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Simultaneous application of ultrasound and sialendoscopy: experience in the management of stenosis and other non-sialolithiasis-related salivary gland disorders

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Abstract. – OBJECTIVE: To assess the value of the simultaneous application of ultrasound and sialendoscopy (US+SE) in several salivary gland diseases not caused either by sialolithiasis or by tumours.

PATIENTS AND METHODS: US+SE are routinely used in patients with inflammatory, obstructive, and other non-tumorous major salivary gland diseases. In patients in whom US and SE as single investigation tools were not conclusive or not useful in the management of several non-sialolithiasis-related conditions (stenoses, ductal anomalies, ductal trauma, space-occupying paraductal lesions), both methods were used simultaneously for diagnosis and treatment.

RESULTS: US+SE were used simultaneously in 44 patients for 56 indications. Stenosis was managed in 36 cases (81.8%) and in thirty-eight of the indications (67.9%) with simultaneous US+SE. The successful opening was achieved in 23 (63.9%), conservative and/or ablative treatment was indicated in 13 (36.1%), and further imaging was indicated in two (5.5%) of these cases. Post-traumatic or postinfectious complications were managed in 12 (27.3%) of all cases, and isolated ductal anomalies and paraductal space-occupying lesions were assessed in three cases (8.3%) each. In all instances, simultaneous US+SE clearly improved the management in diagnosis and/or therapy.

CONCLUSIONS: Simultaneous application of US+SE provided additional information that proved to be valuable for diagnosis, planning and conduction of the treatmen in several non-sialolithiasis-related conditions such as stenoses, ductal anomalies, ductal trauma, and space-occupying paraductal lesions.

Key Words:

Salivary, Glands, Submandibular, Parotid, Ultrasound, Stenosis, Sialendoscopy, Duct anomaly, Ductal trauma.

Introduction

Ultrasound (US) is part of the routine diagnostic armamentarium in many units that deal with salivary gland diseases–particularly various inflammatory and obstructive diseases, or sialadenitis after radiotherapy or radioactive iodine treatment[1-15]. The value of US has been confirmed in sialolithiasis[3,5,16-20] and ductal stenosis[21-26]. Sialoceles, caused by laceration of the duct by trauma or surgery, are mainly observed in the parotid gland (PG) and can be identified by US examination[27,28], but they have also been described as sporadic findings or "idiopathic sialoceles" in the submandibular gland (SMG) as well[29]. The use of US in combination or in comparison with computed tomography or sialography[30,31], magnetic resonance sialography, and sialendoscopy (SE)[31], or as an adjunct to cone-beam computed tomography in combination with sialography[32] for non-sialolithiasis-related conditions has been described in a few reports. SE has been shown to be very useful in the management of patients who present unclear ductal dilation or glandular swelling. This includes non-sialolithiasis-related inflammatory and obstructive salivary gland diseases[14,33-38]. SE plays a dominant role in the management of obstructive sialadenitis associated with stenosis[22-26,39] and in post-traumatic duct laceration[27,40,41]. SE has also been used in combination with MR-sialography, in the evaluation of patients with post-traumatic sialoceles in the PG[42].

No articles describing the role of simultaneous use of US+SE (simUS+SE) in salivary gland diseases had been published. The aim of the present study was to examine the value of simUS+SE in several non-sialolithiasis-related pathological conditions of the major salivary glands. In addi-

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tion to ductal stenosis also duct anomalies/variations, ductal trauma, and unclear paraductal space-occupying lesions were addressed.

Patients and Methods

This retrospective study was carried out at the Department of Otorhinolaryngology, Head and Neck Surgery at the University of Erlangen–Nuremberg, Germany. Patients who presented between 2006 and 2018 for evaluation of salivary gland disorders were included. Approval for the study was obtained from the local institutional Review Board of Friedrich Alexander University of Erlangen–Nuremberg, and informed consent was obtained from all study participants. All patients first underwent a US examination of the major salivary glands (SMG, PG) and of the head and neck region using high-end ultrasound devices (Siemens ACUSON S2000 and S3000; Siemens Medical Solutions USA Inc., Malvern, PA, USA). Video documentation allowed better analysis and validation of the findings. Stimulation of glandular secretion with oral administration of vitamin C was used to enhance the findings in unclear gland swelling or ductal obstruction[43]. SE was performed using the Erlangen set of sialendoscopes (Karl Storz Company, Tuttlingen, Germany)[22,25,27,44]. Stenoses were treated as described previously[21,24]. The criteria for investigation by simUS+SE were pathologies that could not be assessed sufficiently with one method alone. SimUS+SE were used in unclear cases to establish the correct diagnosis, to support treatment decision-making, and/or to conduct the therapy. In cases with a diagnostic indication, one person was active, holding the sialendoscope in one hand and the ultrasound device in the other. If (interventional) therapy was carried out, mostly two persons were needed, with one holding the sialendoscope and working with the instruments and the other maneuvering the ultrasound transducer for navigation.

Statistical Analysis

Statistical analysis was performed using SPSS Statistics for Windows, version 22.0 (IBM, Armonk, NY, USA). Data are given as means plus or minus standard error of the mean.

Results

Forty-four patients were included; the mean age was 52.2±14.16 years (mean 54.5, range 24-76 y). The male-female ratio was 50% (22/44). The PG was affected in 90.9% (40/44) and the SMG in 9.1% (4/44). A total of 56 indications were investigated using simUS+SE, 10.7% in the SMG (6/56) and 89.3% in the PG (50/56; Table I). A single indication was investigated in 33 patients (PG 30, SMG 3), two indications in 10 patients (all PG), and three indications in one SMG.

Twelve patients presented with status post sialolithiasis or with simultaneous sialolithiasis (n=6

Table I. Simultaneous ultrasound and sialendoscopy in patients with stenosis and non–sialolithiasis-related salivary duct pathologies (n = 44).

Gland Indications – non sialolithiasis related	Both/Indications (n)	Submandibular Gl. Indications (n)	Parotid Gl. Indications (n)
Stenosis – total	38	2	36
Indication of supplemental imaging	2	1	1
Indication of therapy	36	1	35
successful (opening + dilation)	23	-----	23#
not successful (conservative, ablative)	13*	1*	12*
Management of complications	12	1*	11
After trauma/operative manipulation	8	1	7
with resulting stenosis	6*	1*	5*#
After infection (abscess) with	4	-----	4
with resulting stenosis	4*	-----	4*
Anatomic duct anomaly	3	2	1
Space occupying lesion next to duct system	3	1	2
Non-sialolithiasis – Indications total	56 (100%)	6 (10.7%)	50 (89.3%)
Non-sialolithiasis – Patients total	44 (100%)	4 (9.1%)	40 (90.9%)

*Indicates combined indications for 9 stenoses not successfully treated and for complications after trauma or abscess formation in 9 patients. #Indicates combined indications for 1 stenosis successfully treated and for complications after post-surgical trauma in one patient.

each). However, the indication for performing simUS+SE was not sialolithiasis. The main indications were high-grade stenoses in ten patients (all PG). In the remaining two an anomaly in the ductal system (SMG) and a sialocele with sialocutaneous fistula after an unsuccessful attempt to remove a stone by open surgery (PG) were diagnosed (Table I). Thirty-six patients with 38 indications were evaluated for stenosis, and US-guided and sialendoscopically controlled opening was achieved in 23 patients. All stenoses were complete and/or therapy-resistant. In two cases, the stenosis was associated with ductal anomalies such as kinking or megaduct. All stenoses could only be opened after simultaneous US was used to guide the maneuvering of the sialendoscope and instruments to avoid ductal perforation and/or paraductal manipulations (Table I, Figures 1A and B, 2, 3A and B). Further imaging was indicated in one patient. After dilation, sialography was performed in one case to obtain a closer view of the course of the duct behind the stenosis, which was in a nearly intraparenchymal location (PG). To decide whether or not a stenosis could be opened, US was used to assess its location within the ductal system and its length, and to measure the exact distance between the visible most distal ductal lumen and the oral mucosa in the distal duct or papilla stenosis[22,24]. In stenoses in the distal ductal system, simUS+SE were used to measure the distance between the position of the sialendoscope at the beginning of the stenotic area and the oral mucosa. If it was located to the middle or proximal duct, the exact position and length of the stenosis was calculated. This was in particular interesting, while sialendoscopic opening was attempted, to calculate the distance to the proximal end of the stenosis. If the proximal lumen of a distal stenosis was located > 1.5 cm from the oral mucosa or the length of a stenosis origin from the middle or more proximal located duct system was > 2 cm, with no visible lumen when interventional SE was tried, further minimally invasive therapy was abandoned and conservative/ablative therapy was indicated. Thirteen stenoses could not be opened or dilated; of these, 11 stenoses were accessible, but could not be opened using simUS+SE. Ten of these patients developed stenoses as complications after ductal trauma or abscess formation (Table I). Two stenoses were not sufficiently accessible and could therefore not be treated. One patient presented with an intraparenchymal stenosis several years after a mandibular fracture with laceration of the parenchyma of the SMG. MR-sialography was indicated to assess this inaccessible stenosis and the post-stenotic ductal system (Table I). Another patient developed a proximal/posthilar ductal stenosis several years after surgical repositioning of a mandibular fracture (PG). It was not possible to reach the stenosis with the instruments adequately, but dilation of the post-stenotic ductal system was observed after intensive irrigation, indicating that passage of saliva was possible. All but one of the stenoses accessible with SE showed heavy scarring extending over the duct-wall borders in some cases, were 2-3 cm long and complete with no visible residual lumen on SE (all PG). Five of these occurred after trauma to the duct, and a sialo-cutaneous fistula was visible in three. All of these patients presented at least several months after surgery in the cheek/parotid region, with partial to subtotal transection of the parotid duct. The ductal lumen was visible in one case, but the stenosis could not be passed adequately even with the smallest SE, and interventional SE was not possible due to a diffusely narrow ductal system. SimUS+SE allowed localization of the stenosis next to the residual fistula-channel and indicated that the proximal duct was open. Conservative treatment was therefore indicated. One patient developed a stenosis after surgical duct manipulation at the papilla and the distal duct system to extract a stone. A complete stenosis with a length >3 cm was diagnosed, and ablative therapy (i.e., Botox-injection) was recommended. Two additional patients developed complete fibrotic stenoses nearly 1-2 cm long in the middle and proximal ductal system, respectively. Neither could be opened with interventional SE. Four patients developed stenoses after abscess formation. One patient had septic granulomatosis and developed a parotideal abscess, and also presented with a visible scar on the cheek. Two patients presented months after spontaneous dislocation of a stone into the paraductal tissue and subsequent expulsion of it through the cheek with a visible scar on the cheek and/or intraorally. All of these patients had complete and long fibrotic stenoses with extension of the fibrotic tissue beyond the borders of the duct wall, which could not be opened. Twelve patients presented after complications due to ductal trauma or abscess formation, 10 of whom had stenoses. Six of eight patients after trauma and/or surgical manipulation had stenoses, and three of them also had fistulas (management as described above). Two patients who presented early after ductal laceration were treated successfully. One patient presented with a sialocele and sialocutaneous fistula after stone extraction was attempt-

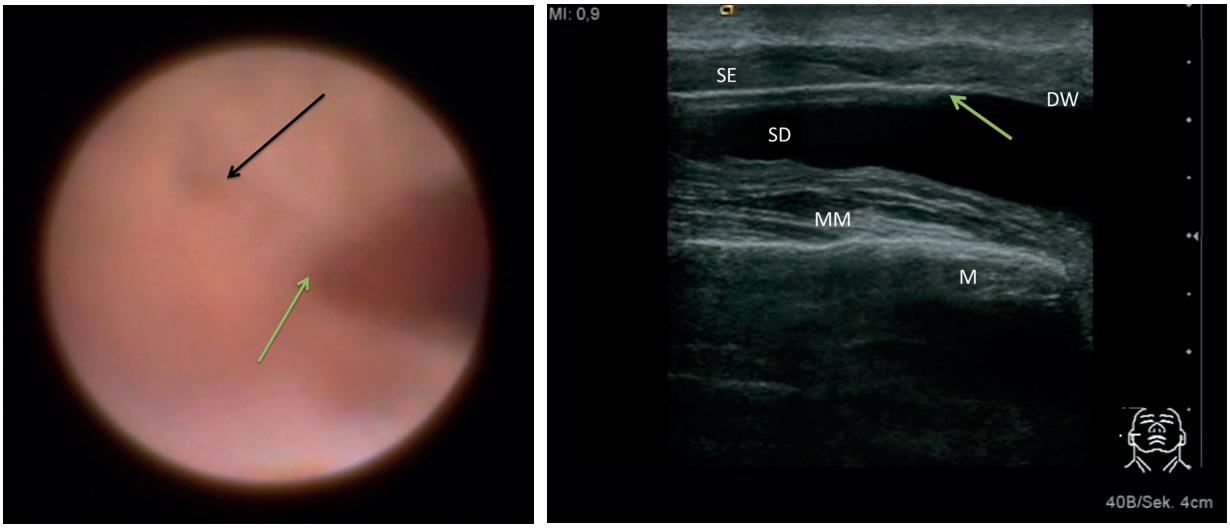

Figure 1. A, Sialendoscopic view to a complete stenosis (middle parotid duct): the micro-drill (*green arrow*) is positioned in front of the suspected residual lumen within the area of the stenosis (*black arrow*). **B,** Ultrasound shows the micro-drill emerging out of the working channel at the tip of the sialendoscope (*green arrow*) and positioned within the duct wall (false passage with the sialendoscope). Abbreviations: SD, Stensen's duct; DW, duct wall; M, mandible; MM, masseter muscle; SE, sialendoscope.

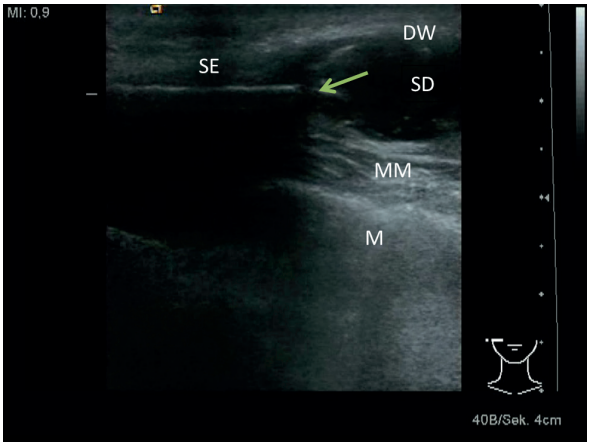

Figure 2. The situation after positioning of the sialendoscope onto the center of the fibrotic tissue within the stenotic area (*green arrow*) and before opening of the stenosis. Abbreviations: SD, Stensen's duct; DW, duct wall; M, mandible; MM, masseter muscle; SE, sialendoscope.

ed with transcervical sialodochotomy (PG). After removal of the stone, simUS+SE allowed detailed assessment of the sialocele and the ductal defect, which were treated using stent implantation. The second patient presented with swelling of the cheek 2 weeks after surgery in the cheek region. SimUS+SE revealed a sialocutaneous fistula due to a complete transection of the duct; the extent of the sialocele was measured and the distal and proximal duct ends and their relationship within the sialocele were assessed (Figures 4, 5 A and B). Ductal reconstruction was performed by open surgery. In both cases, assessment of the sialocele

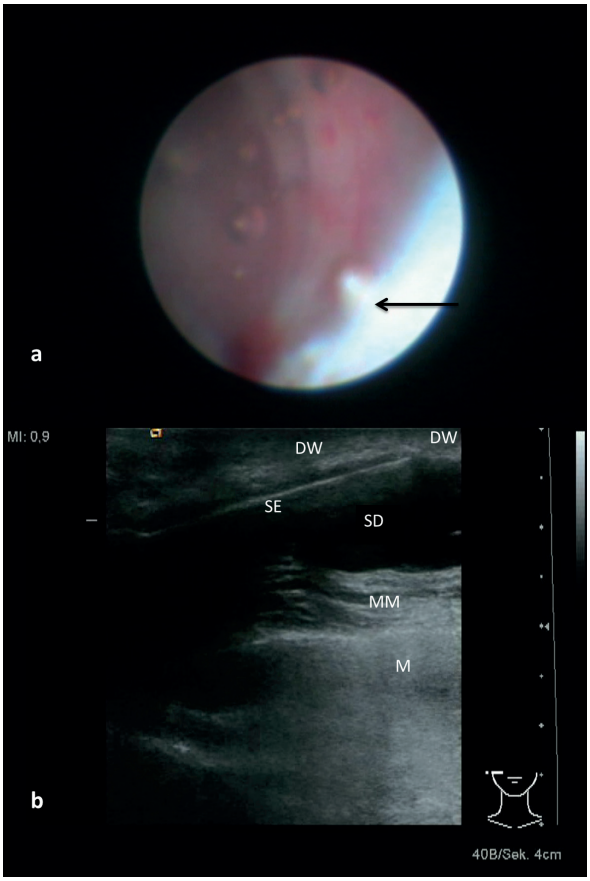

Figure 3. A-B, After opening of the stenosis, its fibrotic tissue (*black arrow*) and the proximal duct-lumen is visible with the sialendoscope (**a**), ultrasound-view shows the sialendoscope now within the ductal lumen (**b**). Abbreviations: SD, Stensen's duct; DW, duct wall; M, mandible; MM, masseter muscle; SE, sialendoscope.

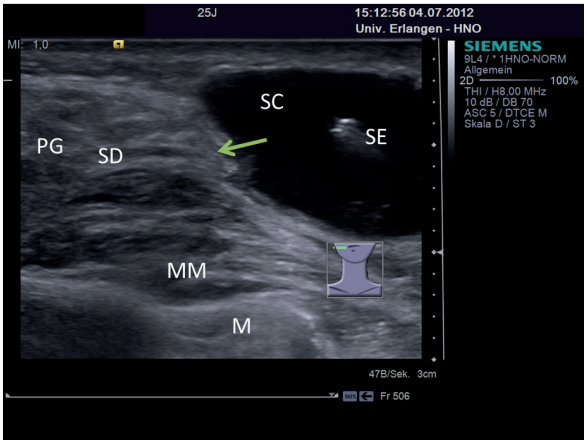

Figure 4. Ultrasound view shows the tip of the sialendoscope within a post-operative sialocele and in opposite to the proximal end of the Stensen's duct (*green arrow*), indicating a complete transection. Abbreviations: PG, parotid gland; SD, Stensen's duct; M, mandible; MM, masseter muscle; SC, sialocele; SE, sialendoscope.

and analysis of the ductal defect by simUS+SE influenced the treatment decision. For detailed data se Table 1.

Ductal anomalies and space-occupying lesions near the ductal system were found in three patients each. One of these patients had an aberrant Wharton's duct in addition to sialolithiasis in the dorsal region of the floor of the mouth. The connection to the gland was identified using simUS+SE, making it possible to adapt the subsequent treatment. Two of the patients had pelvis-like ductal anomalies in the hilar/posthilar duct system, one in the SMG (Figures 6-8) and one in the PG. No further therapy except for gland massage was recommended in these patients, due to the findings obtained with simUS+SE.

Three patients had unclear space-occupying lesions near the ductal system. In one of these patients, the lesion was located to the hilum of the SMG, its puncture allowed amylase to be measured in the lesion, suggesting the diagnosis of a sialocele and conservative therapy was indicated. In the two other patients, simUS+SE were used to rule out any connection to the main duct (both in the PG). In addition to this, the volume of the lesion did not increase after irrigation. The salivary cyst/sialocele was resected with preservation of the residual gland in both cases.

Discussion

The presented results show that simultaneous use of US+SE is of value in the management of several non-sialolithiasis-related salivary gland disorders. Important information for establishing the correct diagnosis was obtained. SimUS+SE enabled treatment to be planned and conducted more adequately: it was possible to avoid surgery or at least opt for less invasive therapy. In addition, more efficient indications for imaging were possible.

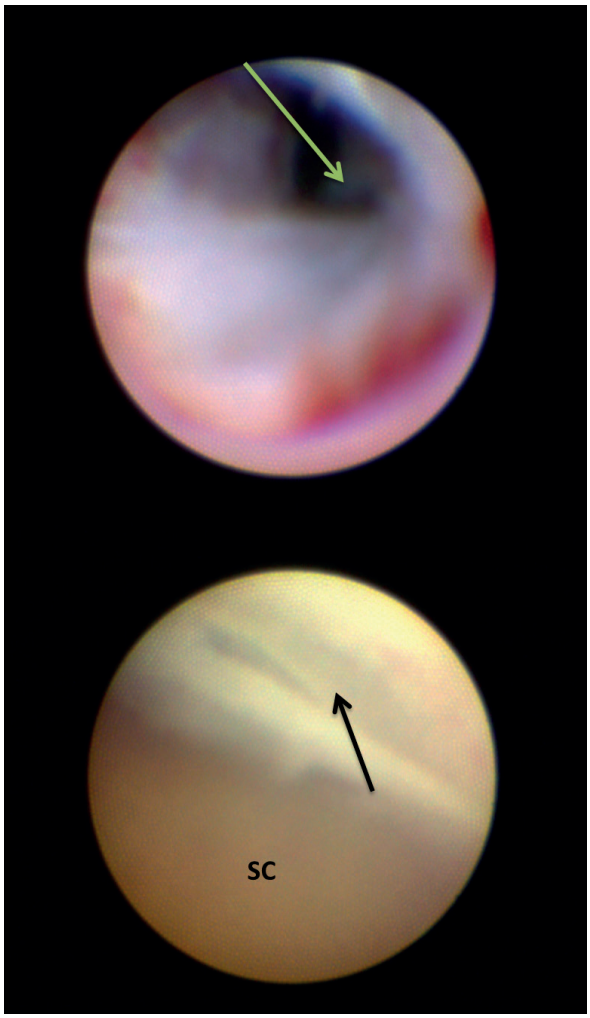

Figure 5. A-B, The simultaneous sialendoscopy provides a view to the end of the distal duct (green *arrow*, (**a**) and into the sialocele with the surrounding interstitial tissue (*black arrow*, **b**). Abbreviations: SC, sialocele;

US is widely used in various inflammatory and obstructive diseases of the major salivary glands[5,23], including stenoses with or without ductal anomalies[6,21,22,24-26,43,44] cases after ductal trauma[27]. It has also been used as an adjunct to or in combination with various other diagnostic tools[28,30,32,42]. US provides an overview of the ductal system and parenchyma from the outside, and the findings can be stored with video documentation.

SE is a less invasive technique that provides visualization of the ductal system, and video documentation is also standardized. Its value has also been demonstrated in many different types of salivary gland disease[14,22-27,33-41,45].

US and SE are routinely applied in all patients managed in our salivary gland center[21,22,24,25,27,44]. Management of the patients described in the present study was improved through simultaneous investigation with US+SE. SimUS+SE provide a unique overview of all the relevant structures and enable the examiner to interpret the findings more precisely. Anatomic structures, or instruments and their positioning within the tissues, can be continuously assessed with a high degree of precision.

Stenosis was the indication in 81.8% of the patients. US can provide information about the localization or position of stenoses within the ductal system. The grade can be estimated by measuring the diameter of the proximal duct and the distance can be calculated by measuring the distance from the proximal lumen to the oral mucosa[6,21,22,24-26,43,44]. SE has proved to be valuable for diagnosis, classification, and treatment of stenosis[21,22,24-26,33,39,44-46].

In distal complete ductal stenoses that cannot be treated using sialendoscopy-based methods alone, US can be used to measure the distance from the proximal duct lumen to the oral mucosa to assess the feasibility of transoral ductal surgery[22,24].

If no residual lumen is recognizable, stenoses from the papilla up to the proximal ductal system can be opened using US-guided SE, as was done in 63.9% of the patients with stenoses. The sialendoscope can be centered onto the fibrotic stenotic area under ultrasound guidance and then steered through the fibrosis into the proximal ductal lumen (Figure 1 A and B, 2, 3 A and B). US-guidance can compensate for the deficiencies of SE, and this clearly increases the feasibility of minimally-invasive treatment in such cases.

US-guided SE can indicate at an early point if successful treatment is not possible and conservative and/or ablative treatment should be recommended. This is the case if the distance between the visible lumen of the distal ductal system and the oral mucosa on US extends for 1.5 cm and/or if no residual lumen is identifiable within a high-grade stenosis longer than 2-3 cm in any part of the ductal system after insertion of the SE[22,24]. In the present study, of the stenoses caused by complications after ductal trauma or abscess formation, 20% were not accessible adequately and 70% not passable with the sialendoscope even with US guidance. Main reasons were heavy scarring with involvement of extraductal tissues and/or long-standing high-grade fibrotic stenoses with a length of >2-3 cm. If simUS+SE indicates that the stenosis is not adequately accessible and/or if the end of the stenosis cannot be visualized on SE due to its length or unfavorable ductal anatomy, then additional imaging may be indicated to visualize the stenosis and plan the further management. This was the procedure used in two patients.

There have only been a few published reports on the management of ductal trauma. Sialography and US were used in one case to diagnose a sialocele[28]. MR-sialography and SE were used in another case for noninvasive management of iatrogenic sialoceles[42]. In a further case report, US was used as the only diagnostic approach in the management of a complete transection of the duct with formation of a sialocele, in order to prepare for SE-assisted ductal repair[27]. There have been no reports on the simultaneous use of US+SE for this indication. In the present study, 75% of patients with ductal trauma presented at a later stage and all were treated conservatively or ablation of the gland was recommended. All patients who presented early were managed successfully, allowing early decision-making in favor of open surgery through precise analysis of the extent of the ductal damage, sialocele, and fistula (Figures 4, 5 A and B). The combination of intra- and extraductal findings obtained with simUS+SE provided more information about the extent of the ductal damage and influenced the subsequent management.

There have been also no published reports on the role of US in ductal anomalies or unclear space-occupying lesions. These conditions may be well assessed with US+SE. The vascular supply can be assessed using Doppler-ultrasound to differentiate the vessels, vascular malformations, benign semisolid tumors or cysts. Any connection to the ductal system can be detected directly or indirectly by measuring changes in the volume of the cavity after irrigation with SE. As a result, unnecessary imaging procedures and even surgery can be avoided.

Ductal anomalies may be associated with distinct types of stenosis[25] or may present as isolated findings. Aberrant ducts, duplication of the ductal system, or a pelvis-like anatomy in the ductal system (mostly within the hilar/posthilar area) can be investigated by US+SE, including video documentation. These conditions can be precisely characterized and the management can be planned more adequately (Figures 6-8). In the

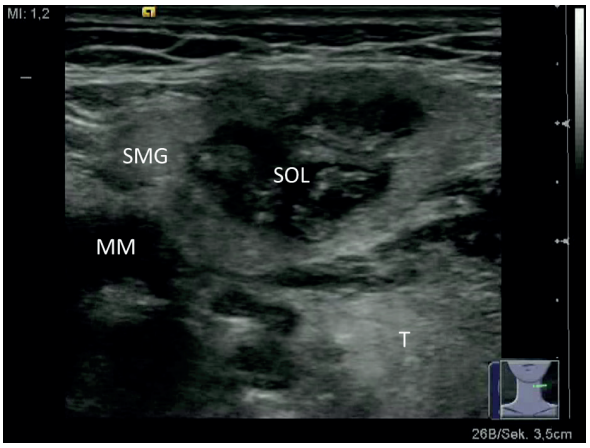

Figure 6. Ultrasound view, showing an unclear, echo-inhomogeneous space-occupying lesion (SOL) within the submandibular gland. Abbreviations: SMG, submandibular gland; MM, mylohyoid muscle; SOL, space-occupying lesion; T, tongue.

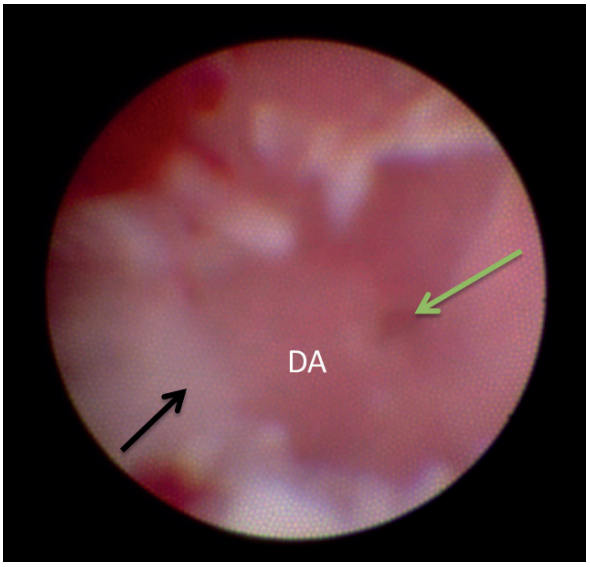

Figure 7. Sialendoscopic view into a cavity filled with fibrinous sludge (*black arrow*), corresponding to the SOL, which proved to a pelvis-like anomaly of the hilar ductal system (small ostium, *green arrow*). Abbreviations: DA, ductal anomaly.

present study, the patients were able to receive adequate counseling and unnecessary more invasive surgery was avoided in all cases.

Space-occupying lesions near the main ductal system may include isolated sialoceles with or without a connection to the ductal system, for example, or other cystic lesions of various origins. In one study, US/Color-Doppler-US and MR-sialography revealed a sporadic parotid sialocele in four cases, and the MR-sialographic findings were confirmed in the patients who underwent SE[31]. SimUS+SE can provide an impression of the relationship between an unclear space-occupying lesion and the main duct and may reveal a connection between these anatomic structures. This can be achieved through comparison of the echogenicity and size of the lesion before, during, and after SE with irrigation of the ductal system (Figures 6-8). It is clear that simUS+SE cannot replace imaging in all of these pathologies, but it may be sufficient for establishing a fast diagnosis in some cases. Particularly with simultaneous sialendoscopic findings, video documentation with ultrasound can provide valuable additional information for counseling the patient. This may allow adequate planning of any therapy, or may even change the treatment prospects.

Many of the diseases of salivary glands can be diagnosed using US or SE alone. However, it has been shown that SE can detect pathologies that are not visualized using other imaging tools, including US[33,45]. US and SE are dynamic real-time

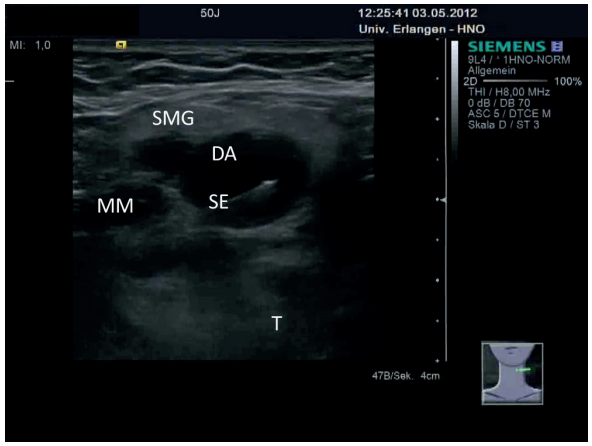

Figure 8. Ultrasound showing the sialendoscope within the ductal anomaly, which is now a hypo-echogenic cavity filled with fluid after irrigation and removal of the sludge. Abbreviations: DA, ductal anomaly; SMG, submandibular gland; MM, mylohyoid muscle; T, tongue; SE, sialendoscope.

methods that do not involve radiation exposure and can be carried out cost-effectively by clinicians/surgeons themselves. The three-dimensional complexity of findings can be assessed more adequately with simUS+SE and the physician's understanding of the pathology can be substantially improved, with an impact on diagnosis, management, and follow-up.

Conclusions

Simultaneous use of US+SE provided valuable additional information in comparison with the single examination tools in all of the patients included in this study. It proved to be valuable for establishing the diagnosis and for planning/conducting the treatment in non-sialolithiasis-related conditions such as stenoses, ductal trauma, ductal anomalies, and space-occupying paraductal lesions. Stenoses were the most important indication (> 80%). Beyond that, simUS+SE can provide a more efficient way of assessing the indication for supplementary imaging.

Conflict of Interests

The Authors declare that they have no conflict of interests.

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