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Lipoosteocartilaginous Choristoma of the Tongue: A Case Report

Rafa A. Elgehani,^{1*} FDSRCSEd, FFDRCSI ; Amna S. Elgehani,² MSc, PhD ; and Najah H. Bisat,³ BDs, MSc 

Abstract

Choristomas, a form of heterotopia, are masses of normal tissues found in abnormal locations. It is rare in the oral cavity and the tongue is the most affected site intraorally. Majority of choristomas were more prevalent in females than males with a wide age range. An 18 years -old female Libyan patient presented to the dental clinic complaining of a painless swelling of the anterior part of her tongue. The clinical examination revealed submucosal mass measuring 1×1 cm just lateral to the tip of the tongue, pink in color with normal mucosal surface. Microscopically, the section showed lobules of mature hyaline cartilage with adipose tissue rimming and foci of osseous tissue. The lesion was surrounded by a fibrous capsule. Choristoma of the tongue is a rare benign lesion that should be kept as a differential diagnosis of tongue lesions not only in posterior one-third. The diagnosis of lipoosteocartilaginous choristoma was giving according to histopathological examination of excisional biopsy to exclude other benign lesions and malignancy.

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1. Department of oral and maxillo-facial surgery, Faculty of Dentistry, University of Benghazi

2. Department of oral medicine, oral pathology, oral diagnosis and radiology, Faculty of Dentistry, University of Benghazi

3. Department of oral medicine, oral pathology, oral diagnosis and radiology, Faculty of Dentistry, University of Benghazi

*Corresponding Author:

ragehani@yahoo.com

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Introduction

Choristomas are masses of normal tissues found in abnormal locations.¹ In contrast to a hamartoma, the growth of a choristoma is normally regulated.² It is different from a hamartoma. The two can be differentiated as follows: a hamartoma is disorganized overgrowth of tissues in their normal location e.g., while a choristoma is normal tissue growth in an abnormal location e.g., osseous choristoma.³

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.⁴ 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.⁵

Methods

A case report and review of relevant literature. Research platforms (PubMed, Scopus, google scholar) were search for clinical literature published in the last ten years in re-

lation to the tongue choristomas. Description of a clinical case treated in a Libyan hospital is provided.

Case Report

An 18 year-old female Libyan patient presented to the dental clinic complaining of a painless swelling of the anterior part of her tongue. The clinical examination revealed submucosal mass measuring 1×1 cm just lateral to the tip of the tongue, pink in color with normal mucosal surface (Figure 1). The lesion was excised under general anesthesia and sent for histopathological examination. The gross macroscopic findings showed an oval yellowish mass, firm in consistency measuring approximately 1.5×1 cm.

Microscopically, the section showed lobules of mature hyaline cartilage containing lacunae with chondrocytes and foci of osseous tissue. There was an adipose tissue rimming the cartilaginous tissue. The lesion was surrounded by a fibrous capsule (Figures 2-4).

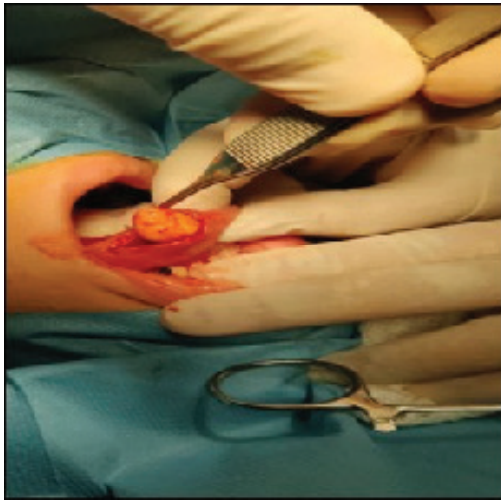


Figure 1: Intraoral photograph showing excision of circumscribed yellowish mass from the tip of the tongue.

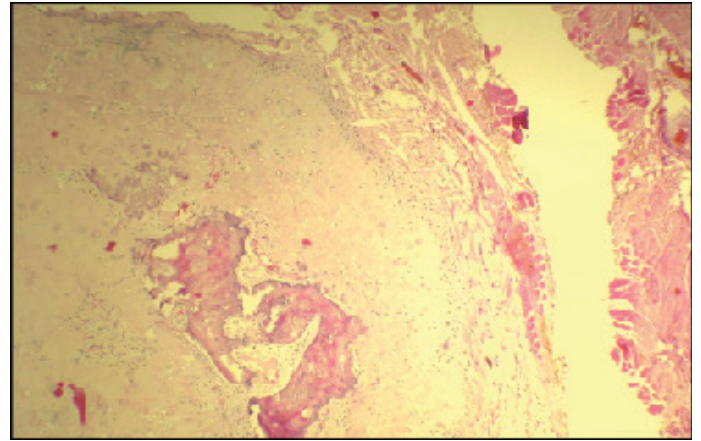


Figure 2: Hematoxylin and Eosin stained section (H&E, 100) shows cartilaginous tissue containing lacunae with chondrocytes mixed with eosinophilic bony calcification

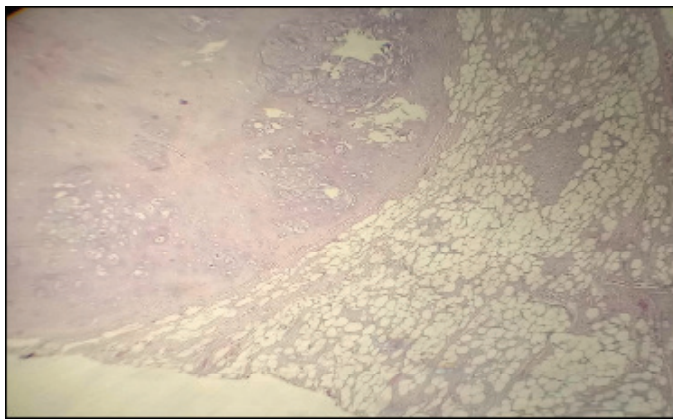


Figure 3: H&E stained section (100x) shows adipose tissue rimming the cartilaginous compartment

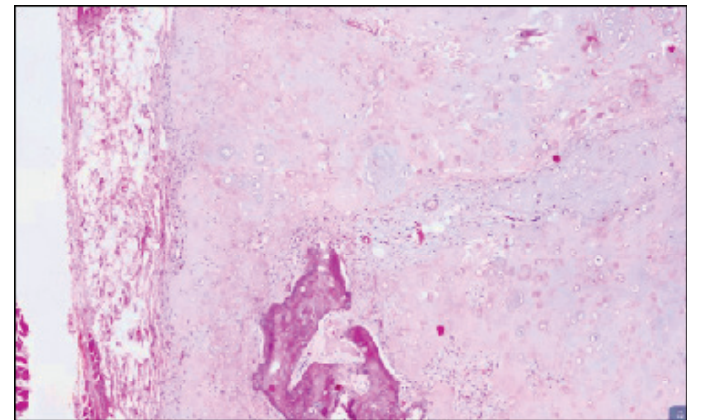


Figure 4: H&E stained section shows the cartilaginous tissue with chondrocytes in lacunae mixed with bony calcification surrounded by fatty tissue

Discussion

Choristomas' rarely present in the oral cavity. However, when they occur in the oral cavity, mucosa of the tongue affected most frequently and less commonly on other sites such as buccal mucosa, alveolar mucosa, and the periodontium.⁶⁻⁸ Until 2020 only 69 confirmed cases of osseous choristoma of the tongue have been reported in the literature.⁹ Majority of osseous choristomas were more prevalent in females than males (3:1), and affected wide age range.^{9,10} This is in accordance with present case which was a female aged 18 years old.

Previous reports have suggested that the posterior one-third of dorsal tongue is the most common location of osseous choristoma with common specific location in (21%) of cases at the apex of the foramen cecum.⁹ The present finding was not consistent with the previous reports as the lesion was found on the tip of the dorsal tongue, which considered as a different new location.

Histologically, choristoma is an island of normal tissue that occurs in an abnormal location. The various tissue types that can occur in the oral cavity as choristomas are cartilage, bone, glial tissue, salivary gland and thyroid tissue.^{11,12} Choristomas with cartilaginous tissue are called as cartilaginous choristomas. It was first described by Berry in 1890. It is usually found in the distal extremities and rarely in the soft tissue of head and neck.¹³ Osseous choristoma is more frequently reported compared to cartilaginous and osseocartilaginous proliferation. The present case containing cartilaginous and osseous tissue in addition to adipose tissue that rimming the other components.⁸

According to the literature, one case of lipoosteocartilaginous choristoma was reported as a pedunculated nodule near the midline dorsum of the tongue, anterior to the circumvallate papillae.¹⁴ Differently, a lesion containing the cartilaginous, and bony tissue with fatty tissue predominance reported as variant of lipoma rather than choristoma.¹⁵

Differential diagnosis of the present study includes pleomorphic adenoma, neurofibroma, lipoma and granular cell tumor can be considered as it is a common lesion on the tongue. There was no epithelial component of pleomorphic adenoma that differentiating it from the present case of choristoma.

The tongue is the typical site for granular cell tumours. Females are mostly affected. It is commonly presented as a painless, well-demarcated, sessile submucosal swelling at the dorsum of the tongue similarly as the present case, but histologically; the granular cell tumor has a characteristic granular cell as a sheet in the lamina propria which is absent in the present case.

Neurofibromas and schwannoma (neurilemmoma) are two common peripheral nerve sheath neoplasms affecting children and adolescents. 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.

Conclusion

Choristomas of the tongue are benign lesions that are rarely reported but should be kept as a differential diagnosis of tongue lesions not only in posterior one-third. The diagnosis of lipoosteocartilaginous choristoma was made according to histopathological examination of excisional biopsy to exclude other benign lesions and malignancy.

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Conflict of Interest

The authors declare no conflicts of interest.

Patient Consent

Written informed consent was obtained from the patient.

Use of Artificial Intelligence

No artificial intelligence tools were used in preparing the manuscript.

CRediT Authorship Contribution

Rafa Elgehani: Surgery, provision of clinical photographs, and manuscript review and editing.

Amna Elgehani: Histopathological interpretation.

Najah: Blsat: Manuscript drafting.

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