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Benign tumors of the superficial lobe of the parotid gland

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Abstract

Tumors of the parotid gland are a rare occurrence among head and neck tumors (only 3-8%), and the vast majority are benign. Most benign tumors of the parotid gland are pleomorphic adenoma (PA), followed by Warthin's tumor (WT) and they occur in the superficial lobe. The only reasonable treatment is considered to be total tumor removal, but the appropriate surgical approach remains a matter of controversy. Over time, it went from enucleation to superficial parotidectomy (SP) and then to extracapsular dissection (ECD). SP is often considered to be the "gold standard"; however, the risk of intraoperative damage to the facial nerve (FN) cannot be ignored. Lately, ECD – which does not expose the main trunk of the FN - has been regarded as a safe technique for the treatment of small benign parotid tumors. Patients, Materials and Methods: The study retrospectively reviewed all parotidectomies for benign parotid tumors of the superficial lobe performed in the Department of ENT, Municipal Emergency Clinical Hospital, Timisoara, Romania, between January 2014 and December 2018. The following patient data were collected: age; sex; follow-up duration; extension of the resection; use of FN monitoring and lesion features, including position, pathology, and size. Preoperative investigation protocol included a computed tomography (CT) scan to assess the size and location of the tumor helping the treatment planning. The same surgical team performed both standard SP and ECD for the treatment of these tumors. All cases were confirmed as benign parotid tumors. Recurrences of any kind and tumors involving the deep lobe of the gland were excluded from the study. All patients underwent a macroscopically complete resection of the tumor. The main outcome was the postoperative complications in the two groups. Data regarding these complications were recorded on follow-up visits at one, six, 12 and 24 months. Results: Ninety-five patients with benign parotid tumors were operated by the same surgical team at the Department of ENT, Municipal Emergency Clinical Hospital, Timişoara, between January 2014 and December 2018 but only 89 (38 men and 51 women) met the follow-up criteria and were included in the study. Sixteen (18%) patients underwent ECD, and 73 (82%) patients underwent SP as a primary intervention. The mean lesion size for ECD group was 2.0±0.9 cm and 4.2±1.7 cm in the SP group, and it was statistically significant (p<0.05). As expected, the most common type of tumor was PA but the distribution between the groups was different: in the SP group, PA represents 83% (61/73) of cases but only 44% (7/16) of cases in the ECD group. Median follow-up time was comparable in the two groups - 33±8 months for the ECD group and 39±11 months for the SP group. There were also differences between the two groups regarding the postoperative complication rate. There were significantly more transient and permanent FN paralysis, transient and permanent great auricular nerve (GAN) dysfunctions and Frey's syndrome (FS) in the SP group. Conclusions: A comparison between ECD and SP as surgical procedures for the treatment of benign tumors of the parotid seems inappropriate as their indications do not overlap. ECD showed similar effectiveness and fewer side effects than SP and can be considered the treatment of choice for small, superficial and/or marginal tumors located in the lateral lobe of the parotid gland.

Keywords: parotid gland, benign tumors, nerve paralysis, pleomorphic adenoma, parotidectomy.

→ Introduction

Tumors of the parotid gland are a rare occurrence among head and neck tumors (only 3–8%) [1–3], and the vast majority are benign. The submandibular and sublingual glands are even less involved by tumors, but they are more frequently malignant. These are smaller salivary glands, mostly involved in inflammatory and infectious pathology. Being such a rare occurrence, tumors of the parotid gland were not a main topic in otolaryngology. Most benign tumors of the parotid gland are pleomorphic adenoma (PA), followed by Warthin's tumor (WT). In the submandibular

and sublingual glands, the most frequent benign tumor is also PA. Most of the benign parotid tumors are located in the superficial lobe [1, 2]. The only reasonable treatment is considered to be total tumor removal [3]. The main problems with parotid gland surgery are the facial nerve (FN) running through the gland and the recurrence of the tumor in case of incomplete removal. Due to its drawbacks – a very high recurrence rate, up to 40–45% –, enucleation is no longer an option. To overcome these problems, the surgical treatment shifted from initial enucleation to a much larger resection – superficial parotidectomy (SP). Despite having a very low recurrence rate (as low as 2%),

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SP presents some dangerous or annoying complications like FN palsy, salivary fistulas, and Frey's syndrome (FS) – (sweating while eating and facial flushing) [4]. The obvious solution was to limit the dissection of the FN – partial superficial parotidectomy (PSP) - but again you need to identify and dissect the main trunk of the FN, and this is usually the dangerous part. It was reported that PSP allows for complete tumor resection with fewer complications then SP. But an exact extension of what is called a PSP is not clearly specified and it is left up to the surgeon. However, when dealing with a small tumor, a SP or even a PSP might look as overtreatment. Minimally invasive surgery made its mark in parotid surgery as well. It started with PSP, but it went further to an extended type of enucleation without the dissection of the main trunk of the FN called extracapsular dissection (ECD). The reported results were very good in the - very experienced, actually - hands of those who proposed this technique but again there are different indications for this procedure, especially regarding the size and location of the tumor. Albergotti et al. (2012) reported similar recurrence rates between ECD and SP, ECD having slightly better results regarding the complication rate [5]. These results were supported by other studies (Xie et al., 2015), ECD being recognized as a good treatment alternative for selected cases [6]. After all, a SP is an extensive ECD (FN dissection being mandatory due to the proximity of the tumor) but the debate over which technique is the "gold standard" continues as each of the aforementioned procedures has its own disadvantages.

Patients, Materials and Methods

The present study was approved by the Ethics Review Board of the Victor Babeş University of Medicine and Pharmacy, Timişoara, Romania, and the informed consent was obtained from the involved patients. The study retrospectively reviewed all parotidectomies for benign parotid tumors of the superficial lobe performed in our Department between January 2014 and December 2018. The following patient data were collected: age; sex; follow-up duration; extension of the resection; use of FN monitoring and lesion features, including position, pathology, and size. Preoperative investigation protocol included a computed tomography (CT) scan to assess the size and location of the tumor helping the treatment planning. The type of surgery was decided according to clinical evaluation and imaging results. The same surgical team performed both standard SP technique and ECD for the treatment of these tumors. ECD was performed if the tumor was small less than 2.5 cm in its larger diameter – or marginally and/or superficially located in the gland. SP was performed if the tumor was more centrally located in the superficial lobe, or it was bigger than 2.5 cm. Total parotidectomy cases were not included in the study nor tumors that were proven to be malignant on histology. Intraoperative FN monitoring was not usually used in cases of SP, but it was always used in cases of non-marginal tumors in which we performed ECD. Postoperative drainage was always used, and it was maintained for three days on average. All cases were confirmed histopathologically as benign parotid tumors and the types encountered were PA, WT, benign myoepithelioma (BME) and oncocytoma (OC). Recurrences of any kind and tumors involving the deep lobe of the gland were excluded from the study. All patients underwent a macroscopically complete resection of the tumor. The main outcome was the postoperative complications in the two groups. Postoperative complications considered were FN weakness/paralysis, FS, salivary fistula, hemorrhage, hematoma, seroma, wound infection, and numbness of the earlobe. Data regarding these complications were recorded on follow-up visits at one, six, 12 and 24 months. The House–Brackmann (HB) FN grading system was used to assess the function of the FN. The alteration of the great auricular nerve (GAN) sensitivity was evaluated in the earlobe and pre-auricular skin, and it was recorded at the same visits. Both nerve dysfunction – FN and GAN – were further classified as persistent or transitory if they were encountered or not at 12-month follow-up. Symptomatic FS was defined as postoperative gustatory sweating described by the patient as frequent and annoying. All patients required a minimum of 2-year follow-up to be included in the study.

There were 95 patients with benign parotid tumors operated by the same surgical team at ENT Department, Timişoara, between January 2014 and December 2018 but only 89 (38 men and 51 women) met the follow-up criteria – primary benign tumors of the parotid gland only, a minimum of 2-year follow-up, visits at one, six, 12 and 24 months – and were included in the study. Mean age at presentation was 49.8 years (age limits between 21 and 77 years). Out of 89 surgeries, 16 (18%) cases were ECD, and 73 (72%) cases SP. FN monitoring was used in five ECD cases – all non-marginal tumors – and in four large parotid tumors (>6 cm in diameter) operated with SP. There were significant differences between the two group features mainly regarding the size of the tumor, the histopathology, and the number of cases (Table 1).

Table 1 – Patient characteristics in the two groups

Parameter	ECD group	SP group
Age, mean ± SD [years]	62±12	42±21
Gender, M/F [n]	6/10	32/41
Tumor size, mean ± SD [cm]	2.0±0.9	4.2±1.7
Location (marginal, superficial/ non-marginal, superficial) [n]	11/16	4/73
Histology (PA/WT) [n]	7/9	61/12
FN monitoring [n]	5/16	4/73
Follow-up period, mean ± SD [months]	33±8	39±11

ECD: Extracapsular dissection; F: Female; M: Male; *n*: No. of cases; PA: Pleomorphic adenoma; SD: Standard deviation; SP: Superficial parotidectomy; WT: Warthin's tumor.

The mean lesion size for ECD group was 2.0 ± 0.9 cm and 4.2 ± 1.7 cm in the SP group, and it was statistically significant (p<0.05). As expected, the most common type of tumor was overall the PA but the distribution between the groups was different: in the SP group, PA represents 83% (61/73) of cases but only 44% (7/16) of cases in the ECD group.

The WT was the other tumor encountered in our study. In the ECD group, the main type of tumor was the WT, representing 56% of the cases (9/16) but it was only 17% of the cases (12/73) in the SP group. There was also one case of OC and one BME in the initial group, but they were

not included in this study as they did not complete the required follow-up. Median follow-up time was comparable in the two groups -33 ± 8 months for the ECD group and 39 ± 11 months for the SP group.

There also were differences between the two groups regarding the postoperative complication rate. We had two cases of permanent FN paralysis (one case HB grade II and one case HB grade III, both involving mandibular nerve) in the SP group but none in the ECD group.

The incidence of transitory FN paralysis was also significantly higher in the SP group [eight (9%) patients vs. none in ECD group]. Out of the eight patients, six cases were HB grade II and two were HB grade III, but they completely recovered before the 12-month visit. Marginal mandibular nerve was involved in five out of eight cases. The rates of transient and permanent GAN hypoesthesia were also different [89% (65/73) transient; 74% (54/73) permanent for SP group and 44% (7/16) transient; 25% (4/16) permanent for ECD group]. Other complications included three cases of seroma (all in the SP group), one hematoma in the ECD group and three symptomatic cases of FS in the SP group (Table 2).

Table 2 - Complications in the two groups

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Postoperative complications	ECD group 16 patients	SP group 73 patients
FN weakness – temporary (<6 months)	none	2 (1 HB grade II, 1 HB grade III)
FN weakness – permanent (>6 months)	none	8 (6 HB grade II, 2 HB grade III)
GAN hypoesthesia – temporary	7	65
GAN hypoesthesia – permanent	4	54
FS – symptomatic	none	3
Seroma	none	3
Hematoma	1	none

ECD: Extracapsular dissection; FN: Facial nerve; FS: Frey's syndrome; GAN: Great auricular nerve; HB: House–Brackmann; SP: Superficial parotidectomy.

→ Discussions

PA is the most frequently encountered benign parotid tumor (40–70%) and has an incidence of 2.4–4.9/100 000 persons/year [7, 8]. In our study, it represented 76% (68/89) of the cases if we only consider the patients who completed the follow-up period. Next in line is the WT, representing around 30% of benign parotid tumors [23% (21/89) in our study]. All other types of benign tumors are very rare [7– 9]. Most tumors present as a palpable lump in the parotid area. Most tumors are located in the superficial lobe. The superficial lobe can be well explored by high frequency ultrasound and with almost the same accuracy by CT and magnetic resonance imaging (MRI) [10, 11] but the FN cannot be positively identified by imaging. The only visual clue regarding the position of the tumor in the superficial lobe is provided by the relative location to the retromandibular vein, which usually runs parallel to the inferior branch of the FN. Imaging and even fine-needle aspiration biopsy (FNAB) can never completely rule out a parotid malignancy. In the past, it was considered that almost one-quarter of benign parotid tumors become malignant [12] but now it is considered that only PA can undergo malignant transformation especially when is recurring. The rate of malignant transformation in recurrent PA was 3.2% -

3.3% in two large populational studies – much lower than reported before [8, 9]. The malignant transformation of WT is rare. These tumors usually have a slow growth rate, but this can be variable and cannot be used as a prognostic factor nor in treatment planning. WT is more often encountered in elderly patients [13]. This means higher risks for general anesthesia; hence conservative management might be reasonable when a WT is suspected in such cases [14]. There are three important factors when planning for surgery in a benign parotid tumor: location, size, and histological phenotype of the tumor [15]. The classical approach for a tumor in the superficial lobe of the parotid is either a PSP (level I or II) or a SP (level I and II). When the patient presents a small mobile superficial tumor, ECD is an alternative if the surgeon has enough experience. Under these circumstances, more than half of all benign parotid tumors can be completely resected using ECD [16]. All parotid surgeons must be proficient with both procedures as they must sometimes change the approach from ECD to PSP or SP, depending on the situation. It is deceiving to compare SP with ECD, if the characteristics (size, location) of the tumors are not comparable in the two groups. Moreover, the indications for ECD varies considerably among different surgeons but usually are much more restrictive than for SP. Thus, we are at risk to compare apples and oranges [17]. However, several metaanalyses did this [5, 6, 18]. A parotid tumor suitable for ECD should be always small, mobile, superficial, or marginally located, whereas the patients who underwent a SP have usually larger and more centrally located tumors. We need to understand these techniques (ECD and SP) are not competing, they both benefit for the patient. SP is FN dissection surgery, whereas ECD is dissecting around the tumor without exposing – if possible – the FN, therefore FN monitoring is more often necessary in ECD. The morbidity is always considered directly related to the extent of surgery, so it seems obvious that ECD should have fewer complications [17]. A very important issue is the risk of tumor recurrence, mainly for PA because of their characteristic pseudopodia and areas without pseudocapsule – especially when very near to the FN branches. When dissecting close to the tumor, it is more likely to cut the pseudopodia and leave rests in situ with ECD, but it can also happen with SP mainly in cases with large tumor adjacent to the FN [17, 19]. Whichever technique is superior continues to divide the surgeons despite the selection criteria for ECD being very confusing. However, many studies considered ECD as an alternative to SP especially in the view of postoperative complications. It has been proven that ECD has a low complication rate, low morbidity, low recurrence rate, short operating time and short hospitalization making it a viable surgical technique in the treatment of the benign superficial parotid tumors. But it cannot be applied – unless you are a very experienced surgeon – to all superficial parotid benign neoplasms. Unfortunately, many of these comparisons are lacking clarity regarding the inclusion criteria for ECD (tumor size, location, or proximity to the FN). And this can be very confusing for the surgeons not very familiarized with parotid surgery and FN dissection. Many studies present significant differences in complication rates between ECD and SP. Usually, they only report slight differences in the rate of permanent paralysis of the FN, but a significant reduction of transitory FN paralysis (21%)

vs. 71%) and FS (20% vs. 71%) in ECD vs. SP cases [16]. However, other studies report significant differences in both permanent (0% vs. 8.9%) and transitory (3.9% vs. 26.8%) FN paralysis, and FS (0% vs. 5.3%) when comparing ECD to SP [20]. Most meta-analyses support these data regarding the risk of transitory and permanent FN palsy (9% and 1%, respectively, for ECD and 23% and 2% for SP) but they do not mention the lack of consistency regarding the indications for ECD [6]. Even in prospective trials, the results are the same – overall FN palsy rate was 24% for SP and 6% for ECD – but the clear including criteria in both groups are rarely presented [21]. Most large studies present temporary FN paresis rates of 15–20% and permanent FN paresis rates less than 5–6% for parotidectomy for benign disease, but they also include total parotidectomies [22– 25]. However, other surgeons demonstrated the opposite in both temporary (22.8% vs. 9.8%) and permanent (9% vs. 5.9%) FN paresis, when comparing ECD to SP [26]. Other studies also presented comparable or even better complication rates with SP – temporary (2.7%) and permanent FN paresis rates (0.9%) and symptomatic FS rates of 1.5% [27]. In our study, the temporary FN paresis rate was 0% in the ECD group and 11% (8/73) in the SP group, and the permanent FN paresis rate was 0% in the ECD and 2.7% (2/89) in the SP group. The size of the tumor seems to be the most important factor to consider when deciding the type of surgery, assuming that the surgeon is familiarized with both procedures. But the tumor size reported in the inclusion criteria seems to vary according to the surgeons' experience: for Dell'Aversana Orabona et al. – tumors smaller than 3 cm underwent ECD [20]; for George & McGurk, this limit raises to 4 cm [16]. As we do not have the experience of the aforementioned authors, we consider that any benign parotid tumor of the superficial lobe that is not marginal, nor superficial, or it is larger than 2.5 cm should be resected using a SP. Other factors associated with increased morbidity, especially of the FN, in parotid surgery are patient's age, tumor location and presence of intraglandular inflammation [28–30]. In our study, most tumors were located near the inferior branch of the FN, in the inferior half of the gland (61%; 54/89), and we consider this might explain the statistical relationship between relative frequent weakness of the marginal-mandibular branch and location, when compared to other branches. Our results suggest that the inferior location of tumor, the size of tumor >2.5 cm and the prolonged operative time are risk factors for FN paralysis. Preserving the GAN is controversial and sometimes – due to the location of the tumor – impossible. The resection of GAN causes hypoesthesia in its territory, which might be felt uncomfortable [31, 32]. In our study, 80% (71/89) of the patients presented hypoesthesia at one month, no matter the nerve was preserved or just stretched, with only 58% (52/89) maintaining their deficit after 12 months. The collateral branches compensate this deficit, along with the auriculotemporal and occipital nerves [32]. Any effort aimed at preserving the branches (the posterior one especially) must be made to improve the sensory deficit – at 12 and 24 months, respectively, 59% and 71% of the patients showed no hypoesthesia in the lobule if the nerve branch was preserved, vs. 24% and 31% if not [33]. FS is another major complication, and its incidence varies from 6% to 96%. Approximately less than 10% of patients complain of FS at 12 months [34, 35]. In our study, we only considered

the diagnosis when the patient complained about it. Thus, we had an occurrence rate of 3.4% for the FS (three cases). This low occurrence rate might be well explained by the lack of complaints. The reported rate is usually much higher even in prospective studies (46% and 43%, respectively, after 12 and 24 months) but they include both asymptomatic and symptomatic patients [33].

☐ Conclusions

In experienced hands, ECD is a very good surgical alternative to SP regarding clinical outcomes. However, ECD cannot be employed in every benign parotid tumor even in experienced hands and requires a proper selection of patients. The selection criteria are not yet standardized and are still largely dependent on surgeons' experience. In our study, tumors treated with SP had short- and long-term FN weakness rates of 11% and 2.7%, respectively. These are acceptably low FN paralysis rates and surgery was usually performed without the routine use FN monitoring. There was also a low occurrence of clinical FS (3.4% – three cases), much lower than usually reported. There are a lot of serious complications related to parotid surgery, so knowing when to employ both techniques is of crucial importance in achieving better results and also in counseling to patient. The results of ECD are very promising, but the importance of appropriate lesion selection and the experience of the surgeon need to be stressed.

Conflict of interests

The authors declare that they have no conflict of interests.

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