

Lingual Osteoma

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Osteomas in head and neck regions are benign, slow-growing bone neoplasms that usually arise in the paranasal sinuses, maxilla, and mandible. An osteoma in tongue is an extremely rare condition, with fewer than 100 cases reported. It usually presents as a mass mainly located near the foramen cecum. Surgical extirpation is the treatment of choice and the prognosis is excellent. Here we present two cases of lingual osteoma that occurred in a 47-year-old woman and 31-year-old man, respectively. The epidemiology, clinical presentation, and pathogenesis are also discussed.

Key words: lingual, tongue, osteoma, extraosseous osteoma

INTRODUCTION

Osteomas are benign, slow-growing tumors being composed of densely sclerotic well-formed bone projecting out from the cortical surface. It has a predilection for the head and neck region including the facial bones, skull, and mandible and is the most common benign tumor of the sinonasal tract¹. An osteoma in the tongue is extremely rare and occurs more commonly in females and in the third and fourth decades of life¹⁻². Most of patients with this disease are asymptomatic but few may present with lump sensation in the throat, evoked gagging, nausea, dysphagia and eating disturbances.

The pathogenesis of osseous lesions of the tongue remains obscure. Two hypotheses, the developmental malformation and the posttraumatic reaction, have been suggested³. The aim of this article is to present two cases of osteoma in the tongue and brief review of literature on lingual osteoma.

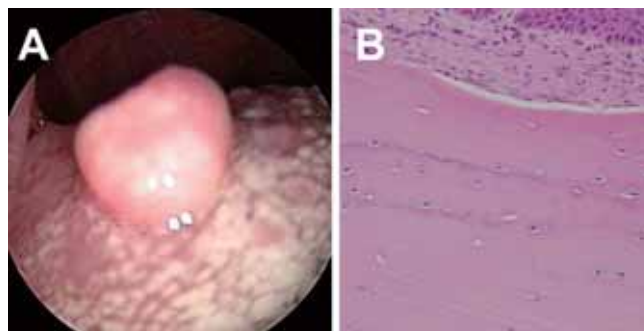


Fig. 1 (A) A lingual osteoma located in the posterior of the tongue and in close proximity to the foramen cecum was demonstrated. (B) Histological findings revealed mature lamellar bone with scattered osteocytes was surrounded by a thin layer of fibrous tissue and squamous epithelium. (hematoxylin-eosin stain, original magnification $\times 100$)

CASE REPORT

CASE 1

A 47-year-old Chinese woman presented foreign body sensation in her throat for three weeks. The physical examination revealed a smooth, firm, light red protruding mass, about $1 \times 0.5 \times 0.4$ cm in size, at the right posterior part of tongue (Fig. 1A). The patient underwent a laser-assisted excision of the mass under local anesthesia with uneventful postoperative course.

Microscopic examination of the lesion showed a picture of mature laminated bony tissue surrounded by fibrous connective tissue located on the submucosal layer of the tongue. The bone was of the compact type, with a normal amount and appearance of osteocytes. (Fig.1B)

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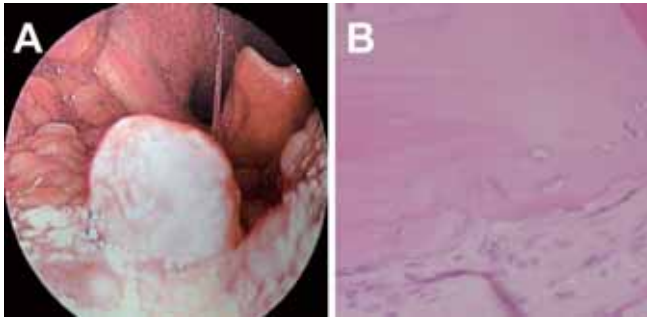


Fig. 2 (A) A protruding mass lesion with normal covering mucosa located in the posterior of the tongue was demonstrated. (B) Histological findings revealed few osteocytes at the periphery of the bone trabeculae was demonstrated. (hematoxylin-eosin stain, original magnification $\times 100$).

CASE 2

A 31-year-old Chinese man came to our institution with mild odynophagia and foreign body sensation in the throat for months. Physical examination revealed an exophytic firm mass located near the midline area of the right tongue base, posterior to the circumvallate papillae (Fig. 2A). The mass, measured $0.7 \times 0.5 \times 0.5$ cm, was mobile and with a narrow base, and covered by normal mucosa. Under general anesthesia, the lesion was excised by laser and the patient's symptoms resolved after surgery.

Microscopic examination of the decalcified tissue showed a well-circumscribed mature compact bone tissue underlying the squamous epithelium. Normal osteocyte and osteoblast activity were noted (Fig. 2B). At 1-year follow-up, there were no signs of recurrence and both patients were symptom-free.

DISCUSSION

Osteomas in head and neck regions are benign, slow-growing bone neoplasms that usually arise in the paranasal sinuses, maxilla, and mandible. An osteoma in tongue is an extremely rare condition with an unclear etiology. According to the literature we've reviewed, only fewer than 100 cases have been reported since 1913⁴. Among those cases, we found that the lingual osteoma was more prevalent in females than in males, and the female-to-male ratio was 3.25 : 1. Age at diagnosis ranged from 5 to 73 years and most common in the third and fourth decades of life. The common location of the lesion was in the posterior third of the dorsum of the tongue³.

In most cases, no symptoms⁵ were associated with the lesion, but occasionally choking, gagging, nausea, and dysphagia were reported.

The differential diagnosis of a protruding mass lesion isolated in tongue includes fibroma, papilloma, salivary gland mixed tumor or osteochondroma and other neoplasm such as osteogenic sarcoma or squamous cell carcinoma⁶. Histologically, the lesions were well-circumscribed and usually composed of compact bone with Haversian systems. The bone was surrounded by a thin capsule of fibrous connective tissue and covered with normal stratified squamous epithelium. In some cases, increased cellular activity of osteoblasts was reported⁵.

The etiology of osseous lesions of the tongue is unknown. Two hypotheses, the developmental malformation³⁻⁵ and posttraumatic reaction, have been suggested. According to the developmental or embryologic hypothesis, osteomas that arise at the foramen cecum, the junction of the first and the third branchial arch, predispose to the development of cell rests and later tumor formation. It is known that certain normal osseous structures are derived from these branchial arches through endochondral ossification. For instances, the incus and malleus is derived from the first branchial arch and most of the hyoid bone from the third arch. It is possible that some multipotential cells congenitally entrapped in the tongue during its development and subsequent development of an osseous lesion in the tongue. A maldevelopment associated with remnants of thyroid tissue has been proposed⁷⁻¹⁰. The foramen cecum is the site where the anlage of the thyroid gland develops in embryologic life, and from this site the glandular tissue descends to the neck to take its normal place. They suggested that remnants of undescended intraglossal thyroid tissue can lead to unusual osseous proliferating lesions later in life, mainly during puberty and adolescence.

The other hypothesis suggests that osseous lesions of the tongue represent a reactive or posttraumatic center of ossification. These types of lesions have been reported in other muscles of the body under the term "myositis ossificans". Several investigators indicate that the posterior third region of the tongue is susceptible to trauma and irritation. The posterior third of the tongue takes an active role in swallowing. Chronic inflammatory cells in the epithelium and the connective tissue of the foramen cecum region are a common finding, suggesting that the tissue in this region is in a constant state of irritations. Thus, it is suggested that soft tissue chondromas, mixed osseous and cartilaginous lesions, and osseous lesions, with progressive maturation of the endochondral ossifi-

cation, may represent continuum of responses to trauma. Soft tissue osteomas may represent the most mature stage of a posttraumatic metaplasia. However, some investigators disagree with this hypothesis. The osseous lesions of the tongue are usually composed of well-developed mature bone, while inflammatory and traumatic lesions with bone formation often appear microscopically as irregular areas of ossification, with neither Haversian systems nor normal bone architecture.

Surgical excision is generally regarded as the mainstay of treatment for symptomatic lingual osteomas. In our two patients, we used KTP laser to excise the lingual osteoma. Care should be taken to excise the tumor with limited margin in order to preserve the tongue function.

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