A Rare Case Report of Mucinous Cystadenoma with Immunohistochemical Analysis and Review of Literature

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Abstract Cystadenomas are rare salivary gland tumours characterised by prominent epithelium-lined papillary projections into the cystic spaces. A 37 year-old female, presented with a swelling on the right side of the hard palate. Clinically, the swelling was soft, fluctuant, sessile and non-tender measuring 2.5×2 cm in diameter. Excisional biopsy was performed. The histopathological and immunohistochemical features were characteristic of mucinous cystadenoma. There has been no recurrence over a 2-year follow up period. The article highlights its clinical, histopathological, immunohistochemical features and differential diagnosis along with review of literature. Cystadenoma of the palate is rare, papillary variety being more common than mucinous with very few cases reported in literature.

Keywords Cystadenoma · Minor salivary gland · Mucinous · Papillary · Immunohistochemistry · Treatment

Introduction

Salivary gland neoplasms represent <1 % of all tumours and 2–6.5 % of all head and neck neoplasms [1]. Minor salivary gland tumours are infrequent accounting for <25 % of all salivary gland neoplasms [1, 2].

Cystadenoma of the salivary glands is a rare benign neoplasm in which the epithelial proliferation is characterized by the formation of multiple cystic cavities containing intraluminal papillary projections [3].

The earliest published reference to the minor salivary gland lesion was made by Skorpil [4, 5] in a paper entitled ‘Papillary cystadenoma of the major and minor salivary glands.’

The Armed Forces Institute of Pathology (AFIP) files contained [1] 96 cases of cystadenomas which constituted 0.7–8.1 % of all benign salivary gland tumours, 7 % of all minor salivary gland tumours and 3.1 % of major salivary gland benign tumours. Almost 58 % of these tumours occur in the parotid gland, 6.6 % occur in the submandibular gland, 0.5 % in the sublingual gland, 10.7 % on the lips, 8.2 % on the cheeks, 7.1 % in the palate and 9.1 % in other intraoral sites [3, 6, 7]. A study by Waldron et al. reported buccal mucosa as the most common site (35 %) followed by the palate (20 %) [9].

In the largest series of intra oral minor salivary gland tumours reported, cystadenomas constituted 6.3 % of all minor salivary gland tumours (benign and malignant) and 10.7 % of benign tumors. Palate was the most common site (50 %), followed by the buccal mucosa (29.2 %), tongue (8.3 %), and one case each in upper lip, lower lip and floor of mouth [9].

Cystadenoma most commonly manifests in eighth decade of life with a mean age of occurrence of 57 years. There is female predilection in the ratio of between 2:1 and 3:1 [3].

Cystadenoma typically presents as a slow growing, painless mass, usually <1 cm in greatest dimension when arising in the minor salivary glands [10] with one case reporting 1.5 cm in its maximum diameter in buccal mucosa [11].

Clinically they are asymptomatic and appearance is similar to mucocele. Surgical excision is the indicated treatment [10].

Here we report a case of mucinous cystadenoma on the palate, with special emphasis on the histopathological

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Fig. 1 Intraoral view of lesion on hard palate

features, immunohistochemistry and differential diagnosis of this tumour.

Case Report

A 37 year-old female, was referred to the Department of Oral and Maxillofacial Surgery, by a local dentist due to the presence of an asymptomatic swelling in the right side of the hard palate since 3 years. Patient also gave a history of pain in the upper right back tooth since 3 months. History revealed extraction of 17 and 18 done 3 years back due to caries and no treatment was done for the swelling on the hard palate. Past medical history was non-contributory.

The intraoral examination revealed a solitary swelling of size 2.5×2 cm in the right side of the hard palate, extending anteroposteriorly from the distal aspect of 14 to distal aspect of 16. Medio laterally the swelling extended 1 cm from midline to the mucogingival junction of the posterior teeth (Fig. 1). The overlying mucosa was intact and showed a bluish hue at the distal aspect of the swelling. On palpation swelling was soft, fluctuant, sessile and non-tender. Pulp vitality testing was done which revealed 14 as non-vital and 15, 16 as vital.

No cervical lymphadenopathy was observed. Routine blood investigations were within normal limits.

CT scans revealed no erosion of bone in the region of hard palate (Figs. 2 and 3).

Based on the history and clinical features, it was provisionally diagnosed as a benign salivary gland cyst/neoplasm.

Complete surgical excision with a 5 mm safety margin was performed under general anaesthesia (Fig. 4). The greater palatine neurovascular bundle was clamped before delivery of the specimen. After excision, the raw surface

Fig. 2 Axial view of contrast CT scan showing no bony involvement

Fig. 3 Coronal view of contrast CT scan showing no bony involvement

was covered with buccal fat pad and collagen-based dermal substitute (Fig. 5). The postoperative course was uneventful. Follow up was done for 2 years (Figs. 6 and 7) with no evidence of recurrence.

Preparation of Specimen for Histopathological Examination

The resected specimen was fixed in 10 % neutral buffered formalin solution, sliced at intervals of 5 mm, and

Fig. 4 Defect on the hard palate after complete surgical excision of the lesion

Fig. 5 Defect covered with buccal fat pad and collagen-based dermal substitute

embedded in paraffin wax. Multiple sections (3 mm thick) were cut and prepared for haematoxylin and eosin, and immunohistochemical staining. Immunohistochemical staining was performed with polymer super sensitive kit. The primary immunohistochemical markers used and their reactivity and distribution are listed in Table 1.

Histopathology and Immunohistochemistry

Macroscopically, the specimen was greyish in colour and soft in consistency.

Fig. 6 Followup after 1 year

Fig. 7 Followup after 2 years

Table 1 Immunohistochemical markers used and their features

Antibodies	Source	Dilution rate	Reactivity and distribution
SMA	Biogenese	1:100	Cytoplasmic reactivity in myoepithelial cell layer
CD 117	Biogenese	1:100	Negative
Calponin	Biogenese	1:100	Cytoplasmic strong positivity in outer and inner layers of epithelial components
CK-8	Biogenese	1:100	Patchy positivity in outer and inner layer confined to cytoplasm
S-100	Biogenese	1:100	Patchy positivity in outer and inner layer confined to cytoplasm

Fig. 8 Multiple cystic spaces filled with pas positive mucin

Fig. 9 Hematoxyline and eosin stained sections showed cystic spaces lined by cuboidal cells and hematoxyphilic material within the cystic space is suggestive of its origin (glycosaminoglycans or mucopolysaccharides)

Microscopically, large multiple cystic spaces of variable size and shapes arranged in haphazard manner was seen. Spaces were lined by single layer of low cuboidal epithelial cells (Figs. 8, 9). Central cystic cavity was filled with amorphous, basophilic material exhibiting mucinous material (Fig. 8). Papillary proliferation is not evident in the present sections. Tumour stroma was fibrous with mild inflammation foci. Capsule was ill-defined but tumour was well-delineated. The surrounding margins were clear. Periodic Acid Schiff (PAS) stain showed positivity for glucose amino glycans, mucins and mucopolysaccharides. PAS stain in the extracellular mucinous material within cystic spaces indicates its origin as a secretory glycos amino glycans of salivary glands (Fig. 9).

Immunohistochemically, expression of pan- cytokeratin in tumour parenchyma is suggestive of origin of tumour tissue from epithelial or epidermal components. Cytokeratins including CK-8 are intermediate filaments expressed in cells of epithelial origin. Mucinous cystadenoma is a proliferation of epithelial and ductal cells of salivary gland. Intense cytokeratin intermediate filament expression was consistently observed (Fig. 10).

Fig. 10 Pancytokeratin: strong cytoplasmic immunoreactivity noted in all tumor cells

Fig. 11 Cd 117: negative in all tumor cells

Smooth muscle actin (SMA) is expressed in myoepithelial cells of salivary gland. It is also expressed in tumours of myoepithelial origin. Lack of SMA expression in the present case was significant to rule out myoepithelial origin of tumour. Thus myoepithelial carcinoma or myoepithelioma was ruled out. In our case, SMA expression was noted in the normal salivary gland acini.

S-100 protein expression was noted within salivary gland tumours. Its positivity and expression in the present case ruled out other tumours of lymphoid, vascular and endothelial cell origin.

CD-117 and calponin were found to be negative in all the cells (Fig. 11 and 12).

Discussion

Salivary gland cystadenomas especially in palate are rare benign tumours, but are well described in many other sites including the ovary, biliary tree, the appendix, the epididymis and the pancreas [3].

Fig. 12 Calponin: negative in all tumor cells

Mucinous cystadenomas of ovary are among the largest tumours known. They account for about 15 % of all ovarian neoplasms. They are typically large, multiloculated measuring up to 50 cm in diameter [12].

Previously, some investigators interpreted cystadenoma of salivary gland to be reactive cystic hyperplasia of the salivary duct rather than a true neoplasm and used corresponding terms such as ‘duct ectasia’, ‘salivary duct cyst’ and ‘intraductal papillary hyperplasia’ [6, 10].

However, now cystadenoma is generally believed to be neoplastic because of its proliferative properties [3].

In the first edition of the World Health Organization’s Histological Classification of Salivary Gland Tumours published in 1972, papillary cystadenoma was sub classified into various types of monomorphic adenomas. In the second classification published in 1991, cystadenoma was more clearly defined as a distinct histopathological entity that is further subdivided into papillary and mucinous types. Cystadenomas exhibit papillary and less frequently mucinous proliferation [13, 14].

Relevant case reports of cystadenomas of minor salivary glands were identified and retrieved by electronic searches in PubMed and Medline up to December 2013 (Table 2). Keywords were used for search with following terms: ‘cystadenoma, minor salivary gland, mucinous, papillary, apocrine, oncocytic, serous, mucous, oral cavity, maxilla, mandible, tumors, lesion, defect, critical size, histology, immunohistochemistry, radiographic, treatment’. Reference lists of all articles were retrieved from PubMed search. To the best of our knowledge, from analysis of the cases in the literature papillary cystadenomas were the most common whereas only one case of mucinous cystadenoma has been reported. The age in the cases reviewed ranged from 28 to 91 years with average of 65.8 years. Among the cases reviewed, majority of cystadenomas occurred in patients above the age of 50 years (Table 2). According to the literature, the cystadenomas are more

frequent in males than in females [5], but occasionally female predominance is mentioned (male to female ratio 1:2.3) [11]. Among the 28 case reports summarized in Table 2, the incidence is higher in males ($n = 15$) compared with females ($n = 12$) with female to male ratio of 1:1.25. The details of age and sex of one case of papillary cystadenoma was not found in the article by Crocker et al. [15]. The palate was found to be the commonest site followed by buccal mucosa, upper lip, lower lip and floor of mouth.

To our knowledge only one case of mucinous cystadenoma has been reported in literature so far in palate in a 64 year old female with the lesion measuring about 1 cm in diameter. In our case, patient was a female in third decade and the site of occurrence was palate. The size of the lesion in our case was 2.5×2 cm which is the maximum diameter reported till date.

The histopathology of cystadenomas has a much wider variation in cellular morphology and in the number of possible growth patterns. They are frequently well circumscribed and may have a thick, encapsulating band of fibrous connective tissue. However, cystic structures are often haphazardly arranged over a background of fibrous connective tissue or salivary gland parenchyma and evidence of encapsulation absent. Cystadenoma usually lacks an extra luminal, solid epithelial component. Foci of lymphocytic cell aggregations are sometimes evident in the fibrous stroma [10].

Although papillary configurations are commonly seen in this tumour, the term Papillary Cystadenoma is applied only when the lesion has multilocular cyst formations with conspicuous multiple papillary projections into the cystic spaces [8, 16]. It is an uncommon benign, well-circumscribed or encapsulated tumour [3].

The cysts contain eosinophilic, protinaecious material, sometimes with a few epithelial and inflammatory cells. Psammoma bodies or crystalloids (tyrosine rich crystals) are rarely present within the luminal secretions. The lining of cystic structures varies from flattened to tall columnar epithelium and cuboidal cells. Mucous, oncocytic, squamous, and apocrine cells are also present in the epithelium focally or occasionally extensively; a mixture of several cell types may commonly be seen [10]. The lining may be one to three epithelial cells thick and may then abruptly become focally thickened or form ramifying papillary projections with central cores of connective tissue [6, 17].

Papillary Cystadenomas account for 2–4.7 % of all minor salivary gland tumours and 4–8.1 % of all benign epithelial minor salivary gland tumours [3].

The review of literature in Table 2 shows that the papillary type of cystadenoma is the most common variety.

If multiple cysts are predominantly lined by mucous columnar epithelium with absence of papillary projections,

Table 2 Cases of mucinous and papillary cystadenomas reported in literature

Type of cystadenoma	Author	Year	Age/sex	Site	Treatment	Immuno histochemistry	Followup	Recurrence
Mucinous cystadenoma	Tange et al. [18]	2001	65/f	Hard palate	Excision of the lesion	Done	No details available	No
Papillary cystadenoma	Anuradha et al. [7]	2012	45/f	Palate	Excision of the lesion	Not done	No details available	No
Papillary cystadenoma	Karthigeyan et al. [38]	2012	52/m	Hard palate	Excision of lesion	Not done	10 months	No
Apocrine papillary cystadenoma	Halbritter SA et al. [23]	2009	46/m	Lower lip	Excision of lesion	Not done	1 year	No
Papillary cystadenoma	Gallego et al. [37]	2008	74/m	Upper lip	Excision of the lesion	Not done	2 years 54 years	No No
Papillary cystadenoma	Ribeiro et al. [36]	2004	54/m	Lower lip	Excision of the lesion	Not done	2 years	No
Papillary cystadenoma	Matsuzaka et al. [19]	2003	35/m	Upper lip	Excision of the lesion	Not done	No details available	No
Papillary oncocytic cystadenoma	Tsurumi et al. [21]	2003	80/f	Palate	Excision of the lesion	Not done	2.5 years	No
Papillary cystadenoma	Masakatsu et al. [28]	2001	82/f	Hard palate	Excision of the lesion	Done	No details available	No
Papillary cystadenoma	Mahler Vet al. [22]	1999	39/f	Buccal mucosa	Excision of the lesion	Not done	No details available	14 years
Papillary cystadenoma	Guccion et al. [39]	1997	62/m	Palate	Excision of lesion	Not done	3 years	No
Papillary cystadenoma	Alexis and Dembrow [24]	1995	73/f	Buccal mucosa	Excision of lesion	Not done	2 years	No
Seromucous papillary cystadenoma	Greene GW et al. [35]	1984	53/f	Maxillary right cuspid area	Excision of lesion	Not done	No details available	No
Papillary cystadenoma	Kerpel et al. [17]	1978	72/f	Buccal mucosa	Excision of the lesion	Not done	27 months	No
Papillary cystadenoma	Whittaker et al. [5]	1976	65/m	Left hard palate	Excision of lesion	Not done	3 years	No
Papillary cystadenoma	Whittaker et al. [5]	1976	84/f	Upper lip	Excision of lesion + radiation therapy	Not done	5 years	No
Papillary cystadenoma	Whittaker et al. [5]	1976	71/m	Right hard palate	Biopsy and radical maxillectomy	Not done	7 years	No
Papillary cystadenoma	Whittaker et al. [5]	1976	53/f	Palate (junction of hard and soft palate)	Excision of lesion	Not done	10 years	Yes
Papillary oncocytic cystadenoma	Whittaker et al. [5]	1976	50/m	Left hard/soft palate	Excision of lesion	Not done	2 years	No
Papillary cystadenoma	Crocker et al. [40]	1972	71/m	Buccal mucosa	Excision of lesion	Not done	No details available	No details available
Papillary cystadenoma	Crocker et al. [15]	1970	—	Palate	Excision of lesion	Not done	No details available	No
Mellanogenic papillary cystadenoma	Goldman et al. [34]	1967	58/f	Soft palate	Excision of lesion	Not done	No details available	No
Papillary cystadenoma	Chaudhary et al. [32]	1961	40/f	Floor of the mouth	Excision of lesion	Not done	No details available	No details available
Papillary cystadenoma	Chaudhary et al. [31]	1960	64/m	Buccal mucosa	Excision of lesion	Not done	No details available	No details available
Papillary cystadenoma	Brooks HW et al. [33]	1956	31/m	Hard palate	Excision of lesion	Not done	2 years	Yes (1, 2 years)
Papillary cystadenoma	Brooks et al. [33]	1956	63/m	Soft palate	Excision of lesion	Not done	2 years	No
Papillary cystadenoma	Skorpil [4]	1941	78/m	Soft palate	Excision of the lesion	Not done	20 years	Yes (16, 18 years)
Papillary cystadenoma	Skorpil [4]	1941	28/m	Soft palate	Excision of lesion	Not done	No details available	No details available

the tumour is termed as Mucinous Cystadenoma [10] which was observed in a case [18] mentioned in Table [2] as well as in the present case. The histopathological findings in our case coincide with those defined for mucinous cystadenoma [10, 18].

Papillary Oncocytic Cystadenoma is composed of papillary-cystic proliferation of a single or double layered oncocytic epithelium, superficially resembling a Warthin's tumour without a lymphoid stroma [6, 19]. It lacks the dense lymphoid stroma characteristic of Warthin's tumour. The lining epithelium of Oncocytic Cystadenoma may focally be admixed with cuboidal or columnar cells [10]. One case of Oncocytic Cystadenoma having prominent signet ring cell component has been reported in literature [20]. Some cases of papillary oncocytic cystadenoma in minor salivary glands have been reported in literature [5, 21].

Papillary Apocrine Cystadenoma is another variant of papillary cystadenoma containing apocrine cells [22]. The appearance of epithelial cells with eosinophilic cytoplasm, tall and columnar cells and blebbing of the luminal surface provides evidence for apocrine secretion. So far only one case has been reported in literature [23].

In view of the complexity and histomorphological diversity of salivary gland tumours, the pathological differential diagnosis of mucinous cystadenoma should include Papillary Cystadenoma, Intraductal Papilloma, Cyst Adenocarcinoma, Low grade Mucoepidermoid Carcinoma, Warthin's tumour and Polycystic Disease which are discussed below.

Papillary Cystadenoma was ruled out on the basis of type of cells lining the multiple cysts and absence of papillary projections.

Intraductal papillomas are unicystic structures characterized by numerous and complex papillary projections with fibro vascular cores and also lack the solid areas [24–26]. These features were not observed in the present case.

Although rare in oral mucosa, low grade cyst adenocarcinomas are morphologically similar to cystadenomas. Pattern of solid growth in focal areas, cellular atypia in some cases, permeation or destruction of glandular parenchyma, breakdown of the glandular lobe architecture as well as infiltration of adipose, muscle or bone tissues [24, 27] are characteristic features of low grade cyst adenocarcinomas. Differentiation of tumour types depends largely on the identification of actual infiltration of salivary gland parenchyma or surrounding connective tissue by either cystic or solid epithelium in cyst adenocarcinomas. Also, low grade cyst adenocarcinoma is a purely intraductal carcinoma lesion [10]. None of these features was observed in the present case.

Low grade mucoepidermoid carcinoma resembles cystadenoma in terms of growth pattern and the cell population

involved. Marked non papillary thickening of cyst lining cells is a more characteristic feature, which often has areas of solid, proliferating, extra luminal cords and islands of tumor. The cell population exhibits a combination of epidermoid, mucosal and to a lesser extent intermediate and basaloid cells. The rare low grade mucoepidermoid carcinoma composed entirely of mucous cells usually contains larger, more irregular mucous cells than those seen in cystadenomas and cystic structures that these mucous cells line are found infiltrating beyond the confines of the salivary gland lobule [3] which was not evident in our case.

Absence of prominent stromal lymphoid aggregates with a germinal centre and double row eosinophilic (oncocytic) cells projecting into cystic cavities [3, 13, 24] ruled out Warthin's tumour as a diagnosis.

Polycystic disease involves the entire gland more diffusely than does the well circumscribed cystadenoma [3].

Immunohistochemical findings suggest that the expression of keratins is closely linked with epithelial differentiation, the composition and patterns of expression in normal epithelium being reflected to a large extent in neoplastic lesions [28]. To our knowledge, immunohistochemistry has been done for only two cases of cystadenoma out of which one was papillary [28] and the other was mucinous [18]. In our case, the immunoreactivity was positive for cytokeratin markers, confirming that the tumor was mucinous and derived from salivary glands of hard palate. Immunoreactivity for S-100 was restricted to outer cytoplasm and absence of immunoreactivity for SMA, CD-113 and calponin ruled out the tumors of myoepithelial, lymphoid, vascular and endothelial origin.

Fine Needle Aspiration Cytology (FNAC)/Fine Needle Aspiration Biopsy (FNAB)/Core Biopsy is one of the diagnostic aids to plan the surgical treatment for a salivary gland tumor. The present lesion was a slow growing tumor on the basis of history, so it was clinically diagnosed to be a benign or a low grade malignant lesion. Also, it was small in size and the CT scan examination showed no invasion of the palatal bone. Hence, it was decided to excise it with clear margins.

In most cases of cystadenoma, the recommended treatment is simple surgical excision [24, 28]. The present tumor was surgically excised with wide margins under general anaesthesia.

A case of malignant transformation to invasive micro papillary adenocarcinoma in mucinous cystadenoma of parotid gland has been reported [29] but no such case has been reported in minor salivary glands.

Follow up was done for 2 years with no recurrence till date. Follow up of the patient is necessary since recurrences, though very few, have been reported [5, 30, 31]. However, the absence of locally destructive behaviour, histological features of a well circumscribed tumor lacking

mitoses, atypia and metastasis rule out its malignant nature. Thus recurrence could be attributed to incomplete surgical excision of the tumor [24, 28].

A final diagnosis of mucinous cystadenoma was arrived based on the histopathological and immunohistochemical analysis otherwise these lesions are often interpreted as a hyperplastic process of salivary ducts which may be the cause for the low incidence in the speciality literature. The present case is the second case of mucinous cystadenoma, in a 37 years old female, occurring in the hard palate. The size of the tumor was around 2.5×2 cm in diameter which is maximum size reported till date in literature to the best of our knowledge.

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Conflict of interest None

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