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OSSEOUS CHORISTOMA OF ORAL CAVITY — REPORT OF TWO CASES AND REVIEW OF THE LITERATURE

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Osseous choristoma of the oral cavity is an extremely rare lesion. Two cases of osseous choristomas of oral cavity, representing two different natures of such lesions are reported. One was situated on the dorsal surface of posterior tongue just anterior to the circumvallate papillae and the other on the right buccal mucosa just below the orifice of the Stensen's duct. To date, about 73 cases of oral osseous choristoma have been reported in the literature including the two present cases, of which, 61 and 8 cases occurred in the tongue and buccal mucosa, respectively. The clinical and microscopic characteristics, and their common sites of these peculiar lesions are presented. The origin and pathogenesis of the lesion are discussed and the literature on the subject is reviewed.

Key words: osseous choristoma, soft-tissue osteoma, lingual osteoma

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Osseous choristoma is a rare tumor composed of normal lamellar bone occurring at a site where bone is not normally found. The majority of this lesion occurred intraorally and were located on the dorsal surface of posterior tongue[1,2]. The most common age group is the 3rd decade. Most of lesions present as an asypmptomatic sessile or pedunculated hard mass but occasionally, dysphagia, gagging, choking, and foreign body sensation are noted. It appears that the lingual osseous choristoma has a predilection for females and occurs almost exclusively on the posterior tongue surface near foramen cecum.

Microscopically, it is a well-circumscribed mass consisting of compact bone surrounded by a thin fibrous connective tissue beneath the oral stratified squamous epithelium. The etiology of osseous choristoma of oral cavity is still unknown and the pathogenesis is debatable[3-6]. It is thought to be a developmental malformation, while the lesions other than posterior tongue is thought to be of traumatic origin. A case of lingual osseous choristoma in a young female and a recurrent case of buccal osseous choristoma in a male are presented and the etiology and pathogenesis are discussed with a review of the literature.

CASE 1

A 21-year-old Chinese woman came to the Department of Oral Diagnosis and Oral Pathology of the Kaohsiung Medical College Hospital with the chief complaint of a mass on the dorsum of the posterior tongue. She had been aware of the lesion for 5 years, but there was neither difficulty with swallowing nor pain. She denied any traumatic injury in that area. On physical examination, the mass was well-circumscribed, and located on the midline of the dorsum of tongue just anterior to the circumvallate papillae. The mass was ovoid, pedunculated and measured 1.2 cm in the greatest diameter (Fig. 1). It was

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hard and covered by a smooth, intact mucosa. The thyroid gland and lymph nodes were within normal limits. Other medical history and physical examination were essentially non-contributory. The clinical diagnosis was an irritation fibroma of the tongue. The patient was referred to the Department of Oral and Maxillofacial Surgery for an excisional biopsy. Under local anesthesia the mass was removed with electrosurgery. The specimen measuring 1.2 × 0.8 × 0.5 cm in size was fixed in formalin. Grossly, it was extremely hard to palpation and was covered by an intact whitish and glistening oral mucosa.

After decalcification, the microscopic examination revealed compact, mature bone with osteocytes in the lacunae. The covering epithelium was a parakeratinized stratified squamous epithelium (Fig. 2). Beneath the epithelium there was a thin dense fibrous connective tissue surrounding the osseous tissue, which appeared to be dense lamellated bone with a good number of Haversian canals (Fig. 3). Osteoblastic activity was prominent in focal areas. A diagnosis of an osseous choristoma of the tongue (so-called lingual osteoma) was made. Post-operative healing was uneventful. After four years, the patient was recalled for check-up and there was no sign of recurrence.

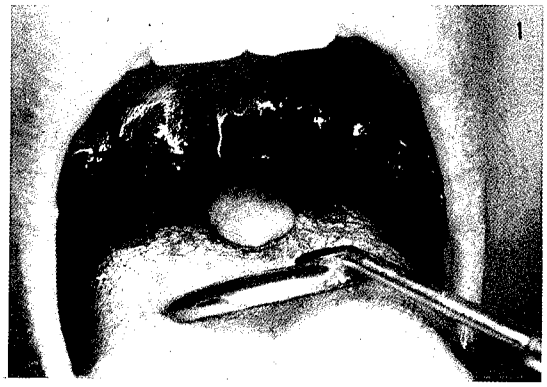

Fig. 1. Case 1. A pedunculated mass on the dorsal tongue just anterior to the circumvallate papillae of a 21-year-old Chinese female

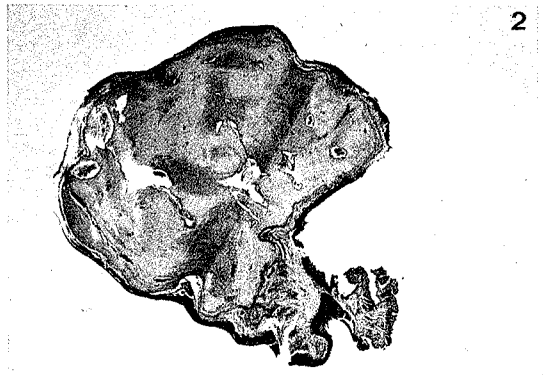

Fig. 2. Case 1. Microphotograph exhibiting a well-circumscribed mass of mature bone covered by stratified squamous epithelium. (H & E stain; original magnification X 2.5)

Fig. 3. Case 1. Microphotograph showing that the mass is composed of dense lamellated bone with osteocytes in lacunae and well-developed Haversian canals. (H & E stain; original magnification X 20)

CASE 2

A 45-year-old Chinese male complained of a painless swelling mass on the right buccal mucosa. He had been aware of this hard mass for about one year. He felt neither discomfort nor swallowing difficulty. He had had surgery twice in the same location but there was no special finding in pathologic examination. The local dentist referred him to our dental clinic for further diagnosis and treatment. He denied any traumatic history in this area. On oral physical examination, a hard movable mass was found on the right buccal mucosa near the orifice of the Stensen's duct measuring 2.3 × 2.0 cm in diameter with normal intact oral mucosa. (Fig. 4) Radiographically, it showed radiopaque shadow from this area by occlusal film. Medical history and physical examination were non-contributory except for reveal-

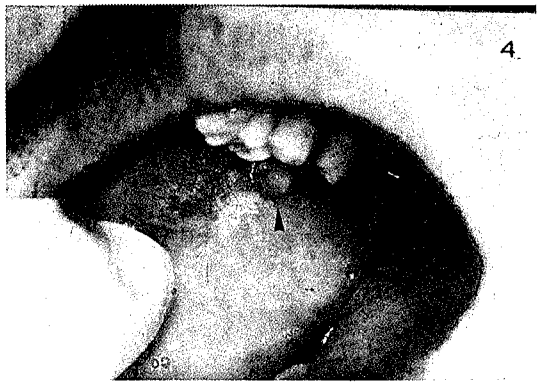

Fig. 4. Case 2. Dome-shaped swelling below the parotid papilla of the right buccal mucosa of a 45-year-old Chinese male (arrowhead)

ing hypertension. A clinical diagnosis of a sialolith in the right Stensen's duct was made. The patient was referred to the Department of Oral and Maxillofacial Surgery for an excisional biopsy.

The specimen was fixed in formalin and measured 2.0 × 2.0 × 1.5 cm in size. On cut surface, there were some whitish nodules with fibrous capsules. After having been decalcified, it was stained with routine H& E. Microscopically, it was composed of round cancellous bone tissue groups empassed by obvious fibrous tissue (Fig. 5). These cancellous bones consisted of bone trabeculae containing osteocytes in the lacunae. Marrow spaces were filled with fibrous connective tissue. The covering epithelium was non-keratinized stratified squamous epithelium. A small amount of metaplastic bone was noted close-by (Fig. 6). The pathologic diagnosis was an osseous choristoma of buccal mucosa. Post-operative healing was good and no recurrence was seen after a follow-up of two years and three months.

DISCUSSION

Since Monserrat[7] reported the first case of osteoma in the tongue, numerous cases of such lesions have been reported in literature. The terms "soft-tissue osteoma" and "extra-osseous osteoma" were used by most authors to describe this lesion[8]. In 1971, Krolls *et al.*[9] published 9 cases of such lesions of oral cavity and coined a new name "osseous choristoma". "Cutaneous ossification", a dermal lesion suggested by Krolls *et al.* is similar to, if not the same, the intra-oral soft-tissue

Fig. 5. Case 2. Microphotograph showing two nodules of cancellous bone beneath the oral stratified squamous epithelium. (H & E stain; original magnification × 2.5)

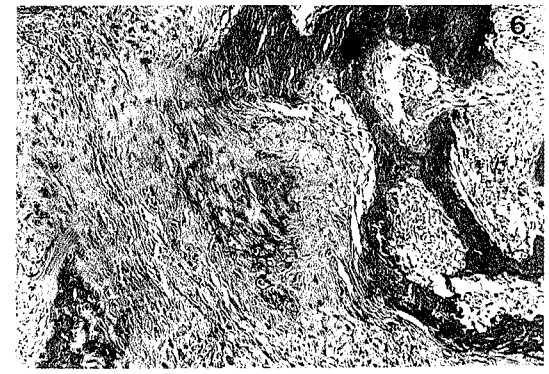

Fig. 6. Case 2. High power view of Fig. 5 showing well-formed bone trabeculae contained osteocytes and areas of metaplastic bone formation. (H & E stain; original magnification × 100)

osteoma. Schweitzer *et al.*[10] reported two cases of extra-oral soft-tissue osteoma in thigh. It is interesting that the intra-oral lesion occurs predominantly in the posterior dorsal surface of the

Table 1. Buccal osseous choristomas reported in the literature

Author	Year	Sex	Age	Size(cm)	Location
Krolls *et al.*	1971	M	40	-	-
Herd	1976	F	75	1.7*1.0	Right
Mesa *et al.*	1982	M	33	1.8*1.7*1.0	Left
Sookasam &	1986	F	43	almond	Right
Philipsen	1986	F	41	hazelnut	Right
Hodder & McDonald	1988	F	12	1.0*0.5	Right
Long & Koutnic	1991	F	50	2.0*1.1*0.6	Right
Lin *et al.*	1998	M	45	2.3*2.0*1.5	Right

tongue, adjacent to the foramen cecum and circumvallate papillae. To date, about 73 cases of osseous choristomas occurring in the oral cavity have been reported including the two present cases. Among the 73 cases, 61 were in the tongue [1-3, 5-9, 11-37], 8 in the buccal mucosa (Table 1)[4, 9, 38-41], one each in the lingual alveolar mucosa[2], palate[42], buccal vestibule[43], and retromolar pad[29], respectively. The ages of patients with lingual lesions ranged from 5 to 73 years with an average of 28.5 years. The most common age group was the third decade (23 cases, 37.7%). Eleven of the 61 patients were males (20. 29%) and 50 were females (79.71%). There were 7 cases of lingual osteomas occurring in Taiwanese[33-37], of which, only one case was found in a male patient. Their clinical features were similar to those reported by others. Of the 8 cases in the buccal mucosa the ages ranged from 12 to 75 years (Table 1). The most common age group was the 5th decade (5 cases, 62.5%). It appeared that the lingual osseous choristoma had an outstanding predilection for females, occurring about four times as often in females as in males, while buccal lesions had only a slight tendency to occur in females (62.5%). Because only eight cases occurred in the buccal mucosa no conclusion could be drawn in age or sex distribution. Why lingual osseous choristoma had a tendency to occur in females is still an enigma.

Most reported lesions presented as a sessile or pedunculated mass with the greatest diameter ranging from 0.3 to 2.5 cm. The average size was 0.96 cm in diameter. The second case reported by Ishikawa *et al.*[3] was 0.3 cm in diameter on the first visit and grew gradually to 0.9 cm 13 years later. This case has the longest period of observation reported in the literature. Only one case occurred as separated masses, one on each side of the foramen cecum[14]. Most cases were asymptomatic. Some patients were unaware of the growth, it was only discovered during routine oral examination. Others knew of its existence but denied any discomfort. If symptoms occurred, they included dysphagia, gagging, nausea, choking, and foreign body sensation[9, 11, 15, 16, 44]. Recently Maqbool *et al.*[6] reported a case with upper airway obstruction and recurrent cough and fever. This was the most serious symptom having been documented in the literature. Among the benign tumors in the tongue close to foraman cecum, fibroma, granular cell tumor, and lingual thyroid should be included in the differential diagnosis from osseous choristoma. Surgical excision was the treatment of choice. Recurrence of lingual osseous choristoma had not been reported. Long and Koutnik[39] reported a case of buccal osseous choristoma which recurred 12 years after surgical removal. In our case 2, the patient had surgery twice before without any definite pathologic diagnosis. It was possible that the patient did not know or the dentist did not tell him the pathologic result of the lesion. We considered the last excision was a recurrent condition.

The etiology of osseous choristoma is unknown and the pathogenesis is debatable. Most authors[1, 4, 16, 20] have thought lingual osseous choristoma to be of developmental origin. The four branchial arches, first seen in early fetal life, play a vital role in the development of the tongue and other related structures. The body and apex of the tongue are derived from the first arch and the base of the tongue is derived from the union of the bases of the second and third branchial

arches. It has been postulated that this lesion developed from proliferation of embryonic rests of the first, second, or third branchial arch and suggested that the pluripotential cells of primitive mesenchymal tissue in the region of foramen cecum differentiate to osteoblasts to form osseous tissue[1, 18, 19, 24]. The great majority of reported cases were located in the dorsal surface of posterior tongue near the foramen cecum (57 cases, 93. 4%). It is still not clear why the osseous choristoma almost exclusively occurs in the tongue, particularly in the region of foramen cecum. This special localization of the tumors must have some developmentally determined anatomic basis. Markaki *et al.*[5], Jahnke and Daly[12], and Cataldo *et al.*[17] suggested that lingual choristoma may arise from ossification of undescended remnants of thyroglossal duct in the tongue. During the embryonic life, the thyroid gland anlage develops along with the thyroglossal duct between the foramen cecum and thyroid gland. This makes it possible that lingual osseous choristoma derivesfrom intraglossal remnants of the thyroglossal duct. Ectopic bone formation cannot explain its origin becaus ectopic bone may be found in many other sites in the body[5]. There was no reason for a bone ectopia to occur in this unique location. Although heterotopic bone might occur in an abnormal location, it arises more frequently from metaplastic changes of mesodermal cells and accompanies with chronic inflammation in that area[29,30]. Trauma has also been discussed to be a possible causative factor. Most authors agree that trauma occurs only in the anterior portion and lateral border of the tongue or other sites in the oral cavity such as the buccal mucosa[4, 8, 26, 39, 45]. Two cases described on the lateral border of tongue were reported by Esguep *et al.*[8] and Tohill *et al.*[29] and a case of osteocartilaginous choristoma on the ventral surface was reported by Wesley *et al.*[46] These three cases were thought to be of post-traumatic origin. Cutright[47] found that trauma or inflammation led to metaplastic bone or dystrophic bone formation in soft tissue. Furthermore, if it was metaplasia induced by trauma or inflammation, there should be some inflammatory reaction, residual cartilage, or the irregular pattern in metaplastic bone formation without Haversian system[5, 9, 26, 30, 40]. These features had been found neither in the reported cases occurring in posterior tongue nor in our first case. Metaplastic bone formation was evident in our second case (Fig. 5,6) although the patient denied any traumatic history. It was possible that the trauma had occurred too long ago to remember. Therefore, we agree with the conclusion made by Sookasam *et al.*[4] that "osseous choristoma" consists of two pathogenetic groups: (a) the lesions in the posterior tongue near the foramen cecum may be a developmental malformation; or (b) the lesions of other sites including anterior or lateral portion of the tongue, buccal, and palatal mucosa may arise from post-traumatic ossification. More case reports of this rare entity should be helpful in clarifying its histopathogenesis.

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舌骨性迷離瘤－病例報告

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骨性迷離瘤(osseous choristoma)是一非常稀少的腫瘤，它包含了正常的骨頭，但卻發生在不正常的位置。大部份的口內骨性迷離瘤多發生在舌背後部近舌盲孔(foramen cecum)附近。到目前爲止，約有73例的口內骨性迷離瘤被發表過，其中61例發生在舌頭，8例發生在頰黏膜。本報告介紹兩病例的口內骨性迷離瘤。一病例發生在後舌背中線輪廓乳頭(circumvallate papillae)正前方。另一病例發生在右頰黏膜剛好在史坦生氏管(Stensen' s duct)開口下方。臨床表徵、顯微鏡下觀、病變來源、和致病原因(pathogenesis)都將討論，並做文獻回顧。

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