Section A-Research paper
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A case report of Benign mucinous cystadenoma of left Angle of Mandible

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ABSTRACT:

Mucinous cystadenoma is considered as a subtype of a rare benign salivary gland tumor, cystadenoma. Adenomatous proliferation of the epithelium is seen in cystadenomas of the salivary glands which is characterized by development of multiple cystic structures. Various morphologic variants of cystadenoma have been mentioned out of which mucinous and papillary type are important. It is very challenging to clinically differentiate it from cases of mucous retention and different benign minor salivary gland tumors. Salivary gland disorders have become a subject of extensive research due to its diverse patterns of presentation. This case report describes a benign mucinous lesion of salivary gland on the left angle of the mandible.

KEYWORDS: benign tumor, cystadenoma, mandible, salivary gland

INTRODUCTION:

Salivary gland tumors include a variety of diverse histologies and sites, such as parotid, submandibular gland, sublingual gland and minor salivary glands of the oral cavity. Although majority of these neoplasms (80%) are benign, they vary in their propensity to recur and/or develop into malignant tumors [1][2]. As a result, the right diagnosis is crucial in determining the best course of action. The most commonly found benign subtypes are pleomorphic adenoma (PA), Warthin's tumor (WT), and myoepithelioma (MYO), followed by rarer histologies

including lymphadenoma (LA), sebaceous adenoma (SA), cystadenoma, oncocytoma (OC), sialadenoma papilliferum (SP), ductal papilloma (intraductal and inverted), basal cell adenoma (BCA), and canalicular adenoma (CA)[2][3].

Salivary gland cystadenomas are benign tumors in which the epithelium exihibits adenomatous growth, which is characterized by the development of many cystic structures. Papillary and mucinous cystadenoma are two significant morprhological forms of cystadenoma that have been reported. According toWHO papillary cystadenoma can be described as "a tumor that closely resembles Warthin'sTumor but without the lymphoid elements, constituting multiple papillary projections and a greater variety of epithelial lining cells."If the population of lining epithelial cells is dominated by mucinous cells, the tumor is referred to as mucinous cystadenoma.[4]

A cystadenoma is an uncommon benign tumor that develops on the salivary gland duct epithelium and displays unicystic or multicystic growth as well as focal intraluminal papillary proliferation of the lining epithelium. [5] It demonstrates as a rare, slow growing, asymptomatic slightly compressible swelling, which most frequently affects minor salivary glands.[4][5] It rarely grows larger than 1.5cm in diameter. It makes up between 0.8% and 6.3 % of benign small salivary gland tumors. [6] Lips and buccal mucosa are common site of occurrence, as opposed to other minor salivary gland tumors, which typically arise on the palate. This is more prevalent in older age group and usually occurs in the eighth decade of life. It is more common in women than in men (2:1). [6][7]

No significant numbers of cystadenomas with follow up data have been documented. Low probabilities of recurrence exist. A minimally invasive surgical technique guarantees total eradication. [6] Recurrence may happen if the slow growing cyst adenoma is not completely removed or if a cystadenocarcinoma is misdiagnosed. For the lesions of oral mucosa, the use of a carbon dioxide laser in oral and maxillofacial surgery has become standard. The carbon dioxide laser has been found to have benefits for oral soft tissue surgery, including minimal injury to surrounding tissue, good hemostasis, less inflammatory reaction, scar formation, and precise cutting. [8]

CASE REPORT

A 22 year old female patient reported with an asymptomatic swelling in the left lower third region of the face. The swelling was present since 7 months and gradually increased to its present size. On inspection, a well defined, ovoid mass, approximately 3cm in diameter was seen on the region of left Angle of Mandible. The overlying skin appeared to be normal. On palpation, it was soft to firm in consistency, non-fluctuant, non-tender and slightly compressible. There was neither sinus opening, nor any relevant intra-oral hard or soft tissue findings present.

A provisional diagnosis of benign minor salivary gland tumor was considered based on the clinical examination. Dentigerous cyst, residual cyst, retention phenomenon, a low- grade minor salivary gland malignancy {acinar cell adenocarcinoma (AcAC), adenoidcystic carcinoma, mucoepidermoid carcinoma}, lipoma and neurofibroma were considered as differential diagnosis.

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Patient was advised for intra-oral periapical radiograph of the left third molar region, to find the presence of any odontogenic cause. She was also investigated with cone beam computed tomography (CBCT), which demonstrated bony erosion present on the left angle of mandible and no tooth anamoly was detected. USG (ultrasonography) guided fine needle aspiration cytology was performed. Smear received showed few scattered epithelial cells and stromal fragments with spindle cells on the background of mucinous material and red blood cells. Before referring the patient for oral surgery, routine hematological investigations were indicated.

Excisional biopsy targeting the mass was carried out under local anaesthesia through the extra-oral periangular approach. The diagnosis of benign mucinous cystadenoma was made after a histopathological evaluation, which revealed epithelial call lining a cystic cavity with mucous discharges. Patient was asked to be under follow-up for reporting of any recurrence.

DISCUSSION

Cystadenoma is a rare benign neoplastic proliferation of salivary gland duct epithelium, and encompasses 0.4% to 1.2% of all salivary gland tumors. [9] In 45% to 60% of instances, it is discovered in the parotid gland, but it can be found intra-orally in minor salivary gland tumors.[10] According to Alexis and Dembrow, 4.7% of benign epithelial salivary gland tumors and 2% of minor salivary gland tumors are cystadenomas.[11] Out of 800 intraoral minor salivary gland tumors, Chaudhary et al. observed 2% incidences of cystadenoma. [12] Waldron et al. observed higher incidence rates of minor salivary gland tumors and benign epithelial salivary gland tumors, respectively, of 4% and 8.1%. [13]

According to Bauer and Bauer, the undifferentiated epithelium of the intercalated ducts of the salivary gland is where the cystadenoma develops. [14] They are distinguished by multiple cystic developments inside a stroma of fibrous connective tissue. The less common mucinous type lacks clearly discernible papillary projections and mostly displays mucous cells in the epithelial lining of the cystic lumens.

Patients between the ages of 17 and 86 have been documented to develop cystadenoma, with a mean age of about 61 years. [15] It is to occur more commonly in 8^{th} decade of life. Females are more prone to develop cystadenomas as compared to males with a ratio of 2:1. [4]In our study, the gender and age of the patient was within the range in accordance with previous records.

Most common site of occurrence are lips, palate, buccal mucosa and tonsillar area.[4] The widespread distribution of minor salivary glands can be used as an explanation for the prevalence of mucinous cystadenoma in subcutaneous layer of the cheek. As in this case, it was found to be present in the lower third region of the face. Mucinous cystadenomas can be found in the oral cavity, sinonasal cavity, throat, middle ear cavity and lungs. Additionally, these are also present in buccal mucosa, labial mucosa, palate and submucosa of the aerodigestive tract. [16][17][18]

Due to lack of distinguishing clinical characteristics, cystadenoma may be difficult to distinguish from other benign salivary gland tumors, extra-vasation /mucous retention phenomenon, along with low-grade minor salivary gland malignancies. Cystadenoma diagnosis is solely histological. The histological characteristics that have been mentioned in this case were typical and generally

in agreement with what have been given in literature.Bearing in mind the differential diagnosis, intra ductal papilloma, Warthin's tumor, low grade mucoepidermoid carcinoma, cystadenocarcinoma, and salivary gland tumors must be considered because of wide range of histomorphological characteristics of the salivary gland tumors.[7][8]

CONCLUSION

This case highlights how difficult it can be to diagnose rare salivary gland tumors. It is due to its varied morphological and histological features. The prognosis of cystadenoma is good and recurrence rate is low. After complete surgical excision, cystadenoma does not reoccur, although there have been a few instances when cystadenocarcinoma has grown from an earlier cystadenoma. To prevent recurrence and malignant transformation, it is advised to undertake a full excision and make a precise histologic diagnosis.[16] As far as the authors are aware, there are extremely few examples of the mucinous type of cystadenoma. The purpose of this case report is to provide a documentation of the uncommon mucinous cystadenoma.

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