was observed in some of our patients with tongue cancer accompanied by a single metastatic node but no extra-nodal invasion.

It is known that lymph nodes occasionally are located along some lymph vessels of the tongue and are known as lingual lymph nodes. The existence of lingual lymph nodes had

received little attention and no studies of metastasis of oral cancer to these lymph nodes had been reported until Ozeki *et al.*⁴⁾ first published their report on three patients with tongue cancer metastasizing to these lymph nodes. The lingual lymph node cannot be removed with usual neck dissection, so that metastasis to this node may cause neck recurrence of oral cancer. We present here two new patients of carcinoma of the floor of the mouth who had metastases to the lateral lingual lymph nodes.

CLINICAL CASES

Case 1

A 62-year old male was referred to our hospital because of pain in the floor of the mouth. Clinical examination revealed a 45 x 28mm tumor with ulceration in the anterior part of the left floor of the mouth (Fig. 1), and an enlarged submandibular lymph node. The diagnosis after histological examination of the biopsy specimen was well differentiated squamous cell carcinoma. CT and MRI images showed an enlarged left submandibular lymph node, and a right lateral lingual lymph node, which were located in the inner- and upper areas of the submandibular gland (Fig. 2). Because of the diagnosis of carcinoma of the floor of the mouth (T3N2cM0), he underwent bilateral functional neck dissection, resection of the tumor, and reconstruction using a free rectus abdominis myocutaneous flap. After resection of the mylohyoid muscle, an elastic hard, lateral lingual lymph node was found on the muscle near the hyoid bone. Histological examination showed that metastasis was evident in two left submandibular nodes, two left upper jugular nodes, and one right lateral lingual node.

There was no evidence of tumor recurrence, but he died of pneumonia 10 months after the surgery.

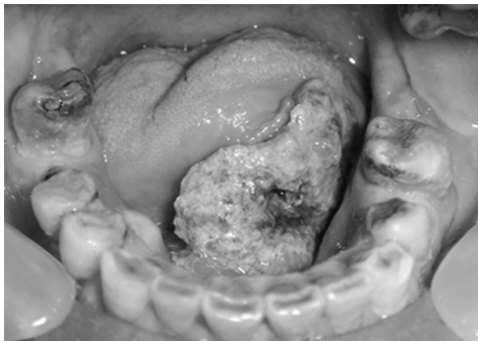


Figure 1 Intraoral findings of Case 1.



Figure 2 MRI image of Case 1 showing the lateral lingual lymph node.

LINGUAL LYMPH NODE METASTASIS OF ORAL CANCER

Case 2

A 62-year old male was referred to our hospital because of swelling of the anterior floor of the mouth. Clinical examination revealed a tumor with ulceration in the median part of the floor of the mouth (Fig 3), and an enlarged left midjugular lymph node. The diagnosis after histological examination of the biopsy specimen was moderately differentiated squamous cell carcinoma. CT and MRI images showed an enlarged left midjugular lymph node, and a right lateral lingual lymph node which were located in the inner- and lower areas of the right sublingual gland (Fig. 4). Because of the diagnosis of carcinoma of the floor of the mouth (T2N2cM0), he underwent bilateral functional neck dissection, resection of the tumor, and reconstruction using a free forearm flap. When en bloc resection of the tumor and the neck was performed, an elastic hard, right lateral lingual lymph node was palpated near the sublingual gland. Histological examination showed that metastasis was evident in two left midjugular nodes and one right lateral lingual node. The patient obtained loco-regional control but died of liver metastasis 16 months after the surgery.

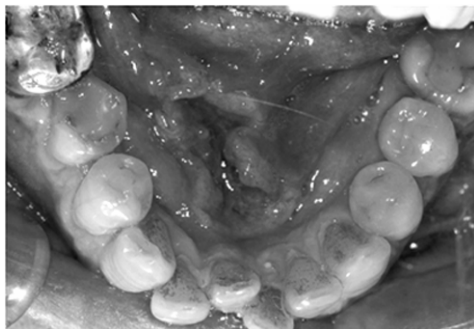


Figure 3 Intraoral findings of Case 2.

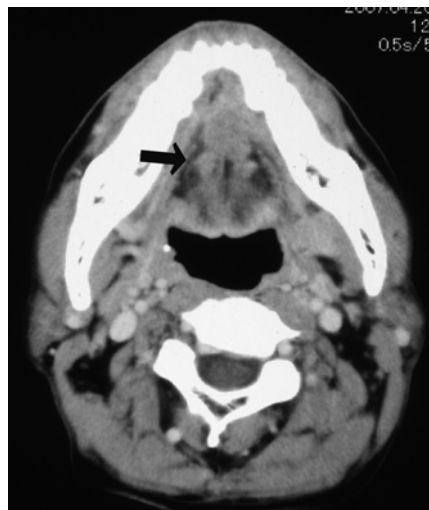


Figure 4 MRI image of Case 2 showing the lateral lingual lymph node.

Surgical approach to the lingual lymph nodes

A neck dissection from levels II to V was performed with the usual method, and before the dissection of level I, the lingual lymph nodes were examined with the following method: after removal of the digastric muscle from the hyoid bone (Fig. 5a) and resection of the mylohyoid muscle near the hyoid bone (Fig. 5b), the geniohyoid muscle was pulled to the median side (Fig. 5c). A finger was inserted along the hypoglossus nerve and fascia of the genioglossus muscle to palpate the lateral lingual lymph nodes (Fig. 5d), after which the finger was moved to the medial side of genioglossus muscle to palpate the median lingual lymph nodes. The superior part of the lingual lymph node is located near the sublingual salivary gland, and the inferior part near the hyoid bone. When metastasis to the lingual lymph node is suspected, the lymph node is included in the resection, but when no lymph nodes are detected, dissection of this area is terminated and proceeds to neck level I.

DISCUSSION

Some lymphatic vessels of the tongue run along the genioglossus muscle and flow out into the submandibular lymph nodes. The lingual lymph nodes are intermediate lymph nodes

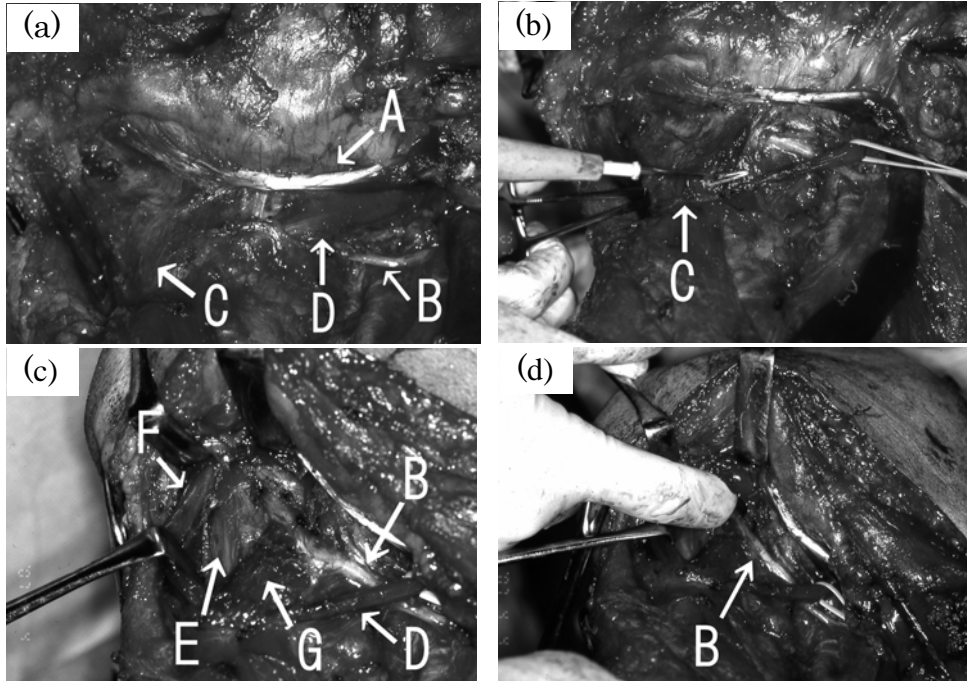


Figure 5 Surgical approach for lingual lymph node. (a): Posterior belly of the digastric muscle is resected near the mastoid notch and the intermediate tendon is removed from hyoid bone. (b) Mylohyoid muscle is resected on the hyoid bone. (c) After the geniohyoid muscle is pulled forward, the hypoglossus and genioglossus muscles become visible. (d) A finger is inserted along the hypoglossus nerve and the fascia of the genioglossus muscle. The presence of a lingual lymph node is easily determined by palpation using a finger. A: digastric muscle, B: hypoglossus nerve, C: mylohyoid muscle, D: stylohyoid muscle, E: genioglossus muscle, F: geniohyoid muscle, G: hypoglossus muscle.

which occasionally are located outside a genioglossus muscle (lateral lingual lymph nodes) or between the bilateral genioglossus muscles (median lingual lymph nodes) (Fig. 6).

Rouviere⁶⁾ first noted the presence of the lingual lymph node and referred to them as the median and lateral lingual lymph node. Omura *et al*⁴⁾ also identified the lingual lymph nodes as inconstant, interrupting nodules placed deeply above the suprahyoid region along the course of lymphatic vessels that drain the tongue and the floor of the mouth. However, according to the TNM classification, the lingual node is not classified as any level of necks, and there have been few reports of lingual node metastasis from oral cancer. Ozeki *et al.*⁵⁾ first reported three cases of tongue cancer metastasizing to the lingual lymph nodes in 1985. Dutton *et al.*¹⁾ also reported on a patient with tongue cancer who had occult metastasis to the lingual lymph node, and stated that a traditional discontinuous neck dissection may not remove all of the lymph nodes at risk of metastasis from the oral cavity. However, the

LINGUAL LYMPH NODE METASTASIS OF ORAL CANCER

significance of this lymph node for cancer of the tongue and floor of the mouth has not been fully documented.

We used to treat early tongue cancer patients with interstitial brachytherapy, but this was sometimes followed by local as well as neck recurrence of the cancer⁷⁾. For the past 10

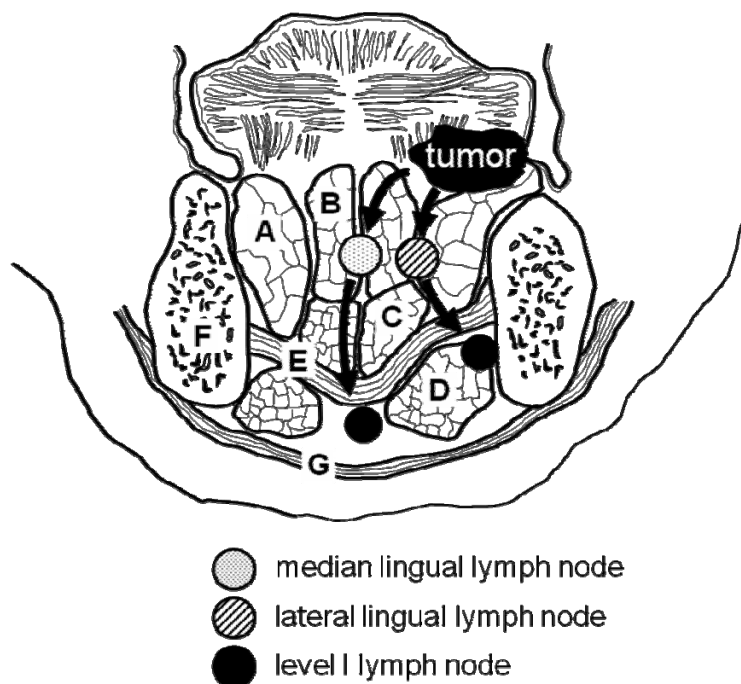


Figure 6 Schematic representation of the median and lateral lymph nodes. The nodes are located along the genioglossus muscle and flow into the level I nodes .A: sublingual salivary gland, B: genioglossus muscle, C: geniohyoid muscle, D: anterior belly of digastric muscle, E: mylohyoid muscle, F: body of mandible, G: platysma muscle.

years, therefore, surgery has been the first choice of therapy for both early and advanced tongue cancer at our hospital, resulting in a substantial improvement in the local cure rate⁸⁾. Currently, the main causes of death from tongue cancer are now neck failure and distant metastasis. Neck recurrence of tongue cancer has been reported in the level II neck, especially in patients with multiple neck metastases associated with extranodal invasion²⁾. However, such neck recurrence has rarely been observed recently, probably because of adequate dissection of the upper- and posterior part of the level II neck combined with resection of the posterior belly of the digastricus muscle and for advanced cases parapharyngeal dissection of posterior invading tongue cancer^{9,10)}. However, some of our patients showed neck recurrence near the hyoid bone.

Of the 58 patients with carcinoma of the tongue or the floor of the mouth who underwent en bloc resection of the primary tumor and neck dissection between 1999 and 2004, we found three patients with neck recurrence probably caused by metastasis to the lingual lymph node. This lymph node may not be removed by normal neck dissection, even in cases of en bloc resection. Whether the lingual lymph node should be resected when it is detected by preoperative imaging or intraoperative palpation in patients with oral cancer who are undergoing neck dissection needs to be discussed. In order to remove the lingual lymph

node, suprahyoid muscles including the mylohyoid, geniohyoid, and genioglossus muscles should be widely resected but this may cause major functional problems with swallowing and mastication. The frequency of metastasis to the lingual lymph nodes among oral cancer patients is relatively low, and we therefore believe that the area of the neck dissection should only be extended to these nodes when CT, MRI, or intraoperative findings lead to a suspicion of metastasis in lingual lymph nodes, or when pathological examination during surgery produces histological evidence of metastasis. Since ours is a report of a small number of patients with metastases to the lingual lymph nodes, further studies are needed to elucidate whether this surgical procedure results in reduced recurrence in the neck after surgery for tongue or floor of the mouth cancer.

REFERENCES

1. **Dutton, J.M., Graham, S.M., and Hoffman, H.T.** 2002. Metastatic cancer to the floor of mouth: the lingual lymph nodes. *Head Neck Surg* **24**: 401-405.
2. **Godden, D.R., Ribeiro, N.F., Hassanein, K., Langton, S.G.** 2002. Recurrent neck disease in oral cancer. *J Oral Maxillofac Surg* **60**: 748-753.
3. **Goto, M., Hasegawa, Y., Terada, A., Hyodo, I., Hanai, N., Ijichi, K., Yamada, H., Fujimoto, Y., Ogawa, T.** 2005. Prognostic significance of late cervical metastasis and distant metastasis in patients with stage I and II oral tongue cancers. *Oral Oncol* **41**: 62-69.
4. **Omura K, Yanai C, Yamashita T.** 1997. Diagnosis and management of lingual lymph node metastases. *Int J Oral Maxillofac Surg* **26**: 45-??.
5. **Ozeki, S., Tashiro, H., Okamoto, M., Matsushima, T.** 1985. Metastasis to the lingual lymph node in carcinoma of the tongue. *J Maxillofac Surg* **13**: 277-281.
6. **Rouviere H.** 1938. *Anatomy of the human lymphatic system* (Tobia MJ, editor and translator). Ann Arbor: Edward Brothers.
7. **Umeda, M., Komatsubara, H., Nishimatsu, N., Yokoo, S., Shibuya, Y., Komori, T.** 2000. High dose rate (HDR) interstitial brachytherapy for stage I-II tongue cancer. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* **90**: 667-670.
8. **Umeda, M., Komatsubara, H., Ojima, Y., Minamikawa, T., Shibuya, Y., Yokoo, S., Komori, T.** 2005. A comparison of brachytherapy and surgery for the treatment of stage I-II squamous cell carcinoma of the tongue. *Int J Oral Maxillofac Surg* **34**: 739-744.
9. **Umeda, M., Minamikawa, T., Yokoo, S., Komori, T.** 2002. Metastasis of maxillary carcinoma to the parapharyngeal space: rationale and technique for concomitant en bloc parapharyngeal dissection. *J Oral Maxillofac Surg* **60**: 408-413.
10. **Umeda, M., Minamikawa, T., Komatsubara, H., Ojima, Y., Shibuya, Y., Yokoo, S., Komori, T.** 2005. T. En bloc resection of the primary tumour and cervical lymph nodes through the parapharyngeal space in patients with squamous cell carcinoma of the maxilla: a preliminary study. *Br J Oral Maxillofac Surg* **43**: 17-22.