Clinical Case Report

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Intraductal papilloma arising from the accessory parotid gland

A case report and literature review

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Abstract

Rationale: Intraductal papillomas of the accessory parotid glands are extremely rare benign tumors that are most commonly derived from minor salivary glands and are easily misdiagnosed as other diseases. Studying these lesions by pathology and immunohistochemistry can raise awareness of the disease, reduce the rate of misdiagnosis, and provide more precise treatments.

Patient concerns: A 35-year-old man first presented to our hospital with a 6-month history of a painless mass on his left parotid gland.

Diagnoses: The patient was diagnosed with intraductal papilloma of the accessory parotid gland by pathology and immunohistochemistry.

Interventions: The mass was completely resected.

Outcomes: After 2 years of postoperative follow-up, the patient recovered well without recurrence.

Lessons: Intraductal papilloma of the accessory parotid gland is very rare, and can easily be misdiagnosed as sialadenoma papilliferum, inverted ductal papilloma, or papillary cystadenoma, among others. It is necessary to analyze its pathology and immunohistochemistry in comparison with other diseases. Early excision and long-term follow-up are necessary to provide optimal treatment and to better understand the pathological processes of intraductal papilloma.

Abbreviations: CK-L = cytokeratin L, CT = computed tomography, FNA = fine needle aspiration biopsy, MRI = magnetic resonance imaging, PAS = periodic acid-schiff, SMA = smooth muscle actin.

Keywords: accessory parotid, intraductal papilloma, salivary gland

1. Introduction

Ductal papillomas are benign papillary neoplasms derived from the neoplastic proliferation of any epithelial layer of the ductal system. The minor salivary gland is more frequently involved than the major salivary gland.[1] The common sites of involvement, in descending order of frequency, are the lip, palate, buccal mucosa, parotid gland, and submandibular glands.[2] According to the cell type, morphology, and location along the salivary duct unit, ductal papillomas are characterized by 3 patterns, namely sialadenoma papilliferum, inverted ductal papilloma, and intraductal papilloma. Sialadenoma papilliferum is a papillomatous exophytic lesion that extends above the adjacent mucosa surface. Inverted ductal papilloma is submucosal and expands or pushes into the surrounding tissue. Intraductal papillomas seem to arise in the duct system more distant from the mucosal surface and present as well-defined unicystic lesions. Intraductal papillomas of salivary glands are extremely rare benign tumors that are most commonly derived from minor salivary glands.[3] Those derived from the major salivary gland are even rarer. Here, we first report a patient with an intraductal papilloma that presented as an accessory parotid mass.

2. Case report

A 35-year-old man with a history of smoking presented with a 6-month history of a painless mass on his left parotid gland. The patient had no family history or previous treatment. An initial physical examination revealed a 1.5×1.0-cm sized, ovoid, movable, firm, and nontender mass on the left parotid area. There was no facial weakness. Ultrasound examination revealed a well-circumscribed hypoechogenic simple cystic lesion of the left parotid gland measuring approximately 1 cm in the greatest diameter (Fig. 1). The patient did not undergo fine needle

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Ethical statement: This case report did not involve human experiments and did not require ethical approval. The professor of Yu Guangyan and patient consent was obtained.

The authors declare that there are no conflicts of interest.

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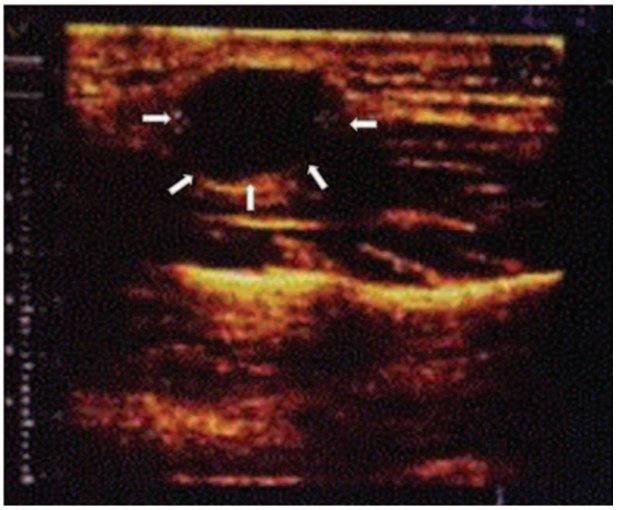

Figure 1. Ultrasound examination revealed a well-circumscribed hypoechogenic simple cystic lesion of the left parotid gland.

aspiration biopsy (FNA), computed tomography (CT), or magnetic resonance (MR) clinical examinations. According to the clinical symptoms and an oral examination, the patient was diagnosed with a cystic tumor of the parotid gland before surgery. Complete blood cell count, urinalysis, blood chemistry, coagulation studies, liver function test, and chest x-ray imaging all provided results within normal limits.

The patient underwent a systemic examination without surgical contraindications. Under general anesthesia, a conventional S incision was made in the parotid gland, which extended upward to 2 cm above the ear. The skin, subcutaneous tissue, and platysma were incised, and the incision went down to the fascia of the parotid masseter. When the flap was opened, parotid, accessory parotid, and accessory parotid gland tumors were revealed. The parotid ducts and facial nerve buccal branches were isolated, and then the accessory parotid gland and its tumor were removed together.

Examination of an intraoperative frozen section revealed a left accessory parotid cyst with partial papillary hyperplasia. The resected specimen showed a well-circumscribed $2.5 \times 1.5 \times 1.0$-cm cyst with clear fluid within its lumen. Microscopically, the cystic tumor had a well-defined outline. The cystic lumen showed evidence of papillary tumor cell proliferation (Fig. 2A). The tumor surface was lined with a single layer of cuboidal epithelial cells (Fig. 2B). Immunohistochemical staining of the resected tissue specimen showed strong positivity for S-100, CK-L, and PAS, and partial positivity for smooth muscle actin (SMA; Fig. 3). These results strongly suggested that the tumor cells were derived from ductal luminal cells. Based on the location of the tumor, plus histopathological and immunohistochemical analyses, the tumor was diagnosed as an accessory parotid gland intraductal papilloma. After 2 years of postoperative follow-up, the patient recovered well without recurrence.

3. Discussion

Accessory parotid gland tumors are rarely found in the clinical setting, accounting for approximately 1% to 7.7% of all parotid gland tumors.[4,5] This is the first report of an accessory parotid gland tumor that developed in a major salivary gland. The first papilloma of the parotid gland was reported by Abrams and Finck,[6] who reported a rare large neoplasm involving the parotid

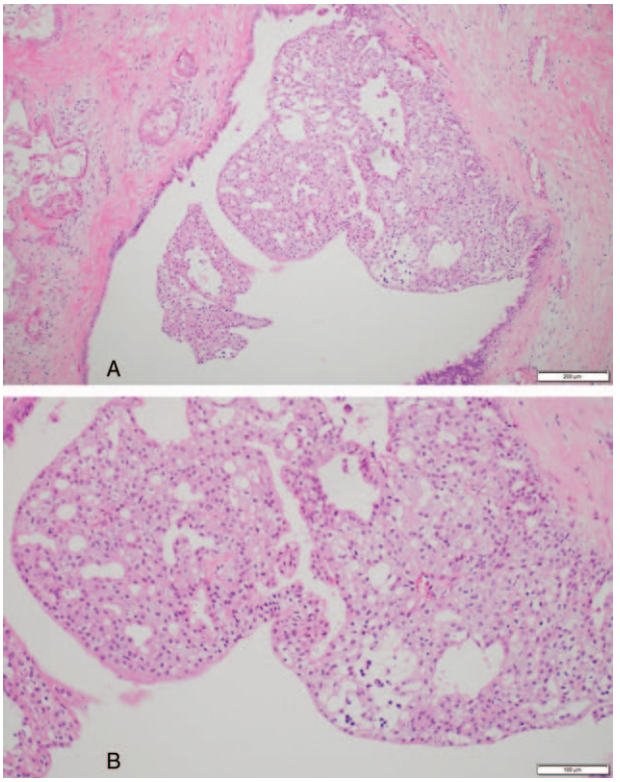

Figure 2. Histopathological features of accessory parotid tumor. (A) The cystic lumen is filled with papillary tumor proliferation (magnification, 100×). (B) The tumor surface is lined with a single layer of cuboidal epithelial cells (magnification, 200×).

gland that extended through the overlying skin. Salivary intraductal papilloma is a subtype of ductal papilloma and considered a rare benign salivary gland tumor.[7] The disease affects patients 8 to 77 years of age, but on average, those aged 50 to 70 years are most likely to be affected. Sex distribution is equal on average.

Clinically, intraductal papilloma of the salivary gland presents as a painless, solitary, small mass. Making an accurate diagnosis of this tumor before surgery is difficult. Warthin tumor, papillary cystadenocarcinoma, papillary cystadenoma, and polymorphous low-grade carcinoma may all present with papillary cystic lesions.[8] According to previous reports, this tumor has been misdiagnosed in cytologic specimens as a cystic tumor of the salivary gland, skin adnexal tumor, basal cell adenoma, and papillary cystadenoma by conventional techniques.[9,10] Ultrasonography cannot provide useful information on this tumor.[11] CT and MRI are very helpful for the assessment of the lesion's location and extension.[12] FNA seems to be the best method for preoperative diagnosis of parotid tumors. Hara et al[13] have described the cytological features of this tumor using FNA. However, there is some uncertainty regarding FNA and whether it is helpful for the diagnosis of intraductal papillomas. Radiography and FNA offer little help in preoperative diagnosis, because intraductal papillomas are very rare and small.[14,15] Histological diagnosis is typically necessary for intraductal papilloma of the major salivary glands.

Characteristically, a salivary gland papilloma consists of papillary intraductal protrusions with connective tissue cores that extend into the tumor's epithelial lining, with single-and double-layered arrays of cuboidal and columnar cells. The lumens are partially or completely filled with many branches of the papillary

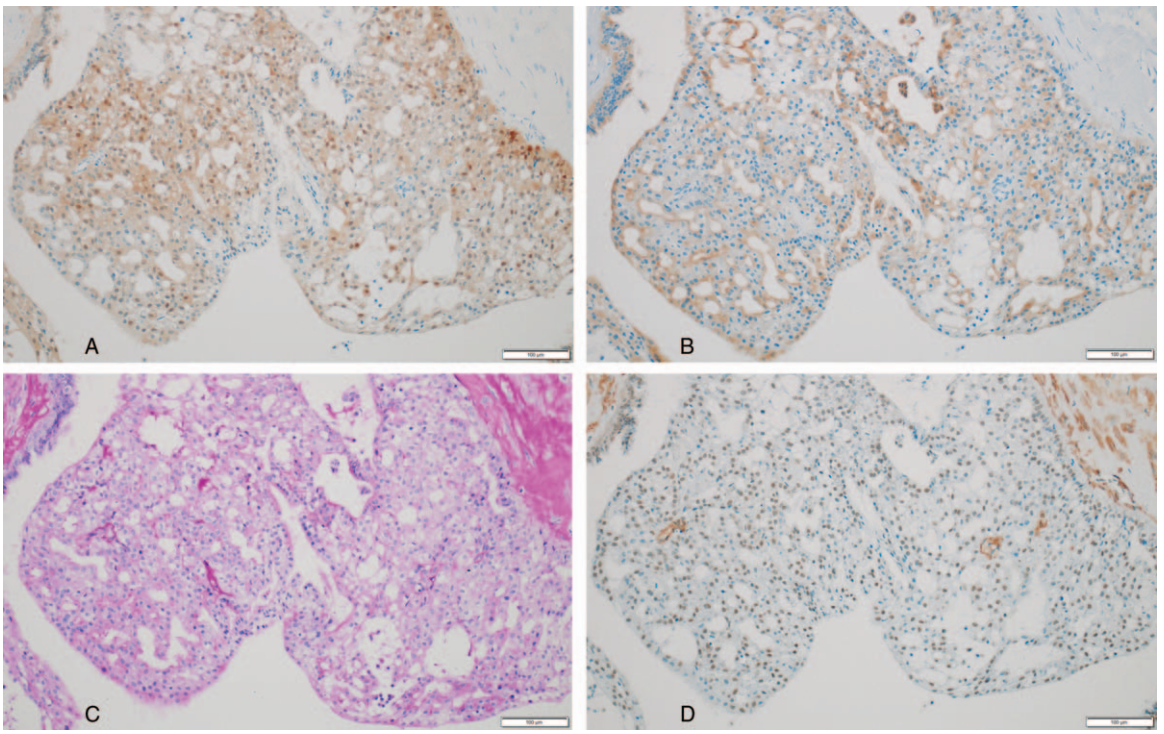

Figure 3. Immunohistochemically, (A) the tumor cells were positive for S-100, (B) positive for CK-L, (C) positive for PAS, and (D) partially positive for SMA (magnification, 200×). PAS = periodic acid-schiff, SMA = smooth muscle actin.

frond, which is covered with a columnar epithelium and sustained by thin cores of fibrovascular tissue. Epithelial cells typically do not have nuclear atypia. Immunohistochemical staining of tumor cells reveals strong positivity for the expression of cytokeratin (CK), an epithelial membrane antigen; partial positivity for the expression of S-100, and negative expression of SMA, P63, or CK14. Nipple surface cells and cyst lining cells have the same immunohistochemical expression.[16] Combining pathology with immunohistochemistry can facilitate making a precise diagnosis while preventing misdiagnosis. While intraductal papillomas of the salivary glands are benign, a small number of parotid intraductal papillomas are malignant.[17,18] Complete resection leads to a good prognosis, and is generally not followed by recurrence or metastasis. Early complete excision is necessary and provides patients with curative treatment. If the tumor area has cellular atypia and mitotic figures, then long-term follow-up of patients is advised.

4. Conclusion

We presented a rare case of intraductal papilloma of the accessory parotid gland. Because it is difficult to diagnose intraductal papillomas, our report adds value to the literature as it can help practitioners in their diagnostic assessments. Studying the pathology and immunohistochemistry of these lesions can raise awareness of this disease, reduce the rate of misdiagnosis, and provide more precise treatments. Complete excision and long-term follow-up is necessary to help us fully understand intraductal papillomas.

Author contributions

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