



# 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 2010 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 was comparable to previous studies reporting a higher rate of malignant tumors in the APG compared with 15-20% in parotid tumors. A standard parotidectomy remains the preferred approach with less risk to the facial nerve compared with direct cheek resection and transoral approaches.

### 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.<sup>1,2</sup> It is an aggressive malignancy that generally affects patients greater than 50 years of age, with a male-female ratio of about 4:1.<sup>1,3</sup> 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.<sup>1-3</sup> 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.<sup>4</sup>

An accessory parotid gland is defined as salivary gland tissue adjacent to Stensen's duct and separate from the main body of the gland; accessory parotid glands have been reported to be present in 21-56% of human cadavers.<sup>5,6</sup> The typical location of this tissue is superior to Stensen's duct and inferior to the buccal branch of the facial nerve.<sup>7</sup> This tissue is histologically similar to tissue of the main parotid gland; it has therefore been postulated that any type of pathology arising in the main parotid gland can also arise in the accessory parotid gland.<sup>5</sup> A small percentage of parotid tumors, from 1.0 to 7.7%, occur in accessory parotid gland tissue.<sup>8,9</sup> 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.

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## 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 she 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 purulence in her sputum. On physical exam, she was noted to have a firm 3.0 cm mass of her left mid-face that was also palpable, intraorally. 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, heterogeneously enhancing, poorly marginated, lobulated mass likely originating from the anterior tail 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.



Figure 1. Computed tomography scan showing a 2 x 4 cm solid mass of abutting the anterior tail of the left parotid gland.

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 overlying the left zygoma distinct from the main body of the parotid gland. It extended towards the anterior wall of the maxillary sinus without any signs 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 intraductal cribriform pattern 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 6). These histological features all support the diagnosis of a highly aggressive, poorly differentiated salivary duct carcinoma in this accessory parotid gland.

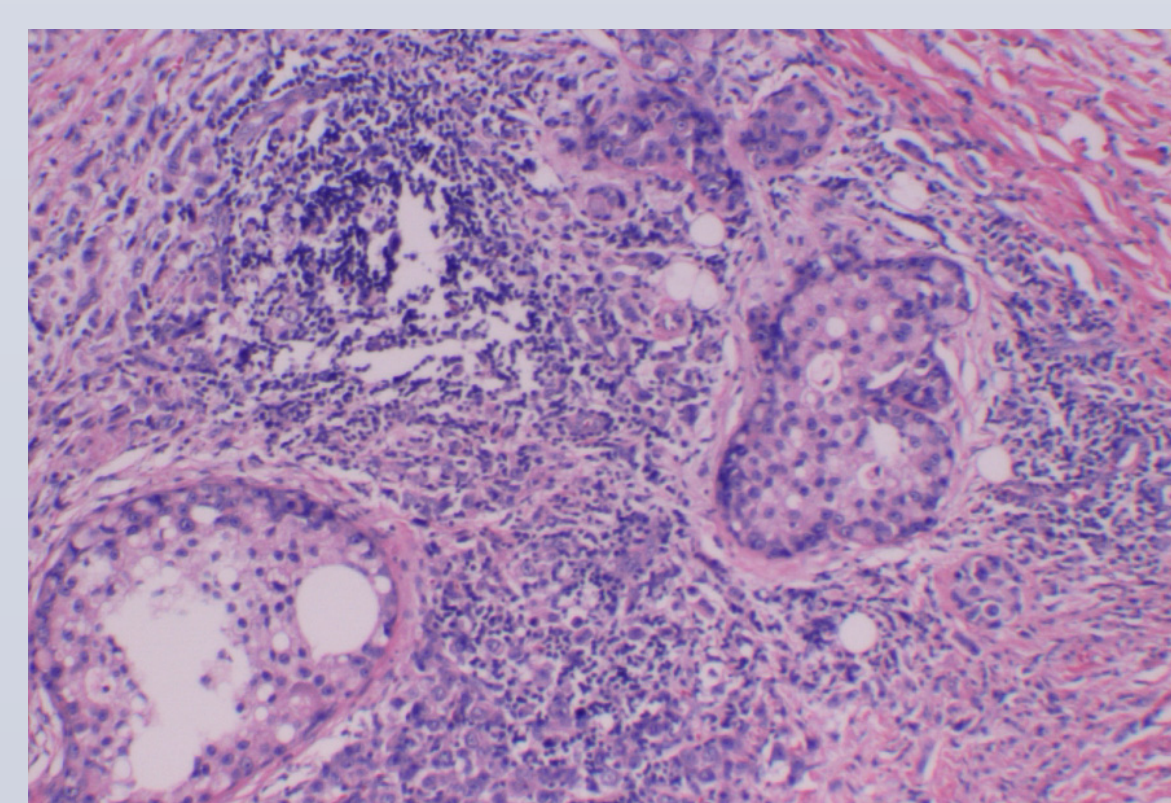


Figure 2. Tumor cells arranged in cribriform (bottom left), papillary (top right), and solid patterns, characteristic of salivary duct carcinoma.

### Figures 4-6: Characteristic histologic findings in salivary duct carcinoma

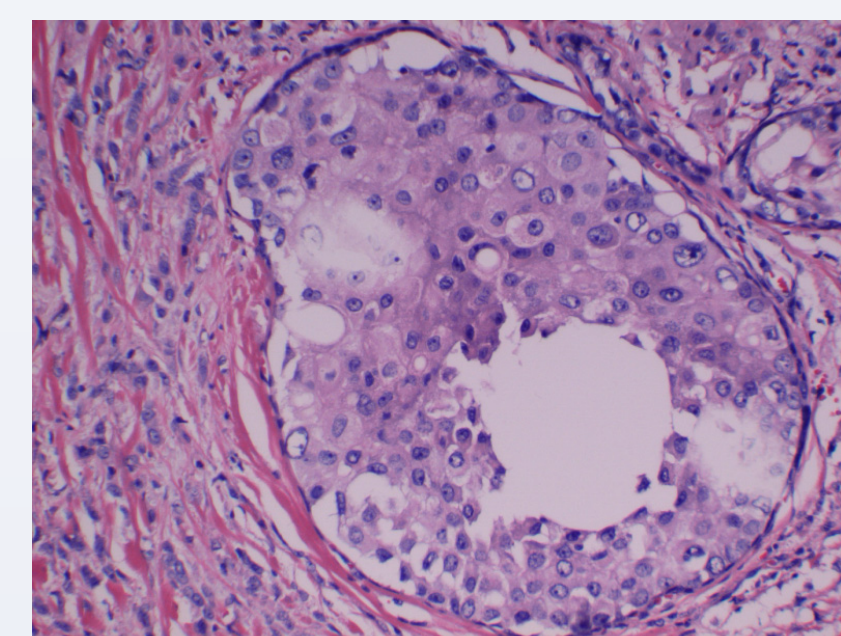


Figure 3. Intraductal cribriform pattern

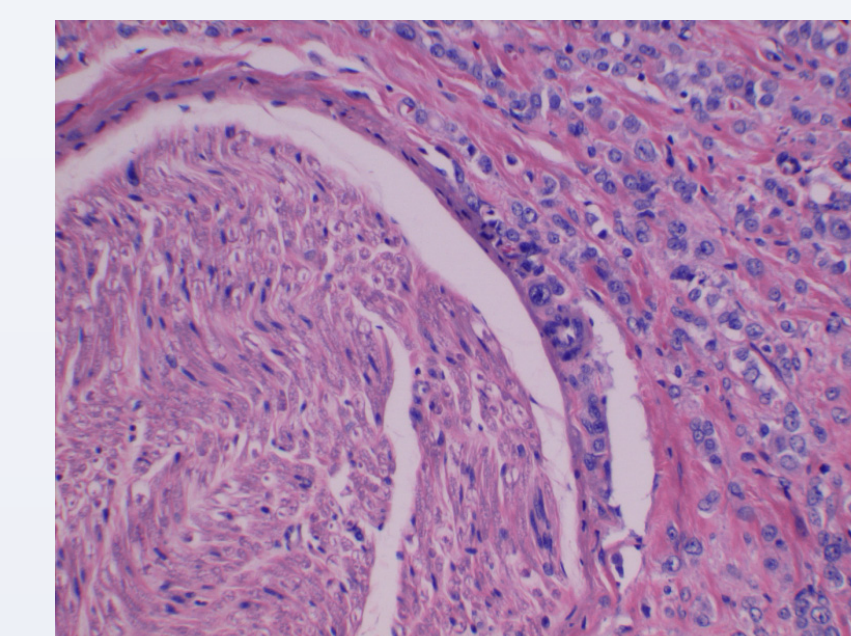


Figure 4. Perineural invasion surrounded by dense collagenous stroma.

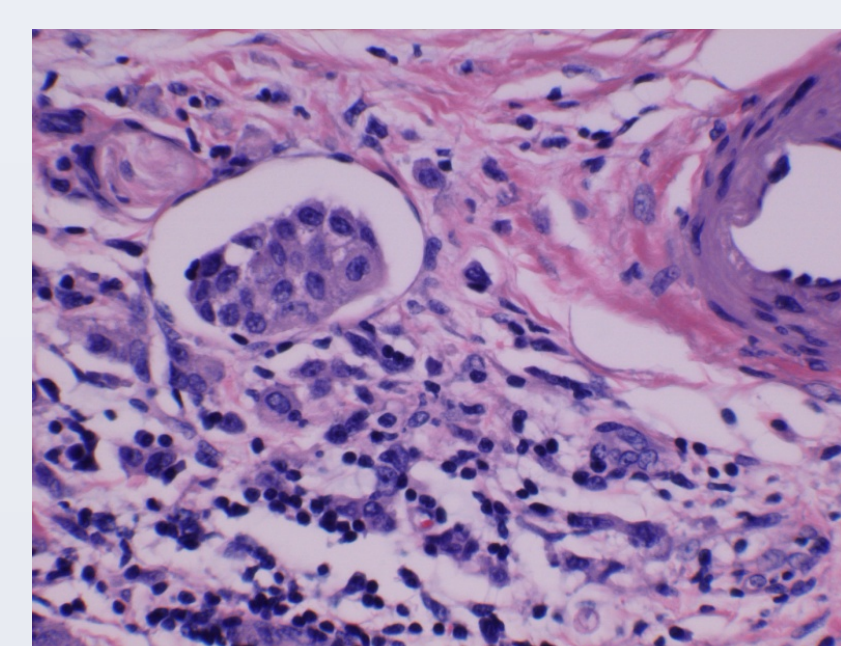


Figure 5. Angiolymphatic invasion

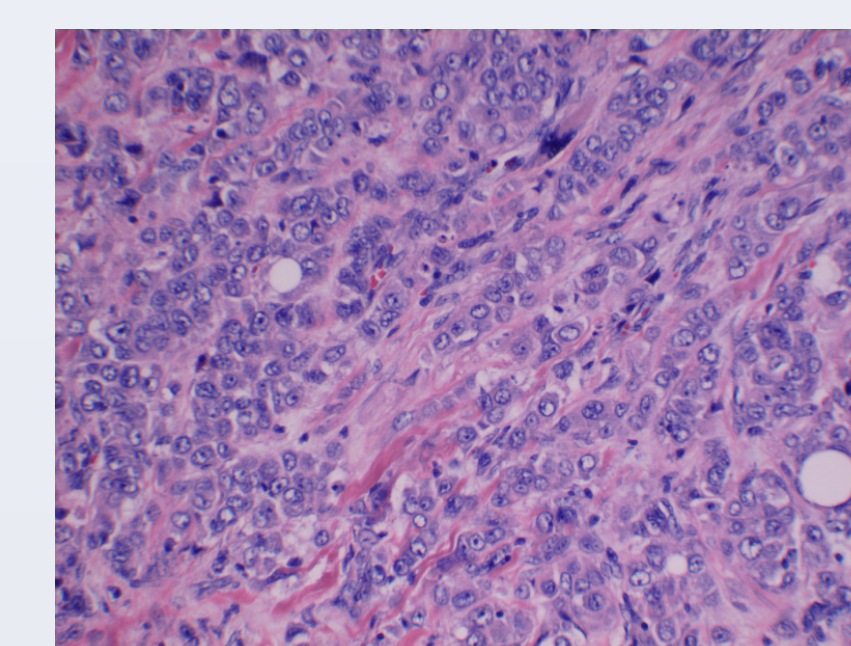


Figure 6. Single-filing of atypical cells, infiltrating fat.

The patient was referred for post-operative radiation therapy. However, she declined any further treatment, choosing to live out the remainder of her life with her daughter in a different state. She ultimately died in November 2010, a little over 2 years after her surgery.

## RESULTS

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 2010. A total of 119 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 cavernous hemangioma, oncocytic carcinoma, myoepithelial carcinoma, and a single case of a metastatic adenocarcinoma, which arose from the prostate. 40% of the neoplasms were malignant.

Tumor type	Number	Percentage
Pleomorphic adenoma	59	49.58%
Mucoepidermoid carcinoma	27	22.69%
Acinic cell carcinoma	6	5.04%
Carcinoma ex pleomorphic adenoma	3	2.52%
Myoepithelioma	3	2.52%
Adenoid cystic carcinoma	2	1.68%
Basal cell adenocarcinoma	2	1.68%
Lipoma	2	1.68%
Monomorphic adenoma	2	1.68%
Neurofibroma	2	1.68%
Non-Hodgkin lymphoma	2	1.68%
Squamous cell carcinoma	2	1.68%
B cell lymphoma	1	0.84%
Cavernous hemangioma	1	0.84%
Metastatic adenocarcinoma (prostate)	1	0.84%
Myoepithelial carcinoma	1	0.84%
Oncocytic carcinoma	1	0.84%
Papillary cystadenoma	1	0.84%
Undifferentiated carcinoma	1	0.84%
All histologic types	119	100.00%

Table 1. Accessory parotid tumors reported in the literature (1966-2010), by frequency.

## DISCUSSION

A high index of suspicion, physical exam, FNA biopsy, and CT scanning are the mainstays of diagnosing the accessory parotid gland (APG) tumor. Most of these tumors present as an asymptomatic cheek mass, as was the case with our patient.<sup>10,11</sup> The generally accepted approach to management is surgical resection via a standard parotidectomy incision.<sup>7-13</sup> This approach has the advantage of being safe by avoiding facial nerve injury, effective in identifying all tumor (with lower rates of recurrence), and is also cosmetically acceptable.<sup>7,8,11,13</sup> Direct cheek resection and perioral approaches are not generally recommended given the difficulties of exposure and facial nerve identification.<sup>10,13</sup>

Klotz and Coniglio recommend that small (T1 and T2) low-grade malignancies may be sufficiently managed by wide excision without concomitant parotidectomy based on the fact that the APG is an independent island of salivary tissue. Total parotidectomy and neck dissection is required for advance-staged disease with a high-grade histological appearance, large tumor size, or locoregional spread. Post-operative radiation therapy is typically reserved for high-grade malignancies and advance-staged disease. First-line radiotherapy is reserved for unresectable cases.<sup>10</sup>

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 mucoepidermoid carcinoma (9%), respectively.<sup>14</sup> The incidence of malignant tumors of the parotid gland in this study by Pinkston, 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.

## 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 literature regarding tumors of the accessory parotid gland suggests that the rate 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 one 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 parotidectomy and the recommendation of postoperative radiation therapy for positive margins and high-grade tumor type.

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