Original Article - Retrospective Study

Long-term Follow-up after Extracapsular Dissection of Parotid Pleomorphic Adenomas - A Retrospective Study

Walter Colangeli, Valerio Facchini, Aleksandr Kapitonov, Fabrizio Bozza, Roberto Becelli

NESMOS Department, Maxillofacial Surgery Unit, Faculty of Medicine and Psychology, S. Andrea Hospital, University of Rome "La Sapienza," Rome, Italy

Abstract

Introduction: Parotid pleomorphic adenoma (PPA) is the most common benign salivary gland neoplasm. Extracapsular dissection (ED) arose as a conservative surgical technique alternative to superficial parotidectomy to reduce complications. **Materials and Methods:** Patients who underwent ED for superficial, <3 cm, mobile parotid lumps (Quer I) between 2004 and 2008 were retrospectively analysed focusing on those with histological diagnosis of pleomorphic adenoma. A retrospective cohort study on 50 patients who accepted to undergo ultrasonography and clinical evaluation for at least 10 years since surgery was performed. Clinical data, surgical reports, and validated questionnaires for the assessment of complications, quality of life (QoL) and aesthetic satisfaction were analysed. Recurrence rate and complications after ED, with their QoL and aesthetic impact, were evaluated. Statistical analysis was performed setting α=0.05 as the level of significance. **Results:** Low occurrence of complications related to ED was seen. Overall QoL after ED was very good (range 1–7, mean 6,86), due to low complications incidence of complications and their low severity, as assessed by patients through Parotidectomy Outcome Inventory-8 questionnaire. Overall aesthetic satisfaction was high (range 1–10, mean 9,78). Aesthetic satisfaction and QoL was statistically related to onset of complications (P = 0.02504 and P = 0.001859). Tumour localization and dimension was not statistically related to onset of complications (P = 0.8207 and 0.7586). After a mean follow-up of 12.5 years, no recurrences were detected. **Discussion:** There is a lack of studies with a long-term follow-up after ED for the evaluation of recurrences and complications. Considering our results, the ED technique should be adopted as the first surgical approach for Quer I PPA without suspicion of malignancy.

Keywords: Extracapsular dissection, long-term follow-up, pleomorphic adenoma, quality of life, recurrences

Introduction

Pleomorphic adenoma (PA) is a benign tumour and is considered the most common salivary gland neoplasm. It accounts for about 85% of all salivary glands neoplasms and involves major salivary glands in about 80% of cases. It mostly occurs in the superficial lobe of the parotid gland.[1]

Proper surgical management of benign tumours of the parotid gland is still debated due to the necessity of minimizing recurrences, complications, and aesthetic outcomes.[2]

Described techniques are enucleation, extracapsular dissection (ED), superficial and total parotidectomy (SP and TP).

First, enucleation was adopted, but over the years, it led to high rates of recurrence due to capsular rupture (205–45%). Therefore, SP was adopted as the treatment of choice for benign lesions at most medical centres[3,4] leading to a drastic decline in local recurrences. Otherwise, complications such as temporary injuries of the facial nerve, Frey's syndrome, and cosmetic deformities did arise.[5]

Over the decades, a conservative surgical approach arose. ED proved useful in reducing complications and maintaining a similar recurrence rate to SP.[6,7]

ED technique concerns the removal of the parotid mass within a thin parenchyma layer, preserving the uninvolved parotid parenchyma and reducing the need for a wide dissection along the facial nerve.[6] Among literature, there is a lack of long-term

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Address for correspondence: Dr. Valerio Facchini, Via San Godenzo 16, 00189 Rome, Italy.
E-mail: valerio.facchini1@gmail.com

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follow-up for the evaluation of recurrences. Moreover, when a long-term follow-up is carried out, it is often only clinical and not radiological.

In this article, a long-term follow-up experience in the surgery of parotid PA (PPA) with ED technique is presented.

The focus of the study is local recurrences rate and complications with their impact on quality of life (QoL) and aesthetic satisfaction.

Materials and Methods

In this retrospective cohort study, we selected 132 patients who underwent ED for benign parotid tumours between January 2004 and December 2008 in our university hospital of Rome. In our practice, ED is performed routinely for mobile neoplasms, located in the superficial lobe, <3 cm in size without preoperative facial nerve dysfunction and with fine-needle aspiration cytology (FNAC) negative for malignancy.

In this study, PPA histological diagnosis was adopted as an inclusion criterion.

From histopathological reports analysis, PPA resulted in 95 of 132 cases.

Of these, we collected data about complications from clinical reports and from questionnaires submitted 1 year after surgery.

For the assessment of symptom-specific outcomes, overall QoL and overall aesthetic satisfaction, two validated questionnaires and a scale ranging from 1 to 10 were routinely submitted to each patient 1 year after surgery [Figure 1]. The first questionnaire is the Parotidectomy Outcome Inventory-8 (POI-8) used for the evaluation of symptom-specific outcomes.[8] Complications analysed by POI-8 are earlobe hypaesthesia, transient facial nerve palsy, Frey's syndrome, depression of surgical site (loss of substance), hypertrophic scar, pain, xerostomia, and fear of reintervention. For each complication, patients assign a value of severity ranging 1–5.

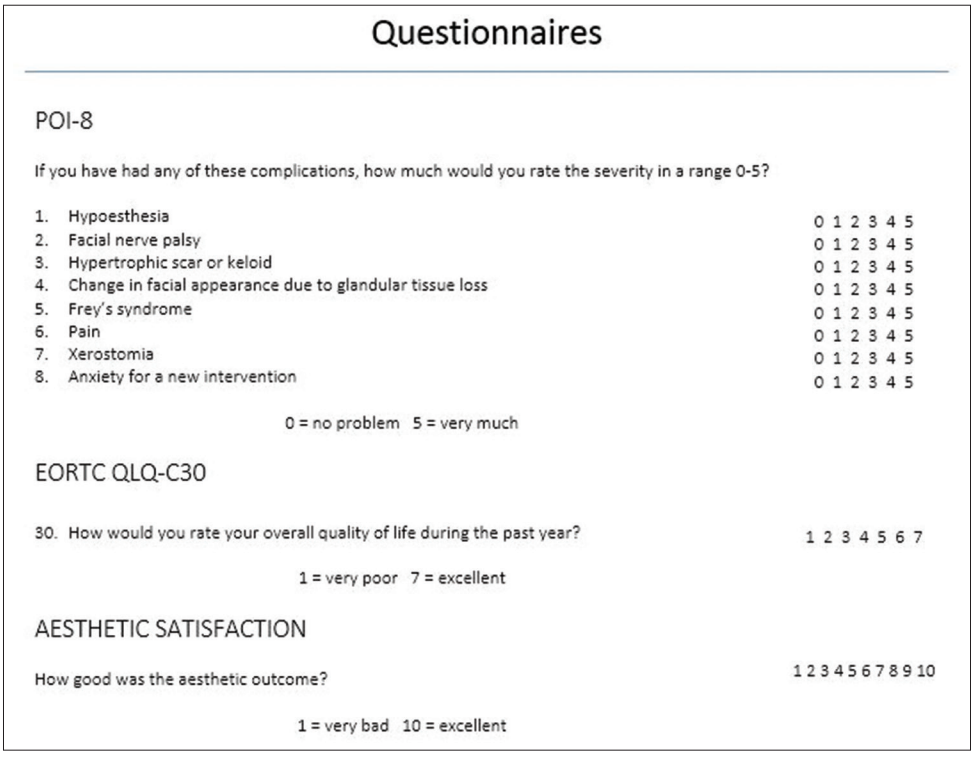
The second one is the QLQ-C30 questionnaire (specifically question 30, referred to the first year after surgery) validated by the European Organization for Research and Treatment of Cancer (EORTC), used for the evaluation of the QoL.[9]

A satisfaction scale ranging from 1 to 10 is used to analyse postoperative aesthetic satisfaction.

In our department, standard follow-up for benign lesions of the parotid gland is performed by ultrasonography (US) (twice a year for 3 years and then once a year for 2 years) and magnetic resonance imaging with contrast (1st, 3rd, and 5th year).

In 2018–2019, we successfully contacted 73/95 patients proposing them the US followed by a clinical evaluation.

50/73 (~68%) patients accepted to undergo the US and the clinical evaluation and were then successfully ruled in for our study. Therefore, a minimum follow-up of 10 years was obtained for every patient (mean follow-up time = 12.5 years; range = 10. 2–14.8 years), to analyse the recurrence rate of the ED procedure.


Questionnaires

POI-8

If you have had any of these complications, how much would you rate the severity in a range 0-5?

1. Hypoesthesia 0 1 2 3 4 5
2. Facial nerve palsy 0 1 2 3 4 5
3. Hypertrophic scar or keloid 0 1 2 3 4 5
4. Change in facial appearance due to glandular tissue loss 0 1 2 3 4 5
5. Frey's syndrome 0 1 2 3 4 5
6. Pain 0 1 2 3 4 5
7. Xerostomia 0 1 2 3 4 5
8. Anxiety for a new intervention 0 1 2 3 4 5

0 = no problem 5 = very much

EORTC QLQ-C30

30. How would you rate your overall quality of life during the past year? 1 2 3 4 5 6 7

1 = very poor 7 = excellent

AESTHETIC SATISFACTION

How good was the aesthetic outcome? 1 2 3 4 5 6 7 8 9 10

1 = very bad 10 = excellent

Figure 1: Questionnaires used for the assessment of symptom-specific outcomes, overall quality of life and aesthetic satisfaction

Data were recorded in an Excel file to perform descriptive and inferential statistical analysis. Qualitative variables were defined by absolute frequencies. Quantitative variables were defined by mean and standard deviation. Age, gender, and lesion localization were analysed in correlation to post-surgical complications. The student's *t*-test was used to evaluate QoL, health state, and aesthetic satisfaction in relation to postoperative complications. A $P < 0.05$ was considered statistically significant.

In conclusion, our study evaluated the presence and the severity of complications 1 year after surgery and the onset of long-term recurrences.

Ethics

Written informed consent has been obtained from patients before the study.

The Ethics Committee of AOU Sant'Andrea of Rome waived the need for ethics approval and the need to obtain consent for the collection, analysis, and publication of the retrospectively obtained and anonymized data for this study. All procedures performed in the study were conducted in accordance with the ethical standards given in the 1964 Declaration of Helsinki, as revised in 2013.

Results

Data from 50 patients were collected: 21 males and 29 females, with an average age of ~46 years (range 27–68). Of these patients with PA, 36% of cases were right-sided tumours and 64% were left-sided tumours. In 34% of patients, lesions were located in the upper pole, and in the remaining 66% were located in the lower pole. The average size of neoplasms was 2.1 cm (ranged 0.8–3 cm). All patients underwent ED performed by experienced surgeons.

The average duration of surgery was 1 h and 25 min (range 47 min - 2 h and 18 min). Statistical analysis was performed through Excel.

The first analysis analysed clinical reports highlighting the low occurrence of complications related to ED intervention as shown in Graph 1.

Analysing POI-8 questionnaire results, we obtained the average perception of the severity of each complication by patients [Graph 2].

Finally, we evaluated both overall QoL during the first year after surgery [Graph 3] and overall aesthetic satisfaction at one year after surgery [Graph 4].

Facial nerve transient palsy onset in only 3 patients after one year was observed with complete clinical recovery in all patients. The average severity of this complication was 366 by POI-8 questionnaire (values assigned by patients 3, 4, and 4).

Frey's syndrome and greater auricular nerve (GAN) deficit (earlobe hypaesthesia) arose respectively in 1 and 6 patients with middle impact on health-related QoL in patients as demonstrated by POI-8 data.

Loss of glandular parenchyma with skin depression (loss of substance, 2 patients), hypertrophic scar (3 patients), and postoperative pain (2 patients) slightly affected QoL. Fear of a new intervention was found in a high percentage of patients (26% of patients) but did not influence QoL. Xerostomia was not observed in any patient of our study.

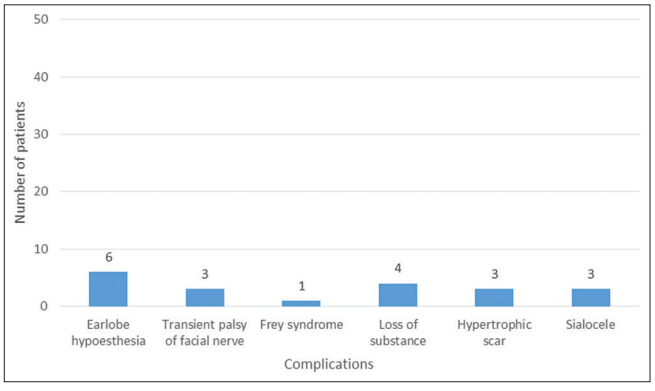
Another data that was detected from clinical reports is the onset of 3 sialoceles.

Statistical analysis revealed that the localization of the lesion did not influence aesthetic satisfaction, health status, and QoL ($P > 0.05$). However, aesthetic satisfaction and QoL were statistically related to onset of complications. Patients with postoperative complications recorded lower satisfaction ($P = 0.02504$) and a worse QoL ($P = 0.001859$). Furthermore, statistically relevant data revealed significant correlation between gender and postsurgical aesthetic perception: Male patients showed a lower satisfaction than female ones ($P = 0.02537$). Evaluating onset of complications, patients' age and the size of the mass did not reveal any statistical differences ($P > 0.05$).

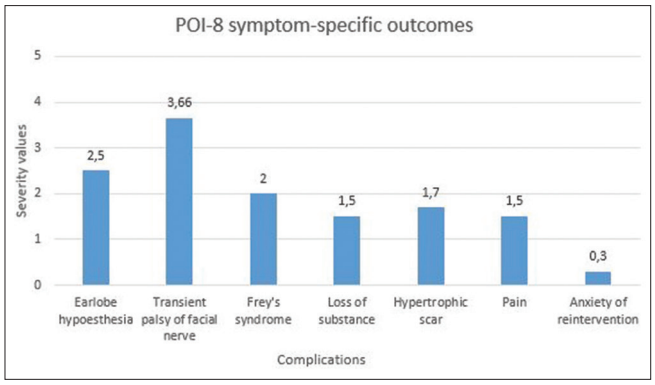
After a minimum of 10 years' follow-up, no recurrences were detected on US and clinical evaluation.

Discussion and Conclusions

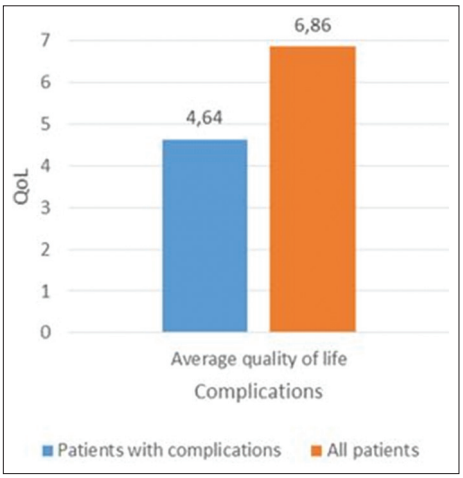
PA is a benign tumour and is considered the most common salivary gland neoplasm, although in some recent papers, the incidence of Warthin's tumour surpassed the incidence of PA.[10] It accounts for about 60% of all salivary gland neoplasms and involves major salivary glands in 85% of cases and minor salivary glands in 15%.[1] It is known as a mixed tumour, because of its dual origin from epithelial and myoepithelial elements.[11] In contrast to myoepithelioma (firstly described as a variant of the PA), a PA can present as a chondroid or an osteoid formation.[12] It mostly occurs in the superficial lobe of parotid glands.[13] Submandibular and sublingual glands are rarely involved.[14,15] Fourth to fifth decades of life are mostly affected, but it can occur over a wide age range.



Graph 1: Number of extracapsular dissection-related complications detected after surgery



Graph 2: Average perception of the severity of each complication by the patient (severity range 0–5)



Graph 3: Average quality of life during the 1st year after surgery (quality of life range 1–7)

PA may be associated with malignant transformation and then an early complete excision of the mass is recommended.[16]

In our practice, ED for tumours up to 3 cm in size, which are mobile and located in the superficial lobe (Quer's category I)[17] was performed by experienced surgeons. It has been observed that capsular invasion is more commonly associated with PA larger than 4 cm.[18] Considering this, tumour size could be considered as a limitation of ED technique application. Selecting patients, we also considered clinical data (lump mobility and facial nerve function) [Figure 2] and FNAC reports, to reduce incongruous treatment for malignant lesions. Even if malignancy was considered as a deterrent for the adoption of ED, McGurk *et al*.[19] observed that ED is a valid and safe approach to the management of clinically benign parotid tumours.

ED surgical techniques are defined as the removal of the neoplasm with a small part of healthy tissue around it, without exposing the main trunk of the facial nerve (a retrograde dissection of peripheral nerve branches is possible) and without performing the long dissection along the nerve itself[20] [Figure 3]. In ED, the incision can be made shorter than in other techniques due to the lower level exposure of the parotid. This could have a good impact on postoperative aesthetic satisfaction.

According to literature,[1,2,7,19] performing ED a low rate of overall complications was observed in our study, including the transient facial palsy associated with a wide and prolonged nerve exposure as reported with other techniques.[6] In SP, preliminary identification and exposure of the facial nerve trunk are mandatory, and then a dissection along the various branches of the nerve removing the superficial portion of the parotid gland with the tumour included is performed.[21] However, a nerve branch could be close to the tumour and could groove it. In this way, the SP technique may involve a step of the ED technique. This close tumour-nerve interfacing is reported in 50% of cases.[22] In our study, transient facial nerve palsy was observed in only three patients after surgery with complete recovery at 1 year. Low rate of transient facial nerve palsy must be considered an adjunctive ED technique advantage, considering that in our study it has been the more severe complication in patients' perception [Graph 2].

In our series, we did not use any nerve monitoring or loupe magnifications.

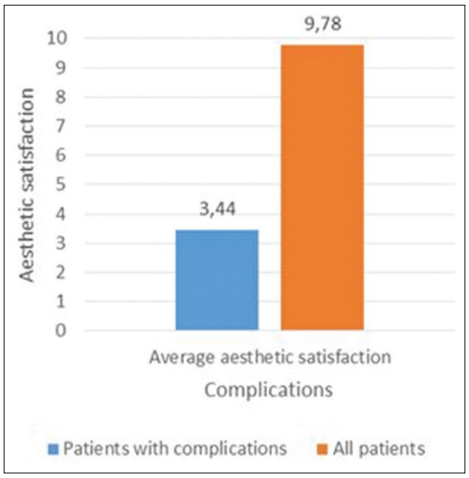
With this technique, GAN could be also easily preserved in a great number of patients (in our series we spared GAN in 88% of cases).[23]

Superficial Muscular Aponeurotic System (SMAS) flap has been routinely used in SP. Some advantages such as the prevention of both Frey syndrome and surgical site depression have been described.[24] In our experience with ED, SMAS flap has not been applied in masses smaller than 2 cm. This is because in these cases, it is often possible to primarily close the parotid gland capsule avoiding depression of the surgical site and at the same time ensuring coverage and protection of the facial nerve. Outcomes related to surgical site depression and Frey's syndrome in our patients supported these data, but larger studies are needed on this aspect.

Sialoceles occurred in three patients in our series and were immediately treated with aspiration, compression dressing, and scopolamine patches.[25] All sialoceles resolved in about 3 weeks after surgery.

One of the key points of ED versus SP debate is the long-term recurrence rate after conservative surgery. Recurrence has been associated with several problems including the increased rate of postoperative complications and malignant degeneration of recurrences itself. Regarding patients treated with ED, there is a lack of studies evaluating recurrence with an instrumental long-term follow-up. Previously, articles with long-term follow-up for PPA treated with SP reported a time interval between primary surgery and the first recurrence ranged from 7 to 24.5 years.[26,27]

Tumour recurrence has been related to histopathological variables such as capsular integrity and thickness, pseudopodia,



Graph 4: Average aesthetic satisfaction 1 year after surgery (aesthetic satisfaction range 1–10)

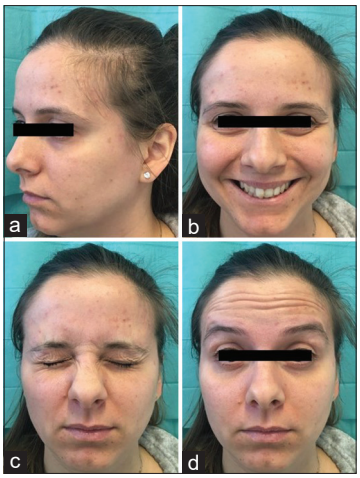


Figure 2: Preoperative clinical evaluation of the patient. (a) Parotid region view; (b-d) VII cranial nerve clinical evaluation

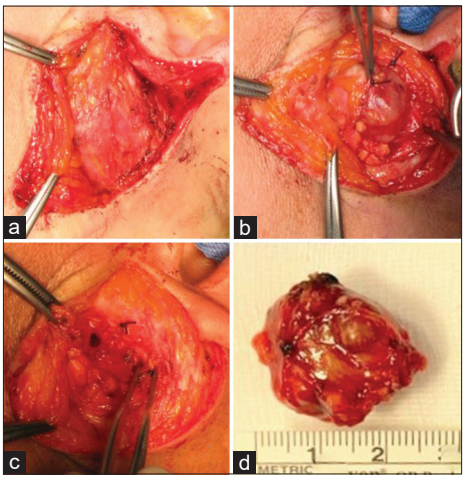


Figure 3: Intraoperative view of the extracapsular dissection. (a) Surgical field after the preparation of the skin and superficial muscular aponeurotic system flaps; (b) Identification of the mass and execution of the extracapsular dissection; (c) Surgical field after the removal of the mass; (d) Surgical specimen with a lining of glandular tissue

Figure 4: One-year postoperative clinical evaluation of the patient. (a) Parotid region view; (b-d) VII cranial nerve clinical evaluation

satellite nodules, and tumour size; other variables could be related to surgery, such as resection margins and tumour puncture and spillage; also younger age at initial presentation may influence tumour recurrence rate.[3,28]

PPA >3 cm are associated with more numerous satellite nodules raising the risk of recurrences. In addition, positive margins and tumour spillage are linked to recurrences.[29] In our study, the capsule rupture was histologically reported in only two cases. Among our patients, we observed no recurrences after a mean follow-up time of 12.5 years.

In conclusion, the application of the ED technique for PPA treatment highlighted a low complication rate and the absence of recurrences after a long-term follow-up performed through US and clinical visit [Figure 4]. Otherwise, literature data showed that ED has similar effectiveness and fewer side effects of SP minimizing the incidence of facial nerve palsy and improving aesthetic results in a short follow-up period.[7,18,30-34] In our opinion, more long-term follow-up comparative studies between SP and ED are needed to evaluate the recurrence rate. Therefore, ED performed by experienced surgeons is a safe option as a first surgical approach for superficial parotid lumps, classified as Quer I, with no suspicion of malignancy.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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