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## **Accessory parotid gland tumours: A 24-year of clinical experience**

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## **Abstract**

Accessory parotid gland (APG) is salivary tissue that is anterior and anatomically separate from the parotid gland. Although the APG is a common anatomical variation, APG tumours are extremely rare. We report on 6 patients having APG tumours with emphasis on diagnosis, clinical features, indications and rationales for different treatment approaches. Patients with primary tumours of the parotid gland or APG tumours that underwent surgical treatment were included. APG tumours comprised 1.23% of overall parotid tumours (6/488) and had a malignancy rate of 33.3% (2/6). There were three males and three females with mean age of 39 (range, 14-70). A total of 5 of 6 parotidectomies entailed superficial lobectomy, while one was total parotidectomy with composite resection of masseter muscle. Concomitant selective lymphadenectomy was carried out in 3 of 6 patients. At 5 years disease-free survival was 83.3%. Mean follow-up was 161 months (range, 14-253). Although nonsalivary diagnoses frequently occur in the buccal area, APG tumours should be considered in every differential diagnosis in patients presenting with a mid-cheek mass. From oncosurgical, cosmetic and functional standpoint, treatment by facelift parotidectomy or "S-incision" with concomitant superficial lobectomy is recommended surgical approach, while high-grade malignancies require total parotidectomy with regional lymphadenectomy.

## INTRODUCTION

The evaluation of a mid-cheek mass represents a significant diagnostic and therapeutic challenge. Benign and malignant lesions in this area may arise from soft tissues of the face, including the skin, lymphatics, skin adnexa, neural, and salivary structures<sup>1</sup>.

The accessory parotid gland (APG) has been described as salivary tissue that is anterior and anatomically separate from the main body of the parotid gland and adjacent to Stensen's duct by way of one or multiple secondary connections. Frommer<sup>2</sup>, in his study of cadaveric dissections, found an APG in 21% of individuals, although some authors reported incidences as high as 56%<sup>3</sup>. Although the APG is a common anatomical variation, APG tumours are extremely rare, with a reported incidence of 1 to 7.7% of all parotid gland tumours, and a malignancy rate from 26 to 50%<sup>4,5</sup>. So far, 157 lesions arising from accessory parotid gland have been reported in the English-language literature, including 54 malignant tumours, 80 benign tumours, and 23 nontumour lesions<sup>6,7</sup>. There are few larger series describing APG tumours<sup>4-9</sup>. To our knowledge, this is second largest European series regarding APG tumours. We report on 6 patients having APG tumours with emphasis on diagnosis and clinical features, as well as the indications and rationale for different treatment approaches.

## MATERIALS AND METHODS

Patients were identified through a database search of salivary gland tumours at the Department of Maxillofacial Surgery, University of Zagreb School of Medicine, University Hospital Dubrava, for the period between January 1, 1985 and December 31, 2008. Patients with primary tumours of the parotid gland or APG tumours that underwent surgical treatment were included. Among these tumours, patients with primary parotid gland tumours and patients with anterior process or extension of the main parotid gland were analyzed separate from patients with primary APG tumours. Patients with normal salivary tissue in accessory parotid lobes and patients with heterotopic (ectopic) salivary gland tissues were excluded. The medical records of patients who were surgically treated for APG tumours were reviewed. Radiotherapy was indicated in cases of malignant disease of the APG. Follow-up intervals were calculated in months from the date of first treatment at our Department to the date of last follow-up or death. The follow-up interval was concluded on December 31, 2011.

## RESULTS

During the 24-year period, 488 patients with primary parotid tumours underwent surgical treatment. APG tumours comprised 1.23% of overall parotid tumours (6/488) and had a malignancy rate of 33.3% (2/6). The clinical features, diagnosis, treatments and outcomes are summarized in **Table 1**.

There were three males and three females with a mean age of 39 years (range, 14-70). All patients except one had undergone no previous treatment. This latter patient is a 48-year-old woman who received surgical treatment for a pleomorphic adenoma elsewhere (mid-cheek incision directly over the tumour) and has been referred to our Department because of local recurrence (**Figure 1**).

Three patients presented with a painless, progressively enlarging buccal mass, while two patients had painful lesions at the same location. One patient had painless mid-cheek lesion with numbness of the cheek and upper lip.

All patients had undergone FNAB preoperatively and the results were identical to those provided by patohistological analysis following operation. Histologically, of the 6 APG tumours, there were three pleomorphic adenomas (one of which was multilocular), one myoepithelioma, one adenoid cystic carcinoma and one high-grade mucoepidermoid carcinoma. Two patients with malignant disease underwent adjuvant postoperative irradiation what included 27 fractions of 2 Gy given over 4 weeks. A total of 5 out of 6 parotidectomies entailed a superficial lobectomy, while one was a total parotidectomy with composite resection of the masseter muscle in case of high-grade mucoepidermoid carcinoma. In four cases of superficial parotidectomy buccal branches were in intimate



relationship with the APG tumour and were sacrificed in order to achieve macroscopic tumour clearance. Apart from these patients in whom the facial nerve was partially sacrificed, two patients developed a facial palsy which fully recovered. Concomitant neck dissection was carried out in 3 of the 6 patients (50%), and comprised two selective neck dissections of level 2 and one selective neck dissection of levels 1 and 2.

Lymphadenectomies were electively performed in suspicious cases, though they yielded negative neck nodes. No postoperative complications were detected.

At the time of follow-up, five patients were disease-free, while one patient developed a locoregional recurrence with distant metastases of a high-grade mucoepidermoid carcinoma 8 months after initial surgical intervention and died 6 months later. At 5 years disease-free survival was 83.3%. Mean follow-up was 161 months (range, 14-253).

## DISCUSSION

The differential diagnosis of a mid-cheek masses includes parotid gland epidermoid cyst or arteriovenous malformation, Stensen's duct stone, lipoma, neoplasm, hemangioma or adnexal tumours, neural tumours, hematoma, benign and malignant adenopathy, metastatic disease, and benign and malignant tumours of the accessory parotid tissue<sup>1,10</sup>. The accessory parotid gland is an isolated cluster of salivary tissue which lies between the buccal and zygomatic branches of the facial nerve on the masseter muscle and occurs typically around the midpoint of an imaginary line drawn from the tragus of the ear to a point halfway between the ala of the nose and the vermilion border of the upper lip<sup>11</sup>. Johnson and Spiro<sup>4</sup> retrospectively reviewed 2 261 parotid gland tumours during a 40 year period, and found that 1% arose in APGs. Of these APG tumours, more than half proved malignant. Perzik et al.<sup>5</sup> reported a 7.7% incidence of APG tumours among all parotid gland tumours with a malignancy rate of 26%. The only study of a metastatic spread to the APG has been reported by Goldberg et al.<sup>12</sup>. Combined with previously reported 157 cases, there are 84 benign tumours, 56 malignant tumours, and 23 nontumours lesions in the total 163 cases (**Table 2**).

According to the literature, APG tumours most often present as an asymptomatic progressively enlarging cheek mass and usually occur in patients with a mean age ranging from 45 to 64.4 years at the time of presentation<sup>1,6,7,9</sup>. In contrast, in this study group, patients were younger, with a mean age of 39 and 50% of patients presented with symptomatic lesions which included different head and neck complaints such as pain and numbness in mid-cheek area. Similar results were reported by Sun et al.<sup>13</sup>. Patients were

evaluated through careful physical examination and additional diagnostic tools including fine-needle aspiration (FNA), computed tomography (CT) and magnetic resonance (MR). Neck dissection was performed in cases of malignant or recurrent APG tumours. In this study, the incidence of APG tumours was 1.23% with a malignancy rate of 33.3% which is in range with prior reports<sup>4,5</sup>. One patient with a malignant lesion of the APG died due to locoregional recurrence and distant disease from high grade mucoepidermoid carcinoma. Similar biological behavior of mucoepidermoid carcinoma arising from APG tissue has been described before<sup>14</sup>.

After differential diagnosis, the second important question concerns the extent of surgical treatment. There are four surgical approaches in management of APG tumours: the standard "S-incision" or facelift parotidectomy approach, intraoral approach and cheek incision directly over the tumour. The latter one is associated with an increased incidence of facial nerve injury because of the superficial location of the buccal and zygomatic branches of the seventh cranial nerve. Johnson and Spiro<sup>4</sup> reported a 40% incidence of facial nerve injury for tumours approached via a direct skin incision over the mass. Furthermore, a limited incision is associated with a high incidence of local recurrence brought on by inadequate excision margins and an unsightly scar. The intraoral approach also places the facial nerve at high risk because of difficulty in optimal exposure<sup>15</sup>. In the context of the nerve-tumour relationship, a standard facelift parotidectomy approach is advisable. Once a standard incision is made, a concomitant superficial parotidectomy is recommended. This allows identification of the main facial nerve trunk with tracing of all its branches. Furthermore, this surgical technique provides excellent exposure which minimizes the risk of complications during excision of these tumours (**Figure 2**). Johnson and Spiro<sup>4</sup> reported no

postoperative facial nerve palsies in 13 patients undergoing a standard parotidectomy approach. Perzik et al.<sup>5</sup> reported no facial nerve injuries in 20 modified preauricular approaches. In addition, this approach hides unsightly scars within natural skin creases and hairlines, giving the best cosmetic result (**Figure 3**). Most importantly, this treatment modality provides adequate resection which minimizes the risk of local recurrence. In our experience, the standard parotidectomy approach (facelift incision) with superficial parotidectomy in conjunction with excision of the APG tumour is the recommended surgical approach (**Figure 4**). According to the recent publications, Xie et al.<sup>16</sup> introduced a minimally invasive endoscopic approach via a small preauricular incision performed on 5 patients with benign APG tumours and no postoperative complications and recurrences were found. Primarily due to oncological reasons, we doubt that the minimally invasive endoscopic resection is the best method of treatment for tumours of the APG. This opinion is supported by previous studies which reported reduced recurrence rates (less than 4%) associated with extended surgical techniques compared with the high incidence of recurrence (25-40%) resulting from enucleation among patients with pleomorphic adenoma of the parotid gland<sup>17-19</sup>. In addition, surgery of recurrent parotid gland tumours carries considerable risk of facial nerve injury which varies from 15 to 30% and the chance of recurrence is high ranging from 15 to 75%<sup>20-25</sup>. Moreover, one year of follow-up is not a sufficient time interval for recurrence detection since recurrent benign parotid gland tumours occur 3 to 9 years after initial surgical treatment<sup>21,25-27</sup>.

A clear weakness of this retrospective study is the small sample size. However, given the rarity of this entity, it's difficult to find larger series. Indications for elective neck dissection

in patients with malignant parotid and APGT tumours remains unclear. Further investigations are necessary in order to improve our better understanding of the subject. In conclusion, APG tumours are extremely rare and represent a considerable diagnostic problem. Although nonsalivary diagnoses frequently occur in the buccal area, APG tumours should be considered in every differential diagnosis in patients presenting with a mid-cheek mass. In this study group, patients with APG tumours were younger than most reports in the literature state and half of them presented with a symptomatic mid-cheek mass unlike the typical asymptomatic cases from the literature. In order to establish the correct diagnosis, we advocate proper preoperative evaluation which includes a detailed physical examination, FNA and imaging techniques (CT and MR scans). Surgical resection remains the mainstay of treatment for both parotid and APG tumours. From oncosurgical, cosmetic, and functional standpoints, treatment by facelift parotidectomy approach or "S-incision" with concomitant superficial lobectomy is the recommended surgical approach, while a high-grade histological appearance requires total parotidectomy.

**Conflict of interest statement:**

The authors declare that they are not in any conflict of interest.

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No author nor any member of their families received any material or financial gain or personal advancement in the production of this manuscript.

**Ethical Approval:**

This work has been approved by the ethical committee of the University Hospital Dubrava and all patients gave informed consent for their inclusion in the article.

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CAPTIONS TO ILLUSTRATIONS

FIGURE (1) Preoperative photograph showing scar (arrow) from mid-cheek incision.



FIGURE (2) Intraoperative photograph.



FIGURE (3) Postoperative photograph (27 months after surgery).



FIGURE (4) Intraoperative specimen showing superficial lobe of the parotid gland (dashed arrow), Stensen's duct (dotted arrow) and masseter muscle with APG tumour (solid arrow).

