Case Report

Papillary cystic variant of acinic cell carcinoma presenting as parotid tail tumor

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Abstract

Acinic cell carcinoma (ACC) is an uncommon low-grade tumor of the salivary glands that constitutes 2.5–4% of parotid gland tumors. Papillary cystic variant (PCV) of ACC is even rarer and can be diagnosed on histopathological examination only. It is important to diagnose this variant as it carries a poor prognosis when compared with other variants of ACC and is known to be universally fatal in 10 years. The present case describes ACC-PCV in a 20-year-old male, which presented as a slow growing parotid tail tumor and was misdiagnosed as a benign lesion both cytologically and radiologically. This case emphasizes the importance of histopathological examination in parotid masses as well as the need to consider malignant lesion in the differential diagnosis of a parotid tail tumor.

Key words: Acinic cell carcinoma, papillary cystic variant, parotid tail tumor
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Introduction

Acinic cell carcinomas (ACC) are usually low grade, uncommon neoplasms constituting 2.5–4% of parotid gland tumors.[1] Papillary cystic variant (PCV) of ACC is a rare tumor, the diagnosis of which is based on histopathological examination. It is composed of tumor with papillary and cystic growth patterns, with varying proportions of one or more cell types.[2] It has mostly been reported in younger patients (16–40 years) when compared to the classic type that characteristically presents in the fifth decade of life.[3] Although an uncommon tumor, it is important to recognize this variant as it has proved to be universally fatal within 10 years.[4] We hereby report an unusual case of ACC-PCV in a 20-year-old male which was considered a benign parotid mass both cytologically and radiologically.

A 20-year-old male presented to the surgical clinic with a complaint of swelling on the left angle of mandible below the ear lobe since 4 months. Swelling was gradually increasing in size. It was 3.5 cm × 2 cm in size, firm to hard with limited mobility and nontender. Skin over the swelling was not fixed. Facial nerve function was normal. His general and systemic examination was normal. Fine needle aspiration cytology done outside was suggestive of a benign cystic lesion. Laboratory investigations revealed hemoglobin 16.2 gm%, total leucocyte count 6,800 cells/µl, differential leucocyte count – neutrophils 74%, lymphocytes 18%, eosinophils 5%, monocytes 3%, platelet count - 1.15 lac/µl and blood sugar 120 mg/dL. Contrast-enhanced computed tomography of neck showed a soft tissue nodular lesion isodense to the parotid (attenuation 40 HU) of size 2.3 cm × 1.6 cm × 1.3 cm with mildly enhancing peripheral walls in the left superficial parotid gland. Few spiculated nodules were also seen in the right lung apart from patches of numerous centriacinar nodules. No obvious significant cervical adenopathy was noted. Both lobes of the thyroid were normal in size, shape, attenuation and enhancement.

Superficial/nerve-sparing parotidectomy was planned. Grossly, the specimen measured approximately 6.5 cm × 4.5 cm × 3.5 cm with attached flap of skin measuring 3.5 cm × 1.5 cm. Cut surface showed a large

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cyst measuring 2.8 cm × 1.5 cm filled with dark brown material [Figure 1]. The cyst was 0.4 cm away from deep resected margin. Histopathological examination showed a large well-circumscribed cystic space lined by numerous papillary projections few of them showing thin vascular cores [Figure 2]. Wall of the cystic cavity was fibro collagenous. Many hemosiderin-laden macrophages and areas of hemorrhage are also seen [Figure 3a]. These papillae were mostly lined by hobnail cells with round, vesicular nuclei, central nucleoli and eosinophilic to vacuolated cytoplasm [Figure 3b]. No atypical mitosis or necrosis was seen. There was no evidence of capsular or vascular/perineural invasion. Adjacent to the tumor normal salivary gland tissue was seen. All the margins were free of tumor. A histological diagnosis of ACC-PCV was made.

**Discussion**

Acinic cell carcinoma is an uncommon salivary gland tumor, making up 1% of all salivary gland neoplasms. They most often arise in the parotid gland, but may occasionally involve the submandibular, minor salivary or seromucinous glands.[3] ACCs are more frequently diagnosed in the fourth to sixth decades of life. It is the least aggressive of salivary gland cancers with low malignant potential, but several recurrences and metastasis have been reported.[6] Few high-grade variants of ACC are known such as papilloctytic carcinoma or carcinomas with undifferentiated cells in the medullary pattern.[7]

Lesions of the “tail” of the parotid gland are difficult to assess clinically and provide a diagnostic dilemma on imaging.[5] Most benign parotid tumors present as slow-growing, painless masses often in the tail of the parotid gland. Hamilton et al. defines the “tail” of the parotid gland as inferior 2 cm of the superficial lobe of the gland.[9] In a study by Hamilton on 117 parotid tail masses, seventeen types of parotid tail masses were identified out of which benign lesions were the most common. Only one case of ACC was identified. In our case also as it was a slow-growing painless mass present in the parotid tail, clinically it was thought to be a benign parotid mass which was also supported by cytological examination. Accurate localization of these lesions on imaging is essential to assist the clinical diagnosis and to prevent inadequate/incomplete excision and complications, especially damage to the facial nerve.

A variety of morphological and cytological appearances are seen in ACCs. The diagnosis of PCV of ACC usually poses a challenge because of the cytoarchitecture that is different from classic type. Cystic fluid in the case of ACC-PCV dilutes the overall cellularity leading to a mistaken diagnosis of benign lesion as was seen in the present case.[2]
Histopathologically, ACC shows a myriad of architectural patterns: Solid, solid-lobular, acinar-microcystic, papillary cystic, tubuloductal, follicular and macrocystic, and dedifferentiated. The solid and microcystic are the most common subtypes and the PCV accounts for one-fourth of ACC. ACC-PCV is histologically composed of tumor with papillary and cystic growth patterns. Varying proportions of one or more of five cell types are seen including hobnail, acinar, intercalated, vacuolated, nonspecific glandular and clear cells. Hobnailing or tombstoning of luminal cells is characteristic of the PCV. This is due to bulging of the apical portions of lumen lining cells into the lumen presumably after release of secretions. The clinical picture is not specific, and diagnosis is based on the histopathologic examination.[10]

The mean age of occurrence is in the fifth decade, but the PCV is reported to occur in younger patients compared to the classic type. Females are involved more commonly unlike in our case. The most significant differential diagnosis of ACC-PCV is papillary carcinoma of the thyroid. Thyroid ultrasonography, hormonal assays, and immunohistochemistry for thyroglobulin are helpful in differentiating these two lesions. Other less significant differential diagnosis includes cystadenocarcinoma and polymorphous low-grade adenocarcinoma. Timely diagnosis and treatment of ACC-PCV is essential as it has been found to be universally fatal within 10 years. Although most parotid masses are benign, removal is required for histopathologic confirmation because of the clinical and radiologic overlap.

To conclude, the present case emphasizes the importance of histopathological examination in parotid masses as the imaging features as well as cytology lack the necessary specificity to differentiate benign from malignant masses, especially in a case of papillary cystic ACC. It also highlights the need to consider malignant lesion in the differential diagnosis of parotid tail tumor.

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