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Case Report

Lipoosteocartilaginous choristoma of the tongue: a case report

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Abstract – Introduction: Choristoma is the proliferation of histologically normal tissue in the ectopic position. Oral choristoma infrequently occurs in soft tissue, with most cases developing on the tongue and displaying osseous histological findings. It is mostly encountered in the fifth decade of life, but the age ranges from 12-to-64 years old, with a female predilection. **Observations**: An 81-year-old man presented with an asymptomatic, firm, mobile and pedunculated nodule on the right dorsum of the tongue, just anterior to the circumvallate papillae, for more than ten years duration. Microscopically, the lesion was covered by a parakeratinised stratified squamous epithelium and central to the lamina propia, large, calcified material that resembles normal bone and cartilage was observed. The osseous basophilic mass appears rounded with the characteristic rim of reversal lines and contains numerous lacunae with osteocytes. Peripherally, chondroid metaplasia was also seen with evidence of mature hyaline cartilage containing chondrocytes. The whole osseous-chondroid mass was surrounded by adipose tissue. **Conclusion**: Oral choristoma is a rare entity, and a mixture of cartilage, bone and adipose tissue is considered an exquisite histological finding. Generally, it has benign behaviour, and surgical excision is the treatment of choice. No recurrence has been reported.

Introduction

Choristoma is defined as a tumour-like growth composed of normal tissue microscopically in an abnormal location and of developmental origin [1]. Monserrat, in 1913 was the first to report a bony lesion in the soft tissue on the dorsum of the tongue and was diagnosed as lingual osteoma. The term osseous choristoma was first introduced by Krolls *et al.* in 1971 [2].

An osseous choristoma in the soft tissue is considered uncommon. Its incidence in the oral cavity is even more infrequently encountered. Histologically, most reported cases contained a mixture of normal structures such as bone, cartilage, gastric mucosa, glial, sebaceous gland, and fat tissue [3].

Observation

An 81-year-old man was presented to the Hospital Universiti Sains Malaysia's Oral and Maxillofacial Surgery unit with a complaint of painless swelling on the tongue that had been present for ten years. Except for minor discomfort during eating and swallowing, the patient denied any history of trauma, bleeding, or paraesthesia. The patient has no known medical conditions. An intraoral examination revealed a well-circumscribed, pedunculated nodule near the midline dorsum of the tongue, anterior to the circumvallate papillae. The lesion was 10 × 10 mm in diameter, mobile, yellowish pink in colour, with a smooth surface (Fig. 1). There was no ulcer noted, and it was firm in consistency.

The lesion was excised under local anaesthesia and sent for histopathological examination. The gross macroscopic finding showed a round to oval yellowish nodule, measuring approximately 10 mm x 13 mm x 10 mm with a hard-rubbery consistency. On the cut surface, the bisected specimen showed a central whitish area surrounded by fatty tissue (Fig. 2).

Microscopically, the specimen was covered by a parakeratinised stratified squamous epithelium and supported by dense fibrous connective tissue stroma. Central to the lamina propia, large, calcified material resembles bone and cartilage was observed. The osseous basophilic mass appears rounded with the characteristic rim of reversal lines and contained numerous lacunae with osteocytes. Peripherally, chondroid metaplasia was also seen with evidence of mature hyaline cartilage containing chondrocytes. The whole osseous-chondroid mass was surrounded by adipose tissue (Fig. 3).

One-week post-biopsy, the patient was reviewed, and the surgical site was healing well. At subsequent 6-monthly and annual check-ups, no recurrence was noted.

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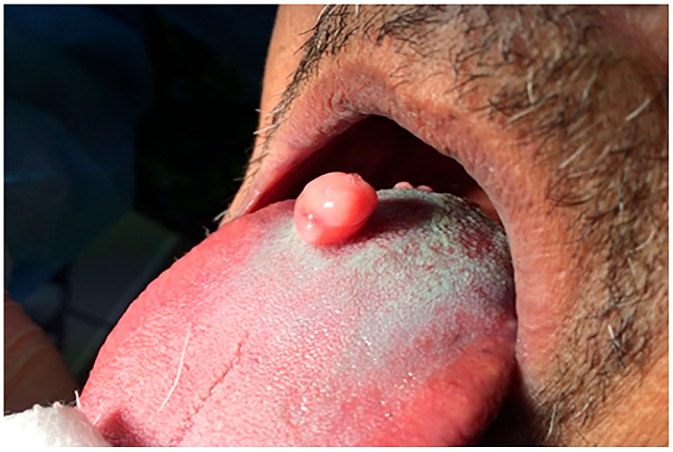

Fig. 1. Intraoral photograph showing a yellowish-pink nodule on the dorsum of the tongue, anterior to circumvallate papillae.

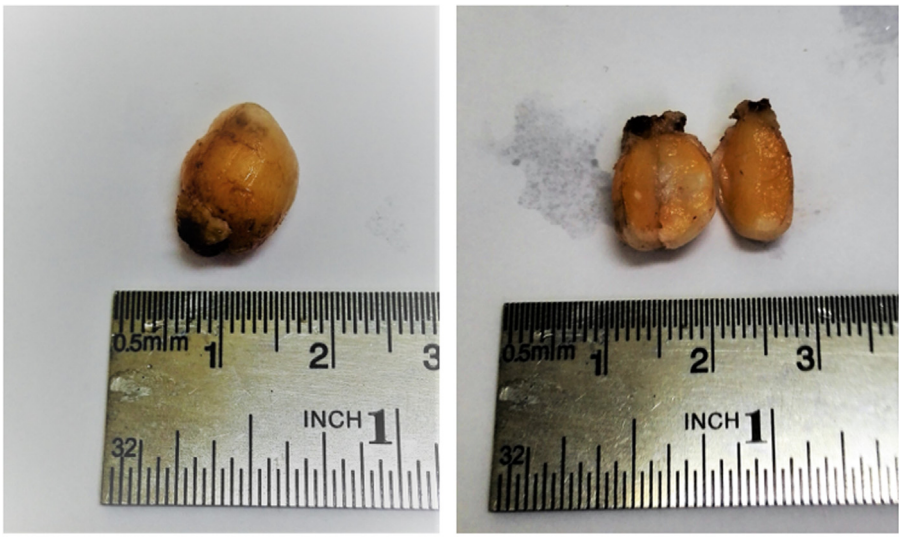

Fig. 2. Gross macroscopic findings show a round to oval yellowish nodule measuring approximately 10 mm × 13 mm × 10 mm with hard rubbery consistency (a). The bisected specimen showed a central whitish area on the cut surface surrounded by fatty tissue (b).

Discussion

Differential diagnoses of the lesion include fibroepithelial polyp, pyogenic granuloma, granular cell tumour, neurofibroma and ectopic thyroid nodule. Fibroepithelial polyp (irritation fibroma) is a reactive hyperplasia lesion, the most common benign swelling in the oral cavity. It is induced by trauma such as sharp tooth edge, overhanging restoration, lip/cheek biting, and calculus when in contact with oral mucosa [1]. However, no history of trauma was elicited in the present case. The buccal mucosa is the most typical site for the fibroepithelial polyp, but it also can occur at the labial mucosa, gingiva, and lateral border of the tongue. Clinically, it may present with a soft to firm nodule. It can occur at any age but is often diagnosed in middle-aged adults. Fibroepithelial polyp and oral choristoma are clinically indistinguishable, except that the former is quite common in the oral cavity. In contrast, the latter is rare and only incidentally found during histopathological examination of the biopsied specimen.

Pyogenic granuloma presents as a pedunculated lesion with usually very reddish and easily bleeds upon touch because of the prominent capillary composition in the hyperplastic granulation tissue. Nevertheless, the lesion in the present case did not bleed easily upon palpation. The lesion of pyogenic granuloma invariably turns pink over time as the capillaries are replaced by fibrous tissue. It also presented as a painless pedunculated or sessile lesion, and the ulcerated surface may be due to secondary trauma [1,4].

The tongue is the typical site for granular cell tumours. It can occur at any age, mainly in the 4th and 6th decade. Females are mostly affected [1]. It is commonly presented as a painless, well-demarcated, sessile submucosal swelling at the dorsum of

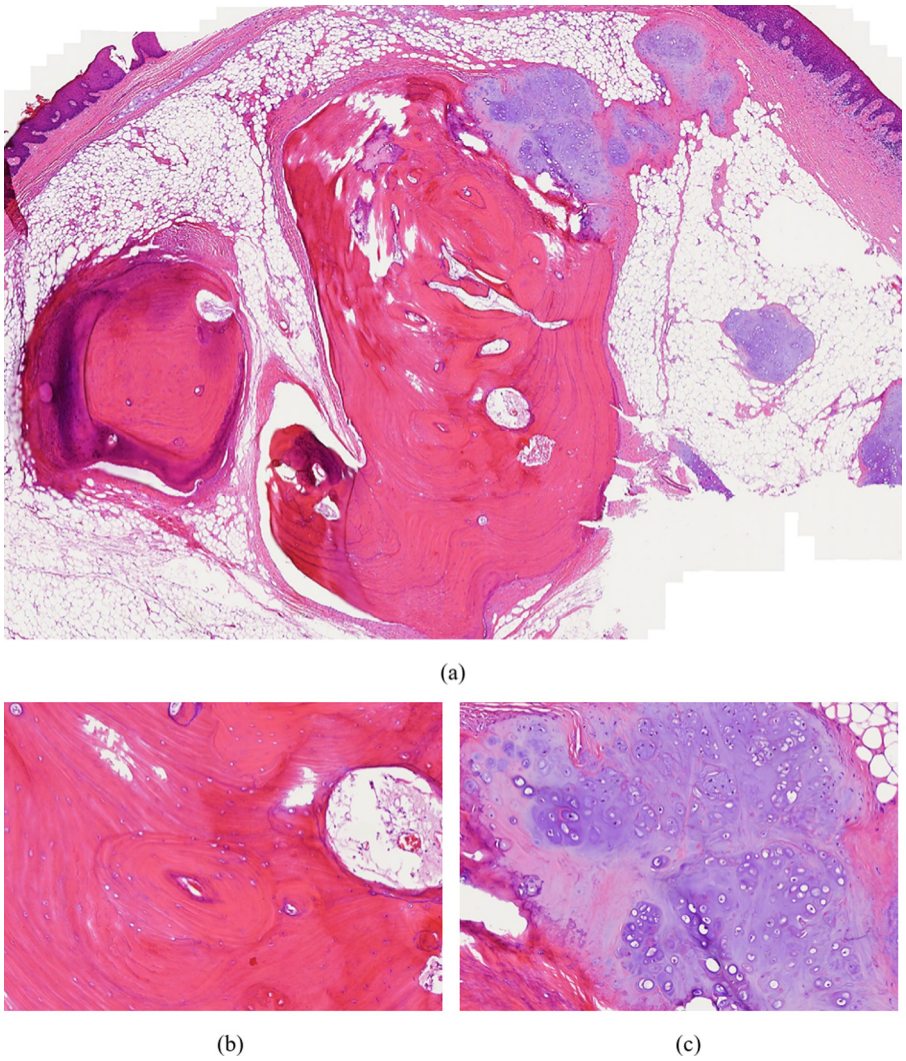

Fig. 3. Hematoxylin and eosin (H&E) stain of lipoosteocartilaginous choristoma of tongue. Low-power photomicrograph revealed multiple extensive calcifications resembling bone and cartilage within lamina propria surrounded by adipose tissue (a) (100×); Higher magnification showed osseous basophilic masses with characteristic rims of reversal lines and contains numerous lacunae with osteocytes (b); some areas of the outer periphery appear chondroid with the presence of mature hyaline cartilages containing lacunae with one to two chondrocytes noted (c) (b-c, 200×).

the tongue as compared to a pedunculated swelling seen in the present case.

Neurofibromas and schwannoma (neurilemmoma) are two common peripheral nerve sheath neoplasms affecting children and adolescents [5]. These lesions usually present as a slow-growing submucosal mass commonly found on the tongue, palate, buccal mucosa, and floor of the mouth. Histopathologically, neurofibroma is composed of an admixture of cells with mixed cellularity, *i.e.*, Schwann cells, fibroblast, perineurial-like cells, and axons, whereby schwannoma comprised spindle cell proliferation of Schwann cells with alternating cellular Antoni A and hypocellular Antoni B areas [6].

Based on the location of the swelling, *i.e.*, midline dorsum and in the vicinity of the foramen cecum, the differential diagnosis of an ectopic thyroid nodule (lingual thyroid) was given consideration. Based on the literature, lingual thyroid is primarily seen in females in the third decade of life who present with symptoms of hypothyroid [7]. In some cases, the lingual thyroid might be the only thyroid tissue in the body.

Lipoosteocartilaginous choristoma was rarely reported in the literature and was described as a variant tissue that presented together with other tissue [8,9]. In the oral cavity, choristoma may develop from several tissues like bone, cartilage, gastric mucosa, glial, and adipose tissue [3,10–13]. The pathogenesis of choristoma is unknown. Several tissue origins and development mechanisms have been proposed in the literature, *i.e.*, a mixed neoplasm (teratoma) with the preponderance of cartilage, a metaplastic process, a derivation

from pluripotential cells, and a development from cartilaginous embryonic rests [14]. If there was a history of trauma, the theory of metaplasia is most accepted. Trauma often occurs during swallowing and articulation, especially at the lateral border of the tongue, causing injuries leading to the development of local inflammation followed by metaplasia of the affected tissues [15].

In 2014, approximately 67 oral cases were reported [3]. Since then, more than 150 cases have been reported worldwide. Osseous choristoma is more frequently reported compared to cartilaginous and osseocartilaginous proliferation. The lesion was more prevalent in females than males (3:1), and the age affected ranges from 5 to 73 years old [3]. Osseous choristoma was found more on the posterior tongue, especially near the circumvallate papillae and foramen caecum. The lesion size varies from 0.5 to 5.0 cm. Besides the tongue, osseous choristoma is often found on buccal mucosa [16].

Conclusion

In the present case, the definitive diagnosis of lipo-osteocartilaginous choristoma of the tongue was revealed by histopathological findings, in which adipose tissue, bone, and cartilage were observed in the lesion simultaneously. As there was no history of trauma, the theory of the proliferation from the embryonic rest is the most plausible to explain the occurrence of the lesion in this location.

Conflict of Interest

The authors have no conflicts of interest to declare.

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Ethical Approval

The ethical approval was obtained from the Human Research Ethics Committee Universiti Sains Malaysia (USM/ JEPeM/ 22050303). This work conformed to the principles of the Helsinki Declaration of 1975 and 1983.

Consent

Informed consent was obtained prior to the preparation of the case report, and the author/s endeavoured all efforts to ensure anonymity.

Authors Contributions

N. A. Rahman: Conceptualisation, Data acquisition and interpretation, Writing final draft. N.A. Ghani: Data acquisition, Writing original draft, M.F.Z. Fikry – Data acquisition, S.A. Rahman, Writing – reviewing and editing.

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References

1. Neville BW, Damm DD, Allen CM, Chi AC. Soft tissue tumours. In Color Atlas of Oral and Maxillofacial Diseases. Neville BW, Damm DD, Allen CM, Chi AC. 1st ed. Elsevier, 2019. pp 328.
2. Krolls SO, Jacoway JR, Alexander WN. Osseous choristomas (osteomas) of intraoral soft tissues. Oral Surg Oral Med Oral Pathol 1971;32:588–595.
3. Gorini E, Mullace M, Migliorini L, Mevio E. Osseous choristoma of the tongue: a review of etiopathogenesis. Case Rep Otolaryngol 2014;2014:373104.
4. Regezi JA, Sciubba JJ, Jordan RC. Oral Pathology Clinical Pathologic Correlations. 7th ed. St. Louis: Saunders Elsevier, 2016, pp. 118–120.
5. Cates JMM, Coffin CM. Neurogenic tumors of soft tissue. Pediatr Devel Pathol 2012;15:62–107.
6. Rodriguez FJ, Folpe AL, Giannini C, Perry A. Pathology of peripheral nerve sheath tumour: diagnostic overview and update on selected diagnostic problems. Acta Neuropathol 2012;123:295–319.
7. Thomas G, Hoilat R, Daniels JS, Kalagie W. Ectopic lingual thyroid: a case report. Int J Oral Maxillofac Surg 2003;32:219–221.
8. Kitazawa T, Shiba M. Osteochondrolipoma of the mandible. Eplasty 2017;17:e35.
9. Roper A, Subar D, Benatar B, Kobbe AER. Osteocartilaginous choristoma: a case report. J Laryngol Otol 2006;120:E23.
10. Toida M, Sugiyama T, Kato Y. Cartilaginous choristoma of the tongue. J Oral Maxillofac Surg 2003;61:393–396.
11. Wetmore RF, Bartlett SP, Papsin B, Todd NW. Heterotopic gastric mucosa of the oral cavity: a rare entity. Int J Pediatr Otorhinolaryngol 2002;66:139–142.
12. Ide F, Shimoyama T, Horie N. Glial choristoma in the oral cavity: histopathologic and immunohistochemical features. J Oral Pathol Med 1997;26:147–150.
13. Hoseini AT, Razavi SM, Khabazian A. Lipoma in oral mucosa: two case reports. Dent Res J 2010;7:41–43.
14. Mosqueda-Taylor A, González-Guevara M, de la Piedra-Garza JM, Diaz-Franco MA, Toscano-Garcia I, Cruz-León A. Case report: cartilaginous choristomas of the tongue: review of the literature and report of three cases. J Oral Pathol Med 1998;27:283–286.
15. Sethi BK, Kali N, Bommareddy A, Subhodaya R. Cartilaginous choristoma of the tongue. J Pediatric Surg Case Rep 2020;62:101583.
16. Chen YK, Shen YH, Lin YJ, Li YT, Tsai KB, Lin LM. Buccal osseous choristoma in a 5-year-old boy. Oral Oncol 2005;41:198–201.