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Extracapsular Dissection for Benign Parotid Tumors: A Meta-Analysis

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Objectives/Hypothesis: Recent studies suggest that extracapsular dissection (ECD) is an option for the resection of certain benign parotid tumors. This study investigates complication rates and effectiveness of ECD versus superficial parotidectomy (SP) for the treatment of primary benign parotid neoplasms.

Study Design: Systematic literature review with meta-analysis.

Methods: Studies available for inclusion evaluated the complications and effectiveness of ECD and SP as surgical techniques for the treatment of solitary, benign parotid tumors. An Ovid/Medline search revealed nine articles that met inclusion criteria. A critical review and meta-analysis of these articles was performed.

Results: The included studies evaluated a total of 1,882 patients. There was no observed difference in tumor recurrence between the ECD and SP groups (odds ratio [OR], 0.557; 95% confidence interval [CI], 0.271-1.147). There was a significantly lower rate of transient facial nerve paresis (OR, 0.256; 95% CI, 0.174-0.377) in the ECD group (59 of 741; 8.0%) compared to the SP group (81 of 397; 20.4%); however, there was no observed difference in permanent facial paralysis between the ECD and SP groups (OR, 0.878; 95% CI, 0.282-2.730). Frey's syndrome was less often observed (OR, 0.117; 95% CI, 0.071-0.191) after ECD (27 of 602; 4.5%) compared to SP (75 of 287; 26.1%).

Conclusions: This systematic review with meta-analysis suggests that ECD has a similar recurrence rate as SP with fewer postoperative complications. ECD may be considered an alternative surgical modality for select benign parotid neoplasms.

Key Words: Parotid neoplasm, parotid tumor, parotid surgery, parotidectomy, superficial parotidectomy, extracapsular dissection.

Level of Evidence: 2a

INTRODUCTION

Salivary gland neoplasms account for approximately 3% of all head and neck tumors.^{1,2} Approximately 80% of these tumors are found in the parotid gland, most often in the superficial lobe, where 80% of the tumors are benign, with pleomorphic adenoma being the most common subtype.² Before the 1930s, intracapsular enucleation, or "shelling out," of the tumor from the parotid gland was considered the treatment of choice for pleomorphic adenomas because of their benign clinical course, in addition to surgeon fear of damaging the facial nerve. It was at this time, however, that McFarland recognized the high recurrence rate of up to 45% following intracapsular enucleation.³ Bailey later attributed this high recurrence rate to incomplete removal of the tumor and therefore advocated superficial parotidectomy

(SP) with special attention to the removal of the capsule to cure pleomorphic adenomas.³⁻⁵

As SP was widely adopted, recurrence rates for pleomorphic adenoma fell, settling at a current rate of approximately 2%. However, while the recurrence rate improved, the number of complications increased, including a higher incidence of temporary and permanent facial nerve paralysis. Because of the frequency of complications from a surgery for a benign condition, surgeons have questioned the need to remove the entire superficial lobe while putting the facial nerve at risk.

Extracapsular dissection (ECD) has been offered as an alternative method to minimize the morbidity of parotidectomy. ECD involves careful dissection around the tumor capsule under magnified visualization without preidentification of the facial nerve.⁶ Several large series suggest that ECD has lower rates of complications without a higher recurrence rate compared to SP.^{7,8} Nevertheless, superficial and/or partial parotidectomy remains the gold standard treatment at most centers.

The objective of this study was to compare the outcomes of SP with those of ECD in the treatment of benign parotid tumors using a systematic review and meta-analysis. Specifically, we tested the null hypothesis that there is no difference between ECD and SP in the rate of tumor recurrence, facial nerve weakness, and Frey's syndrome.

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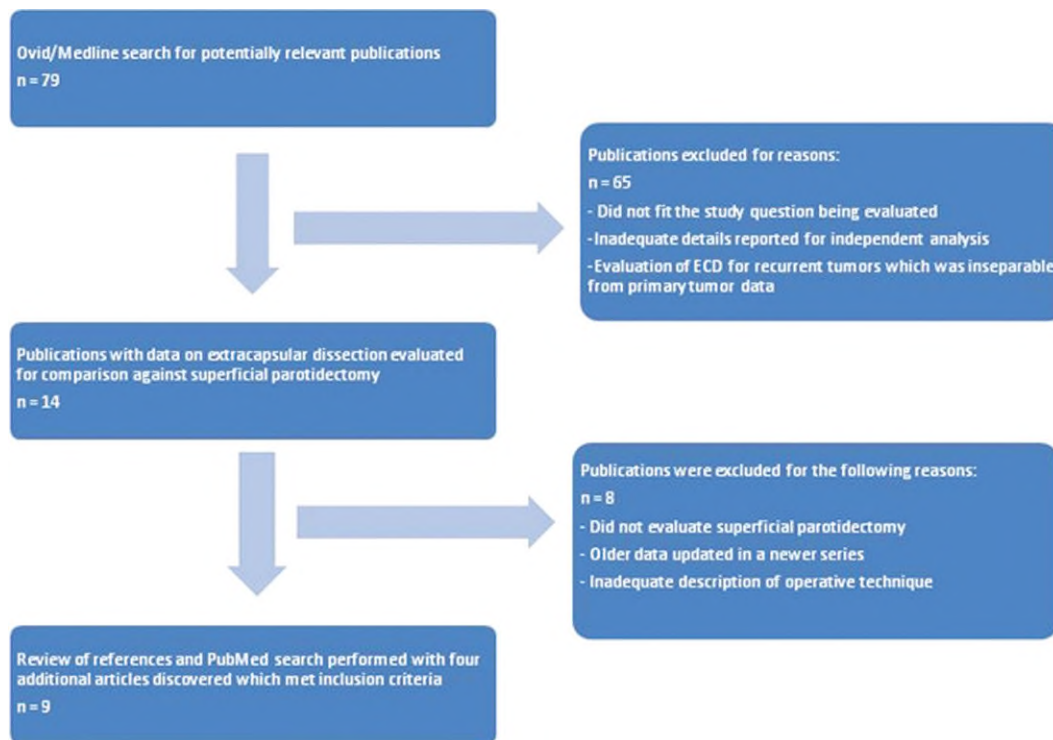


Fig. 1. Steps taken in the determination of papers to be included in the meta-analysis. ECD = extracapsular dissection. [Color figure can be viewed in the online issue, which is available at wileyonlinelibrary.com.]

MATERIALS AND METHODS

An Ovid/Medline literature search was performed using the following search terms: parotid neoplasm, extracapsular dissection, capsule, ECD, and dissection. There were no limits placed on study design. We included studies that compared ECD to SP with regard to at least one outcome of interest (recurrence, facial weakness, Frey's syndrome) in the surgical management of solitary, clinically benign parotid nodules. For the purpose of this study, ECD is defined by capsular dissection of the tumor with a thin margin of surrounding gland without planned identification of the main trunk of the facial nerve. SP is defined as both complete and partial SP, where a portion of the superficial lobe is removed with the tumor after planned identification and dissection of the facial nerve. Studies were excluded if they included recurrent or multiple tumors where the data could not be separated from primary, solitary tumors, involved nonparotid salivary tumors, or included malignant neoplasms. Article abstracts were reviewed to determine whether the inclusion and exclusion criteria were met.

The initial search returned 79 results, of which 14 included original data on ECD for benign, parotid neoplasms (Fig. 1). These 14 articles were obtained and reviewed in detail. A total of eight studies were excluded because there were no data on SP (6 studies) or involved an older series later updated by a single author group (2 studies). An additional three studies were identified that met inclusion criteria through a review of references and a concurrent PubMed search.

A comprehensive, retrospective data review was performed on the included articles with MedCalc 12.1 (MedCalc Software; Mariakerke, Belgium) using a fixed-effects meta-analysis model. The fixed-effects model assumes that all studies come from a common population and that the effect size (odds ratio [OR]) was not significantly different among the different trials. The Mantel-Haenszel method was used for calculating the weighted summary OR under the fixed-effects model. The heterogeneity

statistic was incorporated to calculate the summary OR under the random-effects model (DerSimonian and Laird). The random-effects model gave a more conservative estimate (i.e., with wider confidence interval [CI]), but the results from the two models usually agreed where there was no heterogeneity. The Q value, a measure of the heterogeneity of the included studies, tested the null hypothesis that the studies are homogeneous. If the test of heterogeneity was statistically significant ($P < .05$), then more emphasis would have to be placed on the random-effects model. The following null hypotheses were tested comparing ECD to SP: 1) No difference exists in the rate of Frey's syndrome; 2) No difference exists in the rate of transient facial nerve paresis; 3) No difference exists in the rate of permanent facial nerve paralysis; and 4) No difference exists in rate of tumor recurrence. Effect sizes were calculated using ORs with respective 95% CI. If the value 1 is not within the 95% CI, then the OR is statistically significant at the 5% level ($P < .05$), which means that the study did not demonstrate a significant difference between ECD and SP.

RESULTS

Nine studies clearly met inclusion criteria and were chosen for analysis (Table I). The nine studies evaluated included a total of 1,882 patients, of which 1,102 underwent ECD and 780 underwent SP. The median follow-up time was 12 years, ranging from 2 to 32 years. The majority of the studies were performed with a retrospective cohort design, although one was prospective. Among the studies included, there were five countries that were represented with a disproportionate number of cases being reported from Europe. The indications used by the authors for ECD varied between studies, but the tumors were generally superficial, clinically benign tumors of the parotid gland. Several limited the size of the tumor

TABLE I.
Characteristics of Included Studies.

Authors	Year	Country	Study Design	Randomization	No. of Subjects	Surgical Selection Criteria	Mean Follow-up Time (Range)
Gleave et al. ⁶	1979	United Kingdom	Retrospective cohort	No	369	Decision of ECD vs. SP made at time of surgery; factors that favored ECD included mobility, thin covering, size large enough to allow for digital manipulation	Unknown
Martis ¹²	1983	Greece	Retrospective cohort	No	176	ECD used for superficial tumors, up to 2.5 cm; SP performed for larger tumors or for visualization of the facial nerve	0–2 yr
Prichard et al. ²⁶	1992	United Kingdom	Retrospective cohort	No	46	ECD used for tumors in tail of the parotid; SP used for tumors in the body of the parotid or deep lobe	54 mo (minimum 2 yr)
Natvig and Soberg ²⁴	1994	Norway	Retrospective cohort	No	198	Only considered patients with pleomorphic adenoma; unclear criteria used for the determination of surgical technique	18 yr (11–25 yr)
Hancock ⁷	1999	United Kingdom	Retrospective cohort	No	101	Tumors that were superficial, mobile considered for ECD; converted to SP if boundary was lost between tumor and parotid parenchyma or for deeper, less mobile tumors	10.3 yr for ECD (3–21 yr); 8.3 yr for SP (3–22 yr)
Marti et al. ⁹	2000	Greece	Retrospective cohort	No	249	ECD applied to benign, superficial tumors that were smaller than 2.5 cm; unclear selection criteria for SP	Median, 18 yr; (6–26 yr)
Witt ²³	2002	United States	Retrospective cohort	No	40	Retrospective analysis of outcomes of pleomorphic adenoma treated with ECD vs. SP (all tumors smaller than 4 cm, superficial, mobile)	9 yr for ECD; 8 yr for SP
McGurk et al. ²⁰	2003	United Kingdom	Retrospective cohort	No	662	ECD performed on discrete, mobile tumors less than 4 cm in diameter; ECD vs. SP decision made at time of skin flap based on mobility	Median, 12 yr (range, 5–30 yr)
Uyar et al. ²⁷	2011	Turkey	Prospective cohort	No	41	All patients had FNA-proven pleomorphic adenoma in the superficial lobe of the parotid; unclear criteria used for the determination of surgical technique	194 mo (range, 117–264 mo)

ECD = extracapsular dissection; SP = superficial parotidectomy; FNA = fine-needle aspiration.

TABLE II.
Data Abstraction From Included Studies.

Authors	Temporary Facial Nerve Paresis		Permanent Facial Nerve Paralysis		Frey's Syndrome		Tumor Recurrence	
	ECD	SP	ECD	SP	ECD	SP	ECD	SP
Gleave et al. ⁶							5/257	2/112
Martis ¹²							0/98	0/78
Prichard et al. ²⁶	1/31	2/15	0/31	1/15	0/31	6/15	0/31	1/15
Natvig and Soberg ²⁴							0/5	5/193
Hancock ⁷	2/28	6/73	0/28	0/73	0/28	18/73	0/28	0/73
Marti et al. ⁹	6/139	18/110						
Witt ²³	0/20	4/20	0/20	0/20	0/20	2/20	0/20	0/20
McGurk et al. ²⁰	50/503	48/159	9/503	3/159	27/503	48/159	9/503	8/159
Uyar et al. ²⁷	0/20	3/20	0/20	0/20	0/20	1/20	0/21	0/20

ECD = extracapsular dissection; SP = superficial parotidectomy.

to 2.5 cm, and others used a cutoff of 4 cm. One study suggested that the tumor should be large enough for digital manipulation to be considered for ECD. With the exception of one study, fine-needle aspiration cytology (FNAC) and preoperative imaging were not routinely used unless there was suspicion of a malignant process. The number of patients in each study included in this analysis varied from 40 to 662, with the percentage of patients who underwent ECD in each study also varying widely, from 2.5% to 75% (Table II).

The recurrence rates for ECD and SP were 1.5% (14 of 963 cases) and 2.4% (16 of 670 cases), respectively; these findings were not statistically significant (OR, 0.557; 95% CI, 0.271-1.147) (Fig. 2). There was no difference in follow-up time specified in most of the studies; however, two of the articles mentioned a slightly longer follow-up for ECD cases compared to SP (9 and 10.3 years vs. 8 and 8.3 years, respectively).

The rates of transient facial nerve paresis for ECD and SP were 8.0% (59 of 741) and 20.4% (81 of 397), respectively, demonstrating a mean reduction of 75% in transient facial nerve paresis for ECD when compared to SP (OR, 0.256; 95% CI, 0.174-0.377) (Fig. 3). There was no difference in the rate of permanent facial nerve paralysis for the two techniques; however, 1.4% (8 of 590) of ECD and 1.1% (3 of 268) of SP cases experienced this complication (OR, 0.878; 95% CI, 0.282-2.730) (Fig. 4).

Symptomatic Frey's syndrome was reported by 4.5% (27 of 602) of ECD patients compared to 26.1% (75 of 287) of SP patients, which is an 88% reduction in symptomatic Frey's syndrome in the ECD group compared to the SP group (OR, 0.117; 95% CI, 0.071-0.191) (Fig. 5). None of the reports elucidated the methodology for evaluating Frey's syndrome. The studies likewise provided insufficient detail to determine the timing of the evaluation of Frey's syndrome, which may be relevant because

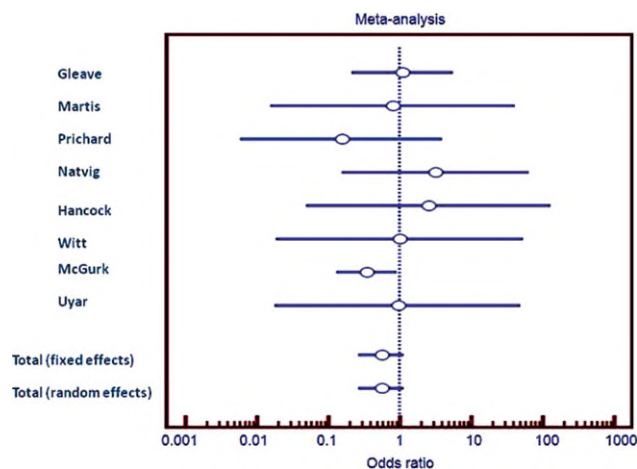


Fig. 2. Forest plot of recurrence rates comparing extracapsular dissection (ECD) to superficial parotidectomy (SP). Dots to the left of the line favor ECD. Any finding that crosses the midline is considered to be a nonsignificant finding. Meta-analysis fails to reject the null hypothesis that there is no difference in the recurrence rates of benign parotid tumors between ECD and SP. [Color figure can be viewed in the online issue, which is available at wileyonlinelibrary.com.]

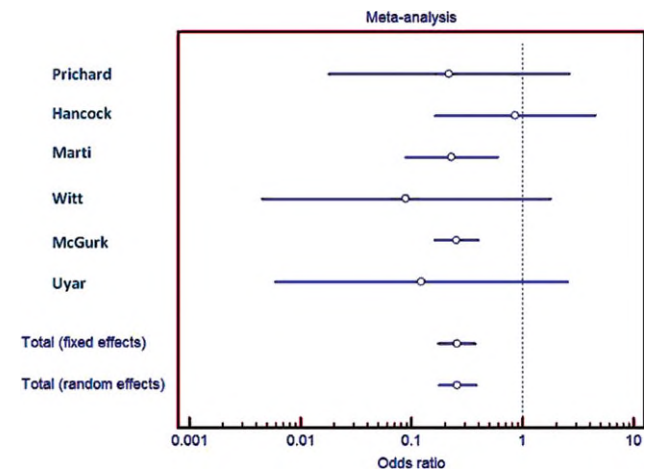


Fig. 3. Forest plot of transient facial nerve rates comparing extracapsular dissection (ECD) to superficial parotidectomy (SP). Dots to the left of the line favor ECD. Any finding that crosses the midline is considered to be a nonsignificant finding. Meta-analysis reveals significantly decreased rates of transient facial nerve damage following ECD as compared with SP. [Color figure can be viewed in the online issue, which is available at wileyonlinelibrary.com.]

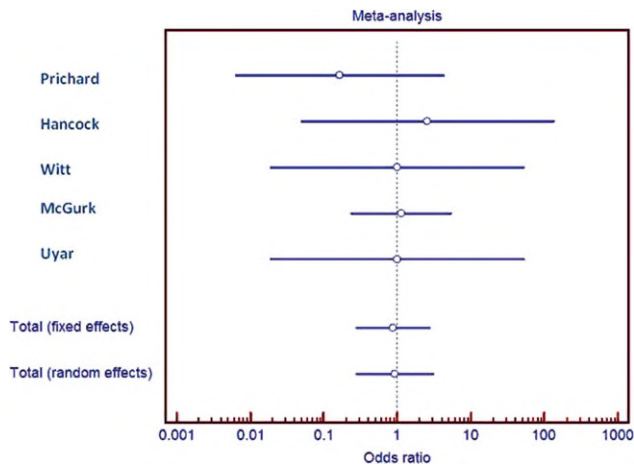


Fig. 4. Forest plot of permanent facial nerve paresis rates comparing extracapsular dissection (ECD) to superficial parotidectomy (SP). Dots to the left of the line favor ECD. Any finding that crosses the midline is considered to be a nonsignificant finding. Meta-analysis reveals no difference in the rates of permanent facial nerve damage following ECD versus SP. [Color figure can be viewed in the online issue, which is available at wileyonlinelibrary.com.]

symptomatic Frey's syndrome in the immediate postoperative period may lessen over time.

DISCUSSION

Surgery remains the mainstay of treatment for both benign and malignant neoplasms of the parotid gland. Surgery for parotid gland tumors has two primary goals: complete removal of the tumor and functional preservation of the facial nerve. In particular, the surgical method should ensure complete tumor removal to prevent recurrent tumor, which is more difficult to surgically cure and involves greater risk of permanent facial nerve injury.⁶ Although a variety of surgical techniques exist, SP remains the most widely practiced despite increasing consideration of minimally invasive techniques.

ECD is a minimally invasive approach that differs from classic enucleation, which involves the incision and shelling out of the contents of the tumor capsule. In ECD, the dense parotid fascia overlying the tumor is sharply incised followed by a blunt dissection to the level of the tumor.^{9,10} Under magnified visualization, a loose areolar plane may be seen 2 to 3 mm adjacent to the tumor and is the preferred plane of dissection. Careful dissection continues along the tumor capsule to prevent rupture of the small outpouchings of the tumor that may be encountered. In this method, unlike other forms of parotidectomy, planned identification of the facial nerve is not performed, although use of a facial nerve integrity monitor is advocated. Branches of the facial nerve may be encountered deep to the tumor and must be carefully dissected away from intervening parotid tissue.

In general, ECD has been applied to small, benign, superficial parotid tumors. Most authors apply the method to smaller tumors with reported cutoffs of between 2.5 and 4 cm.^{11,12} The risk of facial nerve injury increases during ECD with increasing tumor size. One

study found a 4% risk of facial paresis after ECD of tumors 4 cm or less in size compared to a 21% paresis rate in tumors larger than 4 cm.¹³ Although most groups only consider ECD for superficial lobe tumors, several have used it for deep lobe parotid tumors as well.¹⁴⁻¹⁶ This practice should not be widely employed because of an observed recurrence rate of 10% in deep lobe lesions compared to a rate of 3% for superficial tumors.¹⁵ Therefore, if considering ECD, the tumor should be solitary, clinically benign, mobile in two planes, smaller than 4 cm, and located in the superficial lobe. Preoperative imaging is likely to assist in the determination of whether these criteria are met.

Most of the studies included in this analysis did not use preoperative imaging or FNAC for the evaluation of tumor histology; however, it should be noted that the majority of these studies were published before the routine use of these technologies. More recent articles on ECD not included in this analysis have used preoperative imaging and FNAC in their eligibility criteria of tumors for the application of ECD.^{11,14,17,18} We would recommend the use of FNAC and appropriate imaging in the evaluation of tumors being considered for ECD. Preoperative imaging will reveal whether a given tumor is solitary, has a size of ≤ 4 cm, is isolated to the superficial lobe, has no suspicious adenopathy, and is therefore a potential candidate for ECD. FNAC will demonstrate features worrisome for malignancy in up to 80% of malignant tumors; however, the surgeon should have a low threshold for conversion to SP if there are any intraoperative concerns given the relatively high 20% false-negative rate for malignancy on FNAC.

ECD was developed as a surgical modality after it was demonstrated that SP frequently involves an element of ECD in areas where the tumor capsule makes contact with the facial nerve. Although contact between the tumor and the nerve has been reported to be as

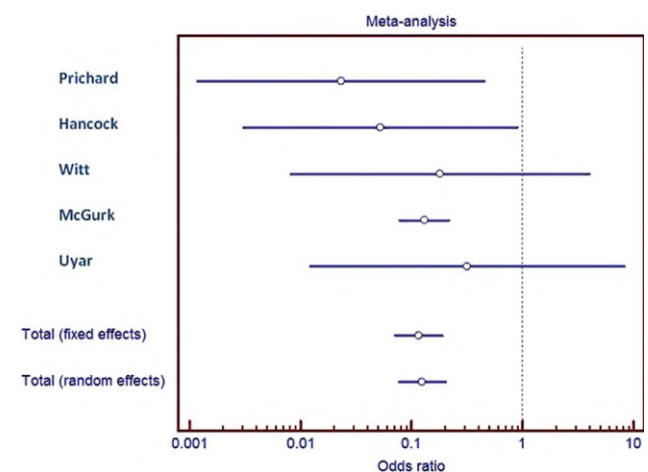


Fig. 5. Forest plot of Frey's syndrome rates comparing extracapsular dissection (ECD) to superficial parotidectomy (SP). Dots to the left of the line favor ECD. Any finding that crosses the midline is considered to be a nonsignificant finding. Meta-analysis reveals decreased incidence of Frey's syndrome in ECD versus SP. [Color figure can be viewed in the online issue, which is available at wileyonlinelibrary.com.]

frequent as 98% of cases, most reports indicate it is in contact 39% to 51% of the time.^{11,18-20} Thus, in theory the risk to the facial nerve should be the same or less with ECD compared to SP, which involves planned dissection of a longer segment of facial nerve. This theory is validated by the finding of a significantly lower rate of temporary facial paresis following ECD compared to SP in this systematic review. It should be noted, however, that the lower rate of facial paresis observed in these studies came from high-volume tertiary referral centers. Therefore, there is the potential that ECD could result in higher rates of facial nerve paralysis in less-experienced hands if the surgeon is less familiar with the common pathways and patterns of facial nerve branches.

Frey's syndrome is a relatively common occurrence following surgery of the parotid gland, and its incidence increases with progressively invasive surgery owing to further disruption of parotid tissue. An incidence as high as 50% has been observed after SP depending on the method used to assess gustatory sweating. Symptomatic complaints of Frey's syndrome, however, are less common, with 5% of patients undergoing SP bothered enough to seek treatment.^{21,22} This review found a significantly lower rate of Frey's syndrome after ECD (4.5%) compared to SP (25%). All the included studies assessed gustatory sweating clinically without starch iodine testing, which could have increased the sensitivity but has questionable clinical relevance. The authors did not clarify whether patients were evaluated for Frey's syndrome in the immediate postoperative period or more distant from the resection. Certainly, the prevalence of Frey's syndrome in this population would be expected to decrease with distance from surgery. It is also not clear from the articles how many of the patients reporting these symptoms actually sought treatment for it.

It is worth noting that this review could not control for the potential that ECD and SP may have been applied to different subsets of tumor. For instance, the largest series by McGurk et al. evaluated a total of 662 patients with clinically benign parotid tumors and determined the type of surgery based upon the tumor's exam once the skin flap had been raised. The likelihood that more challenging tumors underwent SP in that study is apparent when comparing the rate of malignancy in these clinically benign tumors (12.5% in SP vs. 2.4% in ECD).²⁰ Because of this methodology, the meta-analysis was run without this study without a statistical difference in the recurrence rate (1.6% in SP vs. 1.1% in ECD). However, it cannot be overlooked that in all of the studies, SP was likely applied to more challenging tumors. It should also be noted that an ideal follow-up period to truly assess recurrence would be 10 years due to the late recurrence of pleomorphic adenoma. Although most of the studies we evaluated had a mean follow-up of more than 10 years, several did not, and it can be expected that the absolute value for recurrence is somewhat higher than we have reported.

Unlike capsular rupture, capsular exposure does not appear to affect recurrence. Witt studied 60 cases of pleomorphic adenoma and found that microscopic capsular exposure occurs universally and results in no

increased risk of recurrence between ECD and SP.²³ Other authors have found similar results.²⁴

Rupture of the capsule with macroscopic tumor spill during surgery cannot be considered a curative operation.^{24,25} Rupture has been reported to occur in 5% to 10% of SPs.^{23,25} Published rates of capsular rupture with ECD range from 0% to 9.2% with a calculated mean of 5.8%.^{7,13,15,16,18,23} Rupture of the capsule is certainly a surgical complication to be avoided; however, it does not appear that there is any increased risk in this adverse outcome with the use of ECD.²³

ECD should be avoided if there is any concern for malignancy in the preoperative work-up, including a risk of malignancy identified by FNAC or imaging. Intraoperative findings suspicious for malignancy such as tethering, induration, nerve invasion, or suspicious lymphadenopathy should likewise lead to conversion to a superficial or total parotidectomy. Every precaution should be taken; however, it is inevitable that a few cases of malignancy will be subject to resection by ECD. McGurk et al. reported that in their series of clinically benign parotid tumors, 32 of 662 patients were ultimately found to have carcinoma, of whom 12 were subjected to ECD, with 10-year survival rates being broadly similar whether the patient received SP or ECD.²⁰ Although it is not clear that there is any difference in outcomes between surgical techniques in these clinically benign but ultimately malignant tumors, ECD should be avoided. In the case that malignancy is discovered postoperatively, completion SP should be considered and remains a viable option because the main landmarks of the facial nerve are still largely intact.

There are several weaknesses of this analysis, including selection bias to which procedure was performed and lack of randomization. It is thus very likely that patients who underwent the two methods had different types of tumors and that ECD was selectively applied to more manageable nodules. An additional weakness of several studies is the reporting of only the results for pleomorphic adenoma. Although this is the entity most targeted by ECD, it is important to note the recurrence rates and complications for other benign tumors and tumors that were clinically benign but following resection were found to not be benign.

CONCLUSION

No difference in the rate of tumor recurrence or permanent facial paralysis was noted between SP and ECD when applied to solitary, clinically benign nodules of the parotid gland. ECD appears to have a decreased incidence of transient facial nerve palsy and Frey's syndrome, although there is the possibility that the technique was applied to less challenging tumors on average. Although ECD should not be seen as a replacement for SP, it appears to be safe and a potentially advantageous alternative when applied to specific parotid tumors by experienced surgeons. SP remains the standard of care for treatment of benign parotid tumors owing to its proven track record, but ECD may be considered by surgeons trained in its application.

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