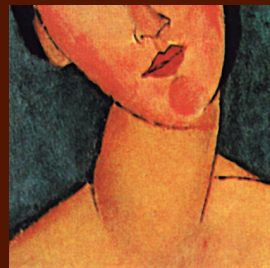


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ORIGINAL ARTICLE

Sialoendoscopy in the management of the salivary gland disorders

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ABSTRACT

BACKGROUND: The aim of this study was to determine the long-term (> 6 month) outcome of patients suffering from obstructive sialadenitis treated by sialoendoscopy.

METHODS: Between 2009 and 2013 seventy-six subjects underwent sialoendoscopy under local anesthesia at the University Hospital P. Giaccone of Palermo.

RESULTS: The mean age of presentation was 50.5 years for male (56.6%) and 44.7 years for female (43.4%). The presenting symptoms were frequent pain in 72% of cases, frequent purulent secretions in 65% of cases, frequent swelling in 62% of cases and frequent bitter taste in 44% of cases. Affected glands were in 54 cases the submandibular unilaterally, in 21 cases the parotid unilaterally and in 1 case the submandibular bilaterally. Preoperative ultrasound showed in 30 cases a sialolithiasis and in 19 cases a ductal stenosis, in 3 cases were both associated. In 46 cases showed a ductal dilatation, variably combined with the previous two and in 13 cases ultrasound gave no indication. Average operating time was 60.7 minutes (range 35-125 min). In 35 cases it was necessary to perform a papillotomies. Presence of stones was shown in 36 patients, in 3 cases these stones were multiple. Endoscopic removal of stones was achieved in 72.2%, with a mean size of 3.6 mm (range 0.7-5.5 mm), only two were larger than 5 mm. In five of this cases was performed a trans-oral combined approach. In 15 cases was highlighted a ductal stenosis, in 6 cases associated with mucus plugs, that were treated with daily irrigations with Thiamphenicol glycinate acetylcysteinate. A percentage of 64.8% of patients had complete resolution of symptoms after sialoendoscopy, with 25% having partial resolution of symptoms and 6.6% required the removal of the gland.

CONCLUSIONS: Sialoendoscopy is the procedure of choice for the diagnosis and treatment of chronic non-lithiasic sialoadenitis, but unlike its apparent simplicity has a significant learning curve, and need appropriate selection of patients.

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Key words: Endoscopy - Salivary gland calculi - Sialadenitis.

Obstructive sialadenitis occurs with acute, painful enlargement of the major salivary gland, usually during mealtime. The most common etiology is calculi within the excretory ducts, which are seen in about 66% of obstructive sialadenitis,¹ representing 50% of major salivary gland diseases.² Lithiasic sialoadenitis affects 1.2% of the general population,³ is more frequent in male,⁴ with a peak of incidence between 30 and 60 years of age.⁵ In 80-90% of cases the submandibular gland is affected⁶ and usually unilaterally.⁴ Strictures, mucoid debris, anatomic ductal abnormalities, scar tissue and foreign bodies are other causes of salivary duct

obstruction. Imaging for the salivary glands is mainly represented by ultrasound,⁷ computed tomography (CT)⁸ and magnetic resonance imaging (MRI).⁹ These are non-invasive techniques with a good accuracy in diagnosing sialolithiasis, but they are limited in their ability to detect non calcified sialoliths, mucus plugs and stenoses. Sialography is still considered one of the most reliable imaging techniques in the diagnosis of non-lithiasic obstruction, but it is invasive and does not show regional anatomy. In contrast to these imaging techniques, purely diagnostic sialoendoscopy allows, in addition to an accurate diagnosis of the place and nature



Figure 1.—Progressive dilatation of Wharton duct with lacrimal probe and conic dilator.

of the obstruction, and a minimally invasive therapeutic approach that in most cases is crucial. Our objective is to review our experiences, compare our outcomes to other groups, and to evaluate technical limitations and ways in which these may be overcome.

Material and methods

We performed a retrospective study evaluating 76 patients with symptoms of obstructive sialadenitis who underwent sialoendoscopy, during a period of 4 years. All the procedures were performed at the University Hospital P. Giaccone of Palermo and were approved by the committee on research ethics.

All patients signed an informed consent, in conformity to the Italian law; respect of individual privacy concerning clinical data was guaranteed.

We collected epidemiologic data, patient age and gender. Clinical data were collected including presenting symptoms, location, recurrence of symptoms and aspects of the patient's medical history that could be relevant to the etiology of the obstructive symptoms. All patients were subjected to preoperative ultrasound

evaluation. In 10 cases where the location of the sialoliths and their relationships with the surrounding structures were not clear even a CT was performed, and in 11 patients in which there was a suspicion of a non-lithiasic obstruction was performed a sialography. Patients with acute sialadenitis, diffuse atresia or stenosis of the main duct, and those with inadequate oral exposure were excluded. The endoscopies were done under local anesthesia. A progressive dilatation of the papilla was performed with lacrimal probes and conic dilator (Figure 1). In some cases it was necessary to perform a papillotomy because of inadequate dilation of the orifice to introduce the sialoendoscope or the excessive size of the stone that could not pass through the papilla (Figure 2). This was usually performed with a radiofrequency scalpel with an extension not greater than 5 mm. The procedures were performed with an all-in-one 1.3 mm Marchal sialendoscope (Karl Storz, Tuttlingen, Germany), which allows the duct exploration and idrodissection through an irrigation port. The irrigation solution was made up to 2 mL of 2% lidocaine and 100 mg of hydrocortisone in 40 mL of saline solution. This maximum volume has been decided to avoid

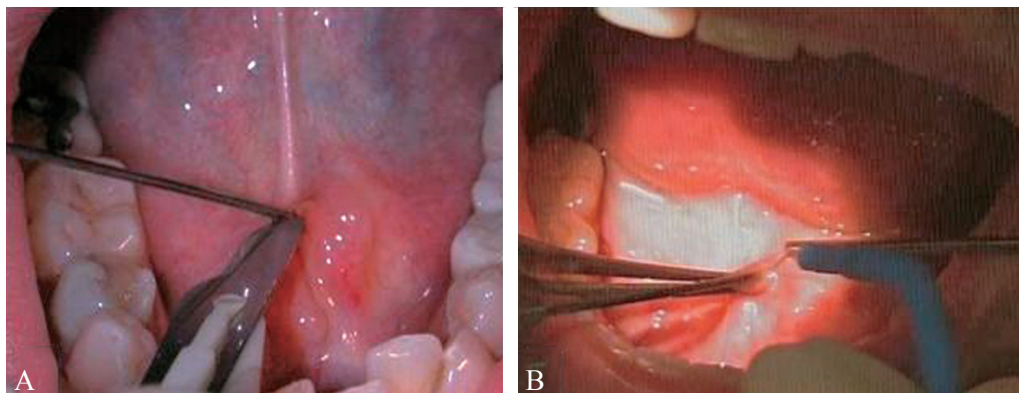


Figure 2.—Papillotomy. A) With cold scalpel; B) with radiofrequency scalpel.

excessive swelling of the gland. The sialoliths were removed with wire baskets introduced through the operating channel, in case of stenosis a mechanical dilation was made through the passage of the sialoendoscope. In case of stones too large to be removed entire, a mechanical fragmentation was attempted with endoscopic forceps and subsequent removal of the fragments with wire basket. Postoperatively patients were counseled to stay well hydrated, use sialogogues and massage their affected salivary gland. In patients in whom it was highlighted the presence of mucus plugs during endoscopy was made postoperative intraductal daily irrigations for six consecutive days to prevent the reforming of the mucus plugs. This was made after cannulation of the duct with a 24-G angiocatheter with a solution of 500 mg of Thiamphenicol-glycinate-acetylcysteinate (TGA) in 4 mL of physiological solution. Antibiotic (Rovamycine) was given prophylactically only in patients that reports gland infection with discharge of pus from the duct. Follow-up in our series was done for a minimum of six months. If patients did not return for clinical evaluation, telephone follow-ups were used to inquire about persistent symptoms or complications and whether any further procedure was indicated or performed.

Results

From January 2009 to January 2013, 76 cases of obstructive sialadenitis were treated with sialoendoscopy surgery at the Department of Head & Neck Surgery, University Hospital P. Giaccone of Palermo. Forty-three patients were (56.6%) males, with an average age of 50.5

years (ranging from 19 to 77), and 33 (43.4%) were females, with an average age of 44.7 years (ranging from 20 to 72). The collection of clinical presentation (Figure 3) has shown frequent pain in 72% of cases, frequent

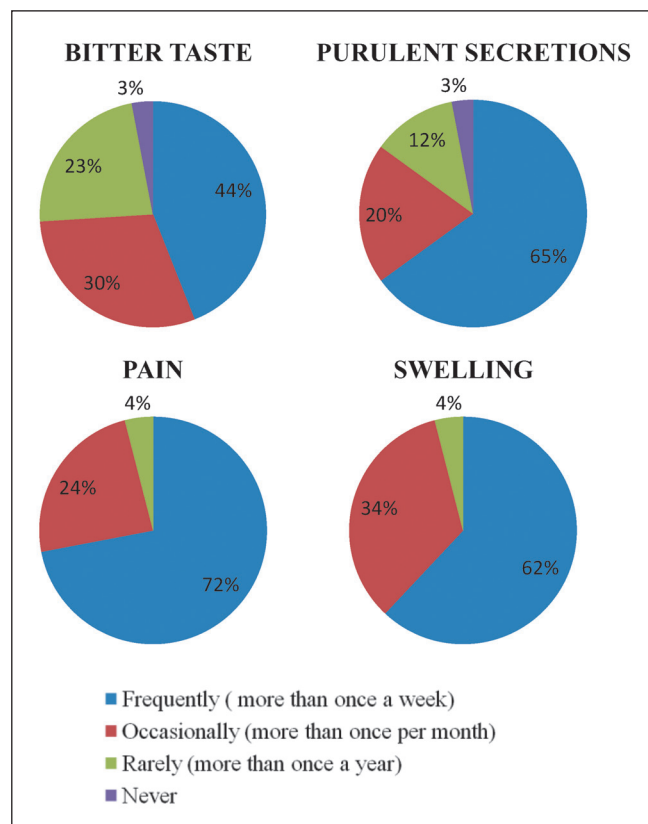


Figure 3.—Clinical presentation.

TABLE I.—*Stone characteristics.*

	N (%)	Mean size (mm)	Range (mm)
Method of removal			
All patients	36 (100)	4.6	0.7-12
Endoscopic removal	26 (72)	3.6	0.7-6.5
Combined removal	5 (14)	6.9	6-12
Failure of removal	5 (14)	5.6	4-6
Location			
Submandibular	26	4.84	2.5-12
Ductal	20 (77)	3.8	2.5-7.5
Hilar	3 (11.5)	6.5	6-7
Hilar - glandular	3 (11.5)	10	9-12
Parotid	10	7.7	4-11
Ductal	7 (70)	7.14	4-11
Hilar - glandular	3 (30)	9	8-10

purulent secretions in 65% of cases, frequent swelling in 62% of cases and frequent bitter taste in mouth in 44% of cases. Affected glands were in 54 cases submandibular unilaterally, in 21 cases parotid unilaterally and in 1 case submandibular bilaterally. The preoperative workup included ultrasound that showed in 30 cases an obstruction due to sialoliths and in 19 cases a ductal stenosis, in three cases both were present. In 46 cases was shown a ductal dilatation variably combined with sialoliths and ductal stenosis. In 13 cases ultrasound gave no indication. In this cases CT confirmed the presence of calculi at the level of salivary duct in seven cases, while in three cases showed ductal dilatation without other evidence of radiopaque stones. Sialography performed in 11 patients, showed the presence of salivary duct stenosis in nine cases and in three of these showed the presence of a post-stenotic obstruction of probable lithiasic nature. Sialoendoscopy was proposed to 81 patients, in 5 cases it was not possible to cannulate the salivary duct due to pre-existing ductal stenosis that made a tortuous course of the duct avoiding the passage of the probes. This patients were excluded from the study and a sialoadenectomy was therefore performed. The average sialoendoscopy operating time was 60.7 minutes, ranging from 35 minutes to 125 minutes. In 35 cases it was necessary to perform a papillotomy with a radiofrequency scalpel (30 submandibular, 5 parotid), in 26 cases due to insufficient dilatation of the papilla to introduce the sialoendoscope and in 9 cases to allow the extraction of a stone too big to pass through the papilla. In four patients in the Stensen's duct were highlighted micro-sialolith of diameter less than 1 mm which are escaped from the duct only with irrigation and massage

of the gland. Sialoendoscopy showed the presence of stones as a cause of obstruction in 36 (47.37%) procedures (Table I), in 3 cases, these stones were multiple. In 26 (72.2%) cases the stones were successful removed endoscopically, 21 submandibular and 5 parotid sialolith. All the removed stones have a maximum diameter less than 5 mm, only in two cases it was possible to remove entire stones larger than 5 mm because they were located distally and were smooth and movable (5.5 mm by 6.5 mm from the parotid and submandibular). In two cases of parotid sialolithiasis with stones >10 mm it was possible to crush them with microforceps and then remove the fragments with wire baskets. In 10 (26.81%) cases it was not possible to remove the stones since they were larger than 5 mm and we could not crush them with microforceps. In five of this cases in which the stone was at the level of the duct it has been possible to perform a trans-oral combined approach. In three cases the stones were at the level of the Wharton's duct and incision and dissection of the duct was performed at the level of the floor of the mouth using the sialoendoscope introduced into the duct as a guide. After removing the stones, a sialoendoscopy was performed through the breach in the duct, which showed no residual sialolith but only mucous plugs, then the duct was marsupialized. The same procedure was applied to the mucosa of the cheek in two cases of large intraductal stones of the parotid gland. In the other five patients the stone was located at a deep level in the gland, 2 submandibular and 3 parotid stones, and therefore it was not possible to remove it with this approach. In fifteen endoscopies a duct stenosis was highlighted, seven cases in the parotid and eight in submandibular gland, in six of these the Wharton's stenosis was associated with the presence of mucus plugs. In a case at the submandibular level and in two cases at parotid level the stenosis was associated with the presence of stones. In the stenosis a mechanical dilation through the passage of the sialoendoscope was attempted. As regards those associated with the presence of mucus plugs, post-operative daily intraductal irrigation was made for six consecutive days, with a solution of 500 mg of TGA in 4 mL of physiological solution. Overall, the presence of mucus plugs has been highlighted in nineteen cases and the same irrigation was practiced. In ten cases were evidenced in Wharton's duct, six in association with stones, while in Stensen's duct mucous plugs were associated with stones in five

cases of nine. In one case in Wharton's duct an intraductal evagination was highlighted. A normal aspect of the ductal system was shown in four cases in the submandibular glands and in six cases in the parotid glands. Antibiotic therapy was prescribed only in two cases due to the intraoperative evidence of intraductal pus with rovamycine for six days. There were no complications such as injury of lingual nerve, infection or perforation of the duct, ranulas, strictures, paresthesia of the tip of the tongue. In the days following the procedure, in three patients in whom it was not possible to remove the large stone there was the recurrence of swelling associated with pain and purulent discharge from the duct, therefore we proceeded to sub-mandibular gland excision. In the other two cases where it was not possible to remove the sialoliths because of the deep level, respectively, in Wharton's and in Stensen's duct.

The average follow-up was 13.2 months, ranging from 6 to 48 months. In a patient who underwent removal of the stone from the Wharton's duct with a combined approach from the floor of the mouth, a year after there was the appearance of a tight stenosis of the duct associated with recurrent symptoms, therefore it was necessary to proceed with the removal of the gland. The thirty patients who underwent removal of stones reported the full and final remission of symptoms. In the 40 cases of non-lithiasic sialoadenitis, during the follow-up in 21 cases (52.5%) there was a complete remission of symptoms after the first treatment. In 14 patients (35%) there has been a modest recurrence of symptoms within the first year, but significantly lower than the preoperative. The symptoms were characterized by intermittent episodes of moderate swelling associated with mild pain, therefore the patients did not wish for further intervention. In 3 cases (7.5%) who had a ductal strictures, there was recurrence of symptoms within the first month; these patients were treated with a series of six days of intraductal irrigation with a solution of 500 mg of TGA in 4 mL of physiological solution, achieving a relief of symptoms. In 2 patients (5%) with diffuse stenosis of the Wharton's duct the recurrence of symptoms occurred within the first week and it was opted for excision of the submandibular gland.

The overall results showed 64.8 % of patients had complete resolution of symptoms after sialendoscopy, with 25% having partial resolution of symptoms and 6,6% requiring the removal of the gland.

Discussion

This technique was introduced for the first time in 1991 by Katz,¹⁰ but it has begun to emerge only in the last years with the improvement and further miniaturization of endoscopic technologies. In our institution we perform sialoendoscopy since 2005, but despite its apparent simplicity it has a significant learning curve and became fully operational only in recent years. Identification and dilatation of the papilla in our experience was often difficult, especially at the level of the submandibular gland, whose papilla is very soft. Furthermore, it was often necessary to practice a papillotomy even after the identification of the duct due to an insufficient dilatation not allowing the introduction of the endoscope. We performed 35 papillotomies and we did not reported post-surgical symptomatic stenosis, according with the low rate of this complication reported in literature.^{11, 12} We reported 5 cases (6.17%) of failing to canalize the duct. This rate of failure in channeling the duct is consistent with what has been shown by other groups in which the frequency of failures ranged from 3.2 to 10.9%.^{11, 13, 14} The average duration of procedures (60.7 minutes) was in line with the statement made by Marchal *et al.*¹ The increased duration of the procedure was influenced mainly by two factors: 1) the difficulty of finding and cannulation of the papilla; 2) the presence of stones, which has required longer procedures.

We have highlighted the diagnostic potential of sialoendoscopy in fact preoperative imaging has found a good correspondence with the endoscopic examination, but in some cases this was not sufficient to predict the type and the site of obstruction. In 6 patients salivary stones have not been highlighted with the preoperative ultrasound, these calculi were smaller than 1.5 mm, soft and slightly mineralized. In addition, the ultrasound showed poor diagnostic potential with both mucous plugs and ductal stenosis.

In our series 86.8% had an immediate improvement in their symptoms after sialoendoscopy. Other groups reported a resolution of symptoms after sialoendoscopy between 83 and 89%.^{1, 11, 14} Patients who undergone successfully the removal of the stones were those with long lasting resolution of symptoms, while in patients with not lithiasic sialadenitis the resolution was in most cases temporary, especially in patients with ductal strictures, although positively influenced by the application of intraductal irrigation with TGA.

In 72.2% (Table I) of cases the stones were successfully removed endoscopically in line with other groups who have had a success rate of between 71 and 90%.¹²⁻¹⁵

For the endoscopic removal of the calculi was fundamental their size, in particular the transverse diameter, that is the one not oriented in the sense of the duct, which affects mainly the extraction.

In case of fragments of less than 1 mm it was possible to remove them only through intraductal irrigation and gland massage. All stones removed endoscopically had the transverse diameter of less than 5 mm, only in two cases we were able to remove endoscopically stones of larger diameter. In one case it was a sialolith of 5.5 mm in the Stensen's duct and in the second case it was a 6.5 mm sialoliths in the Wharton's duct. In both cases these were located distally, appeared to be mobile, oval, free of adhesions and with the greater diameter of the stone oriented parallel to the course of the duct in line with the prognostic factors proposed by Luers *et al.*¹⁶ In the two cases where we crushed large sialolith through the use of microforceps, they were very friable, with irregular surfaces on which it was easy to grip. In the remaining cases, the stones had smooth and compact surface and the consistency was hard, making impossible their crush with microforceps.

There are several techniques for crushing the sialolith, these can be divided into extracorporeal, that use electromagnetic¹⁷⁻¹⁸ or piezoelectric shockwave,¹⁹ and intracorporeal as fragmentation with micro-drills, pneumatic²⁰ and laser-assisted fragmentation,^{1, 21} which is currently the most used. Certainly the aid of these techniques would help in the removal of larger sialoliths, but they are expensive and not always available.

In our series stones with a transverse diameter greater than 5 mm, especially if localized at distal level, were successfully removed through a combined trans-oral approach that results simple and decisive. The stones of large size results localized to hilar and glandular level and for these it was not possible to perform endoscopic removal, but in some cases it was still reported an improvement of symptoms after the procedure, probably due to the removal of the part of obstruction due to the accumulation of mucous plugs.

Patients who did not have any improvement from the procedure were subjected to sialoadenoidectomy, in three cases due to the persistence of symptoms related

to the presence of large stones at the level of the submandibular gland and the other two for the presence of diffuse stenosis at the level of Wharton's duct, which have not undergone improvements with scialoendoscopy. In two cases where it was not possible to remove the sialoliths because of the deep level, the patients reported mild symptoms for about six month and then complete relief, likely to the atrophy of the gland.

One of the patients who underwent removal of a submandibular calculus through trans-oral combined approach reported the occurrence of recurring obstructive symptoms and one year after the procedure has been subjected to removal of the submandibular gland due to a ductal stenosis, that is compatible with the frequencies reported in literature.^{1, 12} Probably postsurgical stenosis may be avoided using salivary stent after ductotomy as recommended by some authors.²² Furthermore, we believe that the combined trans-oral approach is a viable alternative to sialoadenectomy in cases of large stones located in the proximal salivary ducts, having also a lower risk of serious complications as evidenced by Capaccio *et al.*²³ In our series we did not reported any intraoperative complication.

Among the 66 patients experiencing improvement after sialoendoscopy, three patients with stenosis of the ducts have reported modest recurrence of symptoms during the first month after the procedure. Therefore were subjected to daily intraductal irrigation for six consecutive days with a solution of 500 mg of TGA in 4 mL of physiological solution, which led to complete resolution of the symptoms. These recurrences were probably related to the formation of mucus plugs within the ducts causing obstruction of salivary flow at the strictures. The TGA was administered intraductally with the intention of degrading such accumulations of mucus. It has a dual action, the dissolution of the disulfide bonds of the mucus by acetylcysteine and the antibiotic action of thiamphenicol on bacteria that may form at this level a microfilm stimulating the aggregation of mucous plugs and acting as a substrate for the aggregation of salivary calculi, as suggested by Teymoortash *et al.*²⁴

Fourteen of the 40 patients with non-lithiasic obstructive sialoadenitis no longer came to control, at the telephone interview reported a slight recurrence of symptoms after a few months, but due to the weaknesses of symptoms they did not consider necessary to be sub-

jected to other procedures, consequently we have considered them cured.

All patients in the immediate postoperative reported the swelling of the gland subjected to the procedure. This swelling is due to intraductal irrigation and disappeared in all cases spontaneously through the drain of the gland from a few minutes to a few hours after the end of the procedure. In our series we did not have other complications reported in the literature as the creation of false passages (4.5%), temporary lingual nerve paresthesia (0.4 to 2.1%), wire basket blockages (6%), perforation of the duct (0.3-6%), ranula (1%), postoperative infections (2%). But anyway, these reported complications are much less serious and rare than those related to sialoadenectomy.

Conclusions

Sialendoscopy is the procedure of choice for the diagnosis and treatment of chronic obstructive sialadenitis, but unlike its apparent simplicity it has a significant learning curve. In our study, in 93.4% of cases of chronic obstructive sialadenitis it has allowed to save the salivary gland, provided that they are applied appropriate criteria for selection of patients, especially in the case of evidence of salivary calculi. Moreover, in patients with non-lithiasic sialadenitis, that is often due to ductal stenosis, the association of mucous plugs can cause obstructive symptoms by altering what appears to be a delicate balance. So the mechanical action of intraductal irrigation, associated with the mucolytic and antibiotic action of TGA, is often enough to return the gland to a functional equilibrium with the disappearance of the symptoms.

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Conflicts of interest.—The authors certify that there is no conflict of interest with any financial organization regarding the material discussed in the manuscript.
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