Lymphangioma in the Adult Submandibular Salivary Gland.

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Presented In

17\textsuperscript{th} International Congress on Oral Pathology and Oral Medicine  
25 – 30 May 2014  
Joint Meeting the  
British Society for Oral Maxillofacial Pathology  
Istanbul – Turkey  
2014
Lymphangioma in the Adult Submandibular Salivary Gland: A case Report:

Abstract:

Lymphangiomas generally are regarded as a benign hamartomastous lesion represent a developmental malformation of lymphatic vessels, rather than a true neoplasms. A 40 years old black woman was admitted to the clinics of Dental Faculty, Benghazi – University in Libya, complaining of pain especially during eating and a mass in the right submandibular region of the mandible.

Beside the history and clinical examination, CT scan (3D reconstruction) revealed a depression at the lower border of the body of the mandible anterior to the angle. Microscopic examination of the subsequent excisional specimen revealed normal lobules of submandibular salivary gland, within and between which were numerous large and small vascular spaces containing homogeneous, brightly eosinophilic material.

Although a rare case of cavernous lymphangioma has been reported in the parotid gland; but according to the best of our knowledge, this case is of interest in that it is the first case report of a primary cavernous lymphangioma within submandibular salivary gland in adult.

Key words: Lymphangioma, Cavernous, Submandibular gland,
Introduction:

Lymphangiomas are benign proliferative developmental anomalies of lymphatic system\(^1\). The nature of lymphatic malformations (LM) has sparked great interest since they were first described by Redenbacher in 1828. The head and neck region accounts for 40\% and 70\% of all lesions\(^2\), while the most common intr-aoral site of lymphangiomas is the tongue, which when so affected may be enlarged and referred to as macroglossia\(^3\).\(^4\).

Lymphangiomas have been classified into lymphangioma simplex (capillary lymphangioma), cavernous lymphangioma, and cystic lymphangioma (cystic hygroma)\(^4\).\(^5\). Lymphangioma histologically consists of numerous enlarged lymphatic vessels lined by flat endothelial cells. These channels usually are filled with protein-rich fluid with only a few blood cells\(^3\).

Cavernous lymphangioma is a benign congenital lesion that usually appears in childhood. It rarely presents in the adult and it is rarely found in the salivary glands\(^6\).\(^7\). However, Wiegand et al. (2011)\(^7\) state that of all the lymphatic malformations in the salivary glands, the parotid is the most common site.

This study presents and discuss the case of a cavernous lymphangioma in the submandibular salivary gland of an adult female.
Case Report:

A 40 years old African woman was admitted to the Dental Faculty, Benghazi- University Libya, Department of Oral Pathology, Oral Medicine, Oral Diagnosis and Radiology. Complaining of pain especially during eating and a mass in the right submandibular region of the mandible of insidious onset and had apparently increased in size without noticeable obvious symptoms referable to its presence.

On physical examination a non fixed well define submandibular salivary gland mass of about 5.0 x 4.0 cm oval in shape located beneath and anterior to the angle of the mandible, soft fibrous, non-fluctuant, non tender, no any changes in secretion of saliva or in its size during meal time and there was no sings or symptoms of inflammation and during palpation there was depression of the right lower border of the body of the mandible. The overlying skin was normal (Figures 1a and b).

The CT scan (3D reconstruction) revealed clear smooth depression at the right lower border of the body of the mandible (Fig- 3a and b). The radiographical view shows that, the dent extended from the first premolar until the angle of the mandible with lifting up the mandibular canal (Fig. 2a, b and c).

The provisional clinical diagnosis of salivary gland tumour was made.

Microscopic examination of the subsequent excisional specimen with scarified the submandibular salivary gland revealed normal lobules of submandibular salivary gland, within and between which were numerous large and small vascular spaces containing homogeneous, brightly eosinophilic material (Fig. 3a and b). The lymphatic spaces were lined with a single layer of flattened endothelium with underlying walls which varied in thickness depending on the amount of fibrous tissue present. Focal aggregates of lymphocytes were noted in the walls of some of the vascular channels (Fig. 3c). There was no evidence of inflammation or any salivary neoplastic cells, this ruled out inflammation and any other salivary gland neoplasms. The histopathological diagnosis was cavernous lymphangioma of submandibular salivary gland.
Fig. 1a and b: Photographs of the patient’s face shows a swelling in the right submandibular region, located beneath and anterior to the angle of the mandible.
Fig. 2a: Radiographic view shows an extensive depression of the lower right border body of the mandible with superior displacement of the inferior alveolar canal.
Fig. 2b and c: CT-scan (3D construction) shows an extensive dent of the body of the mandible at the level of the lower border in the right submandibular gland region.
Fig. 3a and b: Histopathological views at different powers show small and large lymphatic vascular spaces lined by flat endothelial cells, containing homogenous bright eosinophilic material between normal salivary cells and presences of focal aggregation of lymphocytes around some of these vascular channels in b.
Fig. 3c: Lymphangioma in this section shows some presences of focal aggregation of lymphocytes and variation in the thickness of the walls around some of these lymphatic vascular channels.
Discussion:

Lymphangiomatous lesions are rare congenital malformations of the lymphatic system and most commonly seen during an infancy and childhood periods where in this case the lesion was present in a 40 years female patient.

However occurrence of lymphangioma in the salivary glands are so rare, Wiegand et al. (2011)\(^7\) mentioned that cases of salivary gland involvement of lymphatic malformations have been occasionally reported in the literature and he presented four patients suffered from lymphatic malformations limited to the parotid gland, also Tariah et al. (2000)\(^8\) presented a case of cavernous lymphangioma of adult parotid gland in a non-Caucasian female, but to the best of our knowledge our case is of interest that is the first case report of a primary cavernous lymphangioma within submandibular salivary gland of an adult.

According to the clinical features of this case, we first thought of salivary neoplasm or inflammatory-obstructive lesions, and dependent on the presents of this lesion at an adult age, we felt and considered the developmental malformation as the least expected diagnosis, so we confirmed the diagnosis as a cavernous lymphangioma by histological examination.

The separation of cavernous lymphangioma from cystic hygroma is vaild only in the clinical sense, but they are histologically very similar\(^9\).

There is no encapsulation of lymphatic vessels, even with the tumors which appear well circumscribed clinically, because of the non-encapsulated and "infiltrating" nature of the lymphangioma, complete removal is often inadvisable\(^11\). So; for that the present case treated by surgical removed of submandibular salivary gland to avoid recurrence.

The pathogenesis of the present case could be correlated with the hypothesis that, during embryogenesis, lymphatic tissue lays in the wrong area.
Conclusion:

- To the best of our knowledge, up to now there is no reported data in the literature concerning the presentation of a lymphangioma as a primary submandibular salivary gland malformation.

- Lymphangioma rarely presents in the adult but may be diagnosed at this late stage due to the slow-growing nature of the tumour.

- It is rarely found in the salivary glands and when it does occur, the gland is usually incorporated by lymphangioma of surrounding tissue.
References:


