CASE REPORT

Papillary cystadenoma of anterior one-third of the tongue

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SUMMARY

Papillary cystadenoma is an unusual benign cystic neoplasm with cystic spaces of diverse sizes and intraluminal papillary projections histologically, which is commonly seen in the lips, cheek and palate. We report a case of papillary cystadenoma in a 40-year-old man on the anterior one-third of the tongue with classical histological features. The paper highlights the rarity of the site of occurrence with emphasis on differential diagnosis and the need for considering papillary cystadenoma when benign cystic lesions are encountered in the anterior one-third of the tongue.

BACKGROUND

Rarity of the site of occurrence owing to which it was misdiagnosed as mucocele.

CASE PRESENTATION

A 40-year-old man reported to the outpatient department with the chief symptom of swelling over the tongue since 1 month. It started as a small nodule that gradually increased to the present size. The swelling was associated with a history of trauma and pain. The pain was continuous, non-radiating, interfering with deglutition and speech, with no associated relieving factors. Medical and dental history were insignificant. Clinical examination revealed a 1.6×1.2 cm sized solitary, sessile, nodular swelling over the right half of the anterior one-third of the tongue extending 0.8 cm from the tip of the tongue posteriorly; mediolaterally it extended from the midline of the tongue to 1 cm away from the right lateral border of the tongue. The surface mucosa was erythematous and ulcerated at the centre (figure 1). On palpation, it was non-tender, soft in consistency and compressible. On bimanual palpation, interconnection was noted between the dorsal and ventral swellings. There was an absence of cervical lymphadenopathy.

INVESTIGATIONS

Routine blood investigations were within normal limits. A well-defined, bilobulated, hypoechoic area in the anterior one-third of the tongue was observed on ultrasonography (figure 2). The clinical diagnosis of mucocele was given based on the above findings.

DIFFERENTIAL DIAGNOSIS

Mucocele, pyogenic granuloma, fibroma, neurofibroma, glossal abscess, neurilemmoma, granular cell tumour, cavernous haemangioma, squamous cell carcinoma, intraoral nodular Kaposi's sarcoma.

TREATMENT

Complete surgical excision was carried out under local anaesthesia with an uneventful postoperative recovery, and the resected tissue was sent for histopathological examination. Grossly, the specimen was round, brownish white in colour, measuring 1.5×1 cm in size, soft in consistency with cystic spaces of varying sizes containing mucinous fluid.

Histopathological features

Microscopically, it showed multicystic areas with papillary intraluminal projections containing loose connective tissue cores infiltrated by chronic inflammatory cells, chiefly lymphocytes and plasma cells. Some areas of the cystic epithelium were lined by cuboidal and columnar cells and other areas were lined by pseudostratified epithelium with occasional mucous cells (figures 3–6). No cellular atypia was observed. The glandular parenchyma and lobe architecture was normal (figure 7). Based on the above findings, it was diagnosed as a papillary cystadenoma (PC).

OUTCOME AND FOLLOW-UP

The patient was followed up for over 8 months, initially for every 15 days and later every month with no evidence of any recurrence.

DISCUSSION

Salivary gland neoplasms constitute less than 1% of all tumours and 2–7% of all head and neck neoplasms. Of them, 15% are formed by minor salivary gland tumours. PCs of salivary glands comprise 0.7–8.1% of all benign salivary gland tumours, 3.1% of major salivary gland neoplasms and 7.7% of minor salivary gland tumours. Adenomatous epithelial proliferation with numerous cystic structures is the characteristic feature of cystadenoma. Papillary, serous, mucous and oncocytic cystadenomas are the various morphological variants of cystadenoma that have been described of which the mucinous and papillary variants are common. Usually some cystadenomas have been misdiagnosed as duct ectasia, oncocytic PC, salivary duct cyst, papillary hyperplasia and papillary mucous cystadenoma due to disparity in histological features.^{1–5}

Many investigators have integrated these tumours under the broad heading of monomorphic adenoma or deemed them to be reactive rather than neoplastic. Considering them to be reactive lesions, Chaudhry and coworkers² preferred the term 'intraductal hyperplasia'. Thackray and Lucas proposed the theory of incidental cyst formation of

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Figure 1 Solitary, nodular swelling of 1.6x1.2 cm sized over right half of anterior one-third of the tongue.

cystadenoma.² The WHO defined PC as “a tumor that resembles Warthin’s tumor but differs by the presence of manifold papillary projections, greater variety of epithelial lining cells and by lacking lymphoid elements.” If the lining epithelial cells show mucous cell predominance, the tumour is referred to as mucinous cystadenoma.⁵ Bauer and Bauer suggested the origin of cystadenoma from undifferentiated epithelium of the intercalated ducts of the gland.⁶ They are also described in other sites of the body like the ovary, the epididymis, biliary tree, the appendix and the pancreas where cystadenomas share some morphological resemblance with those of salivary glands. Von Hippel-Lindau gene loss results in the overexpression of ‘hypoxia-inducible factor’, an angiogenic protein, and is considered to be involved in the pathogenesis of PC of epididymis.⁷

Almost 58% of the tumours occur in parotid gland, 6.6% in submandibular gland, 0.5% in sublingual gland, 10.7% on the lips, 8.2% on the cheeks, 7.1% in the palate and 9.1% in other intraoral sites.²⁻³ In a study carried out by Pires *et al*,⁴ out of 42 cases of PC, 11 (26.2%) were reported in the palate, 10 (23.8%) in the buccal mucosa, 9 (21.4%) on the lower lip, 5 (11.9%) in the vestibule and 3 (7.1%) on the upper lip. Waldron *et al*⁸ reported seven in the buccal mucosa, four in the palate, three in the floor of the mouth, three on the lower lip, two on the upper lip and one in the mandibular vestibule. No cases were reported in the tongue in both studies. A review of literature reveals only three reported cases of PC in the tongue. It usually affects patients in the sixth decade. Kacker and de Serres⁹ reported a case of congenital PC. Although it has a female predilection, cases were also reported in men.¹⁰⁻¹² It is usually less than 1 cm in greatest dimension but a case of very huge PC was reported almost occupying the entire floor of the mouth with compression of the tongue towards the craniodorsal direction causing improper visualisation of Wharton’s duct papillae.¹³ Clinically it is generally asymptomatic and presents no distinctive clinical features to mucocele. In the present case, as the lesion was on the dorsal surface of the tongue, it was subjected to trauma as a consequence of pain and also interfered with deglutition.

Clinically, PC was distant from our insight and our differential diagnoses included mucocele, pyogenic granuloma, fibroma, neurofibroma, glossal abscess, neurilemmoma, granular cell tumour, cavernous haemangioma, squamous cell carcinoma and intraoral nodular Kaposi’s sarcoma. The absence of acute symptoms, pus discharge, fever and lymphadenopathy ruled out the likelihood of glossal abscess. Cavernous haemangioma was eliminated as it was fluctuant and even more blanching and other pressure symptoms were absent. Late lesions of intraoral Kaposi’s sarcoma develop nodular appearance but the absence of a contributory history, benign course of the lesion and the absence of necrosis dodged the diagnosis. The cystic nature of

Figure 2 Ultrasonography showing well-defined, bilobulated, hypoechoic area in the anterior one-third of the tongue.

Figure 3 H&E-stained section showing multiple cystic areas.

the lesion as observed under ultrasonography eluded us from aforementioned benign mesenchymal lesions. Malignant tumour-like squamous cell carcinoma was excluded by the well-circumscribed nature of the lesion, and by the absence of decisive factors such as punched out margins, indurated base, invasion of adjacent structures and metastatic lymph nodes. Histologically, the present case displayed the classical histopathological pattern as defined by the WHO. It was not encapsulated. Only 25% of the cases have shown the evidence of a distinct fibrous capsule.¹² A case of PC with cystic spaces containing proteinaceous fluid and focal, scattered psammomatous calcification in the epithelium was reported by Lim *et al.*¹⁴ PC must be differentiated histologically from Warthin's tumour, ductal papillomas such as intraductal papilloma, sialadenoma papilliferum and inverted ductal papilloma, focal oncocytic adenomatous hyperplasia, canaliculic adenomas, low-grade mucoepidermoid carcinoma (MEC) and cystadenocarcinoma. The absence of marked lymphoid component with germinal centres helped us to distinguish it from Warthin's tumour. The presence of multiple cystic spaces with intraluminal papillary proliferation ruled out the possibility of the intraductal papilloma as it classically exhibits a single cystic space with numerous and complex papillary projections. The absence of exophytic papillary projections clinically and histologically covered by stratified squamous epithelium with mild dysplasia and lymphocytic infiltration eliminated sialadenoma papilliferum. Multicystic nature excluded inverted ductal papilloma as it is a single cystic space with thick bulbous papillary projections lined by squamous cells that fill the ductal lumen. Oncocytic change in the ductal epithelium and variable stromal lymphoid component seen in focal oncocytic adenomatous hyperplasia

Figure 4 Complex papillary projections.

Figure 5 Cystic lining partly lined by cuboidal (yellow arrow), columnar (red arrows) and partly by pseudostratified epithelium (green arrows).

were not apparent in the present case. Canaliculic adenomas show large cystic areas with papillary proliferations, most frequently exhibit single-layered cords or columnar or cuboidal epithelial cells with deep basophilic nuclei with prominent vascularity in loose connective tissue stroma. The papillae in MEC are more irregular and complex, when present, than those found in PC, and also the intermediate cells are seen. Canaliculic adenoma and MEC were ruled out because of the absence of these classical features. Cystadenocarcinomas are morphologically akin to PC but some distinctive aspects such as solid growth pattern in focal areas with sporadic cellular atypia, mode of invasion, infiltration or annihilation of the glandular parenchyma, glandular lobe architecture breakdown along with permeation into adipose and muscle tissues helped us to delineate it from PC.

Simple surgical excision is usually recommended as performed in this case. A radiotherapy-treated case called 'papillary cystic adenoma' in the palate was described by Hobock. In 1941, Skorpil reported cases of PC in the palate of which one experienced two recurrences. Similarly, Brooks *et al* in 1945 described two cases out of which one experienced recurrence. Alexis and Dembrow¹⁵ concluded that this lesion has a tendency to recur if not aptly excised, signifying a necessary follow-up. The patient was followed up for over 8 months, initially for every 15 days and later for every month with no evidence of any recurrence.

CONCLUSION

Thus, PC which is an exceptional benign neoplasm with its rare occurrence in the tongue should also be considered in the differential diagnosis of anterior tongue lesions, and the diagnosis

Figure 6 Epithelium showing mucous cells.

Figure 7 Photomicrograph showing normal glandular parenchyma and lobe architecture (at the bottom) and papillary proliferations (at the top).

should be carefully performed ruling out all the histological similarities.

Learning points

- ▶ Papillary cystadenoma is an atypical benign cystic neoplasm very rarely involving the tongue.
- ▶ Clinically it resembles mucocele in most of the cases.
- ▶ It should be considered in the differential diagnosis in anterior tongue lesions.
- ▶ Histologically, it shows multicystic spaces with intraluminal papillary projections.
- ▶ Surgical excision is the suggested treatment option and it recurs if not appropriately excised.

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PHK was involved in drafting the work and revising it critically for important intellectual content. ARA was involved in final approval of the version to be published. VSG was in agreement to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.

Competing interests None.

Patient consent Obtained.

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