Combined endoscopic–transcutaneous surgery in parotid gland sialolithiasis and other ductal diseases: reporting medium- to long-term objective and patients′ subjective outcomes

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Abstract Objective was to assess the medium to long-term results and patients′ perceive of success after combined sialoendoscopic and transcutaneous access in salivary gland diseases. A retrospective analysis was performed in a tertiary referral centre. A total of 19 patients have been treated with a combined sialoendoscopic and transcutaneous access. The main indication was sialolithiasis in 89.5 % of cases ($n = 17$), in 2 of these cases simultaneous complications were treated. Other indications included treatment-resistant stenosis and traumatic transection of the parotid duct. Intraductal stents were placed in 52.6 % of the cases. Patients were evaluated by clinical investigation, ultrasound examination and by a questionnaire to assess patients perceive of success. As a result the treatment was successful in 89.5 % of all cases, and in 94.1 % of the patients with sialolithiasis. Parotidectomy was required in two patients, as reconstruction of the ductal system was not possible intraoperatively (sialolithiasis, $n = 1$) or was unsuccessful (stenosis, $n = 1$). Prerequisites for successful treatment were the endoscopic access to the pathology, the possibility to reconstruct the duct and recovery of gland function postoperatively. A mean follow-up time for successfully treated patients was 40.67 months. All patients were satisfied with the results and reported a significant reduction in symptoms and improvement of their perceived quality of life ($p = 0.001$ each). As conclusion the combined access is a valuable alternative treatment in patients with sialolithiasis. Additional indications may include treatment-resistant stenosis and injuries to the parotid duct. However, the indication in stenosis needs to be carefully weighed up.

Keywords Salivary glands · Obstruction · Therapy · Minimally invasive · Sialoendoscopy · Sialolithiasis · Stenosis · Duct trauma · Combined approach

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Introduction

In patients with obstructive salivary gland diseases, and with sialolithiasis in particular, the most common cause, in 70 % of cases, the trend towards gland-preserving therapy has now largely become an established approach. The introduction of various minimally invasive forms of treatment has significantly reduced the rate of gland excisions, and salivary gland endoscopy plays an important role in a combined treatment approach [1–10]. Although sialoendoscopy is at least a valuable additional method in patients with stones, with success rates of more than 80 % [1–4, 6, 8, 9] it is also an extremely important procedure for ensuring gland preservation in cases of stenoses of the parotid duct (Stensen's duct) [5, 6, 9, 10].

Surgical procedures combining sialoendoscopy and transcutaneous access have been developed in recent years [11–15]. The indications for this approach are treatment-resistant cases, particularly with sialolithiasis. It is often not possible to treat impacted calculi and stones larger than 6–7 mm using sialoendoscopy alone, and even in combination with extracorporeal shock-wave lithotripsy (ESWL) the success rates decline with the increasing stone size [4]. Successful treatment using the minimally invasive treatment methods available is therefore not possible in approximately 5–10 % of patients with parotid stones. In addition to the size, quality, and material of the stones,

other reasons include contraindications for established treatment methods (e.g., status post-cardiac pacemaker implantation for ESWL). Most research groups have thus also reported on the use of the combined approach in treatment-resistant or large parotid duct stones [11–15]. Marchal et al. [13] described the use of this approach in stenoses, but without providing follow-up data. McGurk et al. [11] have questioned the value of this treatment method in stenoses. There has so far been a lack of data in the literature on the acceptability of this operating method to patients. The aim of the present study was to analyze options and limitations with this method using the available medium-term and long-term follow-up data, taking glandular function into consideration. Follow-up investigations also included an analysis of patient acceptance of the method based on a questionnaire survey.

Patients and methods

Patients, indications, and diagnoses

A total of 19 patients have been treated at the Department of Otorhinolaryngology, Head and Neck Surgery at the University of Erlangen-Nuremberg since 2006 using the combined transcutaneous–endoscopic operation ($\sim$1 % of all patients treated for obstructive salivary gland diseases during the period). The patients included 12 men (63.2 %) and 7 women (36.8 %), with an average age of 53.21 years (median 55, range 23–69). The indications were sialolithiasis in 17 cases, and treatment-resistant stenosis and traumatic transection of the parotid duct in one case each. The indication in sialolithiasis was established primarily in two cases (patient's own request following detailed information, $n = 1$; contraindication to ESWL due to cardiac pacemaker, $n = 1$), due to treatment resistance in 12 cases and due to non-compliance in 3 cases. ESWL was carried out an average of 3.38 $\pm$ 0.40 (median 3, range 1–6) times. In three of these cases, the patient did not wish to continue the treatment with ESWL (after 1 ESWL procedure in 1 patient and after 2 in 2 patients). The procedure was indicated after development of complications in two cases: salivary–cutaneous fistula following abscess in sialolithiasis, $n = 1$ (Fig. 1); and stone perforation through the ductal wall with sialocele formation, $n = 1$ (Fig. 2).

One patient was operated because of treatment-resistant stenosis and another was send after trauma of the cheek with signs of a salivo-cutaneous fistula.

The primary diagnostic procedure in all cases was ultrasonography (Sonoline Elegra or Acuson Antares; Siemens Medical Solutions USA, Inc., Malvern, PA, USA). This allowed precise diagnosis of the location and size of the stones or residual stones in all cases (Figs. 1, 2).

Fig. 1 High-resolution ultrasonography. A row of stones is visible in Stensen's duct and in a fistula (*white arrow*) between Stensen's duct and the skin. *Fistel* fistula, *GLP* parotid gland, *MM* masseter muscle, *M* mandible

Fig. 2 High-resolution ultrasonography. A stone has perforated the ductal wall and a sialocele has formed, with no evidence of a direct and patent connection with Stensen's duct. *S* stone, *RF*, sialocele, *GLP* parotid gland, *DS* Stensen's duct

Diagnostic sialoendoscopy was carried out before the procedure to ensure that endoscopic marking was possible. Semirigid endoscopes and instruments forming part of our current set were used (Karl Storz Ltd., Tuttlingen, Germany) [5, 6].

In two cases in which it was not possible to advance the endoscope directly to in front of the stone, a basket was introduced as far as the stone under endoscopic guidance and with simultaneous ultrasound control. This ensured that it was possible to mark the stone intraoperatively (Fig. 3a and b).

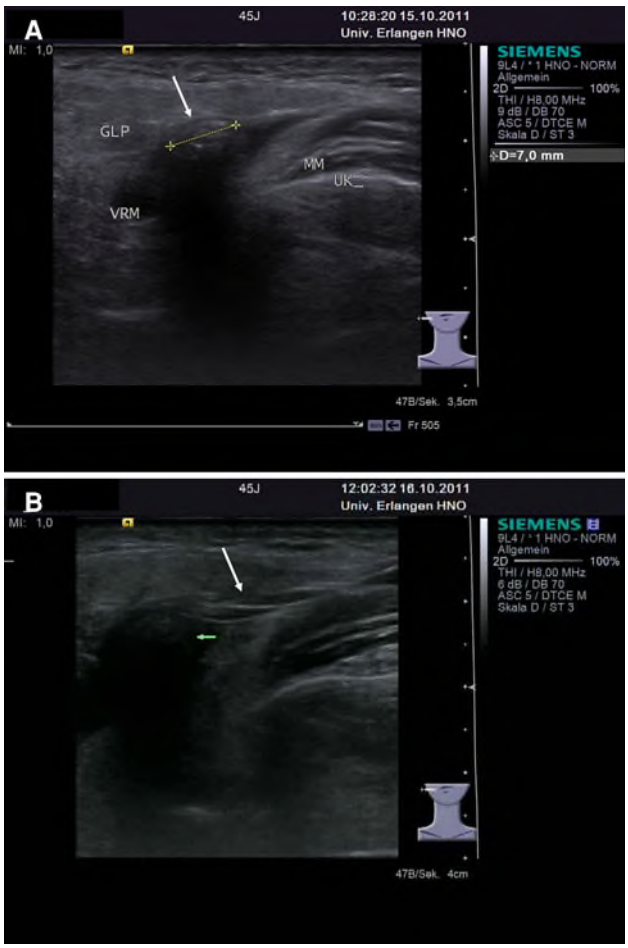

Fig. 3 High-resolution ultrasonography. **a** There is a stone (*white arrow*) in the area of the hilum that was not accessible endoscopically due to a downward bending of the duct after division of the ductal system. **b** Marking of the stone (*green arrow*) using an instrument that has been advanced (basket, *white arrow*) to allow assessment of the stone's intraoperative accessibility. *VRM* retromandibular vein, *GLP* parotid gland, *MM* masseter muscle, *UK* mandible

Surgical technique and modifications

All of the procedures were carried out with the patient under general anesthesia. The basic technique used in our hospital has been described previously [15]. Diaphanoscopy in the ductal system allowed precise dissection and opening of the duct for stone extraction. Buccal branches of the facial nerve usually have to be demonstrated in this process, and facial nerve monitoring was therefore used (two-channel electromyography, Neurosign 100, Inomed Medizintechnik Ltd., Emmendingen, Germany). Residual calculus was excluded sialoendoscopically or removed. If the stone could not be accessed directly with the tip of the endoscope, but marked with an instrument (Fig. 3b), the procedure was modified. The duct was opened at the location of the tip of the endoscope and the endoscope is inserted again directly through the sialodochotomy (Fig. 4a). The stone was then marked again with the sialendoscope and extracted with the basket if necessary by extending the sialodochotomy (Fig. 4b). In such a case of the extended sialodochotomy, stent implantation was indicated (Fig. 4c), as in every case of severe maceration, narrow duct system or simultaneous ductal stenosis (Sialotech Ltd., Ashkelon, Israel) [6, 15]. The stents were placed using two routes: anterograde, from the papilla or retrograde from the sialodochotomy site [15].

In case of a simultaneous sialolithiasis and ductal stenosis, the stenosis was planned to be dilated endoscopically before or during the procedure. If complications like fistula or sialocele were recognizable preoperatively, treatment was adapted and the underlying condition treated.

In case of treatment-resistant stenosis repair of the duct by a vein patch or replacement of the duct by vein interposition were the options. When traumatic damage of the duct was evident, the goal was reconstruction by primary anastomosis.

The closure or anastomosis of the parotid duct was carried out using absorbable monofilament sutures (Monocryl 6–0) or woven sutures (Vicryl 6-0 or 7–0; Ethicon Products, Norderstedt, Germany). After closure of the glandular capsule, a collagen fleece (TachoSil®, Nycomed Ltd., Constance, Germany) was placed over the suture site for prophylaxis against fistulas.

Postoperative follow-up and evaluation of glandular function

The course of wound healing, correct positioning of the stent, and glandular function were checked clinically and with ultrasound at discharge (1 week postoperatively) and after 2 months or during data collection. At the clinical examination, undisturbed glandular function was evident from the lack of symptoms and from clear secretory flow from the papilla after gland massage, increasing after stimulation with vitamin C. Restoration of the physiological function of the gland was evident on ultrasound, with normalization of echogenicity in the glandular parenchyma (from hypoechoic to hyperechoic). The ductal system showed normal to slight dilation. Significant dilation of the ductal lumen after stimulation with vitamin C in combination with clinical symptoms was regarded as a sign of ductal obstruction.

Evaluation of patient acceptance and benefit of the operation for patients

The patients were given a questionnaire 2 months after the operation or at the time of data collection. The questionnaire inquired about the following parameters:

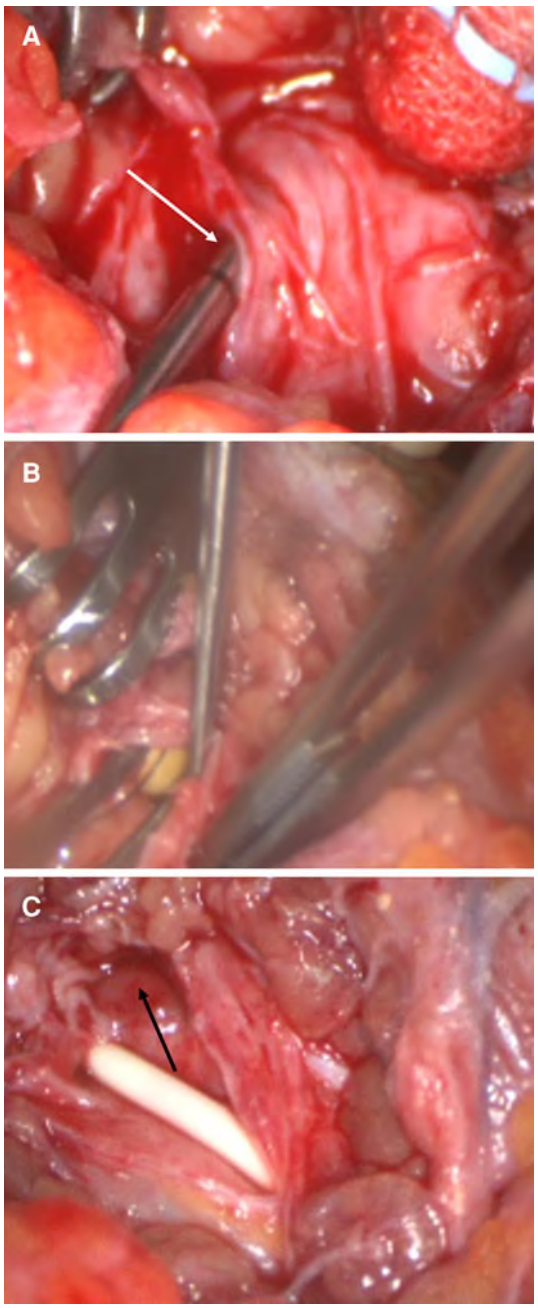

Fig. 4 Sialoendoscopy via the sialodochotomy in a patient with a stone that was not initially accessible endoscopically (corresponding to Fig. 3). **a** The sialendoscope was inserted directly through the sialodochotomy (*white arrow*). **b** When the sialodochotomy was extended (approximately 1.5 cm), stone extraction became possible using various instruments (endoscope, basket and hook). **c** Stent placement was necessary as a result of the extended sialodochotomy and was carried out in a retrograde direction through the sialodochotomy into the lower part of the main duct (the stent is visible in situ, *black arrow* shows the upper part of the duct system)

- Current symptoms or pain: if yes, which, how often, and with an assessment of severity using a visual analogue scale (VAS; 0 = minimum to 100 = maximum).
- Current comorbid conditions.
- Assessment of the value of the operation (negative or positive effect) and change in subjectively perceived quality of life after the operation (much worse—worse—unchanged—better—much better).
- Comparison of subjectively perceived symptoms before and after the operation, each using a VAS (0 = minimum to 100 = maximum).
- Comparison of subjectively perceived quality of life before and after the operation, each using a VAS (0 = minimum to 100 = maximum).
- To obtain evidence of the medium-term to long-term results of the treatment method, patients who had a follow-up period of at least 50 months were evaluated once again separately from the above.

Statistical analysis

Testing for significant differences with regard to preoperative and postoperative symptoms and subjectively perceived quality of life was carried out using the Wilcoxon's rank test for matched samples. The significance level was $P = 0.05$. Data are given as mean ± standard error of the mean (SEM). The software program SPSS, version 18 for Windows (SPSS Inc., Chicago, IL, USA), was used.

Results

The follow-up period in patients with gland preservation ($n = 17$) was 40.67 ± 5.37 months (range 3–67.5).

According to the location of the pathology along the duct system, incision was made along a skin-fold in the cheek area in 2 cases and dissection of a preauricular flap, as in parotidectomy, was performed in 15 patients.

The average size of the stones in 17 patients immediately before the transcutaneous operation was 9.2 ± 0.63 mm (median 9 mm, range 4–16 mm). Stone extraction was successful, and the gland was preserved in 94.1 % including the two cases who presented with complications (16 of 17). Residual calculus was also extracted from the neighboring ductal system endoscopically using a basket or forceps in four cases. Stent placement was carried out in 60 % of the patients (9 of 15) treated for sialolithiasis.

Excision of a salivary–cutaneous fistula was carried out in one case. After the connection with the ductal system had been demonstrated using diaphanoscopy, the fistula in the area of the parotid duct (Fig. 5a) was excised along with several stones (Fig. 5b). A stone that was not accessible via the main duct using the sialoendoscope was removed from a sialocele in one case (Fig. 6a). Diaphanoscopy with the sialoendoscope helped distinguish

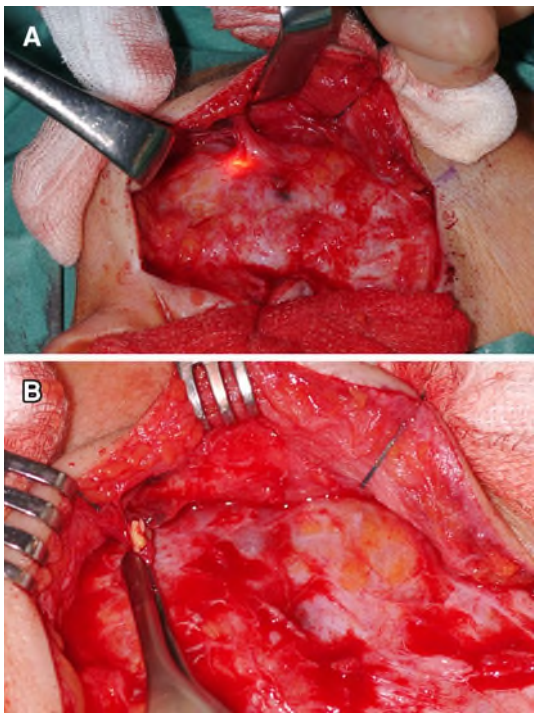

Fig. 5 A fistula between Stensen's duct and the skin. **a** The precise locations of Stensen's duct and the orifice of the fistula were marked using diaphanoscopy. **b** Stone extraction from the fistula and ductal system after opening. A check-up endoscopy in the ductal system proximal to the fistula excluded the presence of any additional stones

between the two structures, and the sialocele was resected (Fig. 6b).

One patient had a simultaneous ductal stenosis proximal to the stone, which was dilated endoscopically before and during the procedure.

In one case parotidectomy was indicated, because the ductal wall appeared to be so severely macerated intraoperatively after stone extraction that ductal reconstruction appeared too risky.

One patient was treated surgically 1 week after a traumatic transection of the main ductal orifice of the parotid gland. Precise assessment of the tissue quality in the area of the ductal stumps assessment of the internal ductal structures was possible using sialoendoscopy. The ductal stumps were anastomosed in this case with 7-0 nylon sutures (Ethilon 7-0; Ethicon Products, Norderstedt, Germany) and an indwelling silicone catheter (22G, 0.9 mm) was implanted as a stent. This was removed after 6 weeks, and the check-up sialoendoscopy showed a slight fibrous stenosis, with no further need for treatment [16].

A completely treatment-resistant fibrous stenosis was treated in one case. The stenosis was located at the junction between the central duct and the hilum and was demonstrated using diaphanoscopy with the help of the sialoendoscope. The duct distal to it was resected and replaced with a venous interposition graft from the retromandibular vein. The graft was anastomosed with the remaining proximal megaduct and was sutured intraorally in the area of the excised papilla (Fig. 7a, b). It was noted intraoperatively even when the duct was opened that the saliva had a very thick, almost jelly-like consistency. After an initially normal postoperative course, a megaduct developed 1 week later. At the check-up endoscopy, the anastomosis was found to be intact and widely patent, but the ductal lumen was obstructed with fibrinous plaques. A stent was therefore implanted. Despite this, obstruction of the stent by fibrinous plaques developed again 2 weeks later: The stent therefore was removed and at the check-up endoscopy

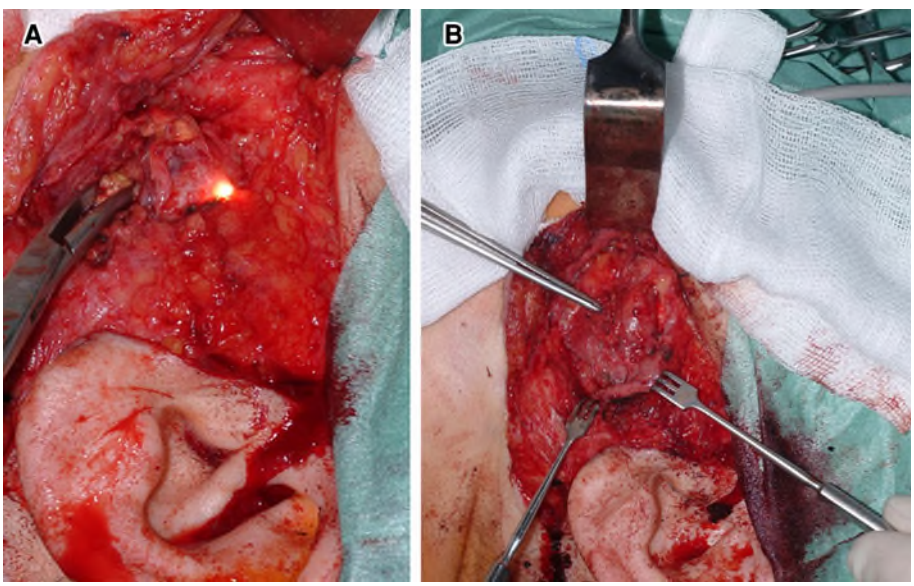

Fig. 6 **a** Stone extraction through an opened sialocele that does not show a direct connection to the main duct and may have arisen due to perforation of the stone through the ductal wall. **b** The main duct is marked by diaphanoscopy from the distal end of the endoscope

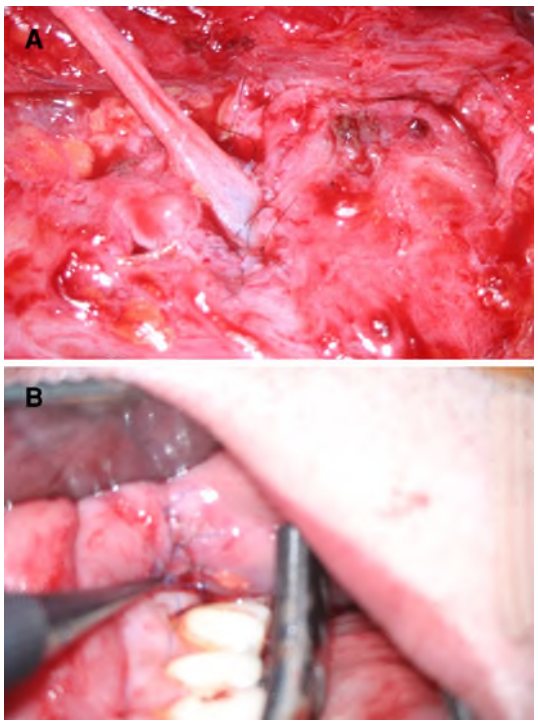

Fig. 7 **a** Vein interposition in therapy-resistant complete stenosis: interponed vein after anastomosis to proximal duct system (**a**) and suturing to the buccal mucosa creating a neoostium (**b**)

an adequate ductal lumen was not visible. Owing to the local situation and insufficient glandular secretion (with a jelly-like salivary consistency), parotidectomy was indicated.

Overall, parotidectomy was unavoidable in two cases, but it was possible to preserve the gland 89.5 % of the patients. Stent implantation was carried out in 58.8 % of the successful procedures (10 of 17). The stent was placed via the anterograde route in seven cases (normally dilated ductal systems) and retrograde in three cases (narrow duct systems). The stents were tolerated without problems in all of the patients and were removed in only two cases after 6 weeks during the planned check-up endoscopy. In all of the other cases (80 %), there was spontaneous dislocation of the stent 2–4 weeks postoperatively.

One complication occurred (postoperative hematoma in a patient with known hepatic insufficiency). No cases of facial nerve paralysis or fistula occurred in any of the patients.

Function was restored in all of the patients with gland-preserving procedures. Clear salivary secretion from the papilla was seen clinically in all of the patients at the follow-up examination, and the ductal system was slightly wider than normal in all cases. Salivary flow increased after vitamin C stimulation, and the ductal dilation visible on ultrasonography increased simultaneously (to a maximum of 2–3 mm); no concomitant clinical symptoms developed, however. In one case after opening of a simultaneous ductal stenosis proximal to the stone, ultrasound showed a ductal dilation (2.5 mm), which increased further after vitamin C administration (to 3.5 mm). However, the glandular parenchyma did not show relevant pathological changes. A check-up endoscopy after 4.5 years showed a residual moderate fibrous stenosis. Considering the absence of symptoms, no additional therapy was required.

The patients' assessments of the benefits and success of the operation were highly positive. No typical symptoms of recurrent obstructive sialopathy occurred during the subsequent course in any of the patients who underwent gland-preserving surgery. The mean VAS scores were 2.94 ± 0.96 (range 0–10) for current symptoms and 0.35 ± 0.29 (range 0–5) for current pain. Two patients each reported weather-related swelling in the scar region or a nonspecific "aching feeling" in the surgical area, but none of the patients described the scar itself as troublesome. All of the patients reported that the operation was certainly worthwhile (highly positive effect, $n = 17$; perceived quality of life much better, $n = 17$). The main reason given for the positive assessment by patients with obstructive salivary gland diseases was ability to eat without glandular swelling, with no pain and with no social stigmatization. The preoperative subjectively perceived severity of obstructive sialopathy symptoms was given as a mean of 77.35 ± 2.94 (range 55–100) on the VAS; postoperatively, in contrast, the mean score was only 2.35 ± 0.96 (range 0–15). The difference was significant ($P = 0.0001$). The preoperative subjectively perceived quality of life showed a VAS score of only 35.0 ± 4.86 (range 5–65); postoperatively, it was 92.94 ± 1.82 (range 75–100). The difference was again significant ($P = 0.0001$). Several patients had comorbid conditions, but only two reported that these made subjective assessment of their perceived quality of life relative to the symptoms of obstructive sialopathy more difficult (epilepsy and hepatic insufficiency, $n = 1$; trigeminal neuralgia, scleroderma, craniomandibular dysfunction, $n = 1$).

Half of the patients, who underwent gland-preserving therapy had a follow-up of at least 50 months, with an average follow-up period of 60.67 ± 2.29 months (median 63, range 50–67.5). The current severity of symptoms averaged 3.88 ± 1.61 (range 0–10) and the current severity of pain averaged 0.67 ± 0.55 (range 0–5). In these patients, the subjectively perceived severity of the preoperative symptoms on the VAS averaged 76.35 ± 4.24 (range 55–100), while postoperatively it was only 3.88 ± 1.61 (range 0–15). The difference was significant ($P = 0.007$). The preoperative subjectively perceived quality of life averaged only 30.0 ± 6.87 (range 5–60) on the VAS, while postoperatively it was 91.11 ± 3.03 (range 75–100). The difference was again significant ($P = 0.007$).

The results for these patients did not differ from those in the overall group.

Discussion

Approximately 90–95 % of patients with obstructive salivary gland diseases can be treated with preservation of the gland using minimally invasive methods [1–10, 17]. There have been several published reports in recent years on a combination of endoscopy with transcutaneous access as an additional treatment option to preserve the gland. Diaphanoscopy with the tip of the sialoendoscope, introduced into the ductal system, is an essential prerequisite for a precise and tissue-conserving dissection technique (Fig. 5). Treatment-resistant sialolithiasis is the indication most frequently mentioned [11–15]. Impacted stones, large stones with a diameter of more than 8–10 mm, and treatment-resistant stones may represent limitations for treatment approaches based on the sialoendoscopy and ESWL [1, 2, 6, 8]. In these cases, the transcutaneous surgical procedure described here may provide an option for treatment with preservation of glandular function. Several studies have reported on the technique, with success rates up to 90 % [11–15]. Our own extended data, with a success rate with parotid gland stones of 94.1 %, confirm the indications for this surgical technique.

The procedure is not possible with intraparenchymal stones which are not safely accessible endoscopically or with endoscopy-assisted methods. In these cases, ESWL seems to be the only treatment option allowing preservation of the gland.

The literature data also indicate that this surgical technique may also be useful in other salivary gland diseases.

Marchal et al. [13] reported on reconstruction of the duct using a venous patch after resection of fibrotic portions. However, precise data on the course and success rates with this modification are not available. McGurk et al. [11] reported that when there was stenosis after excision of fibrotic tissue, adequate reconstruction of the duct was also not possible, leading to ligation of the duct. The case reported in the present study shows that therapy for treatment-resistant high grade to complete stenosis was technically possible in the patient concerned (Fig. 7a, b), but that it ultimately failed due to the quality of the saliva still being produced and the level of residual glandular function still present—evident from the extremely thick, jelly-like saliva with adhesive tendencies. Reconstruction with a more thick-walled vein, such as the cephalic vein might provide additional benefit. In general, with the data currently available it is not yet possible to draw any final conclusions regarding the value of this surgical method in treatment-resistant ductal stenoses.

Traumatic injuries to Stensen's duct are another possible indication. Nahlieli et al. reported on sialoendoscopically controlled diagnosis and treatment of ductal injuries after facelift procedures. The report included not only the identification of strictures and perforations, but also endoscopic dilation of compressions and strictures and endoscopic therapy for ductal perforations using stent placement. Sialoendoscopy appears to be helpful here both in the immediate post-traumatic period and after an interval, as it was the case in our patient [16, 18].

Stent placement in some cases to stabilize the opened ductal system and to avoid cicatricial obliteration is practiced and recommended in all reports [11–15]. Stents were placed in 52.6 % of the patients included in the present study. They appear to be beneficial for complication-free healing, as they stabilize the ductal system and prevent cicatricial stenosis. The indications in our patients were a severely macerated ductal wall, a very narrow ductal lumen, and an extended sialodochotomy. Stents were not associated with any additional symptoms and could be removed without any problems.

Parotidectomy was unavoidable in 10 % of the patients included in the present report, and was carried out either simultaneously or after an interval, in 5 % of cases each. Considering the reported failures, the indication has to established cautiously, particularly with regard to perform a treatment in therapy-resistant stenoses [11, 13]. However, the present analysis also shows that the pathology can be eliminated in 90 % of cases, with simultaneous preservation of the gland and confirms the results reported in the literature [11–14]. There have been several reports on the feasibility of this operation, but no reports are available which reported on the long-term course after this procedure [11–15]. Assessment of whether the operation was successful is based on the elimination of the pathology, long-term freedom from symptoms, and preservation of glandular function. In addition to providing medium-term to long-term results, the present report also evaluated how the treatment was judged by the patients and their assessment of its influence on their perceived quality of life. The mean follow-up period for all of the patients was in the medium-term range at 40.67 months. Half of the patients with gland-preserving therapy had a follow-up of at least 50 months, with an average follow-up period of 5 years (60.67 months). All of the patients were free of stones, and glandular function had recovered in all cases both clinically and on ultrasound. The scores both for the overall group of patients and for those with medium-term to long-term follow-up show that the patients were generally very satisfied with the results of the operation and with the functional results, and they indicate a high level of patient acceptance. There were no cases of current symptoms involving any glandular obstruction, and in particular

current pain symptoms were at a very low level (<1 on a VAS from 0 to 100). The effect of the operation was evaluated as very positive by all of the patients. Significant improvements were reported both with regard to symptoms and also subjectively perceived quality of life both by the overall group (both $P = 0.001$) and by the patients with medium-term to long-term follow-up (both $P = 0.007$).

In conclusion, the combined endoscopic–transcutaneous operation appears to be a valuable gland-preserving treatment option for various diseases of the parotid gland. It is indicated in patients with large and/or treatment-resistant stones, with complications of sialolithiasis, and when there are contraindications against primarily indicated sialolithiasis treatment. A sialendoscopy-assisted transcutaneous approach also appears to be advantageous in cases of traumatic injury to the main efferent duct. The procedure also appears to be at least technically feasible in treatment-resistant stenoses of the main efferent duct.

Prerequisites for success with the procedure include meticulous surgical technique; accessibility of the pathology using the endoscope, or at least with an instrument advanced through the endoscope; and the integrity of the anatomic structures, particularly the ductal system, to be able to carry out reconstruction. In addition, adequate function or regeneration capacity in the salivary gland cells when obstructions have persisted for a long period, particularly in therapy for treatment-resistant stenoses, appears to be from paramount importance.

When the indication is correctly established, this combined surgical method offers a success rate of around 90 %, according to the available data. Assessment of the success of the procedure should include not only achievement of a symptom-free state, but also complete elimination of the pathology and recovery of glandular function. However, another important factor associated with medium-term to long-term success appears to be the degree of acceptance of the procedure by the patients themselves, which was very high in this study. Development of the surgical technique may offer prospects of even higher success rates and also a further widening of the range of indications in the future.

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