

# MRI findings of papillary cystadenocarcinoma of the submandibular gland

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## ABSTRACT

Papillary cystadenocarcinoma is an extremely rare malignant tumor of the salivary glands. Major locations of this neoplasm are the parotid gland, sublingual gland, and minor salivary glands, while occurrence in the submandibular gland is extremely rare. Magnetic resonance imaging (MRI) findings of this lesion have not been reported in the literature. Herein, we present a case of papillary cystadenocarcinoma arising from the right submandibular gland. MRI showed a 5-cm mass composed of solid and cystic components in the mildly enlarged gland. Pathologic evaluation revealed papillary cystadenocarcinoma.

*Key words:* • submandibular gland • cystadenocarcinoma, papillary • magnetic resonance imaging

**P**apillary cystadenocarcinoma is an extremely rare malignant tumor of the salivary glands described by the World Health Organization in 1991. Only a few cases of submandibular gland papillary cystadenocarcinoma have been reported in the literature (1–3). This type of tumor can also occur in the ovary, bladder, bile ducts, pancreas, mammary gland, thyroid, and upper respiratory tract (4). This tumor was classified as an atypical type of adenocarcinoma, and has also been called malignant papillary cystadenoma, low-grade papillary adenocarcinoma, or mucus-producing adenopapillary carcinoma (5). We report a case of papillary cystadenocarcinoma in the right submandibular gland with magnetic resonance imaging (MRI) and histological features.

## Case report

A 74-year-old male patient was admitted to our hospital with right submandibular swelling and pain. The mass had been noted 1 year previously and had grown slowly. Axial T1-weighted neck MRI (Fig. 1) demonstrated a hypointense, non-homogeneous, smooth mass with lobulated contours originating from the right submandibular area, extending anteriorly and posteriorly. The lesion measured 9 × 5 × 3.5 cm. Axial T2-weighted MRI (Fig. 2) showed heterogeneous hyperintensity composed of solid and cystic components. Post-contrast axial and coronal images (Fig. 3) showed contrast enhancement in the solid components.

Fine-needle aspiration provided no diagnostic information; therefore total surgical resection was performed. Histological examination of the surgical specimen showed intermediate grade papillary cystadenocarcinoma of the submandibular gland (Fig. 4).

## Discussion

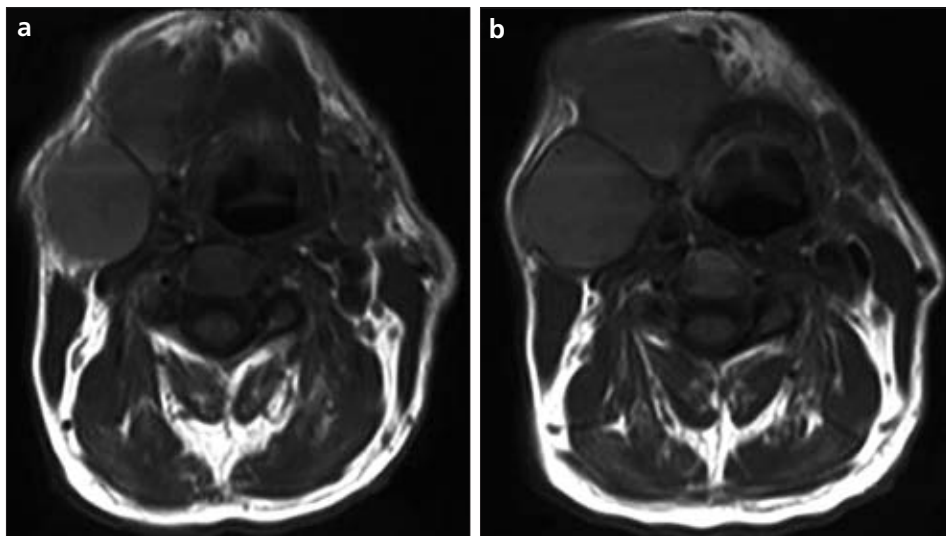
Salivary gland tumors comprise 2% of all adenocarcinomas and 10% of malignant epithelial salivary gland tumors. A large study showed that the peak age of occurrence was in the 7th to 8th decade; 60% occurred in women, 58.5% were located in the parotid gland, 28.5% in the minor salivary glands, 11.5% in the submandibular gland, and only 1.5% in the sublingual gland (6). Solid, tubular, and papillary adenocarcinomas can be distinguished. Solid adenocarcinoma (13%) is predominantly located in the parotid gland. Of tubular adenocarcinomas (52%), 62.5% are located in the parotid gland, 27.5% in the minor salivary glands and 10% in the submandibular gland. Papillary adenocarcinomas (28.5%) are located in almost 50% of cases in the minor salivary glands, 45% in the parotid gland and only 5% in the submandibular gland (6). Clinical features include pain, rapid growth, firmness, or lymph node enlargement, and should alert the physician to the possibility of malignancy.

Contrast-enhanced computed tomography (CT) and MRI are the radiological examinations of choice for evaluating mass lesions of the sali-

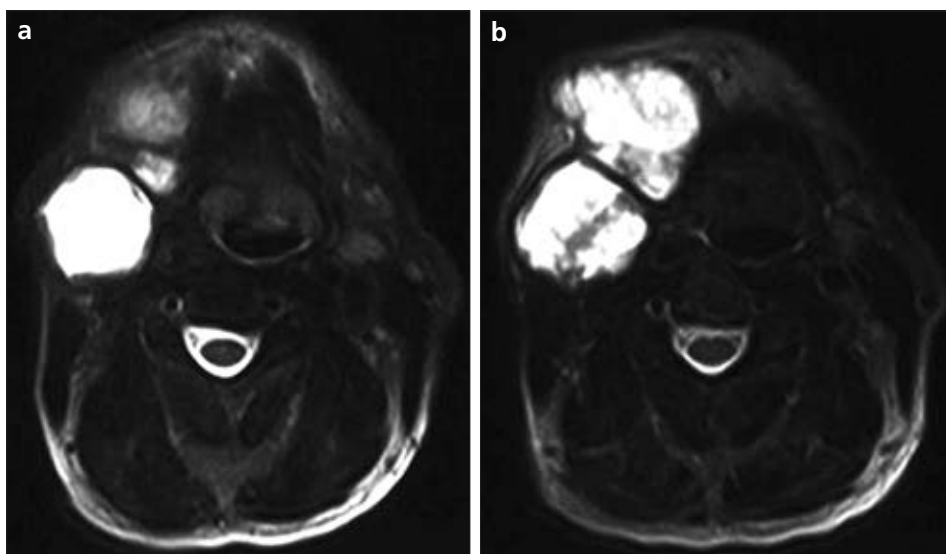
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**Figure 1.** a, b. Axial T1-weighted neck MR images demonstrate a hypointense, inhomogeneous, mass with smooth, lobulated contours originating from the right submandibular gland.



**Figure 2.** a, b. Axial T2-weighted MR images show heterogeneous hyperintensity in the lesion.

vary glands and neck region. With excellent spatial resolution and superior soft tissue contrast, MRI has major advantages over CT. Multiplanar imaging capability of MRI allows it to evaluate the extent of disease and outline the contours of tumors. The tumor-muscle interface is better evaluated with MRI than with CT. Unlike CT scanning and sialography, risks associated with radiation and iodinated contrast are non-existent with MRI (7).

Imaging is very important for confirming the presence of a mass; determining its relationship to the salivary gland and adjacent structures; determining whether the mass is well-circumscribed or infiltrating; ascertaining whether it is solid, necrotic, or cystic in nature; and finding out whether there are bilateral disease and lymph node enlargement (8).

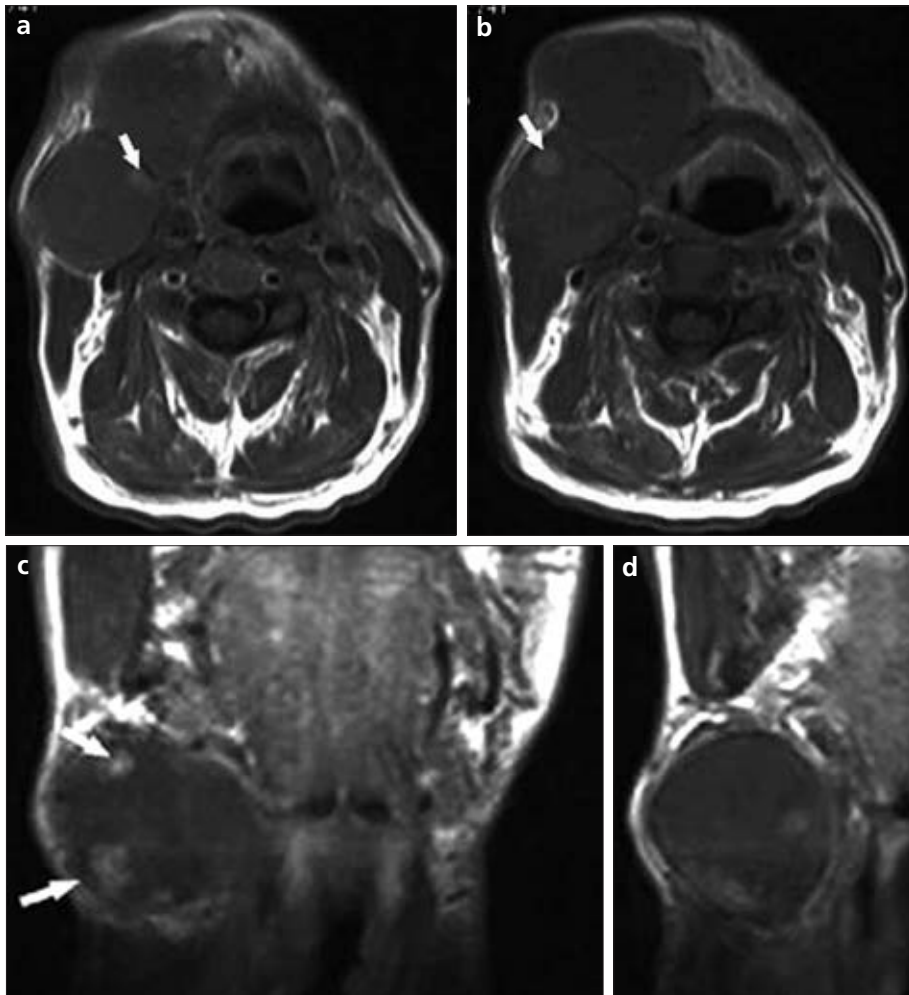
Imaging characteristics of submandibular malignant lesions include an irregular and poorly defined border, heterogeneous internal structure, invasion of surrounding tissues, and lymph node metastases. Although these findings are suggestive of malignancy, they are non-specific. Malignant salivary gland tumors can be solid, cystic, or necrotic. Macroscopic calcification and hemorrhage are rare.

In our case, axial T1-weighted MRI showed a hypointense, non-homogeneous, smooth mass with lobulated contours originating from the right submandibular area. The lesion had a relatively well-defined border, and there was no apparent invasion of surrounding tissues. On axial T2-weighted MRI there was high signal intensity due to cystic components, and heterogeneous signal intensity in the solid com-

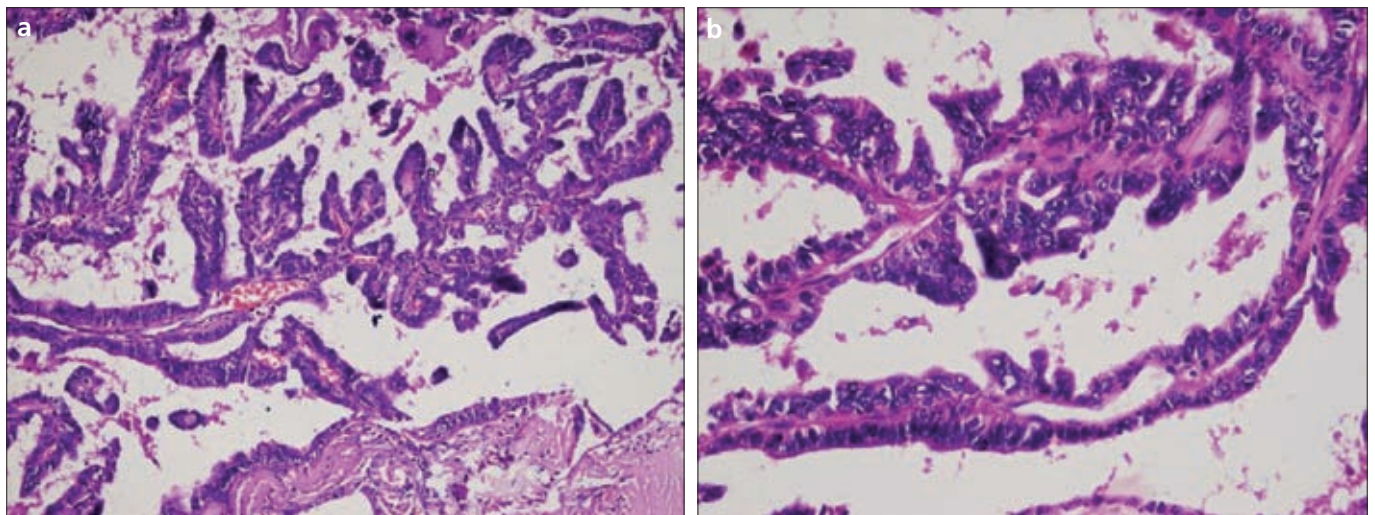
ponents. Post-contrast images demonstrated contrast enhancement in the solid components. Macroscopic calcifications and hemorrhage were not seen in the mass. There was no pathological lymph node enlargement around the lesion and in the neck.

The differential diagnosis of papillary cystadenocarcinoma includes mucoepidermoid carcinoma, acinic cell carcinoma, salivary duct carcinoma, nasal adenocarcinoma, and metastatic carcinoma. It can be distinguished by histopathological findings and histochemical staining.

Histologically, cellular pleomorphism, numerous mitoses, nuclear hyperchromatism, and numerous prominent nucleoli have been reported (9). Although the vast majority of cystadenocarcinomas are low-grade lesions, some are high-grade histological



**Figure 3.** a–d. Post-contrast axial (a, b) and coronal (c, d) T1-weighted MR images show contrast enhancement in solid components (arrows, a–c) of the lesion.



**Figure 4.** a, b. Histologically, the lesion shows papillary projections with cystic lumens (a, HE, x200). Cells of the papillary projections show pleomorphism and nuclear atypical form (b, HE, x400).

malignancies, which are divided into well- and poorly-differentiated tumors. Recurrence and nodal metastases have been observed with the poorly differentiated subtype (10). Intermedi-

ate-grade histological malignancy, as in our case, has been described in the literature; that case showed moderate nuclear pleomorphism in addition to the infiltrative growth pattern (11).

Most of such masses can be managed by surgical excision. Prognosis is related to the histologic grade of the tumor (10).

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