CASE REPORT

The lingual lymph node identified as a sentinel node on CT lymphography in a patient with cN0 squamous cell carcinoma of the tongue

M Saito^{*,1}, H Nishiyama¹, Y Oda², S Shingaki² and T Hayashi¹

¹Division of Oral and Maxillofacial Radiology, Department of Tissue Regeneration and Reconstruction Course for Oral Life Science, Niigata University Graduate School of Medical and Dental Sciences, Niigata, Japan; ²Division of Reconstructive Surgery for Oral Maxillofacial Region, Department of Tissue Regeneration and Reconstruction, Course for Oral Life Science, Niigata University Graduate School of Medical and Dental Sciences, Niigata, Japan

We performed CT lymphography on an 81-year-old female patient with a histologically confirmed squamous cell carcinoma of the tongue with no clinical or radiological evidence of cervical lymph node involvement. The lateral lingual lymph node was identified as a sentinel node, which is the first lymph node to receive drainage from a primary tumour. CT lymphography also showed draining lymphatics passing through the sublingual space, the medial side of the submandibular gland and near the hyoid bone and connected with the middle internal jugular node. Although metastasis to the lateral lingual lymph node is known as one of the crucial events in determining survival outcome in cancer of the tongue and floor of the mouth, very few reports are available on the imaging of the lateral lingual lymph node metastasis. This is the first report regarding the lateral lingual lymph node identified as a sentinel node demonstrated on CT lymphography.

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Introduction

The sentinel node is defined as the first lymph node that receives lymph flow from a primary tumour, and therefore the node that most likely contains metastatic carcinoma.¹ If a sentinel node is negative for metastasis, more distant nodes are also negative. On the basis of the sentinel node concept, sentinel node mapping and sentinel node biopsy have been introduced as a staging method for cN0, where there is no specific clinical or radiological evidence of cervical lymph node involvement in oral squamous cell carcinoma.

Lymph nodes that are located in close contact with a lymphatic channel between the primary tumour and regional lymph nodes are called in-transit, interval or intercalated lymph nodes. Metastasis to an in-transit lymph node is often reported in breast cancer and malignant melanoma. Because of its close positional relationship to the primary tumour, in-transit lymph node metastasis is assumed to be a local recurrence in some cases. There have thus far been no reports debating the significance of metastasis to an in-transit lymph node in cases of tongue cancer. The lingual lymph nodes, first noted in 1938 by Rouviere,² are classified as the in-transit lymph nodes that appear uncommonly in the lymphatic drainage pathways. The lateral lingual lymph node is known as an in-transit lymph node drained from the tongue and the floor of the mouth. The lateral lingual lymph node is located on the outer surface of the genioglossus muscle or on the

*Correspondence to: Dr Mikiko Saito, Niigata University Graduate School of Medical and Dental Sciences, 2-5274, Gakkocho-dori, Chuou-ku, Niigata 951-8514, Japan. Email: micky@dent.niigata-u.ac.jp

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hyoglossal muscle, running along the lingual vein and intervening in the lateral lymph channel feeding into either the superior internal jugular lymph nodes or the anterior submandibular lymph nodes.²

Although metastasis to the lateral lingual lymph node is known as one of the crucial events in determining survival outcome in cancer of the tongue and floor of the mouth, very few reports about the node are available because of its small size and close location to the primary tumour,³⁻⁵ and there have been no reports in which the lateral lingual lymph node is considered as the sentinel node.

Case report

An 81-year-old female was referred to our hospital with a contact pain in the right lateral margin of her tongue. The extraoral examination revealed a single palpable mobile node in her right submandibular region. In the intraoral examination, a white lesion, 22 × 15 mm in size, was observed on the right lateral margin of the tongue. There were no findings of ulceration, induration or haemorrhagic properties. The clinical staging was T2N0. A biopsy revealed a well-differentiated squamous cell carcinoma. There were no distinct signs of lymph node metastases assessed by palpation, CT and sonography. On the initial post-contrast CT, a lingual lymph node was not identified. The primary tumour was not detected on CT because of a metallic artefact. Intraoral sonography showed a hypoechoic mucosal lesion with a thickness of 4 mm (Figure 1).

After approval by our institutional review board and informed consent by the patient, CT lymphography was performed 4 days before surgical treatment of the tongue carcinoma. After induction of local anaesthesia with submucosal injection around the

tumour with 2% lidocaine hydrochloride (without epinephrine) (Xylocaine; AstraZeneca, Osaka, Japan), a pre-contrast CT examination was obtained. 2.0 ml of iopamidol (Iopamiron 370; Bayer Schering Pharma, Osaka, Japan) was then gently administered into the submucosa at four points adjacent to the tumour. CT examination was performed using a single helical CT scanner Xvigor Real (Toshiba Medical Systems Corp., Tokyo, Japan) with a tube voltage of 120 kV, a tube current of 150 mA, a helical pitch of 1.0 and a reconstruction thickness of 1.0 mm at 2 min, 5 min, 10 min, 15 min and 20 min after injecting the contrast agent (Figure 2).

Under general anaesthesia a partial glossectomy was done. Elective neck dissection was not performed. Careful repeated follow-up observation of the regional lymph nodes by palpation, sonograph and CT examinations was carried out. Regional subsequent metastases have not appeared for 14 months (Figure 3).

Discussion

Currently, we perform CT lymphography in cN0 tongue cancer patients when informed consent is obtained from participants followed by careful frequent repeated follow-up observations with physical and radiological examination based on a "wait-and-see" approach.

The sentinel lymph node mapping is commonly performed using the radiocolloid in the radioisotope (RI) method. The sentinel lymph node is identified using a lymphoscintigraphy and gamma probe.

However, if the sentinel nodes are located very close to the primary tumour, RI accumulation to the small nodes might be concealed by the primary tumour because of the "shine-through" phenomenon. CT lymphography, however, allows clear detection of lymph nodes adjacent to the primary tumour and has the advantage of visualization of nodes in an anatomically continuous manner.⁶⁻⁹ Although metallic artefacts sometimes interfere with the view of the level II nodes, CT lymphography is suitable for visualization of small in-transit nodes like the lingual lymph nodes located near the primary tumour.

In cases of T1/T2 and cN0 tongue cancer, there are several options for treating the neck region, including elective neck dissection,¹⁰ follow-up observations based on a "wait-and-see" approach^{11,12} and sentinel lymph node biopsy.¹³⁻¹⁷ Even if a sentinel lymph node biopsy of the lingual lymph node is recommended, the procedure would be highly invasive and could not realistically be considered because of the deep location of the lingual lymph nodes. In the present case, draining lymphatics passing through the sublingual space connected with the middle internal jugular node. If delayed metastasis appeared only in the middle internal jugular node area, the common neck dissection procedure might leave the subclinical metastatic lingual lymph node in the sublingual space. The subsequent lingual

Figure 1 Intraoral sonography: pre-operative examination. A primary lesion involving the submucosal tissue is shown as a hypoechoic area (arrow) with a 4 mm thickness connecting with the surrounding normal mucosal epithelial layer

Figure 2 CT lymphography was taken 10 min after contrast agent injection. (a) Shows axial section of sublingual space; (b) shows the sagittal section where the lateral lingual lymph node passes; (c) shows the coronal section where the lateral lingual lymph node passes. The contrast material was distributed around the primary tumour (P) and ran along the outer side of the right genioglossus muscle and flowed into the lateral lingual lymph node (arrowhead). (d) Shows more caudal level than (c). The lymph flow ran through the sublingual space and the medial side of the submandibular gland (pointed arrowhead). (e) Shows more caudal level than (d). The lymph flow finally drained into the middle internal jugular lymph nodes (pointed arrowhead). (f) and (g) show enhancement pattern of lingual lymph node, lymphatic vessel and tongue

lymph node metastasis might be misdiagnosed as a local recurrence.

There have been recent reports that cases with a recurrent lesion observed near the hyoid bone (parahyoid area) showed poor treatment results and prognosis.¹⁸ The lymph pathway draining the lateral lingual lymph node passes near the hyoid bone, and recurrence in this site may be caused by occult metastasis to the lingual lymph nodes. In other words, our case seems to support the details described by these authors. From the viewpoint of the prediction of the parahyoid recurrence, we can check whether a lymphatic drainage pathway from the tongue does or does not pass through the medial side of the submandibular gland using CT lymphography. Generally, unnecessary neck dissection should be avoided and minimally invasive treatment might be achieved by performing a sentinel lymph node biopsy. However, some cases may require extended surgery such as a “pull-through operation” including the floor of the mouth to determine not only the sentinel “lymph node” but also the

“lymph flow” area. CT lymphography may help to decide an appropriate surgical extent for each individual case.

In summary, we present a case of T2N0 tongue cancer in which the lateral lingual lymph node was demonstrated as an enhancing node close to the primary tongue cancer using CT lymphography. This is the first report in which a lateral lingual lymph node has been identified as the sentinel lymph node. The lymph pathway draining to the lateral lingual lymph node passed through the medial side of the submandibular gland parahyoid area and connected to the middle internal jugular lymph nodes. A common neck dissection procedure might leave the subclinical metastatic lingual lymph node in the sublingual space, which may cause poor prognosis. CT lymphography is a useful technique to visualize the in-transit lateral lingual lymph node for careful repeated follow-up observation in a “wait-and-see” approach without performing a sentinel lymph node biopsy.

Figure 3 The lateral lingual lymph node demonstrated on the post-operative follow-up sonography. A slightly enlarged lateral lingual lymph node was observed in the sublingual space (arrows). However, we could not judge the lymph node metastasis because of no time-course changes in its size, shape and internal structure during a follow-up period. MB indicates the lower margin of the mandible and the mylohyoid muscle (MHM). (a) Coronal section; (b) sagittal section

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