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Case Report

PAPILLARY CYSTADENOMA ARISING FROM THE UPPER LIP: A CASE REPORT

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Abstract

We report a rare case of a papillary cystadenoma arising from the upper lip. This tumor was not distinctly encapsulated and had proliferated replacing the ductal epithelium. Mast cells were found not only in the stroma but also in the oncocytic epithelial layer. There was a strong immunoreaction with mitochondrial antibody in the epithelial layer. Only one case (0.9%) of papillary cystadenoma has occurred among the 110 benign intraoral salivary gland tumors seen in our hospital from 1966 through September 2003.

Key words: Papillary cystadenoma—Oncocytic change—Upper lip—Mast cell

INTRODUCTION

Cystadenoma of the salivary gland is an uncommon benign neoplasm which is further subdivided into papillary and mucinous types^{1,8)}. This tumor closely resembles Warthin tumor but without the lymphoid elements⁸⁾. The origin of this tumor, as suggested by Bauer and Bauer²⁾, is principally the undifferentiated epithelium of the intercalated ducts of the gland. The most frequent clinical

finding is a painless mass beneath the mucosa of the hard palate, cheek, or posterior portion of the tongue^{3,9,10)}. Herein we report a rare case of a papillary cystadenoma arising from the upper lip with oncocytic changes.

CASE REPORT

A 35-year-old male patient with no contributing medical or family history noticed

a painless swelling on his left upper lip in May of 2003 and was admitted to the Chiba Hospital of Tokyo Dental College in June of 2003. The lesion was surgically removed with a clinical diagnosis of lymphadenitis, and the specimen was sent to the Department of Clinical Pathophysiology at the Tokyo Dental College for pathological diagnosis. After the specimen was fixed with 10% formalin, paraffin sections were prepared for light microscopy and were stained with hematoxylin and eosin using routine methods.

PATHOLOGICAL FINDINGS

Histopathologically, predominantly mucous gland tissue with multiple cystic areas was lined by eosinophilic epithelial cells arranged in two layers, a peripheral layer of tall columnar cells with acidophilic cytoplasm and pyknotic nuclei, and a basal layer composed of cuboidal or polygonal cells. The tumor had proliferated replacing the ductal epithelium with polarity. In some areas, there was a papillary proliferation of the lining cells with the supporting connective tissue in the lumens (Figs. 1A, B). No lymphocytic infiltration was evident. Many mast cells were observed not only in the stroma but also in the oncocytic epithelial layer (Fig. 1C). The oncocytic epithelial cells were stained by PTAH (phosphotungstic acid hematoxylin) (Fig. 1D), and their cytoplasm strongly reacted with an anti-mitochondrial antibody (Fig. 1E).

DISCUSSION

Cystadenomas are subclassified into various types of monomorphic adenomas, and they are more clearly defined as distinct histopathologic entities that can be further subdivided into papillary and mucinous types⁸. The present case is classified as a papillary type. Papillary cystadenoma of the minor salivary glands is rare. Chaudhry *et al.* reported that this type of tumor occurs in 2% of 800 intraoral minor salivary gland benign tumors⁵.

Only one case (0.9%) of papillary cystadenoma has occurred among 110 intraoral benign salivary gland tumors seen from 1966 through September 2003 in our hospital. The WHO characterizes cystadenoma as similar to Warthin tumor but without lymphatic tissues. In AFIP files, the frequency of occurrence of cystadenoma is 4.1% of all benign epithelial tumors, and it is relatively more frequent among benign tumors in the minor salivary glands than in the major ones⁹. Waldron *et al.* reported that it occurred in 8.1% of their 245 benign minor gland tumors¹¹. Most cystadenomas are characterized by predominantly unicystic or multicystic growth; there is a focal interluminal papillary proliferation of the lining epithelium⁹. Only 25% of the cases have shown evidence of a distinct fibrous capsule¹¹, and our present case did not have a distinct capsulation. In our case, many mast cells were observed not only in the stroma but also in the epithelial oncocytic cell layer. Although Caselitz *et al.* reported seeing mast cells in cystadenolymphoma in 1984, they noted that the mast cells were related to the IgE of lymphocytes, which is in good accordance, because there is a functional relationship between mast cells and IgE⁴. Therefore, the characteristics of our case are similar to those of Warthin tumor. Guccion *et al.* reported that the oncocytic epithelial tumor cells were eosinophilic, which was revealed in our case by immunohistochemical staining with a mitochondrial antibody⁷. Auclair *et al.* reported that oncocytic differentiation occurred in 16% of their cases of papillary cystadenomas¹.

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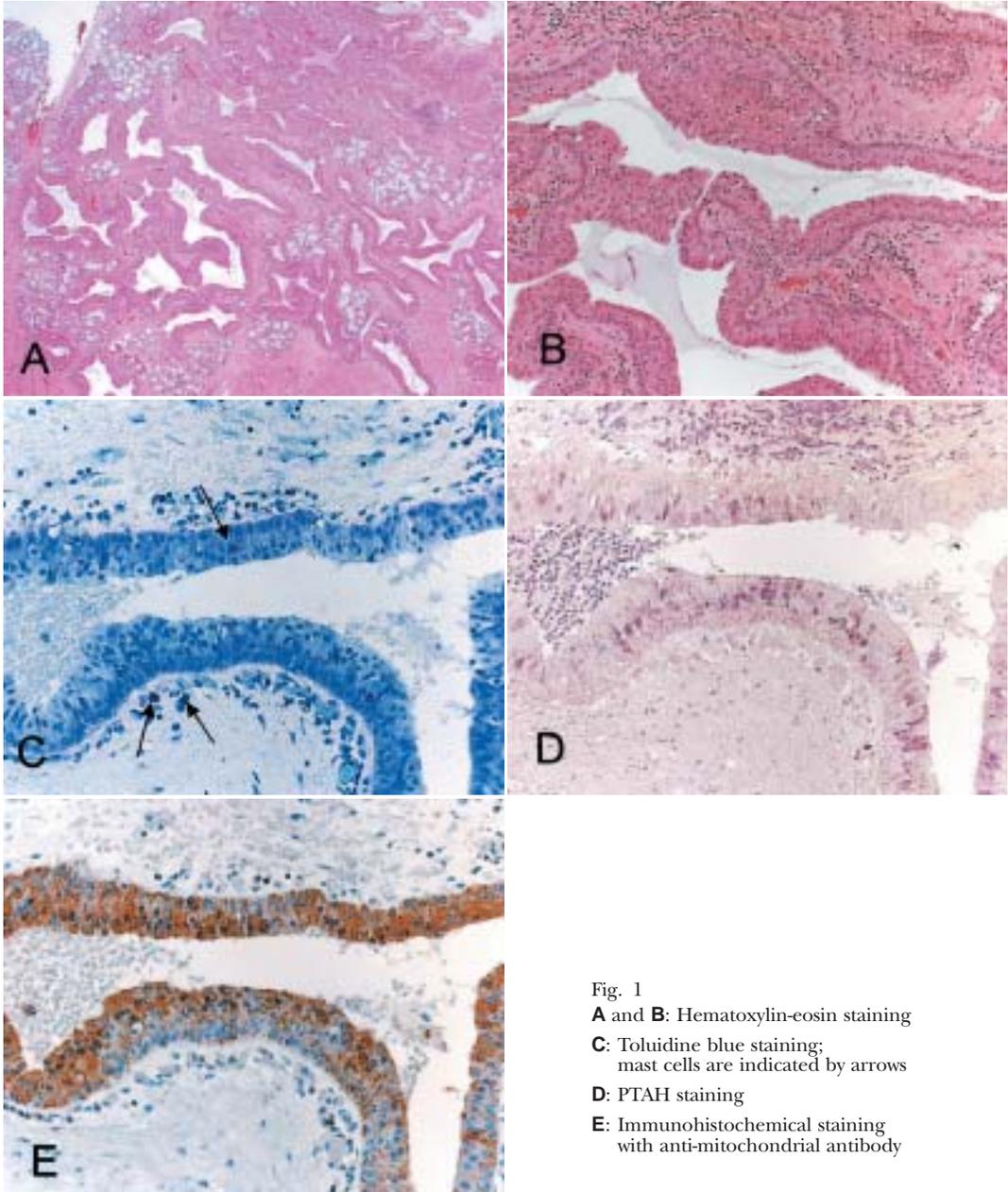


Fig. 1
A and B: Hematoxylin-eosin staining
C: Toluidine blue staining;
mast cells are indicated by arrows
D: PTAH staining
E: Immunohistochemical staining
with anti-mitochondrial antibody

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