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Primary Cutaneous Apocrine Carcinoma of the Neck Involving the Parotid Gland: A Case Report and Review of the Literature

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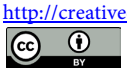


Abstract

Introduction: Primary cutaneous apocrine carcinoma (PCAC) is a rare skin malignant tumor that originates from areas with a high concentration of apocrine glands. The incidence of PCAC in the neck is relatively low. The age of onset in PCAC ranges from 5 to 70 years old. Clinically, PCAC typically shows up as painless nodules or lumps. In immunohistochemistry, positive expression of CK-7, GCDFP-15, AR, and myoepithelial markers are helpful in the diagnosis of PCAC. This case report pertains to a 59-year-old male of Asian descent. Five years ago, a painless mass was discovered behind his right ear, which grew slowly. This patient was identified as PCAC based on clinical symptoms, pathology, immunohistochemistry, and imaging characteristics. PCAC involved the parotid gland in this case; it is easy to be misdiagnosed as a primary malignant tumor of the parotid gland on imaging. Furthermore, a thorough set of clinical, imaging, pathological, and immunohistochemical examinations must be performed to make a diagnosis because it is challenging to differentiate PCAC from metastatic breast carcinoma. **Conclusion:** A well-developed multidisciplinary examination is essential because PCAC can be challenging to diagnose and differentiate.

Keywords

Primary Cutaneous Apocrine Carcinoma, Head and Neck Tumors, Diagnostic Imaging

1. Introduction

Primary cutaneous apocrine carcinoma (PCAC) is a rare malignant skin tumor originating from areas with high concentration of apocrine glands. Although it

can also develop on the scalp, forehead, eyelids, scalp, vulva, inguinal area, axilla, chest wall, and other places, the armpit is the most prevalent [1] [2] [3]. However, PCAC that occurs in the neck is relatively rare. PCAC mainly occurs in Caucasians, with no discernible gender difference. PCAC can begin anywhere between the ages of 5 and 70, with a peak incidence occurring between 60 and 70 [4] [5]. PCAC is typically characterized by a solitary, painless, and gradually enlarging nodule. Due to its slow development, it is often mistaken for a benign lesion. In rare cases, PCAC exhibits a high level of invasiveness and rapid progression [4]. It is challenging to distinguish from metastatic breast carcinoma due to shared apocrine and immunohistochemical characteristics, such as the expression of CK7, GCDFP-15, GATA3, ER, PR, and mammaglobin [6]. This article reports a case of PCAC involving the parotid gland and discusses its differentiation from metastatic breast carcinoma and primary malignant tumors of the parotid gland. We have obtained all appropriate patient consent forms.

2. Case Report

A 59-year-old male patient visited the outpatient clinic due to the discovery of a tumor in the right parotid region for five years. Because the mass grew slowly and without causing pain, the patient did not initially give it much attention. A month ago, herbal medicine was applied externally, resulting in skin ulceration on the surface of the mass. Facial asymmetry is noticeable in the current physical examination. The mass is visible both in front of and behind the right earlobe. The mass in front of the ear is round, approximately 2.0 cm × 1.0 cm, while the one located behind the ear is lobulated, measuring about 1.5 cm × 2.5 cm. The mass involves the earlobe and a portion of the auricular cartilage with a raised and nodular surface and a relatively firm adhesive base. The skin on the mass surface is brownish-black, with slight ulceration and scab formation on the skin behind the ear. Several dark brown mole-like lesions can be seen around the ear. This patient was male, and there was no palpable mass in his breast, with no localized protrusion of the breast skin observed. MRI examination was conducted at our institution. The head and neck MRI revealed a mass in the right neck, exhibiting an hypointense signal on T1-weighted images and a high signal on T2-weighted images. The mass showed infiltrative growth at its periphery, involving the right parotid gland and earlobe. It presented as a local cauliflower-like protrusion towards the earlobe. The contrast-enhanced MRI showed that the lesion had pronounced enhancement with a small, patchy, and non-enhanced area (**Figure 1**). The patient underwent tumor resection. A wide local excision of the lesion was done in the supine position under general anesthesia. The resection area was 0.5 cm beyond the tumor margin. Grossly, the pathology report suggested a lesion with subcutaneous tissue measuring 3.8 cm × 2.1 cm × 2.0 cm. After surgery, two days of negative pressure drainage and five days of traditional anti-infection therapy were administered. The postoperative wound healed satisfactorily. Microscopically, The neoplastic cells exhibited glandular differentia-

tion and had abundant eosinophilic cytoplasms. Vascular and neuronal invasions were present (Figure 2(a)). Immunohistochemical studies showed that the tumor cells stained diffusely positive for gross cystic disease fluid protein-15 (GCDFP-15) (Figure 2(b)), cytokeratin-7 (Figure 2(c)), androgen receptor (AR) antibodies (Figure 2(d)) and Ki67. P63, S100, SOX10, P40, CDX2, and Cytokeratin (CK5/6) stains were negative. After excluding the evidence of breast and visceral malignancy, the diagnosis of PCAC was made based mainly on the histopathologic and immunohistochemical features.

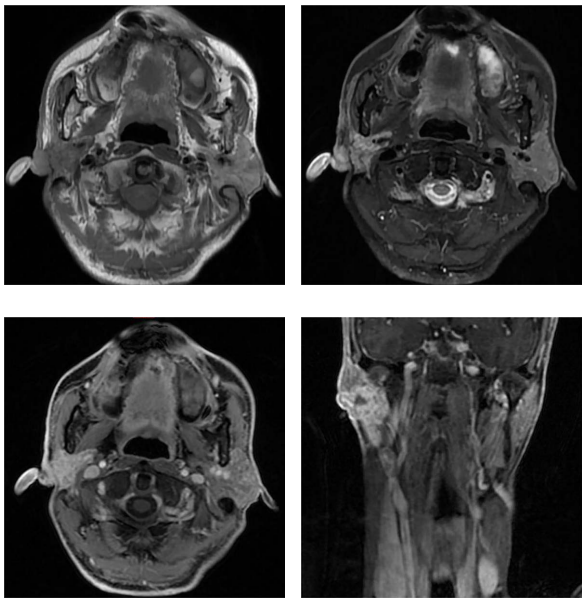
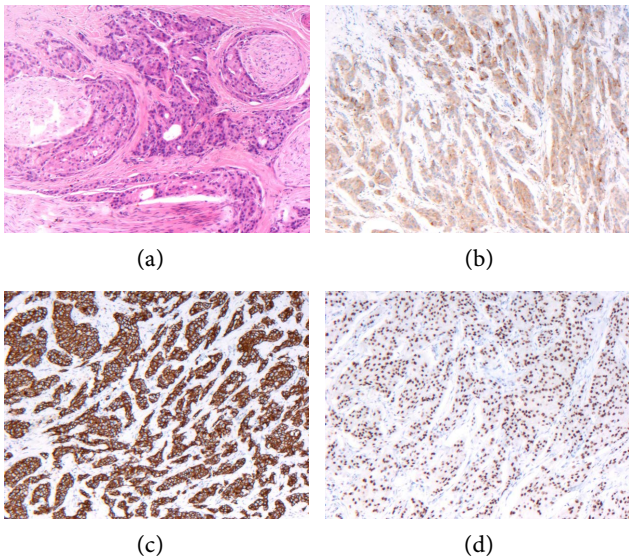


Figure 1. MRI of the head and neck showed that the right neck mass involved the parotid gland.



(a) (b)

(c) (d)

Figure 2. Immunohistopathological examinations showed: (a) tumors cells exhibiting glandular (H & E, ×100), (b) gross cystic disease fluid protein-15 (GCDFP-15, ×100), (c) cytokeratin-7 (CK7, ×100), (d) androgen receptor (AR, ×100).

3. Discussion

PCAC is a rare malignant skin tumor. Its occurrence may have associations with benign lesions such as apocrine adenoma, apocrine hyperplasia, and nevus sebaceous. Certain scholars propose that these conditions could be precursors to apocrine adenocarcinoma [2] [6] [7]. Nonetheless, due to its rare occurrence, its etiologic factors and pathogenesis are poorly understood. Case reports are its primary source of information. In histology, well-differentiated PCAC exhibits distinctive features, including cells with granular eosinophilic cytoplasm, luminal apocrine secretions, and glandular structures. Therefore, the diagnosis of well-differentiated cases is relatively straightforward. However, poorly differentiated PCAC can exhibit similarities in various metastatic cancers, which can complicate the differentiation, particularly with metastatic mammary carcinoma. Clinically speaking, there are no distinctive features that can assist in distinguishing these two entities. Additionally, immunohistochemistry's ability to differentiate is limited. The expression of myoepithelial markers (p63, p40, CK 5/6, SOX-10) can aid in differentiating between PCAC and metastatic breast carcinoma [8] [9]. However, all myoepithelial markers in this case were negative, inconsistent with most previously reported results. One of the nine PCAC cases in the case report published by Portelli *et al.* tested negative for every myoepithelial marker [8]. GATA-binding protein 3 (GATA3) can be expressed in PCAC and metastatic breast carcinoma [10]. Some scholars believe that the negative expression of GATA3 can be a stronger indication that the case is a primary lesion [2]. In a study involving 33 patients with metastatic breast cancer and 18 patients with primary cutaneous sweat gland carcinoma, GATA3 immunoreactivity was expressed positively in 91% of metastatic breast cancer cases, when 71% were positive for sweat gland carcinoma [11]. It is clear that GATA3 has a limited role in differentiating PCAC from metastatic breast cancer. It is hoped that more information about the value of immunohistochemistry in differentiating PCAC from metastatic breast carcinoma will be found in the future. GTAT3 was not tested in this case. Currently, there is a shortage of diagnostic criteria for poorly differentiated PCAC [6]. For most cases, positive results were observed in GCDFP-15, CK7, and AR staining [12]. Among these markers, GCDFP-15 is significant in indicating that the tissue originates from apocrine glands or exhibits apocrine gland differentiation. It proves to be valuable for both diagnosis and differential diagnosis of PCAC. In this case, the positive staining of GCDFP-15, CK7, and AR aligns with previous literature reports and serves as solid evidence in favor of the diagnosis of PCAC. Therefore, a comprehensive evaluation incorporating patient history, imaging examinations, histopathology, immunohistochemistry, and other examinations is essential for confirming the diagnosis.

Given the rare occurrence of this disease, there are relatively few studies on its imaging characteristics at present. Most previous cases have focused on the imaging analysis of PCAC in the scalp. This case occurred in the neck and involved the parotid gland, necessitating differentiation from the primary malignant tumor of the parotid gland. Primary malignant tumors of the parotid gland are

rare, with an incidence rate of 2 - 3 cases per 100,000 people. The most common type is mucoepidermoid carcinoma, while others encompass squamous cell carcinoma, adenoid cystic carcinoma, adenocarcinoma, and so on. Mucoepidermoid carcinoma of the parotid gland is more common in young and middle-aged women. The tumor's boundaries are clear, with single or multiple cystic areas in low malignancy-grade cases. In contrast, the morphology of cancer in high-grade patients is irregular, with unclear boundaries and pronounced enhancement on contrast-enhanced scans, making it challenging to differentiate between mucoepidermoid carcinoma and this case on imaging. However, the peak incidence of cutaneous apocrine carcinoma occurs in individuals aged 60 - 70 years, whereas mucoepidermoid carcinoma is more common in young and middle-aged patients. In addition, it is essential to differentiate this case from squamous cell carcinoma, a rare cancer characterized by features such as a large size, irregular shape, unclear boundaries, central necrosis, and other distinct characteristics. Central necrosis can serve as a characteristic imaging sign of squamous cell carcinoma.

Because PCAC is uncommon, there is a lack of predictive measures as well as standardized management for this disease. Since most lesions are considered low-grade malignancies, wide local excision is the preferred primary treatment when the margins of the lesion are clear and there are no regional lymph node metastases. Patients with positive regional lymph node metastasis can undergo neck lymph node dissection. Radiotherapy or chemotherapy can be used as adjuvant therapy in exceptional circumstances (such as positive margins, positive lymph nodes, and recurrent and metastatic tumors) [13]. The standardization of surgical margins is still controversial, with some experts suggesting a surgical margin of 1 - 2 cm can achieve satisfactory results [14].

4. Conclusion

Due to its rarity and lack of distinct clinical, pathologic, immunohistochemical, and imaging characteristics, PCAC in the neck is difficult to differentiate from metastatic mammary carcinoma. On imaging, it is challenging to distinguish PCAC from parotid primary malignancy when PCAC infiltrates the parotid gland. Based on immunohistochemistry results and a history of malignancy, PCAC can be diagnosed more precisely. Treatment for PCAC of the neck is multidisciplinary, with surgical intervention often yielding positive outcomes. Early diagnosis and management are key factors that influence the prognosis of this condition.

Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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