Heterotopic salivary gland tissue: A report of two cases

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Abstract
The heterotopic salivary gland or salivary choristoma is defined as a tumor-like growth of otherwise normal salivary gland found in an abnormal location. In general, salivary gland tissue is not observed in the gingiva, with only isolated case reports having been found. The occurrence of the case in the midline, especially in the anterior two-thirds of the hard palate, has not been documented so far (PubMed search engine). It is apparent that these reported cases constitute a unique clinico-pathologic entity. Clinically, they appear as soft tissue tumors and are composed of lobules of salivary gland tissue.

Key words: Anterior two-thirds of the palate, choristoma, gingiva, heterotopic salivary gland tissue

INTRODUCTION
This article presents two case reports of heterotopic salivary gland tissue in the oral cavity. It emphasizes the unique nature of the site of presentation of these lesions.

CASE REPORTS
Case 1
A 45-year-old female was referred to our hospital for evaluation of an asymptomatic swelling of 6 months duration. The patient did not have any pain or discomfort other than occasional watery discharge from the lesion.

Clinical examination revealed a swelling on the labial gingiva in relation to the mandibular right canine and mandibular right first premolar. It was about 0.5 cm x 0.5 cm in size, pale yellow in color and firm in consistency.

Access this article online
Quick Response Code:
Website: www.jnsbm.org
DOI: 10.4103/0976-9668.82302

Diagnosis:
Heterotopic serous salivary gland of gingiva.

Case 2
An otherwise healthy 51-year-old male was referred to the Department of Prosthodontics for evaluation of an asymptomatic, multi-nodular swelling, in the midline of the palate. The patient claimed that it had been present since childhood. Clinical examination revealed multiple, well-defined nodules in the anterior two-thirds of the palate along and associated with the midline. An occlusal radiograph revealed a linear radiolucency in the midline of the palate displaced toward the left.
Figure 1: A small swelling in relation to the mandibular canine and the mandibular right first premolar

Figure 2: Intraoral periapical radiograph showing the absence of osseous involvement

Figure 3: Hematoxylin and eosin sections showing numerous salivary gland lobules composed of normal-appearing serous acini and ducts along with sparse chronic inflammatory infiltrate

Figure 4: Multiple well-defined nodules in the anterior two-thirds of the palate along and associated with the midline

Figure 5: Occlusal radiograph showing a linear radiolucency in the midline of the palate displaced toward the left

Figure 6: Hematoxylin and eosin section shows overlying stratified squamous epithelium and salivary gland lobules composed of normal-appearing, mucous acini, few ducts along with a sparse chronic inflammatory infiltrate in the deeper portion of the lamina propria
The microscopic examination from incisional biopsy revealed overlying parakeratinized stratified squamous epithelium and lamina propria. The deeper component of the lamina propria revealed salivary gland lobules. The lobules were composed of normal - appearing mucous acini, and few ducts and exhibited a sparse, chronic inflammatory infiltrate. [Figure 6].

**Diagnosis**

Heterotopic mucous salivary gland - anterior two - thirds in the midline of the palate.

**DISCUSSION**

Heterotopic salivary gland or salivary choristoma is defined as a tumor - like growth of otherwise normal salivary gland found in an abnormal location.[1]

In general, salivary gland tissue is not observed in the gingiva, with only isolated case reports having been reported.[2,3] To the best of our knowledge, this is the first case report of a salivary gland heterotopia in the midline, especially in the anterior two - thirds of the palate. (PubMed search engine).

The age distribution was indeterminable since few articles dealt with autopsy specimens and marrow samples.[1,4] A case of a 9 – year - old girl with gingival salivary gland choristoma in the maxillary incisor region has been reported.[2] Both our reported cases have been noticed in the middle age.

Although various mechanisms for the development and nature of salivary gland tissue heterotopias have been discussed, the precise pathogenesis of this phenomenon is still debatable. It is quite possible in both our cases that minor salivary gland tissue of palate and elements of the sub - mandibular gland may have been mechanically displaced during prenatal development and, subsequently, develop in the palate and gingiva.[2,3]

The most common clinical differential diagnosis for the swelling in the mandibular gingiva is a periapical abscess, cyst or granuloma. It could also be a mucocele. Clinically, periapical abscess, cyst or granuloma are symptomatic. Mucocele has a bluish hue. Radiographically, the mentioned lesions are radiolucent whereas in our case there was no radiolucency.

The clinical differential diagnoses for the multinodular swelling in the anterior midline of the palate are dermoid cyst, nasopalatine duct cyst or any palatally extending periapical abscess, cyst or granuloma. The nasopalatine duct cyst usually manifests as a fluctuant swelling in the anterior midline of the palate. Radiographically, the mentioned lesions are radiolucent, similar to our case. Our case could have been a – long - standing one, which would have been unnoticed due to it being asymptomatic, the duration of which caused the pressure resorption manifesting as midline radiolucency.

The tumorigenic potential of salivary gland heterotopias in these locations needs to be emphasized upon. It is apparent that these reported cases constitute a unique clinico - pathologic entity. Clinically, they appear as soft tissue masses and, histopathologically, they are composed of lobules of normal salivary gland tissue. Clinicians should be aware that such an entity as salivary gland heterotopias exists and should be considered as one of the differential diagnoses of soft tissue masses in the oral cavity. It also emphasizes the need for a proper diagnostic approach and the role of biopsy and histopathology in the diagnosis of such potentially tumorigenic lesions.

**ACKNOWLEDGEMENT**

The authors would like to acknowledge Dr. R. Karthigeyan for all his help.

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How to cite this article: Gheena S, Chandrasekhar T, Ramani P. Heterotopic salivary gland tissue: A report of two cases. J Nat Sc Biol Med 2011;2:125-7.

Source of Support: Nil, Conflict of Interest: None declared.