



ORIGINAL ARTICLE

Surgery in superficial-lobe pleomorphic parotid adenoma

Diane S. Lazard, Bertrand Baujat, Isabelle Wagner, Frédéric Chabolle

ENT and Head&Neck Surgical Department - Foch Hospital - Suresnes - France.

ABSTRACT

Study objective: To review the 1982-2003 literature for data comparing superficial and total parotidectomy in patients with superficial-lobe pleomorphic adenoma.

Material and methods: Original articles selected from Medline were evaluated for data on postsurgical facial nerve dysfunction and recurrences.

Results: All articles provided level IV evidence in the Sackett classification. Facial nerve dysfunction, for which classification was lacked standarization, was more common in patients older than 40 years and occurred with a mean rate of 1.3% and 2.9% after superficial-lobe and total parotidectomy, respectively. Recurrence rates were less than 2% with both techniques. Younger age at initial diagnosis was significantly associated with recurrence; mean ages at initial diagnosis were 30 and 45 years in patients who did and did not experience recurrences, respectively.

Conclusion: Given the discrepancies across case-series, until prospective data are available, total parotidectomy seems preferable in patients younger than 40 years, since their recurrence rate is higher. Superficial-lobe parotidectomy may need to be reserved for patients older than 40 years, to reduce the rate of facial nerve dysfunction.

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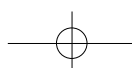
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Corresponding author: Frédéric Chabolle

Service ORL et Chirurgie Cervicofaciale - Hôpital Foch,

40 rue Worth, 92151 Suresnes - France

e-mail: f.chabolle@hopital-foch.org



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INTRODUCTION

Pleomorphic adenoma is the most common tumor of the parotid gland (60 to 70% of all tumors) [1-2]. There is general agreement that pleomorphic adenoma is a benign tumor with a potential for local recurrence [2-4] and an extremely low rate of malignant transformation (1-2% of primary parotid gland malignancy) [5-6].

The hypothesis that recurrences are ascribable to a multifocal distribution of the tumor at diagnosis has been refuted. Fingerlike extensions through the capsule of the primary tumor have been incriminated instead [3-4].

Surgery is the main treatment of pleomorphic adenoma. Enucleation is no longer recommended [1, 4-6]. In patients with pleomorphic adenoma of the superficial lobe, either superficial or total parotidectomy can be used. Superficial parotidectomy may be associated with a higher recurrence rate and total parotidectomy with a higher risk of facial nerve dysfunction. The debate regarding the merits and shortcomings of these two techniques has been active since 1945 [7]. This continuing uncertainty stems from the absence of prospective randomized studies with sufficiently long follow-ups. The objective of the present work was to compare outcomes after superficial and total parotidectomy for superficial-lobe tumors, using data from a systematic literature review.

MATERIAL AND METHODS

Article selection

We searched the international medical-literature databank Medline for articles published between 1982 and 2003. The key indexing terms "pleomorphic adenoma", "surgery", and "parotidectomy" retrieved 124 articles.

To be included in our study, articles had to report on the surgical treatment of pleomorphic adenoma in adults, to be written in English or in French (this last being the authors' native language), and to be published in readily accessible journals. We excluded case-reports, letters to the editor, experimental studies, pathological studies, studies that included parotid gland tumors other than pleomorphic adenoma, and papers written in languages other than English or French.

The 22 articles [1-6, 8-23] that met our inclusion and exclusion criteria were examined as recommended in

guidelines for international literature reviews [24]. Table I shows the grid used to evaluate the articles.

Table I: Grid for evaluating selected articles

Study category	Prospective Retrospective
Subject selection in retrospective studies	Inclusion of all patients who had surgery during the study period Inclusion of only those patients who presented with complications
Epidemiological data	Location of the pleomorphic adenoma Size of the operative specimen
Type of surgical procedure	Superficial parotidectomy Total parotidectomy Enucleation
Complications	Frey syndrome Facial nerve dysfunction Recurrence Malignant transformation
Data collection	Total number of inclusions, number lost to follow-up Study duration, mean follow-up, range

The chi-square test was used to compare complication rates with superficial versus total parotidectomy.

RESULTS

All 22 articles selected for the study were level IV in Sackett's classification for levels of evidence [24] (Table II).

All the studies but one [19] used a retrospective design. Seven studies [1-2, 5-6, 14, 18-19] included all patients treated surgically for pleomorphic adenoma and eight others [3-4, 8-9, 11, 20-21, 23] included only patients who presented with postoperative complications. Four studies [10, 12-13, 22] compared enucleation to superficial parotidectomy and one study compared five surgical techniques (partial and total superficial, partial superficial/deep lobe, selective deep lobe, and total parotidectomy) [16].

Another study used radiation therapy in combination with surgery [17]. Finally, we included a 1953-1979 literature review that reports results without comparing the designs of included studies [15].

Our comparison of facial nerve dysfunction and

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Table II: Levels of evidence from published studies and grades of recommendation (modified from Sackett's classification scheme)

Level of evidence	Grade of recommendations
<p style="text-align: center;">Level I</p> RCTs with strong statistical power Meta analysis of randomized comparative studies Decision algorithms based on carefully conducted studies	<p>A</p> Conclusive scientific proof
<p style="text-align: center;">Level II</p> RCTs with limited statistical power Nonrandomized, carefully conducted, comparative studies Cohort studies	<p>B</p> Scientific presumption
<p style="text-align: center;">Level III</p> Case-control studies	<p>C</p> Low level of scientific evidence
<p style="text-align: center;">Level IV</p> Comparative studies having major sources of bias Retrospective studies Case-series Descriptive epidemiological studies (cross-sectional, longitudinal)	

RCT : Randomized controlled trial

recurrence rates was based only on the seven studies that included all treated patients. Confining a study to those patients who present with complications may lead to underestimation of the complication rate, as patients lost to follow-up are not counted.

Epidemiological data

In over 50% of cases (range, 51.5%-90%), the pleomorphic adenoma was found upon surgery to be located in the superficial lobe [1-2, 14, 16]. At primary surgery, the operative specimen was larger than 2 cm in 87% of patients in one study [1], and in another the mean size was 3.3 cm (median, 2.5 cm; range, 0.5-14 cm) [14].

Three surgical techniques were used.

Enucleation, i.e., dissection along the tumor capsule severing microscopic extracapsular extensions left residual tumor tissue in 30% of cases [5-6] and was associated with a greater than 10% recurrence rate [1, 3-4, 8, 22]. Enucleation is no longer performed and will not be discussed further in this review.

In seven studies [2, 4, 6, 14, 16, 18, 20] including 1305 patients in all, *superficial parotidectomy* was considered the best technique for tumors that did not extend beyond the plane of the facial nerve. Four of these seven studies included all surgically treated

patients [2, 6, 14, 18]. Low recurrence rates and a decreased risk of facial nerve injury were reported. Extension of the adenoma beyond the plane of the facial nerve led to removal of at least part of the deep lobe. The extent of surgical excision was described in four studies [4, 6, 14, 16] : superficial parotidectomy was defined as excision of the part of the gland lateral to the nerve and total parotidectomy as excision of the entire parotid gland including the portion lying under the facial nerve against the masseter and mandibular ramus [16]. Median and mean patient ages were similar across studies, with a range of 44.1 to 48 years.

Total parotidectomy was investigated in five studies including 545 patients in all [1, 3, 5, 10, 19]. Recurrence rates seemed lowest with this technique. Of these five studies, three included all surgically treated patients [1, 5, 19]. Facial nerve injury was more common and more severe after repeat surgery to treat recurrences (facial nerve dysfunction in 14% to 16% of patients [6, 10, 14, 20]), leading the authors to advocate total parotidectomy as the primary procedure. One study report specified that the superficial temporal and medial maxillary vessels were ligated routinely and that the external carotid artery was removed with the deep lobe of the parotid gland in 98.2% of cases [1]. The age distributions of the patients cannot be compared, as two study reports fail to specify the mean or median age [1, 19] and the third provides age data only in patient subsets [5].

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Overall, the two main complications recorded after the immediate postoperative period were Frey syndrome and facial nerve dysfunction (paresis or paralysis).

Frey syndrome occurred in 14% to 30% of patients after superficial parotidectomy [14, 18] and in 70% to 89 % after total parotidectomy [1, 5]. Mean time from surgery to symptom onset was 8 months [14].

Data on facial nerve injury are difficult to compare across studies because of differences in the qualitative and quantitative assessments of the abnormalities. None of the studies used the House and Brackmann classification scheme [25]. Five studies separated partial from total dysfunction and transient from permanent dysfunction [1-2, 14, 18-19] (Table III). Only two study reports provide a definition for paresis (muscle weakness in a well-defined distribution) and paralysis (complete absence of movement in half the face) [1, 14]. The duration used to define transient versus permanent dysfunction varied across studies;

thus Laccourreye et al. stated that recovery could take up to 18 months, whereas Laskawi et al. considered that dysfunction present after 6 months was permanent.

In our comparison, we accepted the categorizations into transient and permanent dysfunction given in each study. We did not separate partial from complete dysfunction, as the variations in definitions were considerable. Table IV reports the outcomes with each surgical technique. The number of patients per study ranged from 60 to 229, and surgery was the only treatment used. Transient facial nerve dysfunction occurred in 32% to 35.8% of patients after superficial parotidectomy and 15% to 70% after total parotidectomy. Mean rates of permanent facial nerve dysfunction were 1.3% (range 1% to 1.4%) after superficial parotidectomy and 2.9% (range 0.8% to 3.9%) after total parotidectomy (Table IV). The difference between the two techniques is not statistically significant ($P=0.7$). Recurrence rates according to follow-up duration and surgical technique are reported in Table V. With

Table III: Definitions of facial nerve dysfunction used in five published studies

	Category	Definition	Distribution	Duration
Laskawi et al. [14]	Persistent	+	-	>6 months
Mc Gurk et al. [2]	Transient vs. permanent	-	-	-
Marandas et al. [18]	Partial vs. total Transient vs. permanent	-	-	-
Maynard [19]	Immediate vs. permanent	-	5	-
Laccourreye et al. [1]	Paresis vs. paralysis Transient vs. permanent	+	4	>18 months

Table IV: Facial nerve dysfunction according to the surgical technique

Superficial parotidectomy						
	N	Lost to FU	Age	Mean/Median age	Transient	Permanent
Laskawi et al.[14]	139	-	14-93	48	-	1.4%
Mc Gurk et al.[2]	95	-	5-87	47	32%	1%
Marandas et al.[18]	81	-	-	-	35.8%	1.2%
Mean	105					1.3%
Total parotidectomy						
	N	Lost to FU	Age	Mean/Median age	Transient	Permanent
Maynard [19]	114	6	13-92	-	15%	0.8%
Laccourreye et al.[1]	229	51	16-81	-	70%	3.9%
Laskawi et al.[14]	60	-	14-93	48	-	3.3%
Mean	134	-				2.9%

FU: follow-up

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Table V: Recurrence rates according to the surgical technique : for the studies by Maynard. and Laccourreye et al, we report only the results in the patients with follow-up durations greater than 5 years and 10 years, respectively. The number of patients lost to follow-up in these subsets is not specified in the study reports.

Superficial parotidectomy							
	<i>N</i>	<i>Lost to FU</i>	<i>Age range</i>	<i>Mean/ Median age</i>	<i>Mean FU</i>	<i>Range</i>	<i>Recurrence rate</i>
Mc Gurk et al.[2]	95	-	5-87	47	12 years	1 - 34	2%
Laskawi et al.[14]	139	-	14-93	48	5 years	-	0.7%
Myssiorek et al. [6]	139	2	15-78	-	-	6 - 39	0.7%
Mean	124						1.1%
Total parotidectomy							
	<i>N</i>	<i>Lost to FU</i>	<i>Age range</i>	<i>Mean/ Median age</i>	<i>Mean FU</i>	<i>Range</i>	<i>Recurrence rate</i>
Laskawi et al.[14]	60	-	14-93	48	5 years	-	0%
Chevalier et al.[5]	293	-	-	-	-	6 - 26	2.4%
Maynard [19]	63		13-92	-	> 5years	5 - 20	1.6%
Laccourreye et al.[1]	100		16-81	-	>10 years	10 - 20	1%
Mean	129						1.7%

FU: follow-up

superficial parotidectomy, the mean recurrence rate was 1.1% (range, 0.7% to 2%) after 1 to 39 years of follow-up. Ranges were not specified in all reports. A similar recurrence rate of 1.7% (range, 0% to 2.4%) was noted after total parotidectomy, after 6 to 26 years of follow-up. The difference in recurrence rate between the two techniques was not statistically significant ($P=0.4$).

The mechanisms underlying recurrence of pleomorphic adenoma are poorly understood. However, the patient subset with recurrences was significantly younger at diagnosis of the first tumor (mean, 29 years; range, 25-33 years) than the subset without recurrences (mean, 49 years; range, 45-54 years). 4-6, 17 Multifocal lesions were present in 34% to 62% of recurrences [4-5]. The most common sites of recurrence were the superficial lobe (67% to 83%) and the cutaneous scar (79%) [3-6]. The mean diameter of the adenoma at recurrence was 3.2 cm (median, 2.5 cm; range, 1-10 cm) [4] in one study and 1.4 cm in another study [20].

Time to recurrence ranged from 5 to 10 years. In one study, 95% of recurrences were diagnosed after a mean of 10.1 years (median, 8 years; range, 2-43 years) [4]. Thus, at least 10 years of follow-up are needed to rule out a potential for recurrence [2-4, 6]. However, among studies of total parotidectomy, only one [1] reports outcomes after more than 10 years. The recurrence rate in this study was 1%.

DISCUSSION

The need for removal of the deep lobe in patients with pleomorphic adenoma of the parotid gland extending beyond the facial nerve is undisputed [4, 13, 20]. In contrast, no statistically reliable data are available for choosing between superficial and total parotidectomy in patients with pleomorphic adenoma of the superficial lobe. We found only level IV evidence in the literature. Data collection, patient recruitment, and treatment strategies varied widely across studies.

Our systematic literature review indicated that Frey syndrome was more prevalent after total parotidectomy than after partial parotidectomy. Transient facial nerve dysfunction was common after first-line surgery with both techniques: 32% to 35.8% after superficial parotidectomy and up to 70% after total parotidectomy.

Caution is needed when interpreting these data, given the wide variations in criteria used to define partial versus total dysfunction and transient versus permanent dysfunction. Improvement in nerve function over time was the rule after superficial parotidectomy. Rates of permanent nerve dysfunction were 1.3% with superficial parotidectomy and 2.9% with total parotidectomy; the difference is not statistically significant. Patient age may play a greater role than surgical technique in determining the risk of facial nerve dysfunction. Laccourreye et al. found a statistically significant

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increase in the risk of facial nerve dysfunction in patients older than 40 years at surgery [1]. This finding is consistent with studies showing age-related decrease in the axonal regeneration rate and in the expression of the main myelin proteins [26-27].

Recurrence rates were low (<2%) with both techniques. Nevertheless, outcomes after superficial and total parotidectomy are difficult to compare in this respect. Indeed, follow-up must be 10 years or more for a reliable evaluation of the recurrence rate, although 80% of recurrences are diagnosed within 5 years of the first surgical procedure [2-4, 6]. A single study [1], which evaluated total parotidectomy, had follow-up durations of 10 years or more for 100 patients. Recurrence rates were probably underestimated in the other studies of total parotidectomy and in the studies of superficial parotidectomy. In addition, none of the studies determined the actuarial recurrence rate, a fact that severely limits the validity of our comparative tests.

The variability in methods used to assess nerve dysfunction and the short follow-up durations preclude a firm conclusion about the superiority of one technique over the other.

A striking finding from our review is that patients with recurrences were significantly younger at diagnosis of the first tumor (mean age, <30 years) than patients without recurrences (45 years) [4-4, 17, 20]. In contrast, the risk of facial nerve dysfunction was lower in the younger patients [1]. Given these statistically validated results and the absence of further data in the literature, it seems reasonable to advocate total parotidectomy in patients who are younger than 40 years at the diagnosis of superficial pleomorphic adenoma of the parotid gland, with the goal of minimizing the recurrence risk. Patients older than 40 years can be treated by superficial parotidectomy in order to preserve facial nerve function, given the smaller risk of recurrence in this age group.

However, there is no proof to date that routine total parotidectomy decreases the risk of recurrence in younger patients with superficial pleomorphic adenoma. Younger age may be associated with aggressive tumor behavior, which may result in recurrences regardless of the surgical technique. A prospective 10-year longitudinal study using reference classification systems for defining facial nerve dysfunction would be the only means of obtaining reliable data and vali-

dating a surgical strategy for patients with pleomorphic adenoma of the parotid gland.

CONCLUSION

Our literature review on surgical treatments for pleomorphic adenoma of the superficial parotid lobe in adults does not provide reliable evidence for choosing between superficial and total parotidectomy.

Patients older than 40 years at diagnosis may be at higher risk for facial nerve dysfunction. Although the potential for recurrence is low overall, it is increased in patients who are younger than 30 years at diagnosis.

This may indicate that total parotidectomy is preferable for treating pleomorphic adenoma of the superficial lobe in patients younger than 40 years and superficial parotidectomy in patients older than 40 years. Prospective multicenter studies providing reliable data regarding the indications of superficial versus total parotidectomy are impatiently awaited.

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