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Management and surgical treatment of accessory parotid gland tumors: 32-year experience from a single institution and systematic review of the literature

Running Head: Treatment of accessory parotid gland tumors

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Abstract

Accessory parotid gland tumors (APGT) are very rare. Regarding their anatomical location an adequate surgical approach is necessary to provide safe resection and satisfactory postoperative result. Aims of this study were to present our tertiary center experience in surgical treatment and management of APGT and to review the literature regarding their treatment, particular in terms of surgical modalities. Data of thirteen patients with primarily surgically treated APGT have been collected and analyzed. Approaches included standard parotidectomy and facelift incision. Well-documented English-literature articles of surgically treated APGT have been extracted from the PubMed, Scopus and Web of Science databases ending in May 2018 and analyzed. Mean age at the diagnosis was 41.1 years. The most common benign histological subtype was pleomorphic adenoma (53.8%), while mucoepidermoid carcinoma (23.1%) was most common in malignant counterparts. A malignancy rate was 38.5%. Postoperative results were satisfying, and follow-up period was uneventful in all patients except one who died of locoregional recurrence. A total of 57 papers with reported series in APGT have been identified with a total of 306 APGT cases. From oncosurgical, functional and cosmetic standpoints approaches through a standard parotidectomy and facelift incision provide satisfactory results.

Introduction

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. It occurs typically around the midpoint of an imaginary line draw from the tragus of the ear to a point halfway between the alae of the nose and the vermillion boarder of the upper lip.[1] Despite the incidence of accessory parotid gland being reported to be 21-56% among human cadavers[2,3], tumors originating from accessories are extremely rare and constitute 1-7.7% of all parotid gland tumors.[1,4] The reported malignancy rate of accessory parotid gland tumors (APGT) is higher than tumors in the parotid, ranging from 26 to 50%.[4,5]

The mainstay of treatment of APGT is surgical resection. Surgical treatment includes preservation of anatomical structures related to the affected region, while respecting oncological principles. Preservation of the facial nerve is the most important aspect in surgical treatment. The selection of an optimal approach used in the surgical treatment of APGT is the subject of the discussion. The most frequently utilized include approaches through the standard parotidectomy incision, facelift incision, and direct transcutaneous or transoral incisions. Recent reports presented the minimally invasive endoscopic-assisted surgery as an alternative method in the treatment of APGT.[6-9]

The aims of this study were to present our tertiary center's experience in surgical treatment and management of APGT and to review the literature regarding it's diagnostics, work-up and treatment, particular in terms of surgical modalities.

Materials and methods

Patients

Data were collected retrospectively from the institutional salivary tumors database for the period between 1 January 1985 and 31 December 2016 at the Department of Maxillofacial Surgery, University Hospital Dubrava, Zagreb. Patients with APGT who were primarily surgically treated, were included in the study. Mid-cheek masses other than APGT were excluded from further analysis. The follow-up interval was calculated in months from the date of a first treatment to the date of a last follow-up or death. The follow-up period was concluded on 31 December 2017.

Well-documented English-literature articles of surgically treated APGT have been extracted from the PubMed, Scopus and Web of Science databases ending in May 2018 and analyzed. The search strategy used a key-word "accessory parotid gland tumors". Search was performed independently by 2 reviewers (M.M., P.S.).

Per the institutional review board of the University Hospital Dubrava, Zagreb, this study met criteria for nonhuman subject research, and as a result, board approval was not required.

Treatment

Pre-operative evaluation included careful physical examination, ultrasound-guided fine-needle aspiration cytology (US-guided FNAC), and multislice computed tomography (MSCT) or magnetic resonance imaging (MRI) (**Figure 1**). After the diagnosis of APGT was established, surgical management was conducted through a standard parotidectomy incision (modified Blair incision) or facelift incision (**Figure 2**). After elevation of the anterior skin flap, initial tracing of the facial nerve main trunk introducing anterograde dissection of its branches was performed

(**Figure 3**). Following identification of the zygomatic and buccal branches, excision of the accessory parotid gland with concurrent superficial parotidectomy was done. Superselective or selective neck dissections were carried out in the cases with preoperative assessed malignant tumor. Postoperative radiation therapy (PORT) was conducted in cases of high-grade tumors or advanced-stage disease. With daily fractions of 2 Gy, a prophylactic dose of 50 Gy to clinically undissected neck levels was given, with a boost of 60 Gy to the tumor bed and metastases confined to lymph nodes. The follow-up protocol consisted of medical history and physical examination every 3, 6, 8, and 12 months, in the first, second, third, and fourth year of follow-up, respectively.

Results

During the 32-year period, 792 patients with parotid gland tumor were primarily surgically treated at our institution. Accessory parotid gland tumors comprised 1.64% of all surgically treated parotid gland tumors (13/792). Clinical features of the study group are summarized in **Table 1**. Mean age at the diagnosis of APGT was 41.1 (range, 15-70) years. A total of 53.8% (n=7) patients were male and 46.2% (n=6) were female. Mean age of patients with benign tumors was 40 years, while in malignant counterparts it was 43 years. Tumors arising in accessory parotid gland presented a malignancy rate of 38.5% (5/13). The most common benign histological subtype was pleomorphic adenoma (PA) (53.8%), followed by myoepithelioma (7.5%). Among malignant tumors the most common subtype was mucoepidermoid carcinoma (MEC) (23.1%), followed by carcinoma ex-PA (7.5%) and adenoid cystic carcinoma (7.5%). Eleven patients (84.6%) were treated primarily surgically, while two patients (15.4%) were referred to our institution due to local recurrence of PA after previous treatment in other institution (both treated using transcutaneous incision). In 2 cases (15.4%), final pathological diagnosis was non-consistent with preoperative FNAC diagnosis (MEC misdiagnosed as lymphoepithelial cyst and carcinoma ex-PA misdiagnosed as skin adnexal carcinoma). Four patients with malignant disease underwent concurrent neck dissection, two of which were therapeutic. Three patients underwent superselective dissection of region II, while one was treated with selective neck dissection of levels I-III. Twelve accessory parotidectomies entailed a superficial parotid lobectomy, while one entailed a total parotidectomy with composite resection of the masseter muscle (case of high-grade MEC). In 2 patients a lymph node within the superficial parotid lobe was involved by APGT metastasis, while in 11 patients the parotid was tumor-free. One patient with a positive intraparotid lymph node had a positive neck dissection

specimen (case of high-grade MEC). Two patients had a positive neck without involvement of the superficial lobe of the parotid gland, with one dissection being elective. In 4 cases of superficial parotidectomy buccal and/or zygomatic branches were in close relation with the APGT and were sacrificed in order to achieve tumor clearance. Apart from patients in whom peripheral branches were sacrificed, two patients (22.2%) developed transient facial nerve palsy. Four patients with malignant tumors underwent PORT. At the time of follow-up, all patients were disease-free, except one with MEC who developed a locoregional recurrence 8 months after initial treatment and died 6 months later. At 5 years, overall survival was 92.3%. Mean follow-up was 69 months (range 14-220 months).

Extracted data from the literature of surgically treated APGT are summarized in **Table 2.**

Discussion

Tumors of the accessory parotid gland are rare and the literature usually depends on individual case reports and limited case series. A total of 57 papers with expertise in APGT have been identified with a total of 306 APGT cases.[1-4,6-58] The series of patients presented in this study is an updated report, previously published by the senior author.[10] According to our knowledge, this is the largest European series of APGT treated at a single institution, and the fifth largest published in the English literature overall.[1,4,11,12]

The neoplasms located in accessory parotid tissue usually present as a slow-growing, painless, mid-cheek masses. In most studies, the age of occurrence ranges between 45 and 64 years.[12-15] On the contrary, in our study patients were younger with a mean age of 41 years. In our series, the most frequent histological subtype of benign APGT was pleomorphic adenoma (PA), while the most common malignant subtype was mucoepidermoid carcinoma (MEC). A similar pattern of distribution with respect to histological subtype has been published in the literature[1,4,12], except for Ma's study in which the most common malignant subtype was lymphoma, followed by lymphoepithelial carcinoma and acinic cell carcinoma.[11]

The incidence of APGT among all parotid tumors primary surgically treated at our institution was 1.64% with a malignancy rate of 38.5% and this is consistent with previous publications.[1,4,11] According to the pooled data extracted from all available studies, the malignancy rate of APGT is 28.8% (88/306). In our study, the 5-year overall survival rate for all patients with APGT was 92.3%, while patients with malignant APGT had a survival rate of 80% in the same time interval. One patient (MEC) developed locoregional recurrence and died due to disease recurrence. While

other series reported a favorable outcome in terms of survival and recurrence among benign tumors, data related to survival of malignant tumors have not been reported.

Pretreatment cytological and radiological findings were the main factors for determining the extent of surgical treatment. In two cases FNAC has not been consistent with definitive patohistological diagnosis presenting preoperative diagnostic sensitivity of 84.6% (two initially benign tumors were later deemed malignant in the definitive histological report). In the first patient (No 11) the diagnosis of MEC on final patohistological report was preoperatively described as a lymphoepithelial cyst, while in second patient (No 12) carcinoma ex-PA was described as a skin adnexal carcinoma (**Table 1**). The FNAC of low-grade MEC is well recognized for its potential false-negative diagnostic pitfall, due to the bland cytological features and hypocellular nature of this histological subtype.[59,60] The differential diagnosis of low-grade MEC includes Warthin tumor, benign salivary gland cysts (lymphoepithelial cysts), branchial cleft cyst, sialolithiasis or chronic sialadenitis complicated by cystic dilatation, and pleomorphic adenoma with excess mucoid stroma.[61,62] Hughes et al. reported that the diagnostic accuracy of FNAC decreased to 48%, with a sensitivity of 73% in detection of malignant and 91% in benign parotid gland tumors.[63] In this study, the benefit of using US-guided FNAC has been demonstrated in patient (No 12) with carcinoma ex-PA in which US-guided FNAC has shown neck metastasis in region II, previously unsuspicious on physical examination and MSCT (**Table 1**). Although in this case FNAC was not consistent with definitive patohistological diagnosis, positive neck lymph node consequently affected planning of the surgery. Given the fact that US-guided FNAC offers additional information regarding enlarged lymph nodes and malignancy in small lymph nodes not identified by other methods, it can be recommended as part of preoperative work-up for most salivary gland tumors, which is supported by the literature.[64-66]

The mainstay of treatment for APGT is surgical excision. However, selection of an optimal approach and the extent of treatment is the subject of debate. Except for one case (No 1), in all other cases concurrent superficial parotidectomy was entailed (**Table 1**). Although it is unnecessary for the management of benign APGT, it was performed to allow better access to APGT. Moreover, considering preoperative inconsistency with definitive patohistological report, superficial parotidectomy may provide an extra margin in oncological terms. The optimal surgical approach should provide safe access to the tumor, flexibility, and easy manipulation. Also, of significant importance is a satisfying aesthetic result. The treatment procedures reported in the literature consisted of traditional surgery (90.8%) and endoscopic-assisted surgical techniques (9.2%) (**Table 2**). Approaches used in both of these procedures were external (transcutaneous) and transoral. The discrepancy between the total number of APGT (n=305) and the total number of reported approaches (n=261) in **Table 2** stems from the lack of information with respect to treatment modalities in published articles.

Surgical approaches used in the present study were the standard parotidectomy incision (61.5%) and facelift incision (38.5%) (**Figure 2**). Both approaches provide optimal visibility of APGT and surrounding structures (**Figure 3**). Wide operative field allows flexibility and safe dissection of the facial nerve trunk with tracing of all its branches and parotid duct. Postoperatively, apart from 4 patients in whom the buccal branches were sacrificed in order to achieve macroscopic tumor clearance, only 2 (15.4%) patients developed transitory facial nerve palsy. No case of salivary fistula or gustatory sweating syndrome was noted. The postoperative scars were hidden in the cervical crease and hair-bearing portion of the scalp providing a satisfying aesthetic result in all patients (**Figure 4**). The facelift approach is considered as a gold standard in facial rejuvenation, so from the aesthetic standpoint, its utilization in oncosurgery is highly acceptable. Interestingly,

the very low rate (2.1%) of facelift incisions according to the reviewed literature has been reported (**Table 2**). Possible reason could be inexperience in the treatment of such a rare tumor through a generally considered aesthetic approach. The preference of using an approach through standard parotidectomy incision was reported by Perzik and White, presenting 20 patients without facial nerve injury and good aesthetic results.[1] Other authors reported similar outcome.[15-18] Both standard parotidectomy and facelift approaches permit frozen sectioning and easy conversion into neck dissection if necessary. These approaches are also appropriate in clinically positive lymph node settings.

The direct approach through transcutaneous mid-cheek incision reduces the damage of the surrounding tissue and shortens the duration of treatment. In our opinion, this approach should be reserved for experienced surgeons, due to high risk of injury of facial nerve.[4] The chance of damaging peripheral branches and Stensen's duct may be increased up to 40%, as reported.[4] Other potential complications include seeding of the tumor, development of a salivary fistula and local recurrence.[4,13]

The main advantages of the direct transoral approach are no visible scars and time-saving compared to the standard parotidectomy incision.[14,19-21] However, it is limited to cases of small-sized tumors located more anteriorly, which reduces the risk of Stensen's duct or facial nerve impairment.[14] Its disadvantage is reduced operative viewing field implicating difficulties in manipulation and bleeding control.[13,17] For these reasons, the direct transoral approach has been described as "ill-advised".[17] Some authors used a nerve monitor to identify small nerve branches.[21,22] Even though the monitor indicate near the nerve, it does not guarantee visualization, and is therefore a routine visualization recommended.[22]

In recent years, endoscopic-assisted surgery has become more frequent in the head and neck. There are few reports on endoscopic assisted treatment of APGT.[6-9] Xie et al. first described such a technique in a series of 5 patients via a 4-5cm preauricular incision and assistance of working space-maker.[6] Li et al. modified the previously mentioned approach with shorter and less visible skin incisions.[7] However, in all series of transcutaneous endoscopic procedures, only benign APGT less than 3.2 cm in size were included.[6-8] To date, only one case of transoral endoscopic-assisted resection of APGT has been described.[9] The advantages of endoscopic-assisted surgery over traditional surgery include reduced tissue damage, low incidence of wound-related complications and minimal scarring. The endoscope provides sufficient illumination and magnification of the operative field.[6,8] Postoperatively, no case of facial nerve palsy, infection, salivary fistula, gustatory sweating syndrome or local recurrence has been noted, all reporting satisfactory cosmetic result.[6-9] The follow-up periods varied between 2 and 14 months. On the contrary, disadvantages of endoscopic-assisted surgery are inability to operate large and malignant tumors, reservation for trained professionals in endoscopic techniques and a time-consuming procedure which is difficult to compare with traditional surgery due a small case series.[6-8] Furthermore, discrepancies between preoperative FNAC and definitive patohistological diagnosis can lead to inadequate resection of an APGT initially considered to be benign, if using an endoscopic approach. Additionally, primarily due to oncological reasons, the feasibility of endoscopic-assisted surgery of benign APGT is questionable. This opinion is supported by previous studies which reported reduced recurrence rates (less than 4%) associated with extended surgical technique compared to high incidence of recurrence (25-40%) following enucleation of the parotid gland PA.[67-69] Two patients referred to the our institution due to local recurrence after previous treatment of PA using direct transcutaneous approach, also support the importance of adequate tumor resection. The re-operation carries a higher risk of facial nerve injury and local

recurrence which varies from 15 to 30% and 15 to 75%, respectively.[70-73] Moreover, since recurrent tumors of the parotid occurs 3-9 years after initial surgical treatment, short follow-up in cases of endoscopic-assisted resection studies is not a sufficient for recurrence detection and analysis of true recurrence rates.[71,73] Endoscopic-assisted approaches may be considered as an alternative in the treatment of small benign APGT, but larger series with an updated and mature follow-up period are needed in order to utilize its full potential in APGT treatment.

Accessory parotid gland tumors are very rare, but should be considered in the differential diagnosis of a mid-cheek mass. According to our results patients with APGT were younger than other authors reported. Detailed diagnostics and pretreatment work-up are needed in order to avoid misdiagnosis and undertreatment. Approaches through standard parotidectomy and facelift incision are recommended for surgical treatment of APGT, with minimally invasive techniques being reserved for benign subtypes. Although series of malignant APGT are limited, the survival is favorable. Further investigations and large prospective and multicenter trials are needed in order to define optimal extent of surgery as well as adjuvant treatment modalities.

Declarations

Funding and Conflict of Interests: We disclose any commercial associations that might pose a potential, perceived or real conflict of interest. These include grants, patent licensing arrangements, consultancies, stock or other equity ownership, donations, advisory board memberships, or payments for conducting or publicizing the study. All authors have viewed and agreed to the submission.

Ethical Approval: The Ethics Board of University Hospital Dubrava has decided that a special ethical approval is not needed because all involved in this study signed written patient consent.

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Captions to Illustrations

Figure 1. MRI and MSCT scans of patients with APGT. Arrows point to the tumor.

Figure 2. Facelift approach skin incision.

Figure 3. Intraoperative photograph.

Figure 4. Postoperative photograph (14 months after surgery with facelift approach).

Table 1. Clinical features of the study group.

Table 2. Review of the published cases of APGT.