



The Role of a Preoperative CT for the Accurate Diagnosis of a Lingual Osteoma: A Case Report

혀의 골종에 대한 수술 전 전산화단층촬영의 역할:
 증례 보고

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An osteoma of the tongue is a rare benign tumor, typically located in the posterior one-third of the tongue. Several etiological hypotheses have been proposed in previous literature reviews. Herein, we report the case of a lingual osteoma in a 39-year-old woman, who complained of a lump in the throat. The lesion presented as a homogeneously hyperdense submucosal mass without any enhancement in the base of the tongue on CT, consistent with previous image findings of lingual osteoma. After surgical resection, the mass was pathologically proven to be a lingual osteoma. The patient no longer complained of the sensation of a lump. Typical CT findings of a lingual osteoma—a well-circumscribed hyperdense submucosal mass seen around the foramen cecum—could be helpful in the diagnosis of lingual osteoma and the prevention of further unnecessary work-up.

Index terms Neck; Adults; Computed Tomography, X-Ray; Tongue; Osteoma

INTRODUCTION

Osteomas, which are benign neoplasms consisting of mature osseous tissue, are uncommon in the soft tissues of the head and neck (1); however, when osteomas do occur in these body parts, they are usually found in certain regions of the head and neck, especially in the localized regions of the paranasal sinuses, maxilla, and mandible (2). Notably, an osteoma of the tongue (lingual osteoma) is rare, and fewer than 100 cases have been reported to date in previous literature. Typically, a lingual osteoma is located in the posterior one-third of the tongue, adjacent to the foramen cecum. It usually oc-

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curs asymptotically in women between 30 and 50 years of age. If symptomatic, patients commonly complain of swallowing difficulties (3).

Owing to the characteristic image findings on CT, the CT scan could be helpful for the diagnosis of lingual osteoma, despite its rarity. Herein, we report a case of lingual osteoma and its CT findings with a review of the existing literature.

CASE REPORT

This report was approved by the Institutional Review Board of Chung Ang University Hospital, and consent was obtained from the patient herself. Ethical approval was not needed as the case report was a retrospective observation of the patient.

A 39-year-old woman visited our outpatient clinic for the evaluation of a mass at the base of her tongue, which was incidentally found approximately one and a half years ago. She complained of a recently developed lump in her throat. Laboratory studies revealed no abnormalities. Laryngoscopy showed a small, whitish submucosal mass protruding from the dorsum of the tongue base, without tenderness, and a well-preserved overlying mucosa (Fig. 1A).

Contrast-enhanced CT of the neck showed a well-circumscribed, hyperdense, oval mass arising from the base of the tongue without any enhancing soft tissue component (Fig. 1B, C). The size of the mass was $0.7 \times 0.5 \times 0.7$ cm, and the mean density was 1152 Hounsfield units, similar to that of the cortex of the mandible. There was a thin mucosal lining above the mass, which suggested a submucosal location. No significantly enlarged lymph nodes were found in the head and neck. Initially, the differential diagnosis of the lesion included calcified lingual thyroid and dystrophic calcification based on the CT appearance of the mass on the tongue.

During excisional biopsy under general anesthesia, the location of the mass was thought to be at the base of the tongue, off the midline to the left side at the palatine tonsil level. Grossly, there were no remaining masses after the surgical excision. Microscopically, the mass was composed of a dense, mature lamellar bone (Fig. 1D, E), which was covered with the normal squamous epithelium of the tongue, and was suggestive of a lingual osteoma.

DISCUSSION

Lingual osteoma is a rare and benign tumor typically arising from the tongue base and notably, with less than 100 reported cases (4) and only a limited number of image findings (5). Most patients are in the third or fourth decade of life, and there is a four-fold female predilection (male to female ratio of 1:4). Approximately 60% of the patients are symptomatic, and a sense of a "lump in the throat," dysphagia, gagging, and nausea are commonly reported (3). There are no correlations between the symptoms and size of the tumor (6). Surgery is the primary treatment for the symptomatic cases, and no recurrences have been reported (7).

There are a few hypotheses on the etiology of lingual osteoma, mainly derived from the characteristic location of the lesion. One theory is that the pluripotent mesenchymal cells from the first and third branchial arches, which meet in the foramen cecum, can facilitate the development of lingual osteoma (2). Another theory is that lingual osteoma may develop

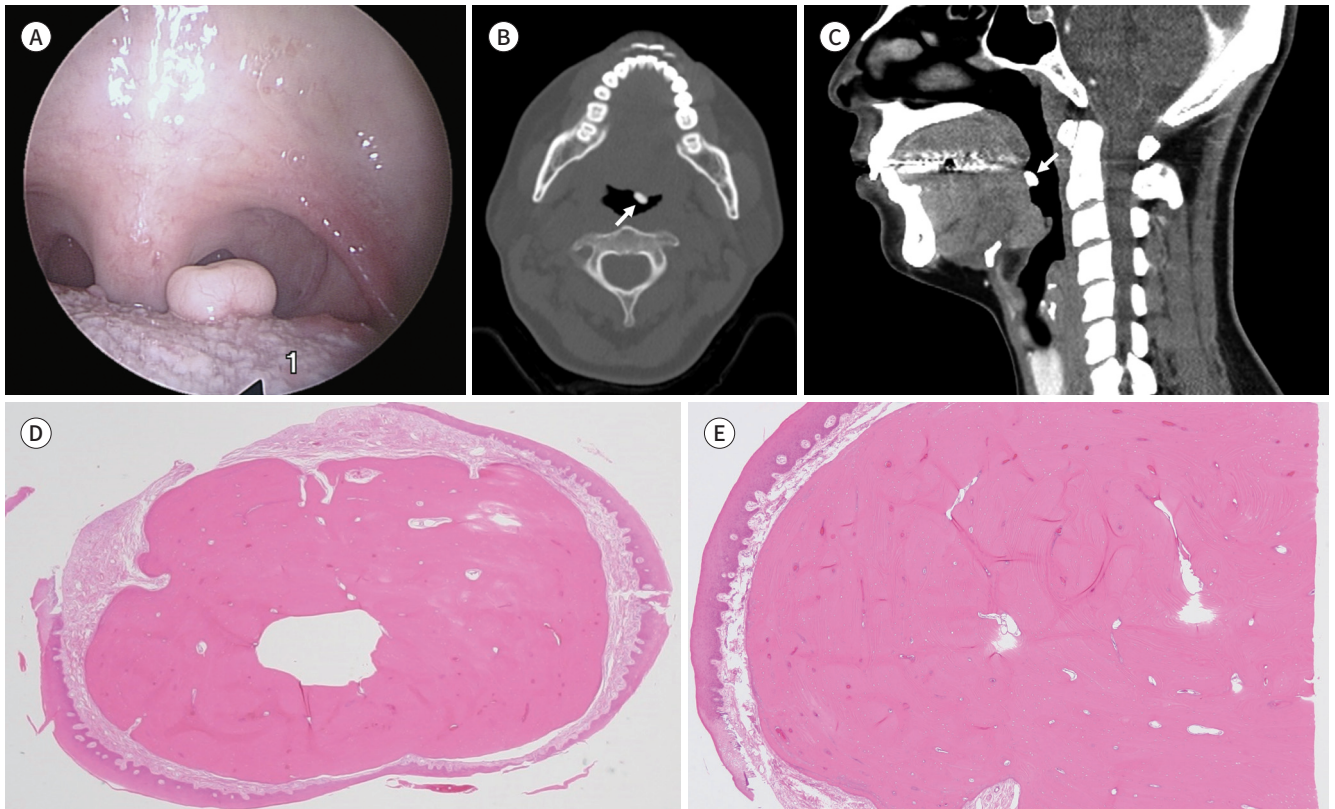
Fig. 1. A 39-year-old woman with lingual osteoma, complaining a recently developed lump in the throat.

A. The laryngoscopy image shows a small, whitish submucosal mass protruding from the dorsum of the tongue base.

B, C. Contrast-enhanced CT axial image with bone window setting (**B**) and sagittal image with soft tissue window setting (**C**) show a well-circumscribed, markedly hyperdense oval mass (arrows) with bone density, arising from the base of the tongue without any definite enhancing soft tissue component.

D. Photomicrograph shows submucosal mass composed of dense mature lamellar bone (hematoxylin and eosin stain, $\times 100$).

E. Photomicrograph shows benign squamous epithelium of the tongue and mature lamellar bone of lingual osteoma (hematoxylin and eosin stain, $\times 300$).



from reactive or post-traumatic changes (2). However, the osseous lesions caused by trauma-induced chronic inflammation usually do not develop into normal bone structures such as the Haversian system or the fatty marrow. Moreover, a lingual osteoma is composed of well-developed mature bone, except in two previously noted cases (3). Lastly, another theory is that the undescended thyroid remnants may cause abnormal bony proliferation of the tongue (1). Not only lingual osteoma, but also lingual thyroid remnants and embryologically displaced intralaryngeal thyroid tissue show a female predilection. Moreover, metaplastic ossification of thyroid tissue is also reported in colloid goiters and thyroid cysts (1). Therefore, lingual osteomas can be considered as the derivatives of thyroid remnants.

On review of the CT findings, the lingual osteoma was noted as a homogeneously hyperdense lesion without enhancement, located at the base of the tongue (5). The margin of the lesion was well-circumscribed, and the attenuation of the lesion was similar to that of the bone. Owing to its typical imaging features, CT could be helpful in differentiating lingual osteoma from other submucosal tumors of the tongue base. Initial differential diagnosis included calcified lingual thyroid and dystrophic calcification. In case of a calcified lingual thy-

roid, thyroid tissue usually shows contrast enhancement on the midline of the tongue base (8). In our case, the lesion was composed of hyperdense bony tissue without any enhancing soft tissue. Dystrophic calcification rarely appears in the injured tissue or at the site of radiation therapy in the head and neck region (9). Our patient had neither a specific medical history nor history of injury. Accordingly, the former two differential diagnoses could be ruled out. Other soft tissue-attenuated tumors near the foramen cecum, such as tonsillar hypertrophy, lingual thyroid, salivary gland tumor, and squamous cell carcinoma, should be ruled out using CT images (1). Furthermore, it can be distinguished from an osteosarcoma with local bone destruction or superficial ulcer, or from osteolipoma with soft tissue density mixed with bone density (3, 5).

The MRI findings of lingual osteoma have also been reported to show a mass with significantly low signal in both T1-weighted and T2-weighted images because of its heavily calcified mature bone structure (1). In contrast, the lingual thyroid, which has an anatomically similar location, can be distinguished by the intermediate signal intensity and the slightly high signal intensity on the T1-weighted and T2-weighted images as normal thyroid, respectively (3).

CT is superior in detecting calcifications in the head and neck area (10). Since lingual osteoma predominantly consists of mature lamellar bony tissue (1), CT is a more suitable imaging modality than MRI for the preoperative imaging of lingual osteoma. Due to the absence of signal from the calcification and cortical bone on MRI, all MRI scans should be preceded by a corresponding CT and radiograph. Therefore, CT would be a better imaging modality than MRI to diagnose a lingual osteoma.

In this case study, we demonstrated the characteristic CT findings of a surgically verified lingual osteoma. It is a well-circumscribed, hyperdense submucosal mass, which appears near the foramen cecum. Although the tumor is rare, knowing the typical CT findings of a lingual osteoma could be helpful for its diagnosis and prevent further unnecessary work-up.

Conflicts of Interest

The authors have no potential conflicts of interest to disclose.

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혀의 골종에 대한 수술 전 전산화단층촬영의 역할: 증례 보고

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혀의 골종은 드문 양성 종양으로, 특징적으로 혀의 뒤 삼분의 일 지점에서 발견된다. 혀의 골종의 병인에 대하여는 기존 문헌고찰로부터 몇 가지 가설이 제시되었으나 아직까지 정확히 밝혀지지는 않은 상태이다. 본 증례 보고에서는 인후두 이물감을 주소로 내원한 39세 여성 환자에서 발견된 혀의 골종에 대하여 보고하고자 한다. 병변은 컴퓨터단층촬영에서 조영 증강 없이 균일한 고음영을 보이는 설근부의 점막하 덩이로 나타났으며, 이는 기존 문헌에서의 설골종의 영상 소견과 합치한다. 외과적 제거 후 병변은 병리학적으로 설골종으로 확진되었고 환자는 더 이상 인후두 이물감을 호소하지 않았다. 설골종은 컴퓨터단층촬영상 특징적으로 막구멍(foramen cecum) 주위에서 발견되는 경계가 뚜렷한 점막하 고음영의 종양으로 나타나며, 이를 앎으로써 설골종의 적절한 진단에 도움을 주어 불필요한 진단적 접근을 줄일 수 있다.

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