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Papillary cystadenocarcinoma: Report of a case of high-grade histopathologic malignancy

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Abstract

Papillary cystadenocarcinoma is an extremely rare malignant tumor of the salivary gland which is cytologically considered to be of low-grade malignancy, showing an indolent biological behavior. Histologically, the tumor is characterized by cysts and papillary endocystic projections. This tumor most commonly arises in the major salivary glands, mainly the parotid gland. We report here a case of papillary cystadenocarcinoma originating from the minor salivary gland, which showed marked growth over a short period of time. Histologically, the tumor was characterized by intense cellular and nuclear pleomorphism and numerous mitotic figures. These findings indicate a tumor of high-grade malignancy.

Keywords: Papillary cystadenocarcinoma; Malignant papillary cystadenoma; Salivary gland tumor

1. Introduction

Papillary cystadenocarcinoma (PC) is an extremely rare malignant tumor of the salivary gland described by the WHO in 1991. Until then, this tumor was classified as an atypical type of adenocarcinoma or was also called malignant papillary cystadenoma, low-grade papillary adenocarcinoma or mucus-producing adenopapillary carcinoma [1]. This type of tumor can also occur in the ovary, bladder, bile duct, pancreas, mammary gland, thyroid, and upper respiratory tract [2]. Cases of PC in the prostate have been reported but are uncommon [3].

Cytologically, PC is defined as a low-grade glandular tumor with an indolent biological behavior which is characterized by cysts and papillary endocystic projections. This tumor most commonly arises in the major salivary glands, mainly the parotid gland, but involvement of the minor salivary glands has also been reported [2,4,5]. Most patients present a mass of slow and painless growth [1].

We report here a rare case of PC arising in the palate of a 79-year-old man, which was histologically considered to be of high-grade malignancy. We found no case of high-grade histologic PC located in the palate in the literature. We describe the clinical and histologic features of the tumor, as well as the immunohistochemical findings obtained using prostate-specific antigen (PSA), thyroglobulin, p53 and Ki 67 as markers.

2. Case report

Patient M.R.N., a 79-year-old black man, was seen at a surgery and oromaxillofacial traumatology service complaining of an asymptomatic volume increase in the palate for 6 months. The patient reported swallowing difficulties.

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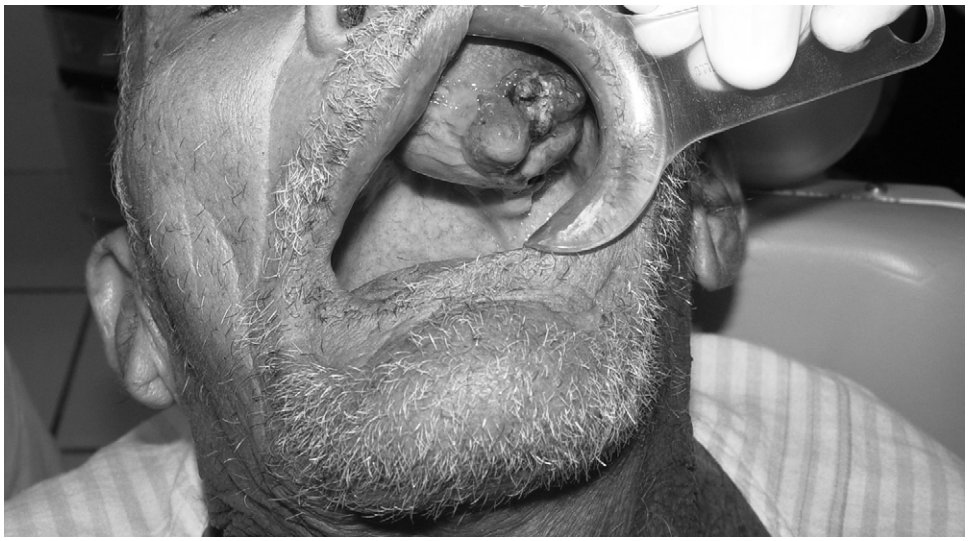

Fig. 1. Ulcerated firm mass in hard palate extending to alveolar ridge.

Review of the medical record revealed no noteworthy systemic alterations. Extraoral physical examination showed discrete facial asymmetry in the left hemiface. No palpable regional lymph nodes were noted.

Intraorally, an extensive exophytic tumor lesion measuring approximately 5.0 cm was identified in the hard palate, extending from the alveolar margin, crossing the midline and provoking expansion of cortical bones. In addition, the tumor presented a lobular aspect and an area of central ulceration (Fig. 1). Axial computed tomography scans with contrast injection showed a tumor mass measuring about 5.0 cm in diameter in the anterior region of the left maxilla and destruction of the buccal cortical bone, with the mass invading the maxillary sinus and extending in the direction of the hard palate (Fig. 2). In coronal sections with contrast injection the same tumor mass was found to be destroying the alveolar margin, body of maxilla and hard palate on the left side. Invasion of the inferior portion of the maxillary sinus and nasal fossa was also noted.

With the diagnostic hypothesis of squamous cell carcinoma, an incisional biopsy was performed whose anatomopathologic analysis revealed malignant salivary gland neoplasm. The tumor exhibited cystic cavities that contained papillary projections consisting of the proliferation of columnar or cubic cells with voluminous nuclei arranged in a single or double layer, as well as stratified areas (Fig. 3). The cells were characterized by atypias such as nuclear and cellular pleomorphism, numerous, sometimes aberrant, mitotic figures, and nuclear hyperchromatism, in addition to prominent nucleoli (Fig. 4). The stroma was scarce and exhibited a moderate, predominantly mononuclear inflammatory infiltrate. The histopathologic diagnosis was PC. The patient was referred to the Service of Head and Neck Surgery, but refused to undergo excision of

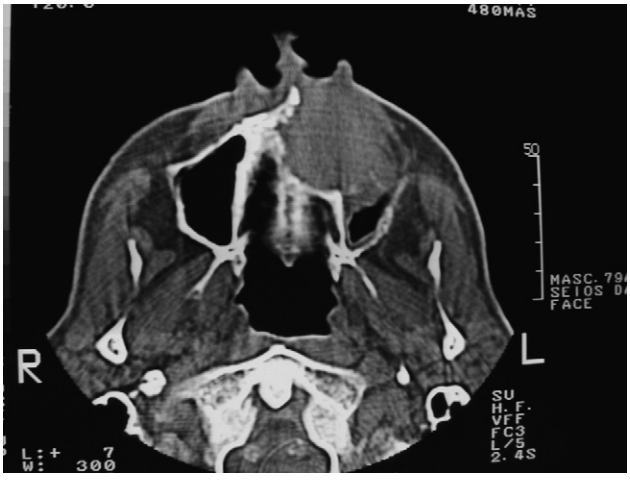

Fig. 2. Axial CT scan showing an expansile mass destructing buccal cortical bone and invading the maxillary sinus.

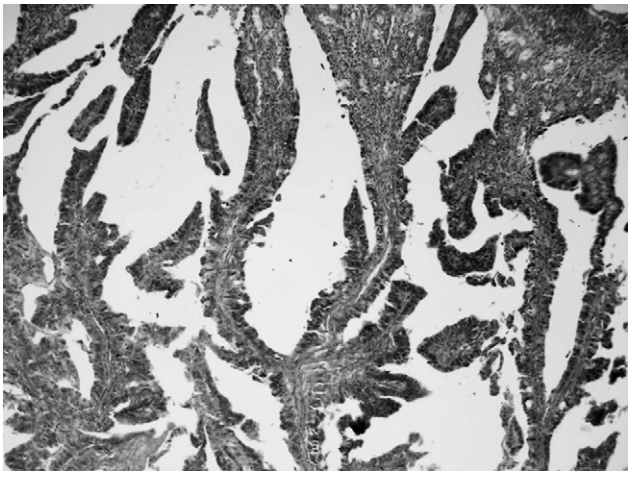

Fig. 3. Photomicrograph of tumor with numerous cystic lumens showing papillary projections consisting of the proliferation of columnar or cubic cells (H & E/100× magnification).