Salivary Duct Carcinoma of the Accessory Parotid Gland

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ABSTRACT

Objectives

Present a rare case of salivary duct carcinoma of the accessory parotid gland

Understand the current prevalence, diagnosis, and management of benign and malignant tumors of the accessory parotid gland

Methods

This study includes a case presentation and literature review. The literature review included articles from October 1966 to January 2020 and included all cases of accessory parotid gland tumors in adults.

Results

One hundred and nineteen cases with 19 different tumor types were reported in the accessory parotid gland (APG). Pleomorphic adenoma and mucoepidermoid carcinoma represented 50% and 23%, respectively. There were no previously reported salivary duct carcinomas of the APG. Forty percent of APG neoplasms were malignant. This malignancy rate is comparable to previous studies in the main parotid and accessory parotid glands.

Conclusion

Most tumor types that have been identified in the parotid gland have also been found in the APG, including this latest finding of salivary duct carcinoma in the accessory gland. The rate of malignant tumors of the APG appears to be higher than that of the main parotid gland.

INTRODUCTION

Salivary duct carcinoma is a rare neoplasm, comprising approximately 1% of salivary gland carcinomas.\(^2\)\(^3\) It is an aggressive malignancy that generally affects patients greater than 50 years of age, with a male:female ratio of about 4:1.\(^1\)\(^4\) The parotid gland is the most commonly involved site, representing 75-88% of cases; tumors of the submandibular, sublingual, and minor salivary glands as well as the maxilla and larynx have also been reported.\(^5\)\(^6\) Histologically, there is a resemblance to ductal carcinoma of the breast. Salivary duct carcinoma is known to be a high-grade malignancy, with an approximately 50% five-year survival rate.\(^7\)

An accessory parotid gland is defined as salivary gland tissue adjacent to Stenson’s duct and separate from the main body of the gland. accessory parotid gland parotid have been reported to be present in 21-56% of human cadavers.\(^1\)\(^8\) The typical location of this tissue is superior to Stenson’s duct and inferior to the buccal branch of the facial nerve.\(^9\) This tissue is histologically similar to tissue of the main parotid gland; it has therefore been postulated that any type of pathology in the main parotid gland can also arise in the accessory parotid gland.\(^1\) A small percentage of parotid tumors, from 1.0 to 7.7%, occur in accessory parotid tissue.\(^1\)\(^4\) Given the rarity of the salivary duct carcinoma as well as the fact that a minority of parotid tumors are localized to the accessory parotid gland, we report this unique case of a salivary duct carcinoma of the accessory parotid gland, as well as a review of accessory parotid tumors that have been reported in the literature to date.

CASE PRESENTATION

A 76 year old woman was referred to our clinic in July 2008 for an enlarging mass of her left cheek. She had first noticed the mass 3 months prior, at which time it was approximately 1 cm in diameter. There was no pain or skin changes associated with the mass, but over time it noted it to be growing larger and was able to feel it with her tongue. She had no numbness, weakness of her muscles of facial expression, or trismus. There was no blood or pus in her saliva. On physical exam, she was noted to have a firm 3.0 cm mass of her left mid-face that was also palpable, intrasysy. Her facial nerve was fully functional, and no cervical lymphadenopathy was appreciated. Fine-needle aspiration biopsy a month prior had demonstrated malignant cells consistent with a poorly differentiated carcinoma. CT scan of the neck and face demonstrated a large, heterogeneous enhancing, poorly margined, lobulated mass likely originating from the anterior tip of the left parotid gland (Figure 1).

Scattered lymph nodes, primarily of left level 1B and 2A were noted, the largest estimated at 1.8 cm in diameter.

The patient subsequently underwent a left parotidectomy with facial nerve dissection via a modified Blair incision, and a left neck dissection. The superior and inferior buccal branches of the facial nerve were identified extending into the tumor and were subsequently sacrificed. The tumor itself was a 5 x 4 x 4 cm smooth mass overlooking the left zygoma distinct from the main body of the parotid gland. It extended towards the anterior wall of the maxillary sinus without any sign of invasion.

Histological examination of the tumor demonstrated several features typically found in salivary ductal carcinoma, including tumor cells arranged in cribriform, papillary, and solid patterns (Figure 2). The cribriform architecture is particularly characteristic (Figure 3). Frequent mitotic figures were visualized, as well as perineural and angiolymphatic invasion (Figures 4-5). Single-filing of atypical cells infiltrating adipose and collagenous tissue are also demonstrated (Figure 4). These histological features all support the diagnosis of a highly aggressive, poorly differentiated salivary duct carcinoma in this accessory parotid gland.

Though salivary duct carcinoma has been found to be highly metastatic, there were no signs of distant spread or regional lymph node involvement in this case. She was subsequently treated with adjuvant radiation therapy and is currently doing well after 3 years.

DISCUSSION

In reviewing the existing literature for incidence of different neoplasms and rates of malignancy in the accessory parotid gland, all case reports and case series written in English with an adult subject population were included. The earliest paper was in October 1966 and the most recent in January 2020. A total of 115 cases meeting these criteria were reviewed (Table 1). Pleomorphic adenoma was the most commonly reported neoplasm, comprising 50% of the accessory parotid gland tumors. Mucoepidermoid carcinoma was the second most common lesion at 23%. Acinic cell carcinoma, carcinoma ex pleomorphic adenoma, and myoepithelioma were other pathologies with several reports in the literature. Individual case reports include caverous hemangioma, oncocytic carcinoma, myoepithelial carcinoma, and a case of a metastatic adenocarcinoma, which arose from the prostate. 40% of the neoplasms were malignant.

Tumor type & Number Percentage

- Pleomorphic adenoma: 10 40.1%
- Mucoepidermoid carcinoma: 27 22.9%
- Acinic cell carcinoma: 6 5.0%
- Carcinoma ex pleomorphic adenoma: 2 1.7%
- Myoepithelioma: 3 2.5%
- Adenoid cystic carcinoma: 2 1.6%
- Basal cell adenoma: 2 1.6%
- Lipoma: 2 1.6%
- Monomorphic adenoma: 1 1.0%
- Neurofibroma: 2 1.6%
- Non-Hodgkin lymphoma: 2 1.6%
- Squamous cell carcinoma: 2 1.6%
- B-cell lymphoma: 1 0.8%
- Cavernous hemangioma: 1 0.8%
- Metastatic adenocarcinoma (prostate): 1 0.8%
- Metaplastic carcinoma: 1 0.8%
- Oncocytic carcinoma: 1 0.8%
- Papillary cystadenoma: 1 0.8%
- Undifferentiated carcinoma: 9 21.9%
- All histologic types: 50 100.0%

Table 1. Accessory parotid tumors reported in the literature (1966-2020), by frequency

REFERENCES


Papillary cystadenoma 1 0.8%

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CONCLUSIONS

Many of the salivary gland pathologies known to occur in the main parotid gland have been found in accessory parotid gland tissue. A review of the literature regarding tumors of the accessory parotid gland suggests that the incidence of malignant tumors is higher than that of benign tumors. Salivary duct carcinoma, which represents fewer than 1% of the tumors found in the parotid gland, is a histologic type that has not previously been reported in the APG. This malignancy can be diagnosed via a high degree of suspicion for a tumor in a firm, rapidly growing mid-cheek mass, thorough head and neck exam, multiple imaging modalities, and FNA biopsy. Subsequent management, as recommended for all accessory parotid gland tumors, involved a standard parotidectomy incision with preservation of the mandibular arcade. In addition, the preservation of the marginal mandibular nerve allows for improved cosmetic outcomes.

The incidence of types of APG tumors in the literature appears to deviate from the data of the incidence and types of tumors found in the main parotid gland. The incidence of pleomorphic adenoma of the parotid gland has been previously reported at 53%, with the second and third most common neoplasms being Warthin’s tumor (28%) and myoepithelial carcinoma (9%), respectively.\(^9\) The incidence of malignant tumors of the parotid gland in this study by Pekinl, et al. was 15%, compared to the 40% we found by literature review of APG tumors. Whether or not this represents an actual higher incidence of malignancy in APG tumors or a bias towards reporting malignant vs. benign tumors of the APG is unclear.