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Osseous Choristoma of the Buccal Mucosa: A Case Report

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Osseous choristomas of intra-oral soft tissue are extremely rare. A case of osseous choristoma in the buccal mucosa is presented. A 55-year-old male was referred to our clinic complaining of a mass formation in the buccal mucosa. A tumor 25 × 25 × 30 mm in size was subsequently removed under local anesthesia. Microscopic examination revealed that the tumor was composed of lamellar bone having fatty marrow and osteoblasts. A diagnosis of osseous choristoma was therefore made.

Key words: buccal mucosa; osseous choristoma; osteoma

Osseous lesions of intra-oral soft tissue are very rare. This tumor is usually found on the dorsal surface (Church, 1964) of the posterior third of the tongue. The proper term for such tumors is in dispute, and variations such as "osteoma" and "osseous choristoma" are both in use. As we have recently experienced a case of osseous choristoma in the buccal mucosa, we report it with a review of the recent literature.

Case Report

A 55-year-old male was referred to Tottori Prefectural Central Hospital in June 1988 with a chief complaint of a pain-free mass formation in the right buccal region. Aproximately 8 years before his first visit to our clinic, an object similar to a needle became stuck in his right cheek at mealtime, and bleeding occurred. Subsequently, red bean-sized tumor formation was recognized at the same spot. The subject ignored the tumor for lack of pain and discomfort, however. Over time, the tumor increased gradually in size. He visited our clinic because it had grown rapidly larger in the previous half year, reaching walnut size.

Extra-oral examination revealed slight swelling in the right buccal region (Fig.1). Intra-oral examination revealed a definite and pedunculated tumor measured 25 × 25 × 30 mm in diameter (Fig.2). On palpation, the tumor was firm and elastic hard on the surface, but a hard mass was palpable in the deeper down. The tumor was covered with normal mucous membrane and toothmark impressions could be recognized in one part. A radiograph revealed a well-circumscribed radioopaque nodule, with a dense opaque border and numerous trabeculae traversing the entire mass (Fig.3).

Our diagnosis was a benign tumor arising in the buccal mucosa, the tumor was then excised under local anesthesia. The tumor was removed at the peduncle in one block along with the surrounding mucous membrane and adipose tissue. The tumor was 25×25×30 mm in size, almost spherical-shaped and covered with normal oral mucous membrane (Fig.4). The cut surface of the tumor was yellow as a whole with partial areas of brown, some scattered hard tissues were also present (Fig.5).

Histological examination demonstrated several osseous lesions in the adipose tisse. The lesions consisted of mature lamellar bone, which was surrounded by thin fibrous tissue. Thin trabeculae and fatty marrow-like structures could be seen inside and some osteoblasts lined the bones partially (Fig.6). Cartilaginoid structures were also partially present. Histologically, this tumor was consistent with osseous choristoma.

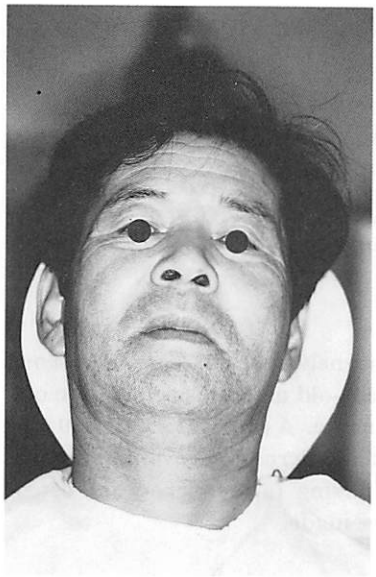

Fig. 1. Facial appearances at initial examination. Slight swelling was recognized in the right buccal region.

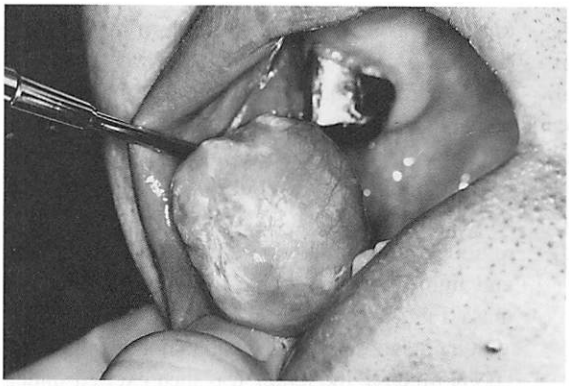

Fig. 2. Intra-oral appearances of pedunculated tumor on the right buccal mucosa.

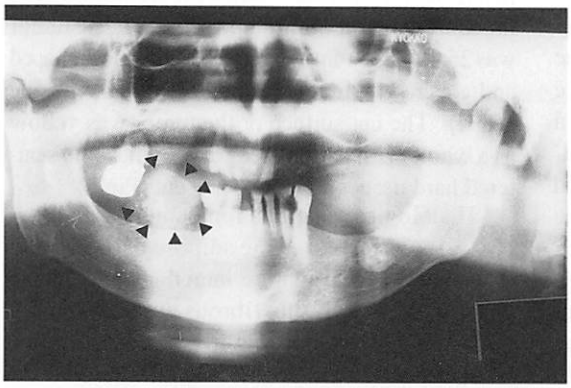

Fig. 3. Radiograph showing a radioopaque trabeculated mass (arrows)

Five years after the operation, there have been no clinical signs of recurrence.

Discussion

Intra-oral osseous choristoma are very rare. In this country, osseous choristoma (osteoma) of the tongue has been occasionally reported (Shimono et al., 1984; Ishikawa et al., 1993), but this was the first report of a case occurring in the buccal region, so far as we could determine. In 1971, Krolls and colleagues (1971) discussed the osseous choristoma (osteoma) of 25 cases, including 9 of their own. According to their report, 24 of these osseous choristomas were found in the tongue and one in the buccal region. Ages of subjects ranged from 9 to 73 years. In thirteen cases, or about half overall, subjects were in their twenties. The sex distribution showed a female : male ratio of 4:1. The size of tumors ranged from 5 mm to 20 mm in diameter. As for clinical symptoms, they reported that many patients with lingual lesion complained of nausea and pharyngeal irritation.

So far as we could determine, osseous choristoma (osteoma) arising in the buccal mucosa has been reported in 7 cases, including that of the authors (Table 1). Age range in the present series was from 33 to 75 years, averaging 47 years. Four of the patients were males. Tumor size ranged from 5 mm to 30 mm in diameter, with our case involving the largest. Osseous choristomas (osteoma) of the buccal mucosa were either asymptomatic or produced only slight discomfort, though those of the tongue produced stronger symptoms.

The histological characteristics of such osseous choristomas are shown in the table. The bone was composed of densely lamellated bone with Haversian canals, and sometimes with bone marrow. This bone was surrounded by fibrous connective tissue. In our case, the existence of

fatty marrow and osteoblasts were remarkable compared with the other cases. We consider that this difference may be related to the increased size of the tumor over the half year before the subject's first visit to our clinic.

Controversy concerning osseous lesions arising in the soft tissue focus mainly on their origin and on terminology. Sookasam and Philipsen (1986) grouped intra-oral soft tissue osteomas into two categories according to the histogenetical concept. The following is their theory: i) The posterior tongue mid-line type is thought to represent a developmental malformation. ii) The type including soft tissue osteomas in intra-oral locations different from that in the first group may be interpreted as a posttraumatic center of ossification (metaplasia).

In addition to this theory, numerous other theories exist concerning the tongue location, including the persistent theory of branchial arch (Monserrat, 1913), ossificated theory of a lingual thyroid (McClendon, 1975), alternatively ossificated theory of fibroma (Roy and

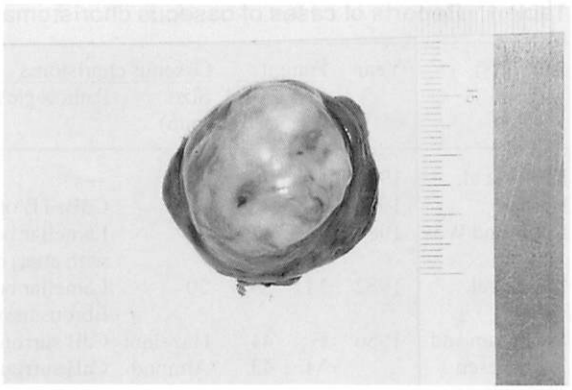

Fig. 4. Gross sepcimen of the tumor after removal.

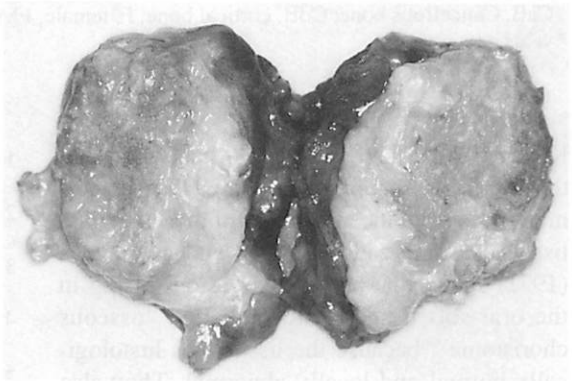

Fig. 5. Cut surface of the tumor.

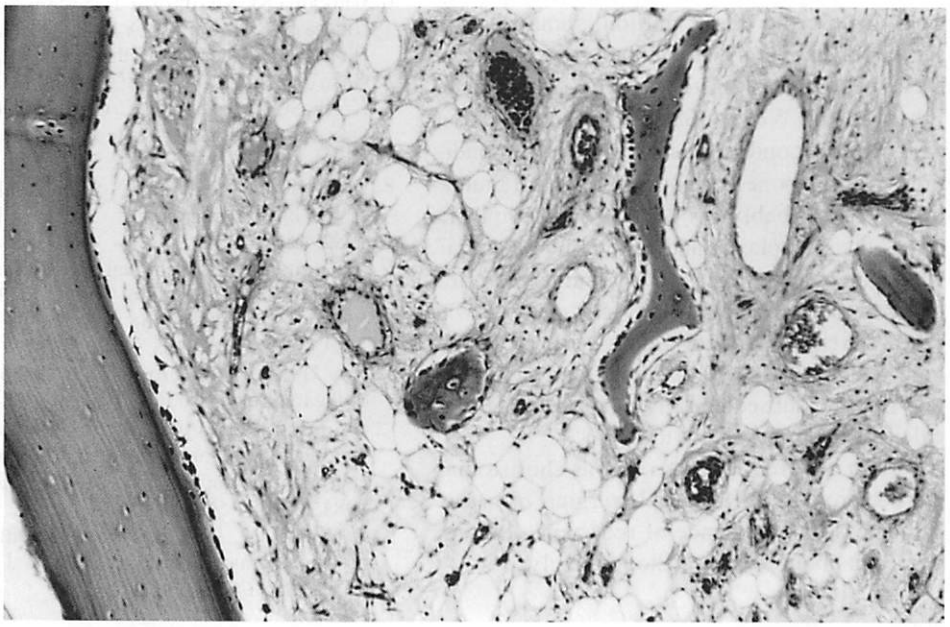

Fig. 6. Photomicrograph showing trabecular bone, fatty marrow and osteoblast surrounded by lammelar bone (hematoxylin and eosin, × 10).

Table 1. Reports of cases of osseous choristomas

Author(s)	Year	Patient		Osseous choristoma	
		Sex	Age*	Size (mm)	Pathologic feature
Krolls et al.	1971	M	40	—	—
Herd	1976	F	75	—	CoB, TB of the center
Davis and Was	1980	M	42	5	Lamellar bone surrounded by fibrous tissue loose TB with areas of FM
Mesa et al.	1982	M	33	20	Lamellar bone with numerous trabeculae surrouded by fibrous tissue, partially myxomatous changes
Sookasam and Philipsen	1986	F	41	Hazelnut	CaB surrounded by cortical plate hematopoietic marrow, FM
		M	43	Almond	CaB surrounded by cortical plate hematopoietic marrow, FM
The present authors	1994	M	55	30	TB surrounded by lamellar bone FM, osteoblast

CaB, Cancellous bone; CoB, cortical bone; F, female; FM, fatty marrow; M, male; TB, trabecular bone.
*years.

Klein, 1970). Ishikawa (1986) has stated that the possibility should not be denied that such tumors arise from the metaplasia of mesenchymal tissue by some cause. Krolls and colleagues (1971) stated that an osseous lesion arising in the oral soft tissue could be called "osseous choristoma", because the tissue was histologically normal and locally abnormal. They also noted that the term "osteoma", most frequently used to describe this condition, should be reserved for an overgrowth of bone closely associated with a part of the skeletal structure. Enzinger and Weiss (1988) stated that a lesion which was secondarily ossified from the condition causing bone formation (e.g., inflammation) was probably produced by metaplasia rather than neoplasia, and the term "osteoma" was inadequate. There were previous histories of trauma or inflammation in four of the seven cases which we studied. In our case, the lesion had the history of the stab trauma, and we speculate that the subsequent granulation tissue was calcified and ossificated further.

None recurrence of the osseous choristomas has been reported after simple surgical excision. In our case, no evidence of recurrence has appeared at about 5 years postoperatively.

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