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Investigation of the surgical strategies for unilateral multifocal cystadenolymphomas of the parotid gland

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ABSTRACT

Objectives: The aim of the study was to compare multifocal extracapsular dissection with complete parotidectomy in the treatment of unilateral multifocal cystadenolymphomas of the parotid gland.
Materials and methods: The records from all patients treated for unilateral multifocal cystadenolymphomas of the parotid gland at a tertiary referral center between 2000 and 2016 were retrospectively analyzed.
Results: 203 patients were included in the study. 96 patients were managed by multifocal extracapsular dissection, 107 patients underwent complete parotidectomy. Metachronous tumors were detected in 7 cases (3.4%) with a significant difference in the occurrence rate between multifocal extracapsular dissection (7/96, 7.3%) and complete parotidectomy (0/107, 0%) ($p = 0.004$). Permanent facial palsy was significantly more common after complete parotidectomy (21/107, 19.6%) than multifocal extracapsular dissection (2/96, 2.1%) ($p = 0.000$).
Conclusions: The functional benefit of multifocal extracapsular dissection was counterbalanced by the advantage of complete parotidectomy, in terms of recurrences. Extracapsular dissection is at its limit in cases of multifocal cystadenolymphomas and the indication for it should be thoroughly verified in each and every case.

Introduction

The multifocal nature of cystadenolymphomas of the parotid gland, also known as Warthin's tumors, has been well documented, with a reported incidence of 6.2–50% in the relevant literature [1–6]. Our therapeutic strategy in treating these tumors is determined by the need for low perioperative morbidity if revision surgery is required for an ipsilateral metachronous tumor. When the primary surgical removal of very large cystadenolymphomas involves dissection of the facial nerve trunk and at least one of its main branches, excessive scar formation around this critical structure is to be expected, increasing the risk of iatrogenic damage to the facial nerve during any revision surgery. In such cases, we therefore choose to perform a complete parotidectomy, in order to reduce the risk of a surgically demanding recurrence. This strategy does, of course, lead to a potential increase in the perioperative morbidity of primary surgery. After 17 years of increasing experience and expertise with gland-preserving surgical modalities (e.g. extracapsular dissection) [7–9] in our department, we wanted to investigate whether perioperative morbidity in cases of multifocal cystadenolymphomas could be reduced by routinely employing less radical surgical procedures, without increasing the risk of an ipsilateral recurrence.

The main aim of our study was to investigate extracapsular dissection of multifocal parotid cystadenolymphomas, in terms of the functional outcome and recurrence rate, in a direct comparison with complete parotidectomy.

Materials and methods

This retrospective study was conducted at an academic tertiary referral center specializing in salivary gland pathologies (Department of Otorhinolaryngology, Head and Neck Surgery, University of Erlangen–Nuremberg, Erlangen, Germany). Cystadenolymphomas were considered to be multifocal if more than one focus was found on definitive histology of the parotid gland specimen or two or more lesions were individually resected from the same side during the same surgical procedure. The records of all patients treated for multifocal parotid cystadenolymphomas between 2000 and 2016 were evaluated. Patients with insufficient data and those undergoing surgical revision of external primary surgery as well as patients with unclear surgical margins on definitive histology were excluded from the study.

Preoperatively, all patients underwent a clinical examination, ultrasonography of the parotid gland and, in some cases, computed tomography or magnetic resonance imaging. The function of the facial nerve was assessed clinically using the House-Brackmann grading

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system and by electromyography of the facial nerve before surgery.

The surgical modalities employed were complete parotidectomy (CP) and multifocal extracapsular dissection (MED). Complete parotidectomy was defined as resection of the entire parotid gland parenchyma, with preservation of the facial nerve. Multifocal extracapsular dissection was defined as removal of the several tumors with a cuff of healthy tissue, without deliberate exposure of the main trunk or the branches of the facial nerve. A metachronous tumor was defined as a histologically confirmed cystadenolymphoma developing at the anatomic site of the primary tumor (parotid bed after CP, residual parenchyma after MED) at any time after primary surgery.

In this study, we compared MED with CP for unilateral multifocal cystadenolymphomas of the parotid gland, in terms of metachronous tumor rate and postoperative complications. Statistical analysis was performed using the chi-square test with 95% confidence intervals. SPSS software version 21 for Windows (SPSS, Chicago, IL), was used for the analysis. A p-value of < 0.05 was considered statistically significant.

Results

A total of 203 patients were included in the study (143 men, 60 women; male:female ratio of 2.38:1). The mean age was 60.6 years (37–89 years). 116 cases had 2 lesions, 44 had 3 lesions, 7 had 4 lesions, 2 had 5 lesions and 34 had 6 or more lesions. The average number of lesions was 3.09. 96 patients were managed by means of MED of all lesions; 107 patients had a CP. Mean follow-up was 60 months. Ipsilateral metachronous tumors were detected in 7 cases (3.4%), all after primary MED, giving a highly significant difference in the recurrence rate between MED (7/96, 7.3%) and CP (0/107, 0%) (χ^2 (1) = 8.081, p = 0.004). Mean time of appearance of a metachronous tumor was 19.8 months after primary surgery. Temporary and permanent facial palsies were significantly more common after CP than MED. Table 1 gives an overview of the postoperative complications of the different surgical modalities and the statistical analysis. According to this table, MED was shown to have a significantly lower incidence of direct postoperative (p = 0.000) and permanent facial nerve palsy (p = 0.000), as well as significantly fewer cases of Frey's syndrome (p = 0.000). No statistically significant difference could be detected in terms of postoperative salivary fistula (p = 0.626) and hemorrhage (p = 0.318) between the examined surgical modalities.

Discussion

A review of the relevant literature reveals that the surgical treatment of multifocal cystadenolymphomas ranges from complete parotidectomy in every case to a simple extracapsular or even capsular dissection of all macroscopic lesions detected [10,11]. In our department, the need to resect all possible foci and to minimize the risk of recurrent ipsilateral tumors with contact to the facial nerve has led to complete parotidectomy being the procedure of choice in the vast majority of cases. This form of treatment undoubtedly warrants excellent results, in terms of the rate of metachronous tumors on long-term follow-up, with the least need for revision surgery. However, performing such radical surgery for a "tumor-like" benign lesion with no relevant potential for malignant transformation could correlate with a significant increase in the postoperative morbidity (facial nerve palsy, Frey's syndrome), a poor cosmetic result (concave facial outcome due to a significant defect in the buccal or parotid region, facial asymmetry), and a reduced quality of life [8,12].

Consideration of these aspects led us to examine the philosophy sustaining our clinic's current treatment strategy. Management of multifocal cystadenolymphomas should aim at giving the patient histological confirmation of the benign status of the disease, removing symptomatic and obvious lesions within the parotid bed, and providing good local control with the best possible postoperative quality of life. Increasing expertise and experience in preoperative imaging (e.g. ultrasound) should help with the accurate localization of the macroscopic lesions, allowing all synchronous lesions to be resected without radical surgery. Moreover, several reports in the literature point to a significant discrepancy between the high rate of clinically irrelevant microscopic lesions (50%) [6,13] and the low rate of clinically relevant macroscopic tumors (21% of all the cases in our department), indicating that only a small number of microscopic foci develop into macroscopic lesions needing treatment [10,11]. The second argument in favor of complete parotid resection for multifocal cystadenolymphomas is the need to minimize the risk of new ipsilateral tumors in contact with the facial nerve, which would then necessitate demanding revision surgery. However, literature reports have found a low overall rate of recurrent tumors after gland-preserving surgical procedures [4,11,14]. Furthermore, when there is a recurrence, the watch and wait approach is becoming more and more widespread in high-risk cases with good patient compliance [10]. These observations provoke the thought that complete resection of the parotid gland for multifocal cystadenolymphomas may constitute overtreatment in some cases, with an overwhelmingly negative effect on the postoperative quality of life.

For these reasons, we took a retrospective look at the possibility of reducing the invasiveness of surgical treatment for multifocal cystadenolymphomas, in terms of adequate surgical outcome and postoperative quality of life. A review from our department of historical data from the 1960s to 2000s showed that the philosophy behind the acceptable surgical minimum for benign parotid lesions followed a sinusoid course between more radical and less invasive forms of treatment, balancing out after 2000 at the level of extracapsular dissection [8]. Multifocal cystadenolymphomas could not have remained unaffected by this paradigm shift over the years: the descriptive analysis of our data shows an impressive increase in the number of extracapsular dissections for multifocal cases, from virtually no cases in the first five years (2000–2005) to consistently more than 10 cases/year in the last five years of our study (2011–2016). It is notable that nearly the half the cases of multifocal cystadenolymphomas in our department after 2006 were managed by means of extracapsular dissection (Table 2, Fig. 1). In routine practice, this procedure was increasingly chosen when preoperative imaging showed conglomerates of lesions with signs of cystadenolymphoma, located within the superficial lobe and ideally in the caudal pole of the parotid gland (Fig. 2). More rarely, multifocal

Table 1
Comparison of postoperative complications after extracapsular dissection and complete parotidectomy for multifocal cystadenolymphomas.

	Incidence (%)			
Complication	Extracapsular dissection	Complete parotidectomy	p-value	Odds ratio
Transient facial nerve palsy	10/96 (10.4)	78/107 (73)	**0.000**	23.131 (10.589–50.529)
Permanent facial nerve palsy	2/96 (2.1)	21/107 (19.6)	**0.000**	11.477 (2.614–50.396)
Frey syndrome	2/96 (2.1)	19/107 (17.7)	**0.000**	10.148 (2.297–44.835)
Salivary fistula	9/96 (9.4)	8/107 (7.4)	0.626	0.781 (0.289–2.113)
Postoperative hemorrhage	4/96 (4.1)	8/107 (7.4)	0.318	1.859 (0.541–6.380)

p-value of < 0.05 were statistically significant are highlighted in bold.