

Adenomatoid Hyperplasia of The Oral Cavity: A Diagnostic Dilemma

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Abstract

This review study presents literature review and discusses the clinical significance of Adenomatoid Hyperplasia, a commonly misdiagnosed lesion. This rare entity has been seldom presented and is not well enough described in the literature. Only 15 reports with 95 cases could be attained during the online literature search using the keywords: Adenomatoid, Tumor, Hyperplasia, Minor salivary Glands. Data revealed a tendency towards the male gender. Age distribution of patients did not reveal a tendency towards a specific age group but presented a peak incidence in the 4th and 5th decades. Location data revealed a tendency towards the palate, especially the hard palate. Most of the presented cases were asymptomatic and the most common initial diagnosis made was salivary gland tumor. It was concluded that, adenomatoid hyperplasia of the oral cavity may resemble a wide range of pathologies and in order to differentiate and to achieve a correct diagnosis, histological evaluation is fundamental.

Key words: Adenomatoid Hyperplasia; Adenomatoid Tumor; Hyperplasia; Minor salivary Glands

Introduction

Minor salivary glands, which is responsible %5 of daily saliva secretion, are found on the walls of the oral cavity and named as labial, buccal, palatal, lingual, minor sublingual, palatoglossal and Ebner glands. Labial glands are found between the oral mucosa and orbicularis oris muscle of lips. Their secretion is directly drained into the oral vestibule. Buccal glands are located between the mucosa lining of cheeks and buccinator muscle. Some of these glands are found around the opening of the parotid gland and sometimes visible are called molar glands. These glands are mixed mucous and serous glands. There has been a wide variety of pathology related to minor salivary glands in which Adenomatoid hyperplasia (AH) is one these pathologies.^{1,2}

AH has been initially reported by Giansanti et al. in 1971.³ Till today AH remain to be an uncommon hyperplastic lesion; comprising neoplasm-resembling lesions.⁴ The etiology of AH is unknown; although it is thought that chronic local trauma

may have a role.⁵ It usually presents as bluish or reddish, asymptomatic masses or elevated nodules.⁶ It may occur in all salivary gland-bearing areas of the body; in addition, reported cases concerning the oral cavity present the palate as the preferred site of occurrence.⁷ Histologically it presents no abnormality other than an apparent hyperplasia of the salivary gland tissue and after adequate surgical excision of this benign lesion recurrence is not expected.⁵ Its resemblance to pleomorphic adenomas and salivary gland tumors makes the lesion clinically important.⁴

Literature Review

The local IRB granted exemption in writing, due to the study's retrospective nature. Data collection was made in PubMed, Science Direct and Google Scholar databases with the keywords

'adenomatoid', 'adenomatoid hyperplasia', 'Salivary gland', 'Minor salivary gland' keywords. Articles only in English language and reports on humans have been selected for the study. Since 1971, to the best of our knowledge, only 15 case reports, presenting 95 individual cases have been published in the English-language literature (Table 1).³⁻¹⁷ A new case has also been added to previously existing data. The review revealed 24 papers, of these papers, 8 could not be fully accessed¹⁸⁻²⁴, one was related to the subject 'Adenomatoid Ductal Proliferation'²⁵ and one involved macaque monkeys.²⁶

Adenomatoid hyperplasia of the minor salivary glands is an uncommonly reported entity in literature. The majority of cases were presented from the USA (55 cases), followed by the UK (21 cases), Colombia (12 cases), Japan (3 cases), Turkey, China, India, Israel (1 case). Nevertheless, a great number of literature in Japanese was also present in literature.

Gender distribution revealed patients to be 39.58% female (38 patients) and 59.37% male (57 patients); for 1 patient (1.04%) gender data was not available. This data revealed a tendency towards the male gender concerning AH.

Features of the Lesion

Age distribution revealed patients to be 1% between the age range of 0-9, 4.3% between the age range of 10-19, 8.3% between the age range of 20-29, 19.8% between the age range of 30-39, 18.7% between the age range of 40-49, 13.5% between the age range of 50-59, 16.7% between the age range of 60-69 and 2% between the age range of 70-79. For three patients (3.1%) age data was not present. For the remaining 12 patients (12.5%) presented by Campos et al. (13), age data was not present for each individual case but the age range for cases was explained as 26-67 years with a mean of 47 years. This data revealed a higher frequency in the in the 4th, 5th, 6th and 7th decades of life compared to other age groups. Although this data did not reveal a tendency towards a specific age group it presented a peak incidence in the 4th and 5th decades.

Data concerning locations of the AHs revealed the lesion as 6.3% in the retromolar area, 71.9% on the palate (presented as 4.2% palate, 15.6% soft palate, 33.3% hard palate, 18.8% hard-soft palate junction), 4.2% on the buccal mucosa, 1% on the upper labial mucosa, 2% on the lower labial mucosa and 13.5% in the sublingual gland. This data revealed that AH had a tendency towards locating itself on the palate, especially the hard palate.

Differential Diagnosis

Among the presented cases, the most common initial diagnosis made was salivary gland tumor involving benign pleomorphic adenoma and adenoma, and malignant mucoepidermoid carcinoma and adenoid cystic carcinoma (21 cases). Salivary gland tumors occur much more commonly in the major glands (80-85%) and the majority is of benign nature (75-80% for major glands, 50-55% for minor glands).²⁷ Pleomorphic adenoma (PA) is a benign, mixed and most commonly encountered salivary gland tumor. It is a slow-growing, painless mass, commonly occurring in the 5th decade of life with a predilection to the female gender. Although the most common localization of PA is the major salivary glands, especially the parotid gland (>80%), it may also appear in the minor salivary glands with the palate being the most common intraoral site.²⁷ With its clinical features very much resembling to AH, it was the most common initial diagnosis made among salivary gland tumors (11 cases).

When clinical impressions were evaluated, it was observed

that other differential diagnosis of AHs involved fibroma, acinic cell tumor, cylindroma, fibroepithelial polyp, lymphoma and lymphangioma, denture hyperplasia, mucocele, and neurofibroma and neural tumor.

Although some cases referred with pain and tenderness, most of the presented cases were asymptomatic. Data of smoking habits and present dentures were not present for most cases.

As result of the present literature review it was concluded that AH of the oral cavity is a rare entity mostly occurring in male gender at the 4th and 5th decades of life, commonly presenting as asymptomatic masses and have a predilection to be localized at the palate. With these features, they may resemble a wide range of benign and malign pathologies. In order to differentiate the lesion from these wide ranges of pathologies and to achieve a correct diagnosis, histological evaluation is fundamental.

Author Contributions

All authors have contributed to; conception and design of the study, data collection and analysis, writing the manuscript, approval of the final version to be submitted.

Conflict of Interest

Authors declare no Conflict of Interests for this article.

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