

Lingual osteoma : report of three cases

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Lingual osteoma is a rare benign lesion of the tongue. Its occurrence is around the foramen cecum and predominates in female patients in the third decade. Lingual thyroid tissue is proposed to be the cause of this tumor, however other hypotheses are discussed. The authors report three cases of lingual osteomas found during 5 years period (Jan 1,1982-Dec 31,1986) at Chulalongkorn Hospital. The tumors were diagnosed and treated by excisional biopsies.

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Osteoma ของลิ้นเป็นเนื้องอกชนิดไม่ร้ายแรงที่พบได้น้อยมากชนิดหนึ่ง และมักจะเกิดขึ้นที่บริเวณรอบ ๆ *foramen cecum* ของโคนลิ้น เนื้องอกชนิดนี้พบมากในผู้หญิงอายุระหว่าง 20-30 ปี พยาธิกำเนิดเชื่อว่าเกิดจากการมีเนื้อเยื่อของ *thyroid* หลงเหลือบริเวณโคนลิ้น แต่ยังมีสมมุติฐานอื่น ๆ อธิบายไว้อีกหลายอย่าง คณะผู้รายงานนำเสนอ เนื้องอก *osteoma* ของลิ้นในผู้ป่วยหญิงจำนวน 3 รายที่มารับการรักษาที่โรงพยาบาลจุฬาลงกรณ์ในช่วง 5 ปี (1 มกราคม 2525-31 ธันวาคม 2529) โดยการผ่าตัดเอาเนื้องอกออกจนหมด

Osteogenous tumors of the head and neck are not as common as in the long bone. They are often found as osteomas in the paranasal sinuses, facial bone, skull and mandible⁽¹⁾. The osteomas of the soft tissue in the oral cavity are extremely rare and they are found originating from either the tongue or the oral mucosa, however the tongue is the predominant site⁽²⁾.

On reviewing the English literatures, nearly 40 cases of the osteoma of the tongue are reported⁽³⁻⁵⁾. The authors found three pathologically proven cases of osteoma of the tongue at Chulalongkorn Hospital in the 5 years period and we think these will be of additional interest.

Case reports

Case 1

A 28 year-old Thai woman presented to the E.N.T. department with complaint of irritation in the throat for four years. During a four years period

she went to see a general practitioner near her house. The symptom was off and on but never did cause severe troubles to her. She denied familial disorders, occupational or chronic diseases. On examination, a round, stony-hard mass of one-cm. in diameter was found on the dorsum of the tongue just to the left of the midline at the junction of anterior two-thirds and posterior one-third of the tongue (Fig 1). The provisional diagnosis of osteoma of the tongue was made and the patient was set up for excisional biopsy under local anesthesia without any pre-operative investigations. Microscopic examination demonstrated a well-circumscribed lesion covered with nonkeratinizing stratified squamous epithelium. The lesion composed of a mass of viable compact bone with osteocytes and lacunas (Fig 2). The pathological diagnosis of osteoma of the tongue was finally made. There was no recurrence after 3 years follow-ups.

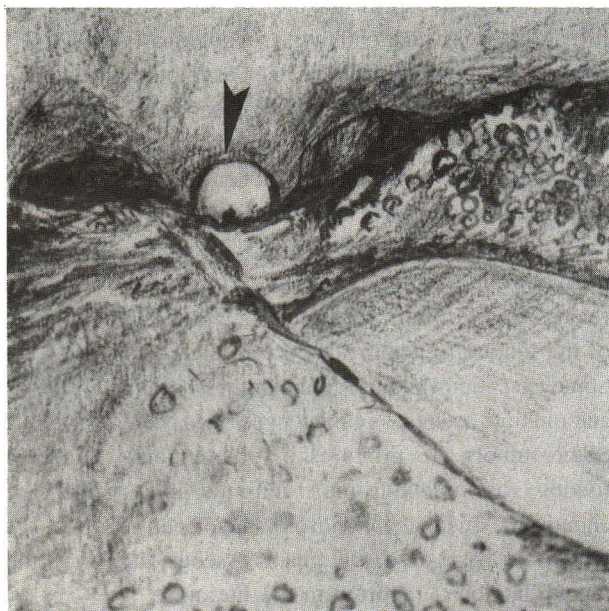


Figure 1. A round stony-hard mass of one-cm. in diameter with smooth and glistening surface is at the base of tongue.

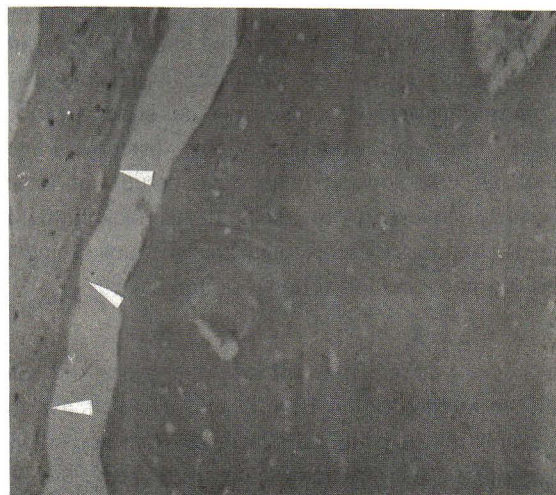


Figure 2. (low power) A mass of viable compact bone covered with non-keratinizing stratified squamous epithelium (arrow heads), (the separation between the mass and epithelium was due to preparation)

Case 2

The patient, a 25-year-old woman presented to the E.N.T. out-patient department for evaluation of a tongue mass found on a routine physical examination by a private otolaryngologist one month earlier. She was suffering from allergic rhinitis for many years and was infrequently being treated by otolaryngologists

in various clinics. One year prior to the examination she was also aware of a lump in her throat. She had negative history to either tuberculosis or any serious diseases. Physical examination revealed a 0.5 cm firm pedunculated mucosal-covered mass originating from the left of foramen cecum at the base of the tongue. The rest of the examinations were normal. The mass

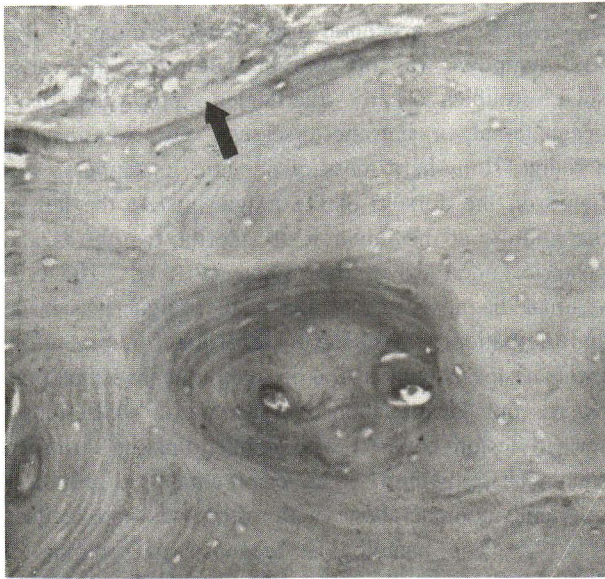


Figure 3. (high power) Dense lamellar bone with well defined. Haversian system and viable osteocyte are noted. A thin zone of connective tissue is located between the mass and stratified squamous epithelium (arrow)

was then excised by blunt dissection under local anesthesia. Bleeding was not significant. Histologically, the mass consisted of dense lamellar bone with well-defined haversian system. Viable osteocytes were noted. A thin zone of connective tissue was located superiorly, covered by a stratified squamous epithelium (Fig 3); extraosseous osteoma was diagnosed and the mass did not recur on follow-ups for 2 years.

Case 3

A young Thai girl of nine years old came with her mother to the E.N.T. out-patient department for evaluation of pain and hearing loss in her left ear. Acute rhinitis with bilateral acute otitis media has been diagnosed and was treated with an appropriate antibiotic, an analgesic and a decongestant nasal spray.

During physical examinations, a mass of 0.5 cm in diameter, pink color and smooth surface was co-incidentally found originating from the right side of the foramen cecum at the base of the tongue. It was bony hard on palpation and the provisional diagnosis of lingual osteoma was made. The mass excised with blunt dissection and electrocoagulation under local anaesthesia as soon as the patient had recovered from the acute nasal and aural symptoms. Histological sections of the tumor showed regular non-keratinized stratified squamous epithelium overlying a thin fibrous tissue layer. The mass was a well developed compact lamellar bone with well-defined Haversian systems.

On follow-up, the mass did not recur 2 years post-operatively.

Discussion

Lingual osteoma is a benign lesion histologically and characteristically. Beside the osteoma, chondroma, osteochondroma, fibroma, cyst, salivary gland tumor, ectopic thyroid tissue and granular cell myoblastoma may be included in the differential diagnosis⁽²⁾. About 70 cases of osteoma, osteochondroma and chondroma were reported in the world literature, but only 40 cases were osteomas⁽³⁻⁶⁾.

Osteoma of the tongue usually presents in female (about 75% of cases) mostly in the third decade (average age = 28 years old) and there is no apparent race predilection^(3,4,6,7). These data correlate with our cases that all of them are women and two of them present in the third decade. Approximately 40% of the patients have complained of either dysphagia or a sensation of fullness or foreign body-like sensation in the posterior third of the tongue. Chronic sore throat, nausea, and choking are noted as complaints. However, most patients have been asymptomatic and the lesion were found during routine ear nose and throat examinations⁽³⁻⁵⁾. This statement is true in our two patients that the osteomas have been found accidentally during otolaryngologic examinations.

The majority of glossal osteoma are about 0.5-2 cm in diameter and are located at or closely related to the foramen cecum or circumvallate papillae of the tongue in pedunculated or sessile fashions^(3,4,8). Some exceptions in those found in the buccal mucosa^(9,10) and multiple osteomas of the tongue have been reported⁽³⁾. Some tumors are mixtures of osseous and chondroid tissues (osteochondroma) and multiple lesions of chondroma and osteochondroma have been reported⁽¹¹⁾. Only one case of a malignant osseous tumor of the tongue has been reported in the literature⁽¹²⁾.

The pathogenesis of an extraosseous osteoma in the tongue is still debated. At least three theories have been proposed to explain their occurrence⁽³⁾. The first one is that osteomas of the tongue result from metaplastic changes that occur in mesodermal cells in the tongue, often related to chronic local irritation from trauma, irradiation and infection⁽²⁻⁴⁾. A second theory is that of the occurrence of ectopic bone formation and the term "osseous choristoma" is used to describe the lesion. Pathologically, choristoma are growths of mature normal cell that are found at sites in the body remote from their normal location^(2,10). The last theory is the congenital cause which includes many explanations of the relationship between the location of osteoma and the foramen cecum. An ossified

branchial arch remnants especially the Meckel's cartilage. It has been hypothesized that the osteoma of the tongue is the unfused, smaller "twin" that joins each other in the oral cavity and that is the "epignathus" formation theory⁽¹⁰⁾.

Of all these theories, not a single one gives us a clear answer. However, the most likely explanation is the ossified lingual thyroid tissue, because most of the cases of lingual osteoma are females in the third decade of life which can be compared with the predominant diseases that are found in the woman at the same age, however, it cannot yet be proved⁽³⁾.

The tumors in our three cases also presented near the foramen cecum. We think the number of tumors are too small to study the exact pathogenesis. Postmortem study has been designed to discover either

the bone or cartilage forming cells in the human tongue but failed to demonstrate even in a single case⁽²⁾.

Summary

Osteomas are benign bony neoplasms commonly found in the long bones and in the skull and facial bones. Osseous growth within soft tissue structures of the head and neck are rare. To date, nearly 40 cases of osteomas of the tongue were reported. In addition, osteochondroma and chondroma, the other two growths that relate to osteoma were also found.

The authors report on three patients with a bony tumor at the base of the tongue, two of whom were asymptomatic and the masses found on routine otolaryngologic examination; the other one complained of irritating sensation in the throat. All cases were diagnosed and treated by excisional biopsies with no recurrence on the follow ups.

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