

# Results of cystadenolymphoma treatment of superficial parotid gland

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## ABSTRACT:

**Introduction:** Warthin's tumor is a non-malignant tumor that occurs in major salivary glands. Diagnostics include an interview and physical examination as well as additional tests – ultrasonography, magnetic resonance tomography, fine-needle aspiration biopsy. Surgical tumor resection remains the method of treatment, the scope of which includes techniques from extracapsular tumor resection to a full range of parotidectomy.

**Material and methods:** A retrospective analysis was conducted for available medical records of 53 patients treated surgically at the Department of Otolaryngology and Laryngological Oncology at the Collegium Medicum of the Nicolaus Copernicus University in Toruń in 2009–2016. Each patient underwent an interview, physical examination and a routine ultrasound examination. Results of treatment of 57 tumors were analyzed.

**Results:** The study showed that in the case of extracapsular tumor excision in 57 patients in the treatment of Warthin's tumors of the lower pole of the parotid lobe, there were no complications in the form of: permanent paralysis or facial nerve palsy, mucocele, symptoms of Frey's syndrome or cosmetic facial defect. Ultrasound examination performed in 8 (14%) patients revealed lesions requiring further diagnostics or periodic ultrasound monitoring.

## Conclusions:

1. Extracapsular tumor excision appears to be an accurate surgical technique in Warthin's tumor of the lower pole of the parotid gland's superficial lobe.
2. After removal of a cancerous lesion of the parotid gland, every patient requires periodic laryngological monitoring.

## KEYWORDS:

benign tumor, parotid gland, treatment, extracapsular tumor excision

## INTRODUCTION

Salivary gland tumors comprise a highly heterogeneous group of tumors in terms of histological construction, which is associated with their complex embryogenesis, as a consequence translating into these tumors' nomenclature. The most frequent non-malignant tumors of the major salivary glands are: pleomorphic adenoma and monomorphic adenoma - lymphatic/lymphadenoma [1]. In the medical literature, lymphadenoma is also called adenolymphoma, cystadenoma papillare lymphomatosum, cystadenolymphoma, as well as the widely used Warthin's tumor. Aldrin Scott Warthin was the first to describe two cases of lymphadenoma in American literature; his work was published in 1929, although there were

already mentions of lymphadenoma as far as 1895 made by dr. Hildebrand. Etiology of the emergence of lymphadenoma not yet been clarified, however, its tobacco-dependent nature has been proven [2]. Until recently, its emergence was mostly associated with predominance in men; current reports indicate trends towards equalization of the prevalence in women and men, which is associated with the dissemination of nicotinism in women. It usually occurs between the 5th and 7th decade of life. In 10–15% of cases, these tumors can be bilateral, both synchronously and metachronically, as well as multifocally [3].

In addition to an interview and physical examination, diagnostic imaging remains an indispensable stage in diagnosing pathology in the salivary glands. An imaging study of choice in

the case of nodular lesions in the deep lobe of parotid gland and in the case of the need to extend diagnosis is magnetic resonance [4]. An advantage of magnetic resonance imaging over computed tomography in the accuracy of tumor imaging in the salivary gland parenchyma and its relation to the course of the facial nerve in correlation with the intraoperative image was demonstrated [5].

The necessity to perform preoperative or intraoperative pathomorphological diagnostics is still controversial. The result of these tests without the whole clinical image cannot constitute a diagnosis. Fine-needle aspiration biopsy (BAC) has the widest application, decreasing the role of open biopsy, core biopsy or intraoperative examination to exceptional clinical situations. This procedure performed under the supervision of ultrasonography increases the accuracy of examination to 85–90% [6–8].

The method of treatment of parotid tumors remains surgical. The extent of resection depends on the location, size of tumor, capsule exposure, as well as histological type of tumor and the entire clinical picture. Among the currently used surgical techniques, according to the nomenclature proposed by the European Salivary Gland Society in 2016, there are two operational techniques: extracapsular dissection and parotidectomies. The method of extracapsular tumor dissection consists in removing the tumor with its capsule, without removing the salivary gland parenchyma [9]. Parotidectomies, in turn, require dissection of the facial nerve (at least the main trunk and one of the two main branches – temporal-facial or cervical-facial). To determine the location of the resected salivary gland, it is recommended to use levels I, II, III, IV, V according to tumor location. Classical clinical classification divides parotidectomies into: superficial (also referred to as lateral or partial parotidectomy) and total (with or without facial nerve) and expanded parotidectomy (resection depending on the extension of tumor may include neighboring structures – including the masseter muscle, mastoid process, external carotid artery) [10].

Contrary to the mixed tumor, the risk of transformation of Warthin's tumor into a malignant form is low and amounts to less than 1%.

## MATERIAL

The study was conducted in a group of 53 people, 22 women, 31 men, the average age is 61 years, the median age is 63 years. The patients underwent excision of Warthin's tumor located in the lower pole of the superficial parotid gland via extracapsular tumor resection (ECD). This group, over the period 2009–2016, included two people diagnosed with metachronous

and in the next two synchronous Warthin's tumor foci in the second parotid gland, which was also qualified for resection via extracapsular technique, which represents the number of tumors equal to 57.

## METHOD

A retrospective analysis was performed based on the collected medical documentation of patients treated surgically for cystadenoma papillary lymphomatosum of the superficial parotid gland in the Department of Otolaryngology and Laryngological Oncology of the Collegium Medicum of the Nicolaus Copernicus University in Toruń in 2009–2016. The following data were analyzed: age, sex, tumor size measured intraoperatively, early and late complications after surgery and clinical and ultrasonographic suspicion of the neoplastic process in the preserved salivary gland. Tumor size and the presence of possible complications were also assessed.

In the period from January 2018 to April 2018, each patient underwent a follow-up physical examination and otorhinolaryngological interview, and an ultrasound assessment was commissioned. In questionable cases, magnetic resonance tomography examination of the salivary glands was commissioned. Drawing on the results of such a study, distant effects of treatment were assessed, i.e., absence or presence of recurrence characteristics in a clinical and radiological study, and the results were compared with gender, age, tumor size, form of surgery, concurrent factors/diseases before the procedure, presence of possible postoperative complications.

## AIM OF WORK

The analysis aimed to assess the results of treatment of Warthin's tumor in the lower pole of the parotid gland using extracapsular tumor dissection.

## RESULTS

Based on data obtained from the medical history, it was found that at the time of hospital admission, patients reported a number of symptoms of the ongoing neoplastic process. Among the most common complaints, they mentioned the presence of palpable tumor, noticeable face oval asymmetry and paresthesia. Less frequently there was detection of a salivary gland tumor in the course of an episode of acute inflammation, pain on palpation and rapid tumor growth. None of the respondents reported such symptoms as: acceleration of initially slow tu-

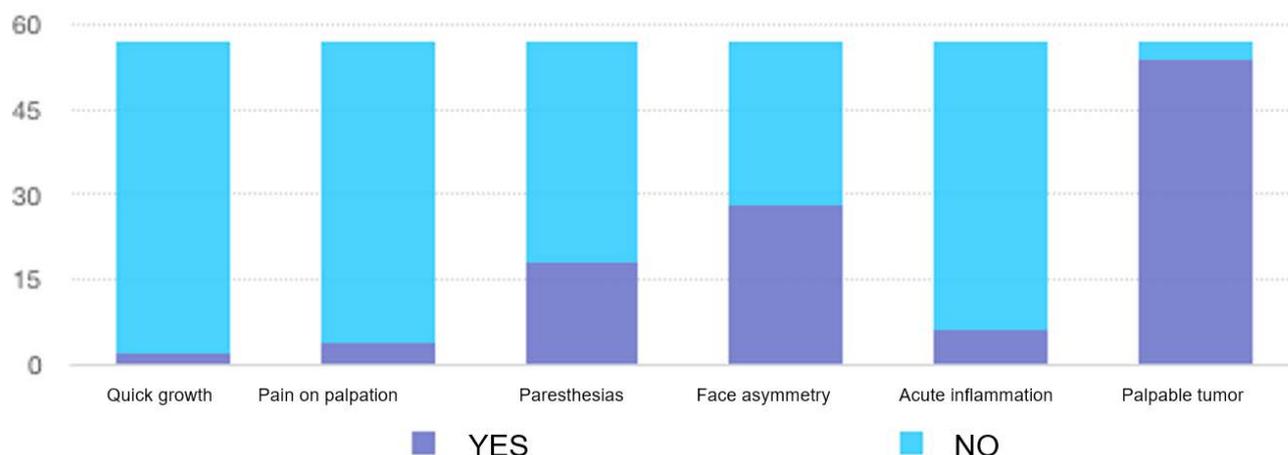


Fig. 1. Pre-operative symptoms reported by patients.

Tab. I. Distribution of the number of tumors dissected in individual years, broken down by gender.

YEAR OF SURGERY	NUMBER OF PROCEDURES	NUMBER OF WOMEN	NUMBER OF MEN
2009	3	0	3
2010	3	2	1
2011	7	5	2
2012	4	3	1
2013	4	0	4
2014	9	2	7
2015	11	4	7
2016	16	6	10

Tab. II. Pre-operative symptoms reported by patients.

DE-SCRIPTION	QUICK GROWTH	PAIN ON PALPATION	PARE-STHESIA	FACE ASYM-METRY	ACUTE INFLAM-MATION	PALPABLE TUMOR
YES	2	4	18	28	6	54
NO	55	53	39	29	51	3

mor growth, spontaneous pain, immobility of tumor against substrate, characteristics of facial nerve damage, change of skin tone over tumor and skin infiltration, oral bleeding, trismus and otalgia, tinnitus or hearing loss. Symptomatology of patients treated with extracapsular tumor dissection is presented in Table II, and illustrated in Figure 1.

Pre-operative imaging diagnostics in 12 tumors was extended to include magnetic resonance imaging.

Surgical protocols allowed to obtain the size of removed tumors

measured intraoperatively as well as data on complications found intraoperatively. In turn, data obtained from the medical history of hospital treatment and the history of posthospital follow-up enables determination of the nature of early postoperative complications.

Size of removed tumors measured intraoperatively ranged from 2–4 cm in 61%.

In the group of failures and complications found intraoperatively during 57 procedures, only one surgery was described as a complication in the form of clinically significant bleeding, which was initially supplied by ligating the bleeding vessel. After the procedure there was no decrease in the parameters of the erythrocytic system which would require supplementation with blood-borne products.

Early complications observed, which include complications associated with surgical treatment used and appearing within 28 days after surgery include: paresthesia, facial nerve paresis and salivary fistula. Presence of paresthesia was determined on the basis of the patient's message and was defined as improper sense of touch and pain, or their spontaneous occurrence. Twenty-eight patients reported complaints of such nature. Paraparesis of the marginal branch of the facial nerve was observed in one patient on the first day after surgery. Pharmacological treatment and early rehabilitation were immediately implemented, resulting in the resolution of paresis on the 7th day after surgery. Complication in the form of salivary fistula was observed in the 4th postoperative day in one patient. It was decided that the patient should be hospitalized. On postoperative day 13, intensive conservative treatment allowed to close the salivary fistula. None of the patients showed complications such as facial nerve paralysis, postoperative wound

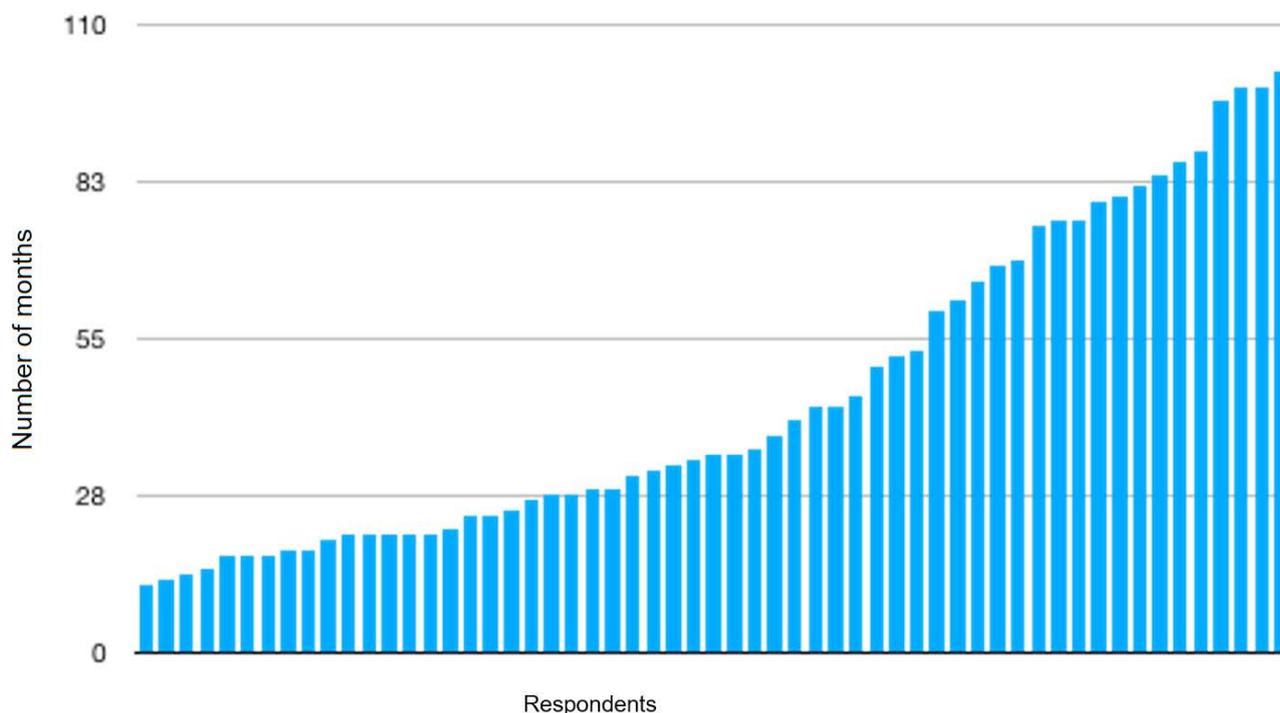


Fig. 2. Period of observation expressed in months.

bleeds, hematoma in tumor bed secondary to dissected tumor, poor healing, skin flap necrosis, salivary cysts or ailments suggesting Frey syndrome.

The observation period of the study group, counted in months as the difference between the date of surgery and the date of ultrasound examination, ranged from 12 months to 102 months, with an average of 45 months and a median equal to 35 months.

Graphical distribution of the observation period is shown in Figure 2.

During follow-up visits, none of the respondents reported complications such as paresis or facial nerve paralysis, fistulas or salivary cysts, symptoms of Frey syndrome. Paresthesia persists in 5 patients without reducing the quality of life in the assessment of subject. Facial oval asymmetry or unsatisfactory scar appearance was determined based on the patient's subjective assessment. In a situation in which the subject would express dissatisfaction with scar appearance and symmetry of the face oval, these factors would be considered positive. However, none of the operated patients with extracapsular tumor dissection reported information about the existing cosmetic defect to the researcher. Two people from the study group reported that they detected lesions by palpation, which caused their anxiety in the projection of the tumor bed after parotid

tumor dissection. Among them, there was one patient who reported pathologies in both salivary glands (he underwent Warthin's tumor resection of both right and left parotid glands).

Ultrasound assessment performed in all subjects showed pathologies in 7 patients in the range of 8 salivary glands that may correspond to recurrence of the neoplastic process. It was decided to further extend the diagnosis of magnetic resonance imaging in these patients.

One of the subjects, who reported bilateral lesions that raised suspicion of tumor recurrence, underwent magnetic resonance imaging which confirmed the presence of multifocal, multiple, bilateral nodular lesions of the salivary glands. However, this patient does not agree to fine-needle aspiration biopsy, nor to surgical treatment remaining under regular monitoring of the Otolaryngologic Outpatient Clinic. Another patient, in the absence of subjective and objective features of the neoplastic process, consciously decided to discontinue further diagnosis. Both clinical examination and magnetic resonance imaging performed in three consecutive patients with the described pathology in ultrasound assessment did not show pathologies in the salivary glands. Two people are awaiting further diagnostics.

The relationship between the date of surgery and suspicion of tumor recurrence in ultrasound is shown in Table IV.

**Tab. III.** Late postoperative complications.

NATURE OF LATE COMPLICATION	NUMBER (%)
Late postoperative complications.	0
Facial nerve paralysis	0
Paresthesia	5 (8,77%)
Salivary fistula	0
Mucocele	0
Frey Syndrome	0
Face oval asymmetry	0
Unsatisfactory scar appearance	0
Clinical suspicion of recurrence of neoplastic process	3 (5,26%)
Ultrasound suspicion of recurrence of cancer process	8 (14%)

**Tab. IV.** Relationship between date of surgery and suspicion of neoplastic process in ultrasound assessment.

ULTRASONOGRAPHY RESULT	2009	2010	2011	2012	2013	2014	2015	2016
No pathology	3	3	5	4	4	5	10	15
Suspicion of recurrence	0	0	2	0	0	4	1	1

The size of pathology described in ultrasound examination may correspond to recurrence of the neoplastic process, as shown in Figure 3.

## DISCUSSION

The history of surgical treatment of benign tumors of the salivary glands begins with the enucleation technique. This method, initially widely used, consisted of simple enucleation of tumor with the capsule, without resection of the surrounding macroscopic unlesioned tissue of the salivary gland parenchyma [11]. After using this technique, the presence of residual disease and frequent recurrences of the neoplastic process were quickly observed [12]. Use of this surgical technique was commonly abandoned in treatment of parotid gland tumors, while implementing an entire range of parotidectomy. This resulted in reducing the risk of recurrence of the neoplastic process, and parotidectomy techniques became the gold standard for treatment of salivary gland tumors in many centers [13].

When removing benign tuberous lesions, two basic goals of this procedure should be borne in mind, which can attest to the success of treatment. Apart from radical dissection of tumor, the priority is to preserve undisturbed function of the facial nerve [13]. In addition, complications such as Frey syndrome or a cosmetic defect associated with the presence of visible scars

or change in facial contours cannot be disregarded. Hence, in recent years there have been discussions on the selection of the optimal surgical technique for benign tuberous lesions of the parotid glands, and opinions are voiced by supporters of preserving techniques – extracapsular dissection (ECD) and partial superficial parotidectomy (SPS) [14].

Performance of extracapsular dissection performed half a century ago and today's meticulous surgery under supervision of the microscope and a facial nerve monitor differ strongly [15]. In publications there are often phrases such as "local excision", "enucleation" and "curettage" used as synonyms for the extracapsular technique. Increased risk of damage to the facial nerve and recurrence of the neoplastic process is observed secondary to these procedures. What is more, these procedures were often carried out as part of the General Surgical Wards, under local anesthesia, without control of the facial nerve monitor [16]. Therefore, extracapsular dissection as a method of removing a tumor with its capsule and the margin of unlesioned salivary gland tissues [11] should be categorically distinguished from the historical technique of enucleation.

Based on a meta-analysis of 14 retrospective studies comparing the failures and complications following treatment of benign tumors of the parotid glands with various surgical techniques, involving 3194 patients, Xie S. observed that supporters of superficial parotidectomy abandon a high percentage of cancer recurrences via extracapsular dissection. On the other hand, proponents of extracapsular dissection argue that with ECD, patients demonstrate a lower number of postoperative complications lowering the quality of life, with a similar percentage of recurrences with superficial parotidectomy [14].

Our group of patients consisted of patients qualified for extracapsular dissection of papillary cystadenoma in the Department of Otolaryngology and Laryngological Oncology of Collegium Medicum of the Nicolaus Copernicus University in Toruń in accordance with the following principles:

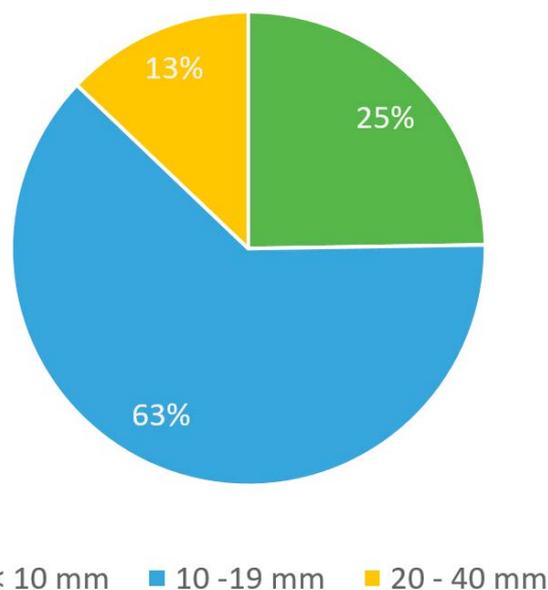
- Tumor located in the lower pole of the superficial parotid lobe, which speaks for the exposure of the tumor's capsule and provides for convenient intraoperative conditions,
- Lack of clinical and ultrasound characteristics that raise suspicion of a malicious process,
- Using fine-needle aspiration biopsy to confirm the benign nature of lesion and/or ensuring availability of intraoperative histopathological examination.

Mean size of tumor operated via superficial parotidectomy is 23.6 mm, and via extracapsular dissection 13.9 mm [17].

Amongst the researched group, the average size of removed tumor is 26.5 mm (range from 13 to 47 mm). The size of tumor was not an absolute factor determining the qualification or lack thereof for the extracapsular procedure.

It has been demonstrated that intraoperative disruption of the tumor capsule correlates with increased risk of tumor recurrence. A significant part of research confirming this relationship is based on a group of patients treated for pleomorphic adenoma [18–20]. In the course of surgery performed via extracapsular dissection, the frequency of this failure is determined at 3.4% regardless of the histopathological type [21]. Our material did not include any case of intraoperative capsule rupture. However, due to the different histopathological character of mixed tumors and adenolymphoma, and above all, the different structure of tumor capsule, we depart from comparing our results with the results available in literature.

Facial nerve palsy in the group operated via ECD ranges from 0–9.7%, and its paralysis from 0–13.3% [17, 21–26]. Our study included only one patient (1.75%) who experienced transient facial nerve paresis - its marginal mandibular branch, which disappeared in the 7th day after surgery as a result of pharmacological treatment and early rehabilitation. Complications in the form of facial nerve paralysis were not observed in any patient. The literature also describes other complications after extracapsular dissection. These include: symptoms of Frey syndrome (2%), formation of salivary cyst (10.1%), postoperative bleeding (0.5%) [16, 17]. These complications were not observed in our material. Salivary fistula occurred in one patient (1.75%), which is similar to reports from Kadletz L. where the frequency of this complication was described at 1.5% [17]. In view of the purpose of surgical treatment, which is radical dissection of cancer, face oval deformity is not considered as a complication of treatment, but rather as its marginal inconvenience. However, with extracapsular dissection, risk of deprivation of face contours is minimized, and even not observed [16]. Our results are consistent with this stance. None of the respondents reported a noticeable cosmetic defect associated with loss of part of the salivary gland or an unsatisfactory postoperative scar appearance



Ryc. 3. Size of pathology described in ultrasound assessment.

Among 57 tumors treated via extracapsular dissection, our study revealed clinical suspicion of recurrence of the neoplastic process in 3 patients, and ultrasonographic imaging performed in these patients revealed hypoechoic focal lesion requiring further diagnosis. Routine ultrasound examination performed in 5 subsequent patients without clinical symptoms showed lesions of unclear nature which required further regular monitoring or extension of diagnosis. Therefore, recurrence of the neoplastic process was suspected among 14% of performed ECD procedures.

## CONCLUSIONS

1. Extracapsular tumor excision appears to be an accurate surgical technique in Warthin's tumor of the lower pole of the parotid gland's superficial lobe.
2. After removal of a cancerous lesion of the parotid gland, every patient requires periodic laryngological monitoring.

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**Competing interests:** The authors declare that they have no competing interests.

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