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Case Report

Mucinous Cystadenoma of Palate: A Rarecase Presentation

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ABSTRACT:
Cystadenoma is an uncommon, non-lymphoid benign salivary gland tumor that chiefly originates in the minor salivaryglands as a cystic growth with papillary projections into the cystic lumen. WHO has classified it into two histopathological variants, viz. papillary and the mucinous forms ofcystadenoma.Mucinous cystadenoma is very rare when compared to papillaryvariant. Clinical presentation of this entity mimics mucous retention phenomenon and other common benign salivary gland neoplasms and often poses a diagnostic challenge. This article reports a caseof mucinous cystadenomain a 56-year-old female patient who presented with an asymptomatic swelling on the hard palate of four years duration.
Keywords: Benign tumor, hard palate, minor salivary gland tumor, mucinous cystadenoma

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INTRODUCTION

Tumors of salivary glands constitute a heterogeneous group of lesions of great morphologic variation.Annual incidence of salivary gland tumors around the world is 1-6.5 cases per 100,000 people[1]Minor salivary gland tumours (MSGTs) represent 9–25% of all salivary gland tumours[2] as well as 2-3% of all head and neck tumours[3] with a female preponderance. Cystadenomas of salivary glands are benignneoplasms, in which adenomatous proliferation of epithelium is demonstrated that is characterized byformation of multiple cystic structures. Severalmorphological variants of cystadenoma have been described of which papillary and mucinous cystadenoma are important. World Health Organization (WHO) described papillary cystadenomawhich closely resembles War thins tumor, but without the lymphoid elements. If mucous cells predominate in the cell population of the lining epithelial cells, the tumor is termed as mucinous cystadenoma.[1]The most frequent clinical finding of salivary gland cystadenoma is a painless mass beneath the mucosa of the palate, lip or buccal mucosa .[4]Size rarely exceeds 1.5 cm in diameter. Cystadenoma most commonly manifests in eighth decade of life with a mean age of occurrence of 57 years with a female predilection .The majority of cystadenoma cases are treated by simple excision, and recurrence is extremely rare.[5]

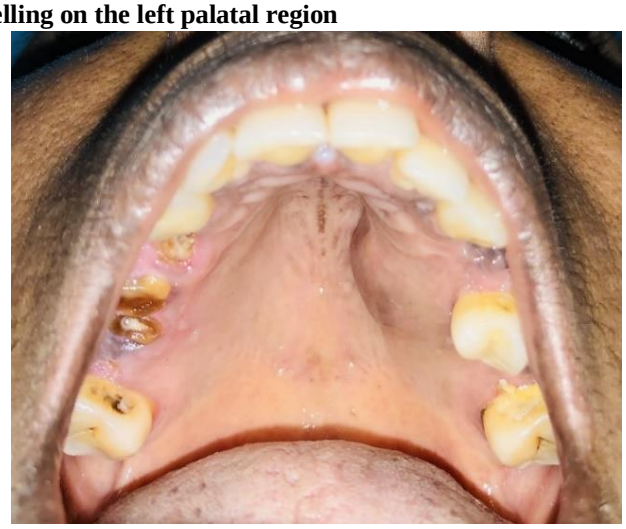
CASE REPORT

A 56-year-old female patient came to the outpatient department of Oral Medicine and RadiologyGovernment dental college, Kottayam with a chief complaint of swelling in the left side of roof of mouth in the past4 years.Swelling was asymptomatic and was not associated with pain or pus discharge.It was initially small and gradually increased and reached the present size.Patient stated about undergoing extraction of decayed tooth adjacent to the swelling one year before. Recently, a physician whom she consulted for head ache, observed the swelling on palate and was referred to Government Dental College Kottayam for further evaluation and management.

Shewas under medication for dyslipidemia for last 3years.Her family,social and personal history were unremarkable.Her vital signs were with in normal limits.

Extra oral examination did not show any significant alterations. On intra oral examination a well-defined solitary ovoid shaped nodular swelling of size approximately 1.5 x1.2 cm was seen on the left side of the hardpalate in relation to maxillary canine and premolars.Surface of the swelling was smooth with no secondary manifestations or visible pulsations.On palpation, the swelling was non-tender, non-pulsatile and firm in consistency.Conditions considered in the differential diagnosis included pleomorphic adenoma, residual cyst and benign connective tissue neoplasms, viz.fibroma and schwannoma.

Figure 1: intra orally swelling on the left palatal region



Panoramic radiograph did not show significant bony alteration in the area of the swelling. Computed tomography scan in soft tissue window setting showed a well-defined, oval shaped hypodense lesion of size approximately 1.8x1.1cm on left side of the hard palate.No evidence of erosion or destruction of adjacent bone were evident.Aspiration from lesion yielded purulent aspirate,however, culture did not yield any bacterial growth.Routine haematological investigation results were within normal range. Patient underwent excisional biopsy of the swelling under local anaesthesia.

Figure 2: coronal CT showing softtissue mass on the left palatal region

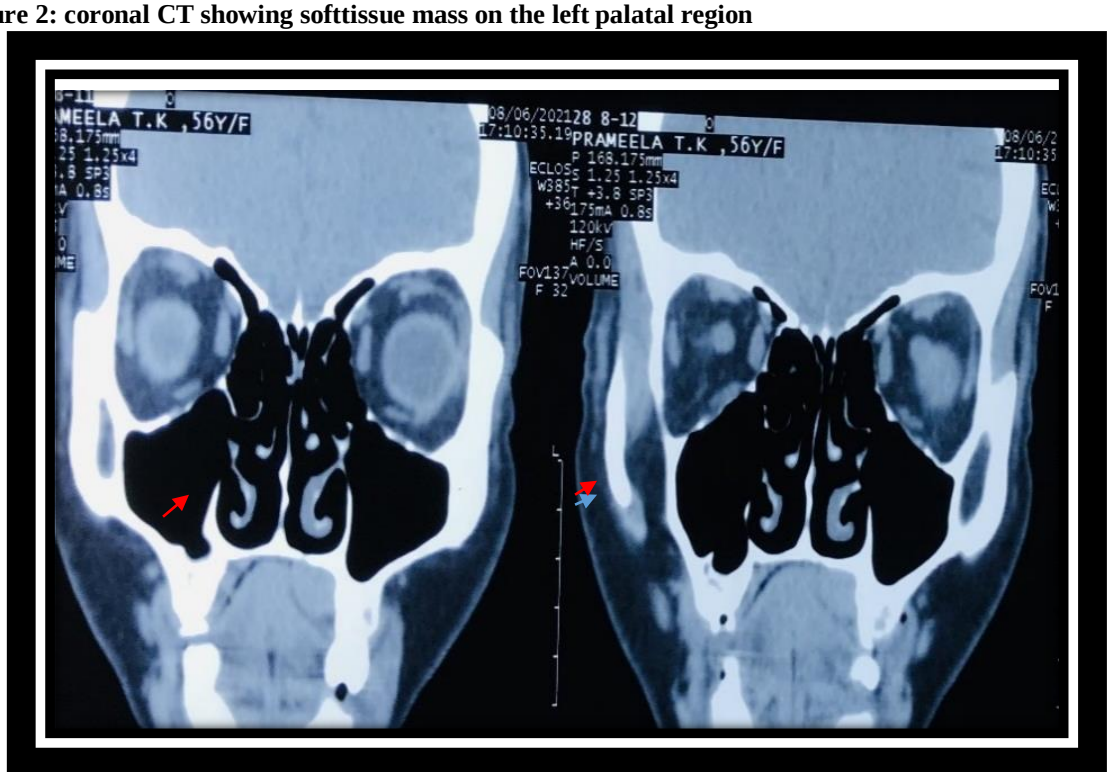
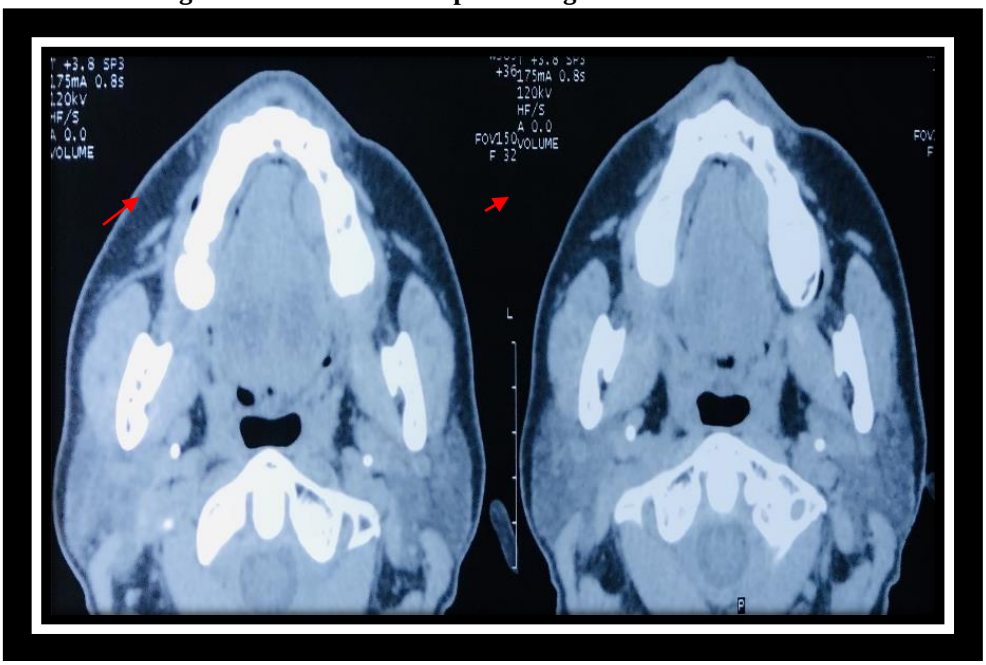
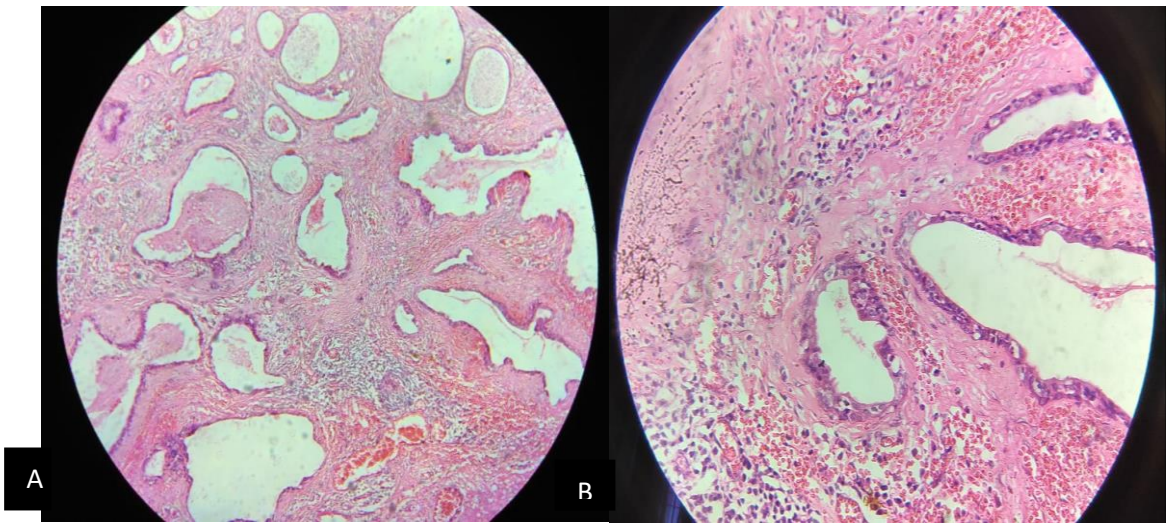


Figure 3: axial CT showing soft tissue mass in left palatal region.



Histopathological examination revealed moderately fibrous connective tissue stromainterspersed withcystic spaces.Some cystic spaces were lined by single layer of flattened epithelial cells along with few mucous cells. Cystic spaces were filled with eosnophiliccoagulam. Connective tissue stroma exhibited dense chronic inflammatory infiltration, chiefly lymphocytes and plasma cells.Microscopic features were suggestive of mucinous cystadenomaand patient is under regular follow up.

HISTOPATHOLOGY



A.H &E Staining (x10) B. (x40) showing fibrous stroma interspersed with cystic spaces lined by single layer flattened cells,multilayered epithelial cells along with mucous cells .Cystic spaces filled with eosnophiliccoagulam.

DISCUSSION

Salivary gland cystadenomas, especially, in palate are extremely rare, but are common in many other sites including the ovary, biliary tree, the appendix, the epididymisand the pancreas.[6] Mucinous cystadenomas of ovary are among the largest tumors known. They account for about 15 % of all ovarian neoplasms.[7]In the first edition of the World Health Organization Histological Classification of Salivary Gland Tumorscystadenoma of the salivary gland was first subclassified into various types of monomorphic adenoma. In the second edition, which was published in 1991, cystadenomas were more clearly defined as a specific histopathological entity that was further

subdivided into papillary and mucinous types.[8]However, in the third classification published in 2005, cystadenomas were only subdivided into papillary and mucinous types. Chaudhry *et al*. reported2% cases of cystadenomas out of 800 intra oral minor salivary gland tumors studied.

Clinically it is usually presented as a slow growing asymptomatic swelling.As such, cystadenoma presents no distinct clinical features and may be impossible to differentiate clinically from other, mucous retention or extravasation phenomenon,benign salivary gland tumors and low-grade minor salivary gland malignancies such as papillary cystic variant (PCV) of adenoid cystic carcinoma and mucoepidermoid carcinoma.[9]Cystadenomas of the major salivary glands are usuallypainless, slow-growing swellings, whereas those of the minor salivary glands are often compressible and mayappear as mucoceles. Initially this lesion was consideredas a type of ductal hyperplasia and later after a careful study of the histopathological features, it was classified asa benignneoplasm.[10]Differential diagnosis must include mucocele, other benign salivary gland tumours like pleomorphic adenoma andconnective tissue tumours like fibroma, lipoma, and Schwannoma.Palatal cystadenomas may mimic odontogenic infections and cysts like residual cyst and dentigerous cyst. Hence, when cystadenoma occurs on the palate, such as in our case, it must be differentiated from radicular/ residual cyst and palatal space abscess.A possible odontogenic origin must be ruled out by appropriate clinical and radiographic examinations.

Bauer and Bauer suggested that the cyst adenoma arises principally from the undifferentiated epithelium of the intercalated ducts of the salivary gland.[11] They are characterized by multiple cystic growths within a fibrous connective tissue stroma. The papillary variety of cyst adenoma exhibits papillary proliferations that project into the cystic lumens and may closely resemble the Warthin's tumor with the absence of the lymphoid element. The less frequently seen mucinous variety predominantly shows mucous cells in the epithelial lining of the cystic lumens, with absence of distinctly visible papillary projections. Due to the abolition of these variants, the rare appearance of mucous cells in cystadenoma was documented in the 2017 WHO classification.

Histopathological differential diagnosis includes ductal ectasia, Warthintumour, and low-grade mucoepidermoid carcinoma. Ductal ectasia occurs secondarily to salivary obstruction and is characterized by oncocytic metaplasia of the epithelium. Warthintumour has a pronounced lymphoid stroma and bilayeredoncocytic epithelium, whereas in cystadenoma, the epithelium is usually not bilayered and lacks lymphoid stroma. The low-grade mucoepidermoid carcinoma is prominently cystic. Its salient features are infiltrative borders and noncystic epithelial proliferation.Cystadenocarcinomas are morphologically similar to cystadenomabut its solid growth pattern in focal areas with sporadic cellular atypia,mode of invasion, infiltration of the glandular parenchyma, glandular lobe architecture breakdown along with permeation into adipose and muscle tissues make it delineate from cystadenoma. [11]Immunohistochemically, the positive staining of papillary cystadenoma for pan cytokeratin and S100 suggests its epithelial origin and rules out other tumorsof lymphoid, vascular, and endothelial origin. It stains negative for smooth muscle actin (SMA) and rules out the possibility of myoepithelial carcinoma.[6]Most cases of cystadenomas, including our case, are treated by simple surgical excision. Follow up of the patient is necessary since recurrences, though very few, have been reported.

CONCLUSION

Very few cases have been reported of mucinous variety of cystadenomaintra-orally.This variant mimic mucous retention phenomenon and other common benign salivary gland neoplasms and often poses a diagnostic challenge to the clinician. Hence, a thorough clinical examination followed by appropriate investigations is mandatory for accurate diagnosis and management of the condition.

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