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Case Report

Adenoid cystic carcinoma of the parotid gland ☆

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ABSTRACT

Adenoid cystic carcinoma (ACC) is a slowly progressing malignant tumor of the salivary glands that mostly affects minor salivary glands. ACC of parotid gland is exceptionally rare. In this article, we aimed to provide a case report of parotid gland ACC in a 55-year-old female that was misdiagnosed as benign mixed tumor. The patient was successfully treated by surgery without any complications. We recommend that ACC of parotid gland should be considered in differential diagnosis of benign mixed tumor since there still existed overlapped imaging characteristics.

Introduction

Robin and Laboulbene published the first description of adenoid cystic carcinomas (ACC) in 1852 [1]. Adenoid cystic carcinomas account for around 1% of the oral and maxillofacial area; however, they are less common in all locations with secretory glands (such as the breast, cervix, colon, prostate, etc.) Although it can develop in any salivary gland location, about 50%-60% do so in the smaller salivary glands [2]. The ACC makes up just 2%-3% of all tumors in the parotid gland, making it very uncommon [3]. The majority of female experience the highest occurrence of ACC between the fifth and sixth decades of life [4]. Due to its modest size and gradual development, which conceals its widespread subclinical penetration aggressively into neighboring structures, it is clinically misleading. The 3 histopathologic patterns of ACC are solid, cribriform, and tubular [1–4]. Herein, we intended to describe a 55-year-old female with ACC of parotid gland that misdiagnosed as a benign mixed tumor.

Case Presentation

The main complaint of a 55-year-old female was swelling and mild discomfort on the right side of the parotid gland area, which had been present for one month. Five years prior, the parotid gland tumor was first noted by the patient. After that, she just required yearly examinations and follow-ups. The edema had grown larger during the previous month, causing skin redness and agony. The patient had no prior history of fever or other general health issues.

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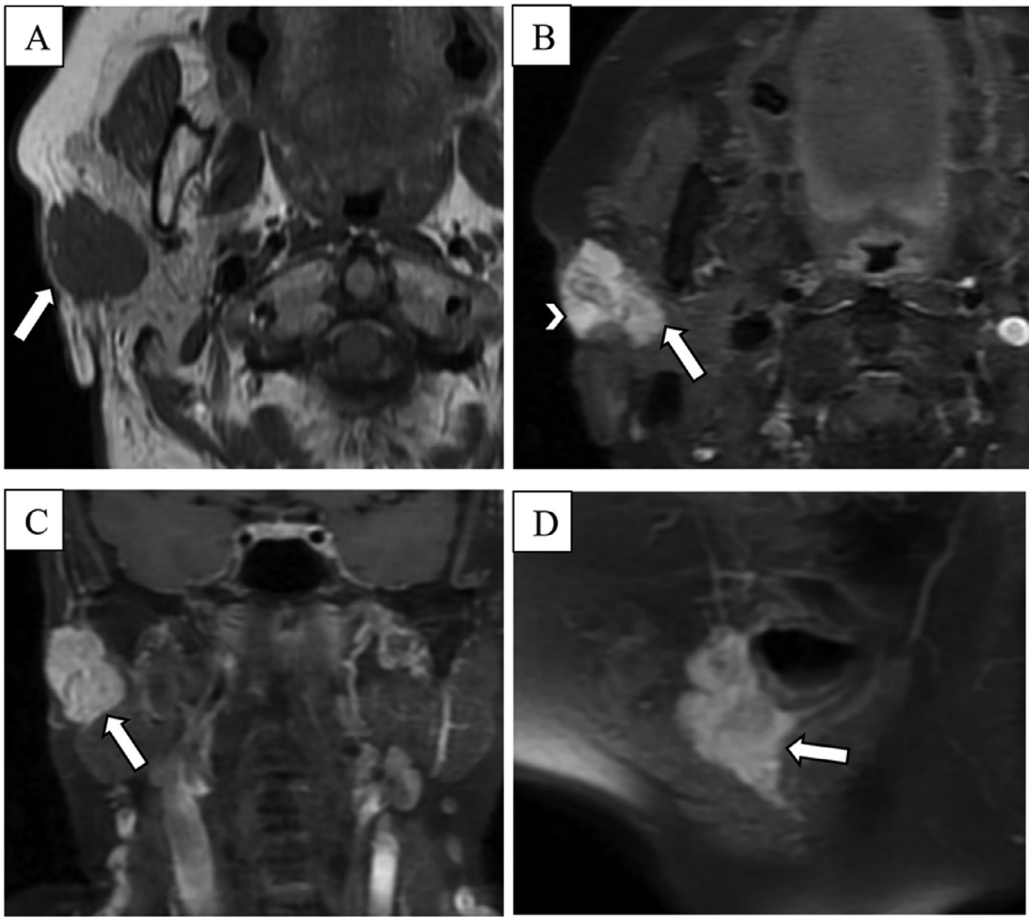

Fig. 1 – A solid mass located in the right parotid gland, low signal intensity on axial T1-weighted image (arrow, A). The mass appears hyperintensity on axial T2-weighted fat-saturation image (arrow, B), and invasion to skin and subcutaneous fat in front of the right auricle (arrow head, B). Axial contrast-enhanced fat-suppressed T1-weighted MR image shows homogeneous enhancement of the mass (arrow, C and D).

Upon physical examination, a mass that was adhered to the skin above was found to be hard and immobile. The skin that covered the tumor had a glossy, erythematous appearance. It was 3 cm superior-inferior and 3.5 cm antero-posterior diameters. The facial nerve was operating normally. There were no larger lymph nodes seen by palpation. No abnormality linked to the edema was seen intraorally. The results of the laboratory tests were within normal bounds.

Magnetic resonance imaging identified a neoplasm located in the superficial lobe of the right parotid gland (Fig. 1). Lesions have well-defined, multilobulated margins and spread to the skin and subcutaneous fat in front of the right auricle. The size of the mass was 34 mm × 22 mm. A homogeneous solid mass showed low signal intensity on T1-weighted imaging (Fig. 1A), appeared hyperintensity on T2-weighted fat-saturation imaging (Fig. 1B), and showed homogeneous enhancement on contrast-enhanced fat-suppressed T1-weighted imaging (Figs. 1C and D). In diffusion-weighted imaging, this showed appeared subtle hyperintense (Fig. 2A), and showed isointense on apparent diffusion coefficient (ADC) imaging (Fig. 2B). There were no signs of hemorrhage or invasion related to the mass. The mean ADC values of the mass and normal parotid gland were 1.2 and 1.49 × 10^{-3} mm^2/s, respectively (Fig. 2). MRI identified no pathological lymph nodes.

Based on clinical and radiological data, the preliminary diagnosis was a benign mixed tumor. The primary surgical treatment for the patient consisted of a total parotidectomy. However, after a radical dissection, the histopathological evaluation of the excised sample tissues was fully congruent with an ACC of the parotid gland. She experienced no complications during postoperative healing. One month after surgery, postoperative radiation therapy (60 Gy; 2 Gy, twice a day, 5 days a week) was applied to the surgical bed and the right neck area due to the aggressive nature of the tumor. The patient was discharged finally without any adverse events.

Discussion

Less than 1% of cervicofacial malignancies are ACC, a relatively uncommon kind of malignant epithelial tumor. They mostly grow in the paranasal sinuses, accessory and primary

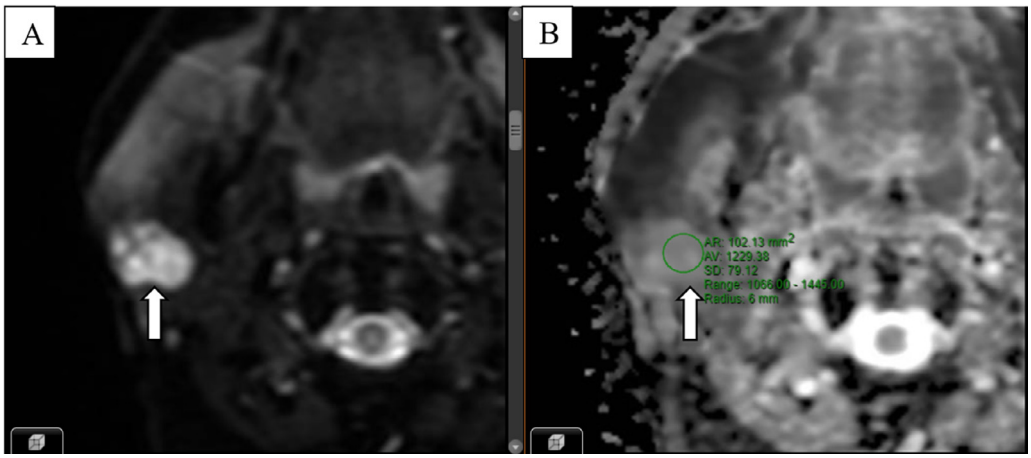

Fig. 2 – A mass (arrow) appeared hyperintense on diffusion-weighted imaging (DWI, A), and showed isointense signal intensity with ADC value on apparent diffusion coefficient imaging (B) images.

salivary glands, lacrimal glands, and mucous glands of the nasal cavity. Of 5% primary salivary gland cancers are these tumors [3]. Outstandingly, ACC of parotid gland is very rare.

Histologically, ACC can be divided into 3 types: cribriform, tubular, and solid. Most cancers exhibit a combination of these patterns. The poorest prognosis may be seen in solid patterns, which are more frequently used to represent mitotic figures. Three grades are assigned to ACC. Grade I is made up of cribriform and tubular designs with no solid regions. Grade II (less than 30%) and Grade III exhibit solid sections (more than 30%). In Grade III, abnormal cells and mitoses are seen. Our instance fell into Grade III since it had a predominately solid pattern (more than 30%) [5,6].

When contrast agents are injected, ACC often shows up as a homogeneously increased mass; however, necrosis may cause heterogeneous enhancement [7]. It can also show up as an ill-defined mass with widespread infiltration of its surrounding tissues. On T2-weighted MRI imaging, the solid and more cellular histological subtype of ACC shows a smaller signal [8]. Salivary gland cancer is characterized by irregular margins, surrounding tissue infiltration, and low intensity in T2-weighted sequences, with diminishing predictive value [9]. Although it can discriminate between pleomorphic adenoma and ACC, ADC has a poor prognostic value for malignancy [10]. Perfusion-weighted imaging sequences improve sensitivity but not specificity of MRI for carcinomas; ACC frequently exhibits a quick wash-in form plateau, which is also typical of pleomorphic adenoma but with substantially lower ADC values [10].

The primary form of therapy for ACC is surgery, however in some instances it has been effectively combined with radiation. Due to its high propensity for recurrence and metastasis if a patient survives long enough, even despite severe excision, ACC is still a very challenging disease to treat [11].

Conclusions

The parotid gland malignant tumor known as ACC is uncommon. ACC is a secretory gland tumor that grows slowly but exhibits a remarkable propensity for local dissemination and recurrence. Surgery is the only effective treatment; however, radiation may be used as an adjuvant or in more severe instances. Because metastasis can appear very slowly, early diagnosis is essential for a better prognosis and quality of life.

Authors' contribution

Ho Xuan Tuan and Nguyen Minh Duc contributed to write original draft. Nguyen-Hoang Thanh Tu and Nguyen Minh Duc contributed to undergo diagnostic procedure, collect, and interpret the imaging. Nguyen-Hoang Thanh Tu and Nguyen Minh Duc made substantial contributions to collect patient data and clinical data analysis. All authors have read, revised, and approved the final published version of the manuscript. All authors were responsible for submission of our study for publication.

Statement of ethics

Ethical approval was not necessary for the preparation of this article.

Data availability statement

All data generated or analyzed during this study are included in this article and/or its online supplementary material files. Further enquiries can be directed to the corresponding author.

Patient consent

Written informed consent was obtained from the patient for publication of this case report and any accompanying images.

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