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Adenocarcinoma, NOS of the Accessory Parotid Gland

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Abstract

Although accessory parotid glands (APGs) in humans are not uncommon, the occurrence of APG malignancy is rare. Here, we report such a rare case of a 41-year-old man with pathologically confirmed adenocarcinoma, not otherwise specified (ANOS) in a left APG. The patient had been aware of an indolent and slowly growing mass in his left mid-cheek area for three years before visiting our clinic. Clinically, the mass showed good mobility, was elastic and hard, had a smooth surface, and was 10 mm long in diameter. Facial paralysis was not recognized. Imaging studies including ultrasonography, computed tomography, and magnetic resonance imaging revealed a well-circumscribed solitary mass, 6 × 12 mm in size, anterior to the front edge of the left parotid gland and on the outer layer of the masseter muscle. The fine needle aspiration cytology result was classified as class III. The mass was extirpated under a tentative diagnosis of a benign tumor of the APG. A standard parotidectomy incision was made for complete and safe exposure of the mass, parotid duct, and facial nerve branches. The parotid duct and facial nerve branches were successfully preserved. The postoperative pathological diagnosis was intermediate-grade ANOS. Because microscopic invasion of the tumor into the surrounding salivary tissue was not observed, no additional therapy was administered and no sign of recurrence was observed two years after surgery. We concluded that the possibility of malignancy must be considered in cases of APG tumor excision, even if preoperative examinations indicate benign lesions.

Keywords: Accessory parotid gland; Mid-cheek mass; Adenocarcinoma NOS; Surgery

Introduction

The accessory parotid gland (APG) commonly exists just anterior to the main parotid gland and along the parotid duct, adhering to the masseter muscle. APGs in humans are not uncommon, with a reported incidence of 21–61% [1]. However, tumors of the APG are rare, accounting for only 1–7.7 % of all parotid gland tumors [2,3]. Because an APG tumor often presents as a mass in the mid-cheek region anterior to the main parotid gland, it may sometimes be misdiagnosed as a subcutaneous tumor.

Adenocarcinoma, not otherwise specified (ANOS) is not common among malignant salivary tumors, accounting for 4.3% to 17.8% of all malignant parotid gland tumors [4], and an APG tumor with an ANOS histology is even rarer. Here, we report a rare case of intermediate-grade ANOS arising from the APG, which was treated by surgery alone.

Case Report

A 41-year-old man was referred to our university clinic by a local physician for further examination of a left mid-cheek mass, suspected to be a benign subcutaneous tumor. The patient had noticed the indolent mass three years previously, which had slowly increased in size. Clinically, the mass in the left mid-cheek area showed good mobility, was elastic and hard, had a smooth surface, and was 10 mm in the greatest dimension. Cervical lymph nodes were not palpable. Facial paralysis was not recognized. The results of laboratory examinations, including serum and urine amylase levels, were within normal ranges. Imaging studies, including ultrasonography, computed tomography (CT), and magnetic resonance imaging, revealed a well-circumscribed and homogeneously well-enhanced solitary mass, 6 × 12 mm in size, anterior to the front edge of the left parotid gland and on the outer layer of the masseter muscle (Figure 1 and 2). These findings suggested a benign tumor derived from the APG, facial nerve, or blood vessel. Ultrasound-guided fine needle aspiration

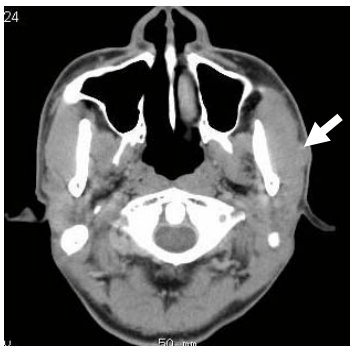

Figure 1: An axial contrast-enhanced computed tomography image showing a faintly enhanced nodular lesion (arrow) located anterior to the front edge of the left parotid gland and on the outer layer of the masseter muscle.

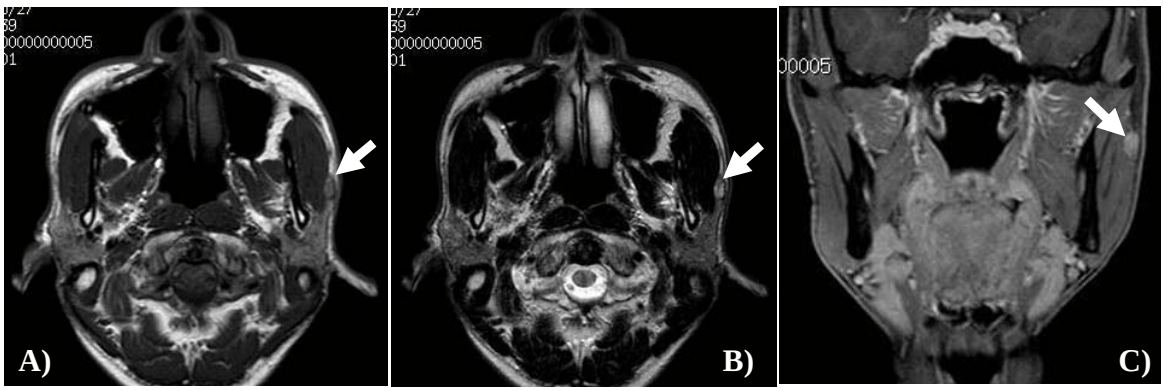

Figure 2: Axial and coronal magnetic resonance imaging scans (a. T1-weighted image b. T2-weighted image c. contrast-enhanced T1-weighted image) revealing a well-circumscribed solitary mass (arrow) with isointensity on a T1-weighted image and hyperintensity on a T2-weighted image, compared to the main parotid gland. The mass was homogeneously well-enhanced on a contrast-enhanced T1-weighted image.

cytology (FNAC) revealed atypical cell clusters, and the result was classified as class III. No remarkable cervical lymphadenopathy was detected. The tentative preoperative diagnosis was a benign APG tumor such as pleomorphic adenoma; however, we could not completely rule out the possibility of a low-grade mucoepidermoid carcinoma. Complete surgical excision of the mass was performed under general anesthesia through a standard parotidectomy incision, i.e., an S-incision, for complete and safe exposure of the mass, parotid duct, and facial nerve branches in the anterior portion of the left parotid gland front edge. The salivary tissue surrounding the mass was completely detached from the main parotid gland and was connected to the parotid duct via a ductule. On intraoperative pathological examination of a frozen specimen, a low- or intermediate-grade ANOS was suspected, and therefore, the entire APG was removed with a 10-mm surgical margin. The parotid duct and facial nerve branches were successfully preserved, and neither facial paralysis nor salivary fistula was observed postoperatively.

Postoperative microscopic examination showed that the tumor was composed of large eosinophilic atypical cells with relatively marked pleomorphism, forming ductal structures (Figure 3). Tumor invasion into the surrounding salivary tissue was not observed. These tumor features did not meet the diagnostic criteria for any other known types of salivary gland carcinomas, and hence, the diagnosis of intermediate-grade ANOS was confirmed, according to World Health Organization classification of salivary tumors published in 2005. Since neither lymph node metastasis nor distant metastasis was detected by ^{18}F-fluorodeoxyglucose-positron emission tomography/CT, the final clinical stage was determined to be stage I (pT1N0M0). No additional therapy, including radiotherapy and chemotherapy, was administered, and no sign of recurrence has been noted two years after surgery.

Discussion

Because APG tumors are rare and often occur in the mid-cheek region, they are often mistaken for subcutaneous tumors, especially in cases of small lesions. According to previous reports, mucoepidermoid carcinoma and pleomorphic adenoma are the most common subtypes of malignant and benign APG tumors, respectively [5]. In this case, the preoperative tentative diagnosis was a benign APG tumor such as pleomorphic adenoma, although we could not completely rule out the possibility of low-grade malignant lesions. However, the postoperative pathological diagnosis was intermediate-grade ANOS.

ANOS is a subtype of malignant epithelial tumors arising in the salivary gland. It is defined as a carcinoma with glandular or ductal differentiation that does not show features characteristic of other known salivary malignant epithelial tumors. ANOS is classified as being of low grade, intermediate grade, or high grade according to the degree of differentiation. Tumor grade and clinical stage may significantly influence the outcome of the tumor [4]. ANOS of the salivary gland commonly occurs in the main parotid gland [6], but rarely does in the APG. The differential diagnoses of ANOS include acinic cell carcinoma, polymorphous low-grade adenocarcinoma, cystadenocarcinoma, salivary duct carcinoma, and metastatic adenocarcinoma. In addition, the possibility of carcinoma ex pleomorphic adenoma must be ruled out when diagnosing ANOS.

Little is known about the clinical behavior and treatment strategies for ANOS of the APG because of its rarity. Therefore, we managed the present case with reference to clinical data on ANOS of the main parotid gland. Recently, Newberry et al. reported a case of ANOS of the APG, measuring 2.0 × 2.0 cm in size, in a 52-year-old man. However, this report did not include information on the tumor grade, and FNAC results were non-diagnostic. The tumor was treated by complete excision with facial nerve dissection and subsequent radiation therapy, and the patient was followed up for three years, without evidence of recurrence [5].

Low- and high-grade ANOS are assigned to low- and high-risk categories, respectively. However, the clinical behavior of intermediate-grade ANOS is controversial [7] and the optimal

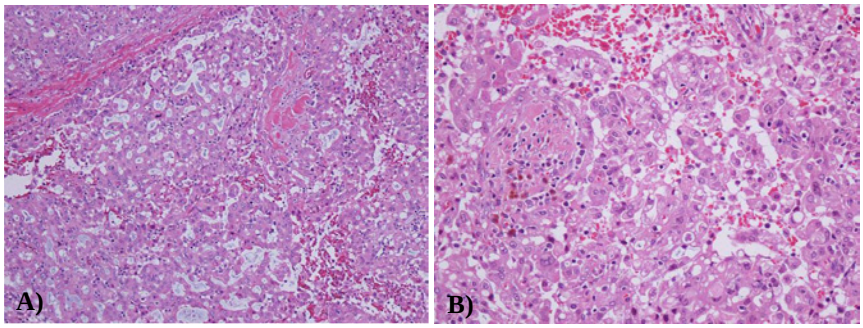

Figure 3: Microscopic appearance of the tumor (hematoxylin and eosin staining) (a. ×100 and b. ×200). The neoplastic cells with abundant eosinophilic cytoplasm and large nuclei containing small but significant nucleoli grow forming large islands. Duct-like structure with mucin production is evident. The cellular atypia is relatively prominent.

treatment approach for low- or intermediate-grade T1 ANOS remains unclear. In general, the most appropriate therapeutic strategy for salivary ANOS is surgery followed by radiation therapy in cases of advanced-stage tumors, high-grade lesions, positive surgical margins, and lymph node metastasis [4]. Lin et al. reported that radiation therapy should be considered for APG malignancies in cases of unresectability and as postoperative treatment in cases of advanced-stage or high-grade disease [8]. Yang et al. however, recommend surgery followed by radiation therapy even in patients with T1 and T2 APG malignancies [9]. The incidence of occult lymph node metastasis of salivary ANOS is not low even in cases of early T stage disease [10], and hence, neck dissection should also be considered for high-grade tumors. The predisposed levels for occult lymph node metastasis are level II and III [10]. However, for early-stage ANOS lacking an aggressive course, as in our case, complete surgical excision of the tumor alone may be the optimal treatment of choice. Although we could preserve facial nerve branches in the present case because the tumor was small with noninvasive features, we would have dissected facial nerve branches with the tumor to ensure a surgical margin of more than 15 mm if the tumor was of a high grade [4].

Although we used a standard parotidectomy incision for complete and safe exposure of the mass, the parotid duct, and facial nerve branches, various approaches have been reported for APG tumor excision, including the intraoral, a direct skin incision, a standard parotidectomy incision or facelift, and endoscopic approaches [5]. Of these methods, the standard parotidectomy incision has been recommended for complete tumor excision, avoidance of facial nerve damage, and cosmetic results [1]. In addition, an endoscopic approach was recently found to yield satisfactory results in cases of benign APG tumors [11,12]. This endoscopic technique could be adapted for a low- or intermediate-grade small malignant tumor, as was the case in our patient. The optimal approach should be selected after evaluating the advantages and disadvantages of all methods.

In the present case, the tumor was suspected to be benign because of the clinical findings of slow growth, small size, and good mobility. However, the tumor was pathologically proven to be malignant. The incidence of malignant tumors in the APG is higher than that of parotid gland tumors [1,5]; therefore, the possibility of malignancy must be considered in cases of APG tumor excision, even if preoperative examinations indicate benign lesions.

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