

# Papillary cystadenoma and cystadenocarcinoma of salivary glands: Two unusual entities

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## Abstract

Cystadenoma and cystadenocarcinoma are rare salivary gland tumours histologically characterized by prominent cystic and frequently papillary growth. We present two cases of cystadenoma of a minor salivary gland (upper lip) and parotid cystadenocarcinoma respectively, captured between 834 salivary gland tumors studied in our hospital from 1980 to 2004. The authors review the clinical, histological, and biological features of these two unusual tumours, and differential diagnosis with other salivary glands neoplasms. Both entities usually reveal papillary proliferation of the epithelial lining and are composed of cells that possess bland cytomorphologic features. 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 cystadenocarcinomas. Step sections of a borderline tumour may yield unequivocal evidence of invasion. The authors discuss the problematic diagnosis between these rare neoplasms and with other tumours and compare histological findings of these two entities.

**Key words:** *Papillary cystadenoma, cystadenocarcinoma, salivary gland tumours.*

## Introduction

Papillary cystadenoma of salivary glands is an uncommon benign neoplasm. In two large reviews, it constituted 2% and 4.7%, respectively, of all minor salivary gland neoplasms, and 4% and 8.1%, respectively, of all benign epithelial minor salivary gland neoplasms (1,2). Cystadenocarcinoma is a rare malignant tumour histologically characterized by prominent cystic and frequently papillary growth, but lacking features that characterize cystic variants of several more common salivary gland carcinomas: polymorphous low-grade adenocarcinoma, mucoepidermoid carcinoma, and the papillary cystic variant of acinic cell carcinoma (3).

This study presents two cases of these unusual neoplasms, captured between 834 salivary gland tumors studied in our hospital from 1980 to 2004. The objective is compare histological findings of these two entities.

## Case Reports

### - Case one

A 74-year-old man presented with an asymptomatic mass of the upper lip mucosa that had been present since 5 years. Clinical examination revealed a 1.2-cm-diameter, well-circumscribed, solid, round mass (Figure 1). A provisional clinical diagnosis of pleomorphic adenoma was made, and the lesion was excised uneventfully. The pathological

diagnosis was papillary cystadenoma (Figure 2). There has been no recurrence over a 2-year follow-up period.

- Case two

A 34-year-old male without personal antecedents of interest presented with a parotid tumour mass for the past 10 years. Exploration revealed the presence of a well delimited lesion measuring some 6 cm in diameter, of cyst-like consistency and located in the caudad portion of the right parotid region (Figure 3A). The skin overlying

the tumour showed no changes in color or evidence of infiltration. No neck masses were palpated, and no synchronous pathology was observed in the oropharyngeal region. According to the patient, the lesion had grown in size in the previous 6 months, causing slight pain. Fine needle aspiration biopsy (FNAB) diagnosed Warthin's tumor, and computed tomography (CT) identified a cystic tumor measuring 6 x 4 cm in diameter and located in the caudal portion of the right parotid gland (Figure 3B). A



Fig. 1. Well-circumscribed mass on the upper lip mucosa.

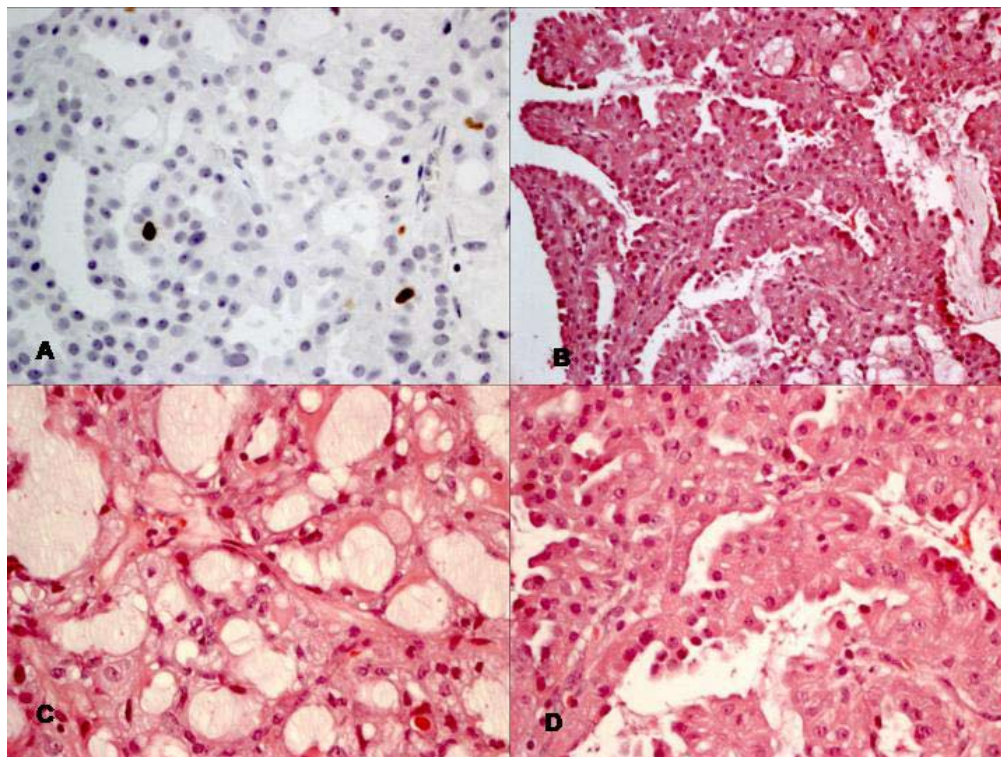
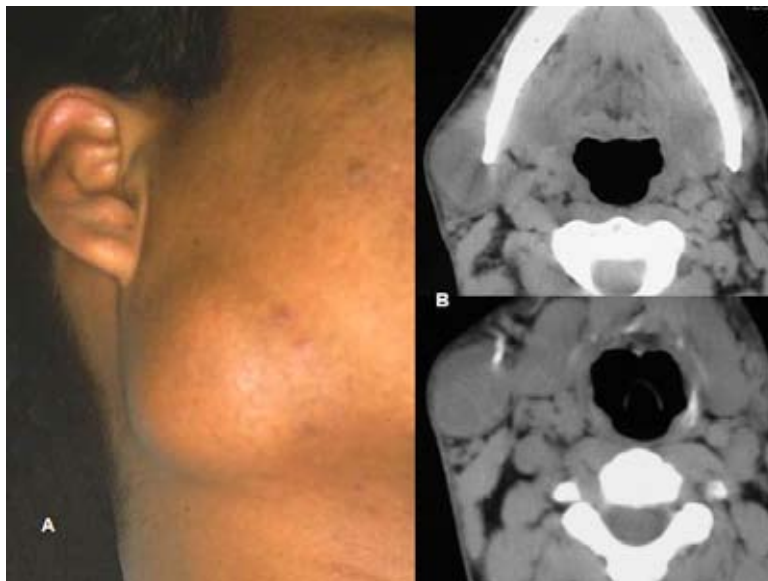
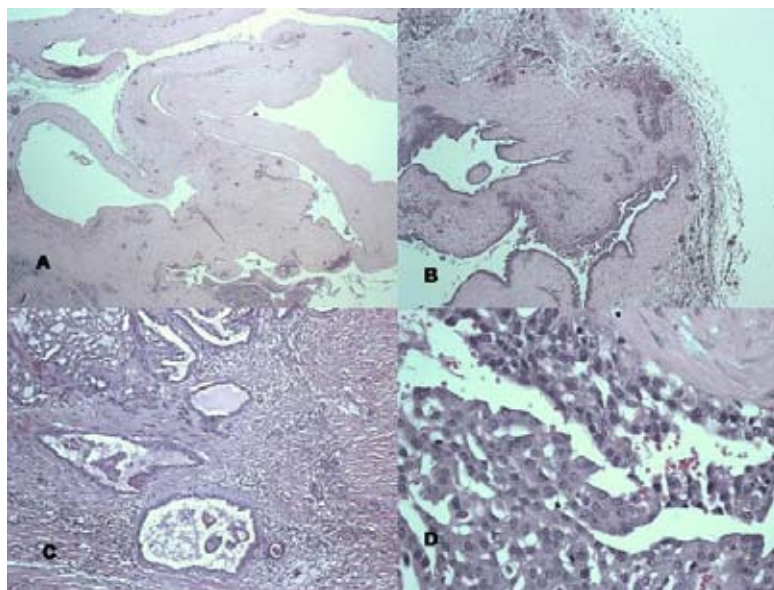


Fig. 2. A. Immunohistochemical study with Ki67, showing less than 5% of cellular activity. B. The papillae are lined with pseudostratified cuboidal to columnar epithelium (H&E stain, original magnification x 150). C. Multiple small cystic spaces surrounded by eosinophilic cells (H&E stain, original magnification x400). D. The epithelium consisted of cells with eosinophilic cytoplasm admixed with clear cells and occasional mucinous cells (H&E stain, original magnification x400).



**Fig. 3.** A. Facial appearance of the patient showing the right parotid tumor. B. Computed tomography examination showing a rounded and well delimited lesion within the right parotid gland.



**Fig. 4.** A. Cystadenocarcinoma is relatively well circumscribed (H&E stain, original magnification x25). B. Medium ovoid cysts form the vast majority of the tumor (H&E, original magnification x100). C. Papillary epithelial proliferation is focally prominent (H&E stain, original magnification x200). D. The epithelium reveals enlarged nuclei and prominent nucleoli (H&E stain, original magnification x400).

conservative superficial parotidectomy was performed under general anesthesia. The marginal branch of the facial nerve was included within the tumor mass. A diagnosis of papillary cystoadenocarcinoma was made based on the characteristic clinicopathologic findings, as shown in figure 4. There were no postoperative neurological definitive alterations, and no tumour recurrence was observed five years after surgery.

### Discussion

First reviews of the clinical, histologic, and biologic features of the papillary cystadenoma (4) show that it appears to occur more frequently in women; most patients have been older than 50 years of age, with several in their seventies. The most common sites are the palate and buccal mucosa; however, tumors in the lip and tongue also have been described. The usual presentation is an asymptomatic

mass. However, in the files of the Armed Forces Institute of Pathology (AFIP) (5) the lesion are widely distributed among major (65 percent) and minor (35 percent) salivary glands. In our center, any case of cystadenoma of major salivary gland was found over 693 tumours sample.

On microscopic examination, the neoplasm is usually well circumscribed and may be surrounded by a rim of fibrous tissue; there are solid areas (usually limited in extent) and cystic areas into which project papillae lined by cuboidal to columnar cells usually two layers thick. The cells usually have eosinophilic cytoplasm, and goblet cells may be present (6,7).

Perhaps the most important entity in the differential diagnosis of papillary cystadenoma is cystadenocarcinoma; sometimes the distinction may be difficult because the neoplasms have similar architecture, and also because cystadenocarcinoma often shows little atypia (8). Both neoplasms usually reveal papillary proliferation of the epithelial lining and are composed of cells that possess bland cytomorphic features. 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 cystadenocarcinomas. Step sections of a borderline tumour may yield unequivocal evidence of invasion.

The infrequent presentation of the cystadenocarcinoma, together with the associated terminological and histological confusion, has precluded the documentation of important series of cases. The files of the Armed Forces Institute of Pathology (AFIP) (8) have recorded 57 cases of cystadenocarcinoma. The mean patient age in this series was 59 years, and both sexes were affected equally. About 65% of the documented tumours occurred in the major salivary glands, and most of these comprised the parotid glands. The preoperative diagnosis of cystadenocarcinoma is complex. When located in the parotid gland, the presentation is usually that of a slowly growing, asymptomatic mass. In a series of 56 cystic lesions of the salivary glands including only two cystadenocarcinomas, Layfield and Gopez (9) reported an overall accuracy of 84%. Other authors describe a FNAB diagnostic efficacy of 80% in application to cystadenocarcinomas (10). In the literature, the lesions most often confused with cystadenocarcinomas when performing FNAB are Warthin's tumour or salivary gland cysts (11).

In our case, the evolution of the lesion, its specific location, the sex of the patient and the information provided by FNAB in the preoperative phase suggested Warthin's tumour, despite the unusual age of presentation. During surgery, the direct relation of the lesion to the marginal branch of the facial nerve may have been suggestive of some other histological type of tumour.

The management approach suggested for cystadenocarcinoma to date is similar to that advocated for other low-grade salivary gland adenocarcinomas (8). A number

of high and low-grade cystadenocarcinomas have been documented in the literature in both animals and humans (12-14) in which complementary radiotherapy is recommended. High grade tumours frequently show perineural infiltration, vascular or lymphatic channel invasion, infiltration of surrounding connective tissues, and regional lymph node metastasis. In this cases, the mitotic activity was high and there were occasional abnormal mitotic figures. In our case, and considering the low histological malignancy of the tumour and the absence of lymph node metastases, annual revisions were decided for a minimum period of five years.

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