The incidence of recurrent pleomorphic adenoma of the parotid gland in relation to the choice of surgical procedure

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ABSTRACT

Aim To investigate an incidence of recurrence of pleomorphic adenoma of the parotid gland in relation to the choice of surgical procedure.

Methods All patients with a diagnosis of pleomorphic adenoma of parotid gland who underwent surgery at the Department of Maxillofacial Surgery (MFS) of the Clinical Center of the University of Sarajevo (CCU), Bosnia and Herzegovina, in the period 01 January 1992 – 31 December 2010 were included in the study. The incidence of recurrence and complications in relation to the choice of surgical procedure (enucleation, excision of the tumor, superficial parotidectomy and total parotidectomy) as well as the choice of diagnostic procedures were compared. For all variables measured the arithmetic mean (x), standard deviation (SD), the correlation coefficient (r), and χ² test were used.

Results A total of 81 operations were performed on 60 patients. The highest recurrence rate was after enucleation (88.9%), then after excision (46.9%), and the least after superficial (4%) and total parotidectomy (0%). The highest number of complications was noted after the surgery of recurrence (8.3%), because it was difficult to preserve facial nerve in the previously treated area.

Conclusion Both total or superficial parotidectomy were optimal surgical procedures which, if applied at the first surgery of pleomorphic adenoma, provided almost 100% certainty of healing. All subsequent surgeries carry a higher risk of complications, e.g. severe lesion of facial nerve.

Key words: parotidectomy, facial nerve, Frey syndrome
INTRODUCTION

Pleomorphic adenoma is a benign tumor of the salivary glands with cytomorphological diversity. In 80% of cases it occurs in the parotid gland (1). Second localization is submandibular and palatal salivary glands, in 7% of cases (2). It has an incomplete capsule, and multifocal growth, especially when relapse occurs. Isolated islands of tumor tissue can be formed, tied with a thin trace of the main mass (3). The function of facial nerve is always preserved (3). Pleomorphic adenoma has a tendency to malignant alteration (4). The World Health Organization (WHO) recognizes the entities of carcinoma in pleomorphic adenoma, carcinosarcoma and metastatic pleomorphic adenoma, which are not different histologically from benign pleomorphic adenoma. This tumor metastasizes to lymph nodes, bones, lungs, and kidneys, and can kill the patient (4). Recurrent pleomorphic adenoma is difficult to treat. The best chance for complete removal of the tumor is total parotidectomy at the first operation. Recurrent pleomorphic adenoma has more chances for next recurrences (5). Removing multifocal recurrence, spreading beyond the borders of parotid gland, increases the risk of facial nerve injury (3). The greater possibility of malignant alteration is in recurrence, and the fact that recurrence can occur after ten or 20 years, may falsely encourage conservative treatment (6). Widely accepted operative procedure for benign parotid gland tumors is parotidectomy with preservation of facial nerve, but many surgeons still use simpler operative methods (7). Enucleation was performed as “scaling” of well-circumscribed encapsulated tumor, without removing healthy tissue. Tumor was generally easily peeled off from falsely appearing healthy tissue. An excision was made by removing the tumor with a cuff of healthy tissue, and without preparation of the main trunk of facial nerve and without its lesions. Sharp meticulous preparation around the tumor was done, still leaving a few millimeters of clinically healthy tissue on the tumor, with retrograde preparation of branch of facial nerve if it was found. By superficial parotidectomy the superficial lobe of the parotid gland was removed. After locating the facial nerve (near stilomastoid foramen), continuous removal of the tissue of parotid gland above the nerve was performed. Total parotidectomy was performed by removing complete parotid gland. Once superficial parotidectomy was done, removal of the rest of the tissue beneath facial nerve continued.

The most common diagnostic test performed preoperatively was ultrasound of salivary glands. Some of the patients also had MRI and/or CT scans, and some did not have any diagnostic procedures prior to the surgery.

The aim of this study was to investigate an incidence of recurrence of pleomorphic adenoma of the parotid gland in relation to the choice of surgical procedure, and to prove the necessity of total or superficial parotidectomy at first appearance of tumor.

PATIENTS AND METHODS

A total of 60 patients (81 surgery) who underwent surgery at the Clinical Center of the University of Sarajevo (CCUS), Department of Maxillofacial Surgery (MFS), during the period 01 January 1992 – 31 December 2010, whose histopathological diagnosis was pleomorphic adenoma of parotid gland, were analyzed.

Prospective part of the study included all patients who underwent surgery of parotid gland tumor during 2010, with histopathological diagnosis of pleomorphic adenoma, and retrospective part patients from the period 1992-2009 with the same diagnosis. There were 10 patients in prospective and 50 in the retrospective part of study.

Some patients had several surgeries. They were grouped by ordinal number of operational methods, e.g., 1st, 2nd, 3rd and 4th operation, and the total number of operations was always compared with operating method in relation to recurrence of pleomorphic adenoma.

Enucleation was performed as “scaling” of well-circumscribed encapsulated tumor, without removing healthy tissue. Tumor was generally easily peeled off from falsely appearing healthy tissue. An excision was made by removing the tumor with a cuff of healthy tissue, and without preparation of the main trunk of facial nerve and without its lesions. Sharp meticulous preparation around the tumor was done, still leaving a few millimeters of clinically healthy tissue on the tumor, with retrograde preparation of branch of facial nerve if it was found. By superficial parotidectomy the superficial lobe of the parotid gland was removed. After locating the facial nerve (near stilomastoid foramen), continuous removal of the tissue of parotid gland above the nerve was performed. Total parotidectomy was performed by removing complete parotid gland. Once superficial parotidectomy was done, removal of the rest of the tissue beneath facial nerve continued.

The most common diagnostic test performed preoperatively was ultrasound of salivary glands. Some of the patients also had MRI and/or CT scans, and some did not have any diagnostic procedures prior to the surgery.

An influence of the type of first operation on recurrence was analyzed and set as a predictor of recurrence in patients using a binary logistic regression. As an independent variable (predictor) the type of the first operation was used, since all patients (n = 60) had the first operation.

Further data processing including statistical analysis was done according to the type of surgery
regardless of the number of operations (1st, 2nd, 3rd, 4th) and the patient. The sample was increased from 60 (patients) to 81 (operations). During the transformation of data the type of surgery was used as a predictor, regardless of the order of that operation.

Omnibus test, Hosmer and Lemesh test and Cox & Snell R Square and Nagelkerke R square test were done. This study has approval of the Ethics Committee of the School of Dental Medicine, University of Sarajevo, Bosnia and Herzegovina.

RESULTS

A total of 60 patients (81 operations) were included, 32 female (53.3%) and 28 (46.7%) male patients. The average age at first operation was 38 years. In 28 patients an ultrasound was done without other diagnostic methods, 13 patients had combined diagnostic methods, e.g. ultrasound and magnetic resonance imaging, or ultrasound and CT, or all three methods (CT, MRI, and ultrasound). Ultrasound as a diagnostic tool was done in the total of 41 patients; 18 patients did not have any preoperative diagnostic procedure (Table 1).

Among 60 patients who underwent the first operation, 25 (41.7%) had excision of tumor, superficial parotidectomy was performed in 20 (33.3%) patients, enucleation in eight (13.3%), total parotidectomy, in seven (11.7%) patients. After the first surgery, during the years of follow-up in the period 1992-2011 recurrence was diagnosed in 15 (25%) patients. In relation to the type of the 1st surgery all recurrences happened after enucleation or excision. There was no recurrence in the cases of superficial or total parotidectomy as the first operation (Table 2).

Of 15 operations of the first recurrence, excision was done in six (40%), superficial parotidectomy in four (26.7%), total parotidectomy in four (26.7%), and only one enucleation (6.7%). After the second operation, five patients had second recurrence of the tumor, total parotidectomy was performed in three, one superficial parotidectomy and one excision. One patient had the third recurrence of the tumor, and she underwent fourth operation, total parotidectomy.

Taking into account the total number of 81 operations, 60 (73.4%) were performed for the first occurrence of tumor and 21 operations (26.6%) were performed after a recurrence. Enucleation was done in nine patients, and eight (88.9%) of them had recurrence. Excision was done in 32, recurrence occurred in 12 (37.6%) patients. Superficial parotidectomy was performed in 25, recurrence occurred in one patient (4%) Total parotidectomy was performed in 15 (100%) patients and no recurrence was observed. The highest percentage of recurrence occurred after enucleation in nine (88.9%) patients, and after total parotidectomy no recurrence was noted (Table 3).

Table 1. Diagnostic procedures

<table>
<thead>
<tr>
<th>Diagnostic procedure</th>
<th>No (%) of patients</th>
</tr>
</thead>
<tbody>
<tr>
<td>Ultrasound</td>
<td>28 (46.7)</td>
</tr>
<tr>
<td>Ultrasound and CT</td>
<td>8 (13.3)</td>
</tr>
<tr>
<td>Ultrasound and MRI</td>
<td>2 (3.3)</td>
</tr>
<tr>
<td>Ultrasound CT and MRI</td>
<td>3 (5)</td>
</tr>
<tr>
<td>None</td>
<td>18 (30)</td>
</tr>
<tr>
<td>No data</td>
<td>1 (1.7)</td>
</tr>
<tr>
<td>Total</td>
<td>60 (100)</td>
</tr>
</tbody>
</table>

Table 2. Incidence of tumor recurrence after the 1st surgery according to type of surgery

<table>
<thead>
<tr>
<th>Type of first surgery</th>
<th>No recurrence after first surgery</th>
<th>Recurrence after first surgery</th>
<th>Total</th>
</tr>
</thead>
<tbody>
<tr>
<td>Enucleation</td>
<td>1 (12.5) 7 (87.5)</td>
<td>8 (100)</td>
<td></td>
</tr>
<tr>
<td>Excision</td>
<td>17 (68) 8 (32)</td>
<td>25 (100)</td>
<td></td>
</tr>
<tr>
<td>Superficial parotidectomy</td>
<td>20 (100) 0</td>
<td>20 (100)</td>
<td></td>
</tr>
<tr>
<td>Total parotidectomy</td>
<td>7 (100) 0 7 (100)</td>
<td>14 (100)</td>
<td></td>
</tr>
<tr>
<td>Total</td>
<td>45 (75) 15 (25) 60 (100)</td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

Table 3. Incidence of recurrence after each type of surgery (total number)

<table>
<thead>
<tr>
<th>Type of surgery (all operations)</th>
<th>No (%) of patients</th>
</tr>
</thead>
<tbody>
<tr>
<td>No recurrence</td>
<td>9 (100)</td>
</tr>
<tr>
<td>Recurrence</td>
<td>32 (100)</td>
</tr>
<tr>
<td>Total</td>
<td>41 (100)</td>
</tr>
</tbody>
</table>

Among 60 patients who underwent the first surgery, clinically normal function of facial nerve was noted in 56 (93.4%). Postoperative transient paresis of facial nerve function was noted in three (5.0%) patients, while in one (1.7%) patient a permanent lesion of buccal branch was noticed.

After 21 surgeries of tumor recurrence, two patients had permanent paralysis of facial nerve branches. Of the total number of 60 patients, no one had Frey’s syndrome. There were no salivary fistulas in our patients after any type of surgery for...
pleomorphic adenoma of parotide gland (Table 4). There was a negative correlation between the type of the first operation and the recurrence ($p = 0.000, r = -0.609$). Increasing radicalism of operation reduced the chance of recurrence by about 26 times (OR=0.038, 95%CI=0.005-0.293; $p=0.002$).

There was a good negative correlation between the type of the surgery regardless of the (ordinal) number of surgeries and recurrence ($p=0.000, r = -0.618$). Increasing radicalism of operations resulted in less chance of recurrence 4-62 times (OR=0.064, 95%CI=0.016 to 0.262; $p = 0.000$).

| Complications after each type of first surgery, and after surgery for recurrence |
|------------------------------------------------|----------------|----------------|----------------|----------------|----------------|
| Facial nerve palsy | After 1st enucleation | After 1st superficial parotidectomy | After 1st total parotidectomy | After surgery for recurrence |
| Facial nerve paralysis | 2 | 1 | 0 | 0 | 0 |
| Frey syndrome | 0 | 0 | 0 | 1 | 2 |
| Salivary fistula | 0 | 0 | 0 | 0 | 0 |
| Total | 2 | 1 | 0 | 1 | 2 |

Table 4. Complications after each 1st surgery, and after surgery for recurrence

DISCUSSION

Mild prevalence of the female population with pleomorphic adenoma is expected and described in the literature (8-14), results of this study are the same. The average age of the patients with pleomorphic adenoma in this study at the time of the first surgery was 38 years. The similar age range was reported in previous studies (8,10,12,15).

The authors that advocate for a less invasive approach in the type of surgery are numerous (13,15-23).

Authors who are proponents of radical surgical treatment of pleomorphic adenoma in terms of total and superficial parotidectomy at the first appearance of the tumor are more numerous (1-3, 5, 11, 12, 14, 24-40).

Arshad has published the results of operations of 96 pleomorphic adenoma of the parotid gland, of which 16 were already recurrent, all operated by the subtotal parotidectomy, without recurrence (24). Witt has published the data on 60 patients, of which 20 were treated with extracapsular dissection, 20 with total parotidectomy and 20 with a partial superficial parotidectomy, without any recurrence after eight years (15). Of 106 pleomorphic adenomas operated by total parotidectomy, Web reported four recurrences. McGurk reported that 491 patients underwent extracapsular resection, of which eight had a recurrence (1.7%), and two recurrences (1.8%) after 139 superficial parotidectomy after 15 years (17). Stennert has reported 31 patients with recurrence, in which previous initial treatment was enucleation in 17 (55%) patients, superficial parotidectomy in ten (32%), and subtotal parotidectomy in four (13%), and recurrence was treated with total parotidectomy with a preparation of facial nerve and scar excision (14).

Orvidas recommended a radical treatment if patients were children. Of 21 patients with pleomorphic adenoma of the parotid gland initially treated at the Mayo Clinic, with superficial or total parotidectomy, only one had recurrence (40).

Differences among compared studies are related to the initial treatment of pleomorphic adenoma, where some recommend wide excision or partial superficial parotidectomy, and others recommend superficial or total parotidectomy. (1-3, 5,11,12,14, 24-40). This study recommends superficial or total parotidectomy as initial treatment.

When it comes to the treatment of recurrence, most recommend total parotidectomy (1-3, 5, 11-40) Ultrasound was the most common diagnostic procedure with our patients. Some authors have reported fine needle biopsy and ultrasound done in all patients (11). Other diagnostics, CT and MR, were done in large neoplasms or recurrence (8).

It has to be mentioned that fine needle aspiration (FNA) biopsy with ultrasound is mandatory because both patient and surgeon preoperatively know if they are dealing with benign or malignant tumor, and FNA biopsy usually gives the exact diagnosis (39).
Up to 100% facial nerve palsy (temporal) was reported after total and superficial parotidectomy (27). However, this complication is not severe, it usually lasts for a couple of weeks (27). Facial nerve paralysis (definitive) is frequent after surgery for recurrent tumor (after enucleation and excision done as the first surgery) (27).

Frey’s syndrome was not observed in our patients, and the same results were reported by others (8), while some authors found up to 100% of Frey’s syndrome after total parotidectomy (15), and other authors reported Frey’s syndrome frequently with more radical procedures (18,24,35).

There were no salivary fistula in our patients, and some other authors reported the same results.

REFERENCES

11. Redaelli de Zinis LO, Piccioni M, Antonelli AR, and (24,35). McGurk had 0.6% of salivary fistula after extracapsular dissection, and no one after superficial parotidectomy (18).

In conclusion, recurrent pleomorphic adenoma was more frequent after enucleation and excision, while superficial and total parotidectomy have significantly less recurrence. Complications of parotid surgery were more frequent after surgery for recurrence, therefore, everything must be done to prevent recurrence of pleomorphic adenoma.

FUNDING

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TRANSPARENCY DECLARATIONS

Competing interests: none to declare.
Čaušević Vučak et al. Pleomorphic adenoma of the parotid gland


Učestalost recidiva pleomorfog napadenja parotidne žlijezde u odnosu na izbor operativne metode

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SAŽETAK

Cilj Ispitati učestalost pojave recidiva pleomorfog napadenja parotidne žlijezde u odnosu na izbor operativne metode.

Metode Ispitanici su bili svi pacijenti koji su operisani na Klinici za maksilofacijalnu hiruršicu (MFH) Kliničkog centra Univerziteta (KCU) u Sarajevu, u periodu od 01. 01. 1992. do 31. 12. 2010, s dijagnozom adenoma pleomorphe gl. parotis. Ispitana je učestalost recidiva i komplikacija u odnosu na izbor operativne metode (enukleacija, ekscizija tumora, superficijalna parotidektomija i totalna parotidektomija), kao i izbor preoperativih dijagnostičkih metoda. Za sve mjerene varijable određena je aritmetička sredina (x), standardna devijacija (SD), koeficijent korelacije (r) i χ2 test.

Rezultati Ukupno je urađena 81 operacija na 60 ispitanika. Najviše recidiva zabilježeno je nakon operativne metode enukleacija (88.9%), zatim ekscizije (46.9%), a najmanje nakon superficijalne (4%) i totalne parotidektomije (0%). Rezultati su pokazali najveći broj komplikacija nakon operacija recidiva (8.3%) jer je bilo teško sačuvati facijalni nerv u prethodno tretiranom području.

Zaključak Totalna i superficijalna parotidektomija su najoptimalnije operativne metode koje, ako su primjene pri prvoj operaciji pleomorfog adenoma, pružaju gotovo stopotstnu sigurnost izlječenja. Svaka sljedeća operacija recidiva bolesti nosi veći rizik od pojave komplikacija od kojih je najteža lezija glavnog stabla facijalnog nerva, što može ostaviti težak invaliditet.

Ključne riječi: parotidektomija, n. facialis, Frey sindrom