

# รอยโรคกระดูกโคริสโตมาของลิ้น: รายงานผู้ป่วย 1 ราย และทบทวนวรรณกรรม

## Osseous Choristoma of the Tongue: Report of a Case and Review of Literature

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### บทคัดย่อ

รอยโรคกระดูกโคริสโตมา (osseous choristoma) เป็นรอยโรคในช่องปากที่พบได้ไม่บ่อยนัก เป็นรอยโรคที่ไม่ร้ายแรงและส่วนมากจะพบได้ที่บริเวณลิ้น ลักษณะที่พบจะมีลักษณะที่เป็นก้อนแข็งคล้ายมะเร็งที่มีโครงสร้างของกระดูกปกติในตำแหน่งที่ผิดปกติ เรายังรายงานผู้ป่วย 1 รายที่พบรอยโรคกระดูกโคริสโตมาบริเวณลิ้นในผู้ป่วยชายชาวญี่ปุ่น อายุ 27 ปี รอยโรคมีลักษณะเป็นก้อนแข็งสีชมพู, ผิวเรียบ, มีก้าน และ ตำแหน่งอยู่ทางด้านซ้ายเล็กน้อยของเส้นกึ่งกลาง, หลังต่อเซอร์คัมวัลเลท พาพิลเล (circumvallate papillae) และใกล้ต่อฟอราเมน ซีกัม (foramen caecum) รอยโรคถูกตัดออกภายใต้การดมยาสลบโดยปราศจากการเป็นโรคเดิมอีกเฉพาะที่.

**คำสำคัญ:** กระดูกโคริสโตมา ช่องปาก ลิ้น

### Abstract

Osseous choristoma is a rare, benign lesion of the oral cavity and usually found in the tongue. The presents as a tumor-like mass of normal bony structure with mature cell in an abnormal position. We reported one case of osseous choristoma of the tongue in 27 year-old Japanese man. The lesion was pink, hard, smooth, pedunculated and located slightly to the left of midline, posterior to circumvallate papillae and close to foramen caecum. The lesion was excised under general anesthesia without local recurrence.

**Keywords:** osseous choristoma, oral cavity, tongue

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## Introduction

Choristoma is a tumorlike growth of microscopically normal tissue in an abnormal location. Osseous choristoma of the oral cavity is an extremely rare lesion. The term “ osseous choristoma ” was introduced by Krolls et al. to describe a tumorlike growth of normal, mature, and lamellated bone occurring in the soft tissue of the oral cavity.

The lesion most frequently develops in the tongue area and less commonly in other sites such as buccal mucosa, soft palate , gingiva, tonsil, and submandibular region.

Osseous choristoma of the tongue occurs predominantly on the dorsal surface of the posterior third in the area of circumvallate papillae or close to the foramen caecum<sup>(1)</sup>. The pathogenesis of the lesion remains obscure.

In this article, we report an additional case of osseous choristoma of the tongue, with a brief review of the literature, pathogenesis and differential diagnosis.

## Report of a case

A 27-year-old Japanese man was referred to the Department of Oral and Maxillofacial Surgery, Tokyo Medical and Dental University for evaluation and management of the retained root of the left mandibular third molar. In panoramic radiograph, we found a radiopaque mass at base of the tongue as well as the retained root in the floor of the mouth (Fig. 3).

Computed tomography showed the retained root and a radiopaque mass at base of the tongue (coronal section) (Fig. 4).

Physical evaluation revealed no apparent abnormalities.

Intra oral examination showed a pink, pedunculated and hard mass which was covered with normal mucosa (Fig. 1).

It was approximately 8 mm. in diameter and located slightly to the left of midline and posterior to the circumvallate papillae (Fig. 2).

The lesion was asymptomatic, and he had no history of trauma around the region.

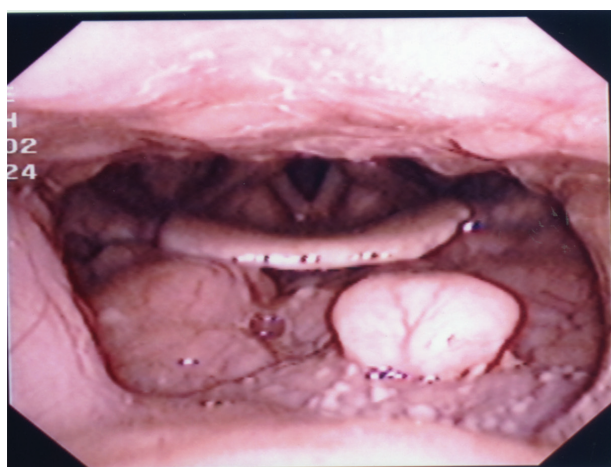
The clinical diagnosis was osseous or cartilaginous choristoma.

The lesion was excised perorally, and the retained root was removed under general anesthesia.



ภาพที่ 1 แสดงลักษณะทางคลินิก พบลักษณะรอยโรคก้อนแข็งสีชมพู มีก้าน บริเวณด้านท้ายของลิ้น

*Figure 1 Clinical appearance of pink, pedunculated and hard mass of posterior of tongue.*



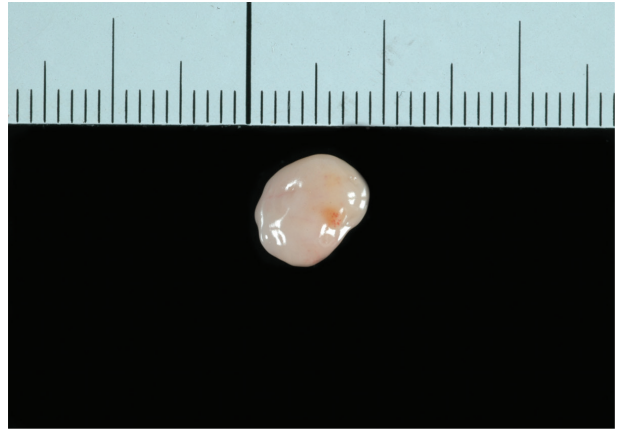
ภาพที่ 2 ภาพจากกล้องไฟเบอร์ออปติกของรอยโรคก้อนแข็ง

*Figure 2 Fiberscopic view of the mass.*



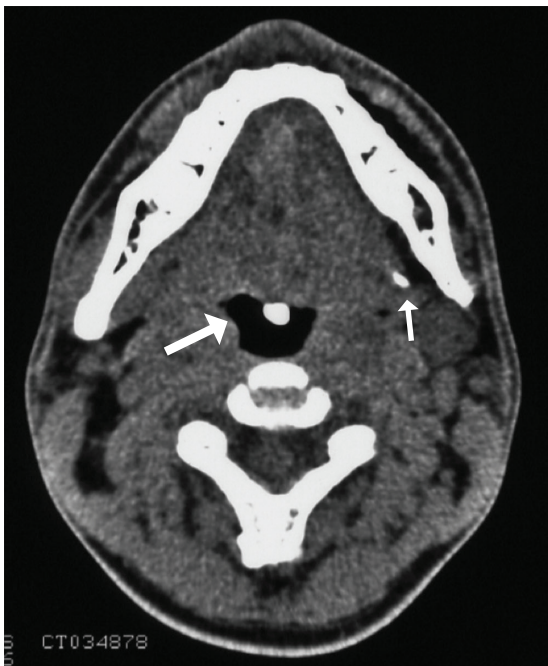
ภาพที่ 3 ภาพรังสีปริทัศน์ แสดงก้อนที่บรังสีบริเวณด้านท้ายของลิ้น (ลูกศรชี้)

**Figure 3** Panoramic radiograph showing radiopaque mass located at posterior of tongue (arrow).



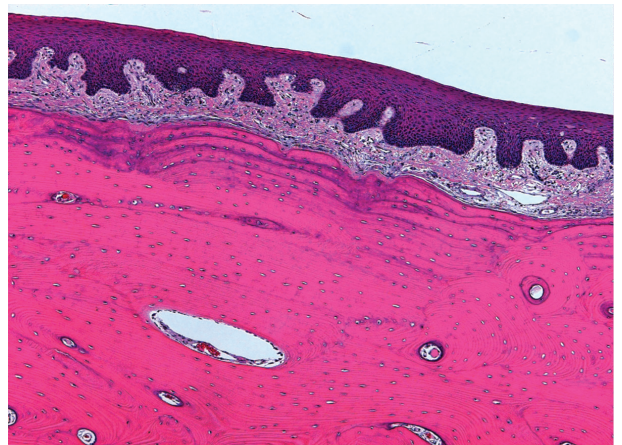
ภาพที่ 5 ภาพถ่ายแสดงรอยโรคก้อนแข็งของลิ้นที่ถูกตัดออกมา

**Figure 5** The surgical specimen of the tongue.



ภาพที่ 4 ภาพรังสีซีที แสดงก้อนที่บรังสีบริเวณด้านท้ายของลิ้น (ลูกศรใหญ่ชี้) และรากฟันที่หลงเหลืออยู่บริเวณพื้นของช่องปาก (ลูกศรเล็กชี้)

**Figure 4** Computerized tomograph imaging showing radiopaque mass at posterior of the tongue (big arrow) and retained root at the floor of the mouth (small arrow).



ภาพที่ 6 ภาพถ่ายจากกล้องจุลทรรศน์ของกระดูกโค-ริสโตมาของลิ้นแสดงกระดูกชนิดเดนส์ลามลลา (dense lamellar bone) ใต้เยื่อบุผิว สเตรทไฟยัด สความัส (stratified squamous epithelium) (ย้อมด้วยสี ฮีมาทอกซีลินและอีโอซิน (Hematoxylin and eosin) กำลังขยาย 40 เท่า)

**Figure 6** Photomicrograph of osseous choristoma of the tongue showing dense lamellar bone underlying surface stratified squamous epithelium. (Hematoxylin and eosin stain; original magnification  $\times 40$ .)

The surgical specimen of the tongue lesion measured 9×8×6 mm. in size (Fig. 5).

Microscopic examination of the specimen revealed a well circumscribed nodule of mature and dense lamellar bone with Haversian canals surrounded by thin fibrous connective tissues with the stratified squamous epithelium (Fig. 6).

The final histopathologic diagnosis was osseous choristoma of the tongue. The patient had periodic follow-ups and has remained free of disease for 6 months.

## Discussion

Majority of osseous choristoma were asymptomatic, though symptoms of pain, dysphagia, foreign body sensation, disturbance in swallowing, throat irritation, gagging, vomiting, and nausea have been reported in some patients<sup>(4,7,8)</sup>. In most cases, the tumor was located at the posterior third of the tongue in the area of foramen caecum and circumvallate papillae.

The cause of osseous choristoma is obscure, but several histogenic hypotheses have been proposed<sup>(3,9,10)</sup>

(1) Congenital abnormality

(2) Epignathous formation

(3) Ossification of branchial arch remnant, thyroid gland remnant, lymphatic tissue, or degeneration fibroma

(4) Developmental malformation from multi potential cell enclaved during the development of the tongue

(5) Metaplastic formation based on the transformation of mesenchymal cell under traumatic or chronic inflammatory stimulation

Several investigators indicate that the posterior third of the tongue is susceptible to trauma and irritation<sup>(6,8)</sup>. Chronic inflammatory cells in the epithelium and connective tissue of the foramen caecum region are commonly seen, suggesting that the tissue in

this region is in a constant state of irritation . The posterior area of the tongue also takes an active role in swallowing , especially in the late stage of deglutition.

In our case, the metaplastic theory seems unlikely because we could not find any irritant agents nor traumatic history.

We are in substantial agreement about the developmental malformation theory, because some osseous choristoma were found at birth, infant or childhood.

Monserrat firstly suggested the developmental malformation theory, basing his theory on anatomical location of the lesion in foramen caecum area. During the embryologic development of the tongue, the union between its anterior two thirds and its posterior third takes place in the region of the foramen caecum and sulcus terminalis. The anterior two thirds of the tongue originates from the first branchial arch , and the posterior third originate from the third branchial arch. It is known that certain normal osseous structures are derived from these branchial arches through enchondral ossification the incus and malleus from the first arch and most of the hyoid bone from the third arch. Therefore, the possibility of enclavement of mesenchymal pluripotential cells originating from these embryonic branchial arches, and subsequent development of an osseous lesion in the tongue, is possible.

This theory is strongly supported by Begal et al., Engel and Cherricle, and Peimer et al.

The clinical differential diagnosis of osseous choristoma depends on the location.

In the tongue, it should be distinguished from a thyroid nodule, hyperplastic tonsil, salivary gland neoplasm, fibroma, granular cell tumor, neural neoplasm, and phlebolith. In the gingiva, the differential diagnosis are peripheral odontogenic fibroma, and giant cell granuloma. In the buccal mucosa the differential diagnosis are salivary calculi, salivary gland

neoplasm, fibroma and calcified lymph node. In the submental region, the additional possible differential diagnosis are giant cell granuloma, thyroglossal duct cyst, calcified lymph node, and myositis ossificans<sup>(2,12)</sup>.

The osseous choristoma shows a benign behavior, and usually does not recurrence.

Simple surgical excision of the tumor and follow-up would be satisfactory in the majority of cases.

However, Long and Koutnick registered a case of recurrence 12 years after surgical excision<sup>(11)</sup>. The lesion was located in the buccal soft tissue connected to the mandibular ramus. Dalkiz et al. reported a case of osseous choristoma in the masseter muscle which recurred 1 year after excision<sup>(5)</sup>

## Literature Review

The literature review by Supiyaphun P et al. revealed 58 cases of osseous choristoma of the tongue, including his own cases. Our search of the English language literature (1913-2006) revealed an additional nine cases. As of March 2006, there were published report of 67 cases of lingual osseous choristoma in addition to our one case. Of these 68 patients, 53 (77.9%) were females and 15 (22.1%) were males, with ages ranging from five to 73 years (most in their third and fourth decade of life).

The osseous choristoma were located in posterior third of the tongue in 55 cases (80.9%), in the lateral border of the tongue in 8 cases (11.8%), in the middle third of the tongue in 3 cases (4.4%), and at the ventral surface of the tongue in 1 case (2.9%).

The lesions in the posterior third of the tongue were at or in close proximity to the foramen caecum and circumvallate papillae. Lesion size, reported in 50 cases, ranged from 0.5 cm. to 2.5 cm. at the greatest dimension. The duration of the lesion was unknown in half of the patients.

Among the 68 documented cases, symptoms include a growth or swelling of tongue or a lump in the back of the tongue (11.8%), dysphagia (7.4%), gagging (7.4%), throat irritation (2.9%), choking (1.4%). Twenty patients (29.4%) were asymptomatic and symptoms were not specified in twenty-seven (39.7%) patients.

## Conclusion

The authors reported one case of osseous choristoma of the tongue in 27 year-old Japanese man. The lesion was pink, hard, smooth, pedunculated and located slightly to the left of midline, posterior to the circumvallate papillae and close to the foramen caecum. The lesion was excised under general anesthesia. The osseous choristoma shows a benign behavior, and no recurrence has been reported, although 2 recurrent cases after surgery were reported in the English literature. Therefore extended clinical and radiographic follow-up is necessary after surgical excision of an osseous choristoma.

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