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CASE REPORT

Buccal osseous choristoma in a 5-year-old boy

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KEYWORDS

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Summary Choristomas are defined as proliferations of histologically normal tissue located in an abnormal position. Osseous choristomas of oral soft tissue are rare lesions, especially in children, occurring most frequently in the tongue, and less commonly in other sites such as buccal mucosa, palate and alveolar mucosa. It has been suggested that osseous choristomas have developmental or traumatic origins. Definite diagnosis can only be determined after histologic examination, whereupon the treatment of choice is surgical excision. Here we present, what is to our knowledge, a second case of buccal osseous choristoma in a pediatric patient, along with a brief review of the current, pertinent literature.

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Introduction

Osseous choristoma is a term used to describe the growth of a tumor-like mass of normal bone in an abnormal position.¹ The occurrence of osseous choristoma in buccal mucosa is relatively uncommon, especially in children. To the best of our knowledge, only 13 cases of buccal osseous choris-

toma have been previously reported,^{1–11} including just one case in a pediatric patient.⁶ This report presents an additional case of buccal osseous choristoma, which occurred in a 5-year-old boy, along with a brief review of the pertinent literature.

Case report

A 5-year-old male, accompanied by his mother, was referred to our institution with a complaint of a swelling in the buccal mucosa. The mother

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of the child reported that she had noted a swelling that had been slowly-enlarging for about 1 year, which was not associated with any obvious trauma. Generally, the patient was in otherwise good health, and his medical and family history was non-contributory.

Oral examination revealed the presence of a 2.5 cm circumscribed swelling, located on the right side of the buccal mucosa. Upon palpation, this non-ulcerated lesion was found to be firm. Furthermore, the swelling was not tender and, reportedly, painless. The clinical impression was of a benign tumor, of mesenchymal origin, and the entire mass was subsequently excised with both sharp and blunt dissection under local anesthesia (Fig. 1). The specimen was then sent for histopathological examination. The well-circumscribed, smooth-surfaced mass was pinkish in color and measured $2.5 \times 2.5 \times 1$ cm in size (Fig. 2). Histopathologic examination of the decalcified specimen revealed a well-circumscribed, non-encapsulated mass consisting of multiple osseous foci of varying diameter and maturation, surrounded by loose fibrovascular connective tissue (Fig. 3(A)). Examination of some of the foci revealed viable osteocytes within the lacunae (Fig. 3(B)). The ultimate diagnosis was osseous choristoma of the buccal mucosa. The excised area healed uneventfully, and was free of recurrence for one year.

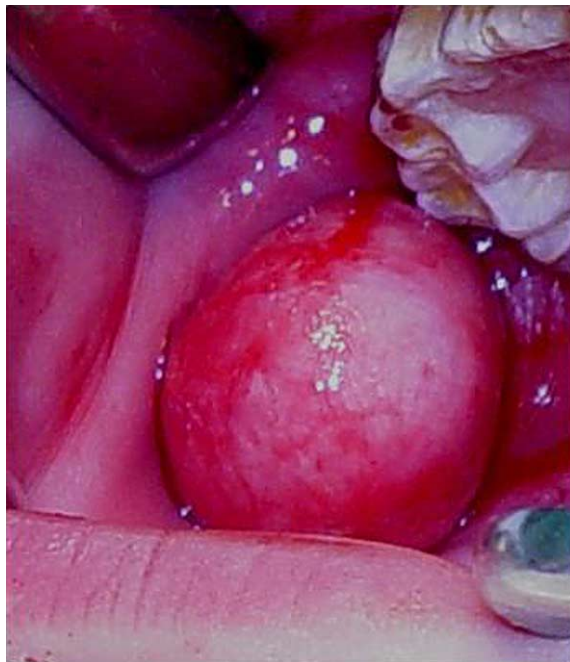


Figure 1 Intra-operative appearance of a swollen mass in the buccal mucosa.



Figure 2 Photograph of the surgical specimen.

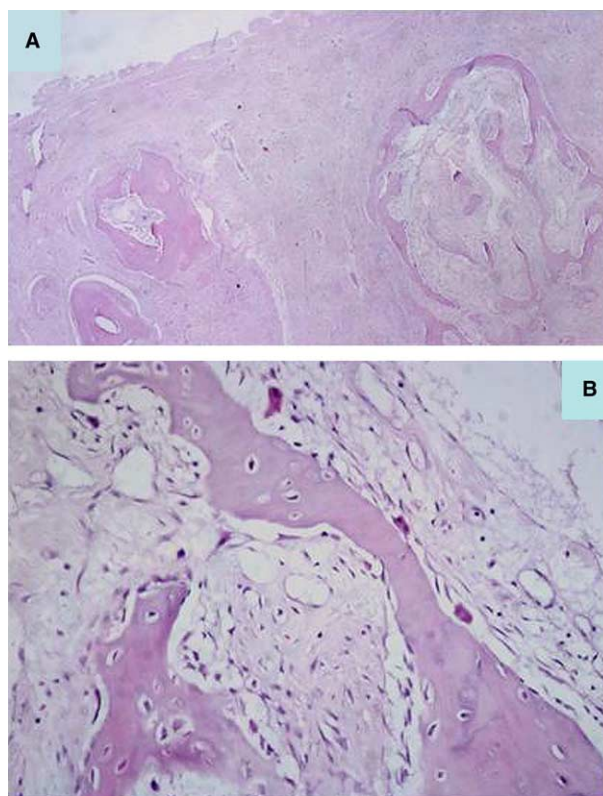


Figure 3 Photomicrograph of decalcified specimen showing multiple bony foci of varying dimensions within the loose fibrovascular connective tissue in low-power view (A, Hematoxylin eosin $\times 10$). Higher magnification photomicrograph demonstrating the bony trabeculae with viable osteocytes within the lacunae (B, Hematoxylin eosin $\times 100$).

Discussion

The term osseous choristoma was introduced by Krolls et al. in 1971.¹ "Cutaneous ossification", a dermal lesion suggested by Kroll et al.¹ is similar to, if not the same, as intra-oral soft-tissue osteoma. Osseous choristoma is a more suitable term to denote these lesions, as it refers to histologically normal tissue occurring in an abnormal site, whereas the term osteoma indicates a proliferation of bone involved with normal skeletal structures.¹

The most frequent topographic region for osseous choristoma is the posterior third of the tongue.¹² Localization of such a lesion in the buccal mucosa is relatively uncommon, indeed review of the literature showed just 14 cases of osseous choristoma in the buccal mucosa, including the present case (Table 1). Of these 14 cases, the mean patient age was 37 years, with a range from 5 to 75 years. The most common age group for occurrence of buccal lesions seemed to be the fifth decade.^{1,3,5,6,9} Furthermore, while it appears that lingual osseous choristoma has a clear predilection for females, occurring about four times more commonly than in males,⁹ equal sex distribution was noted for buccal lesions (7 female: 7 male). Nine lesions were found in white patients^{1-5,7,8,11} and

two in Chinese patients⁹ (and the present case), with the remaining two lesions occurring in Black⁴ and Turkish¹⁰ patients; one affected patient's race was not stated.⁶ Most lesions,^{1,3-5,7-9} including the current case, were present for 1–2 years, although one was present for 40 years² and one had been present for an undetermined duration.⁶ Almost all lesions, including this case, were within 2.5 cm in diameter^{1-5,7-11} but a case of up to 5 cm in diameter has been reported.¹⁰ In addition, the patients reported varying symptoms ranging from asymptomatic^{3,4} or merely aware of presence^{5,6,9} to having difficulty in mouth-opening or closing.⁷ Eleven cases were found in the right buccal mucosa^{2,4-9,11} and only two lesions in the left site^{3,10}; the site of one case was undetermined.¹

The current case is only the second case reported of buccal osseous choristoma occurring in pediatric patient, and represents the youngest-reported case of this lesion in buccal mucosa. It is noted that the second case reported by Ishikawa et al.¹³ was a 5-year-old girl who presented with a 0.3 cm lingual lesion on the first visit that gradually grew to a diameter of 0.9 cm 13 years later. This case has the longest period of observation reported in the literature. On the other hand, Negow et al.¹⁴ also reported a case of lingual osseous choristoma

Table 1 Reported cases of buccal osseous choristoma

Author(s) (year reported)	Sex	Age (yrs)	Race	Duration (yrs)	Size (cm)	Symptom	Site
Krolls et al. (1971)	Male	40	White	1	Not stated	Tumor	Not stated
Herd (1976)	Female	75	White	40	1.0 × 1.7	"Lump"	Right
Davis (1980)	Male	42	White	1	0.5	None	Left
Mesa et al. (1982)	Male	33	Black	1	1.8 × 1.7 × 1.0	None	Right
Sookasam and Philipsen (1986)	Female	43	White	2	Almond sized	"Lump"	Right
	Female	41	White	Not stated	Hazelnut	Aware of presence	Right
Hoddler and McDonald (1988)	Female	12	Not stated	0.5	1 × 0.5	Aware of presence	Right
Long and Koutnic (1991) ^a	Female	50	White	2	2 × 1.6 × 1.2	Difficulty in opening and closing mouth	Right
Mintz et al. (1995)	Female	16	White	1	2.5 × 2 × 2	Painless swelling	Right
	Male	67	White	1	2.5 × 2 × 1.5	Painless swelling	Right
Lin et al. (1998)	Male	45	Chinese	1	2.3 × 2.0 × 1.5	Aware of presence	Right
Dalkiz et al. (2001) ^b	Male	22	Turkish	5	5 × 3 × 3	Painless, slowly enlarging swelling	Left
Gaitan-Cepeda et al. (2003)	Female	28	White	4	0.5 × 0.5	Painless swelling	Right
Chen et al. (present case)	Male	5	Chinese	1	2.5 × 2.5 × 1	Painless swelling	Right

^a Lesion recurred 12 years after excision.

^b Lesion recurred 1 year after excision.

with a long observation period by the patient concerned, who first noticed the swelling when she was about 10-years-old but was unaware of it until a routine dental check-up at the age of 23-years-old. Therefore, taken together, and as indicated in this case, osseous choristoma, though uncommon, may occur in pediatric patients. It is, therefore, important for both pediatric dentists and otolaryngologists to include this entity, amongst other benign tumor-like lesions, such as salivary gland neoplasm, fibroma, lipoma or neural tumor, as a possible differential diagnosis for buccal swelling in a child.

With regards to the pathogenesis of buccal osseous choristomas, some have suggested that buccal lesions are developmental malformations in which the osseous foci derived from ossification of branchial arc remnants, or osseous differentiation and development of ectopic mesenchymal cells.¹² Multiple osseous foci with varying diameter and different maturation, within a fibrovascular stroma, were observed microscopically in the current case, appearing to support the aforementioned etiology. On the other hand, lesions were also thought to appear as a result of post-traumatic ossification.⁶ Although our patient has no clinical record of history of trauma to the maxillofacial region, there remains the possibility of an unnoticed or ignored trauma in an active male child.

The treatment of choice for such a lesion involves surgical excision, after which recurrence is not usually expected. Interestingly, recurrence has been reported for two buccal osseous choristomas^{7,10} whilst no recurrence has yet been found for lingual counterparts. The explanation for a recurrence had been attributed to a new fibrotic region, which might have arisen as a result of the surgical trauma, and underwent ossification, or to an uncalcified lesion that subsequently ossified.^{7,10} However, since there have been only two such cases of recurrence,^{7,10} no definite conclusion can be drawn as to the cause of recurrence. Neverthe-

less, a longer period of observation should be adopted for osseous choristomas, particularly for a buccal lesion. Furthermore, the current case is of interest due to the rare occurrence of the osseous choristoma in pediatric patients.

References

1. Krolls SO, Jacoway JR, Alexander WN. Osseous choristomas (osteomas) of intraoral soft tissues. *Oral Surg Oral Med Oral Pathol* 1971;**32**:588–95.
2. Herd JR. Extra-osseous osteoma. *Aust Dent J* 1976;**21**:469–74.
3. Davis GB. Intraoral osseous choristoma: report of case. *J Oral Surg* 1980;**38**:144.
4. Mesa ML, Schneider LC, Northington L. Osteoma of the buccal mucosa. *J Oral Maxillofac Surg* 1982;**40**:684–6.
5. Sookasam M, Philipson HP. The intra-oral soft tissue osteoma: report of two cases. *J Dent Assoc Thai* 1986;**36**:229–34.
6. Hodder SC, MacDonald DG. Osseous choristoma of buccal mucosa: report of a case. *Br J Oral Maxillofac Surg* 1988;**26**:78–80.
7. Long DE, Koutnik AW. Recurrent intraoral osseous choristoma. Report of a case. *Oral Surg Oral Med Oral Pathol* 1991;**72**:337–9.
8. Mintz S, Anavi Y, Barak S, Nalley K. Osseous choristomas of the buccal mucosa. *J Mich Dent Assoc* 1995;**77**:30–2, p. 55.
9. Lin CC, Chen CH, Chen YK, Shen YH, Lin LM. Osseous choristoma of oral cavity-report of two cases and review of the literature. *Kaohsiung J Med Sci* 1998;**14**:727–33.
10. Dalkiz M, Hakan Yurdakul R, Pakdemirli E, Beydemir B. Recurrent osseous choristoma of the masseter muscle: case report. *J Oral Maxillofac Surg* 2001;**59**:836–9.
11. Gaitan-Cepeda LA, Quezada-Rivera D, Ruiz-Rodriguez R. Osseous choristoma of the oral soft tissue. Case report. *Med Oral* 2003;**8**:220–3.
12. Chou LS, Hansen LS, Daniels TE. Choristomas of the oral cavity: a review. *Oral Surg Oral Med Oral Pathol* 1991;**72**:584–93.
13. Ishikawa M, Mizukoshi T, Notani K, Iizuka T, Amemiya A, Fukuda H. Osseous choristoma of the tongue. Report of two cases. *Oral Surg Oral Med Oral Pathol* 1993;**76**:561–3.
14. Cheong NW, Boon LC, Shanmahasuntharam P. Osseous choristoma of the tongue. *J Nihon Univ Sch Dent* 1996;**38**:49–51.