SUBMUNDIBULAR DIAGNOSTIC AND INTERVENTIONAL SIALENDOSCOPY: NEW PROCEDURE FOR DUCTAL DISORDERS

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We present our initial experience with submandibular sialendoscopy, a new therapeutic approach for disorders of Wharton’s duct. We review the sialendoscopes used and discuss their respective merits. We evaluated and treated 129 consecutive patients with suspected ductal disorders. Diagnostic sialendoscopy was used for classifying ductal lesions as sialolithiasis, stenosis, sialodochitis, or polyps. Interventional sialendoscopy was used to treat these disorders. The type of endoscope used, the type of sialolith fragmentation and/or extraction device used, the total number of procedures, the type of anesthesia, and the number and size of the sialoliths removed were the dependent variables. The outcome variable was the endoscopic clearing of the ductal tree and resolution of symptoms. Diagnostic sialendoscopy was possible in 131 of 135 glands (97%), with an average (±SD) duration of 28 ± 15 minutes. Interventional sialendoscopy was attempted in 110 cases, with an average duration of 71 ± 41 minutes, with a success rate of 82%. Multiple sialendoscopies were necessary in 25% of cases. General anesthesia was used in 12% of cases. Submandibular gland resection was performed in 4%. The average size of the stones was 4.9 ± 2.9 mm. Multiple sialoliths were found in 31 cases (29%). Sialolith fragmentation was required in 26%. Larger and multiple stones often required longer and multiple procedures and general anesthesia, and more often resulted in failures. Semirigid endoscopes had a higher success rate (85%) than flexible sialendoscopes (54%). Complications were mostly minor, but were encountered in 10% of cases. Diagnostic sialendoscopy is a new technique for evaluating salivary duct disorders that is associated with low morbidity. Interventional sialendoscopy allows the extraction of sialoliths in most patients, thus preventing open gland excision.

KEY WORDS — salivary duct, sialendoscopy, sialolith, Wharton’s duct.

INTRODUCTION

Salivary gland disorders are traditionally divided into neoplastic and non-neoplastic disorders, the latter being further subdivided into inflammatory and noninflammatory ones. The advent of new endoscopic techniques allows a complete exploration of the salivary ductal system and a precise evaluation of its disorders. Therefore, the present study supports the division of non-neoplastic salivary gland diseases into parenchymal ones, which require traditional treatments, and ductal ones, which can be handled endoscopically in the majority of cases.

Most salivary ductal disorders are obstructive, with sialolithiasis being the most frequent cause, although its exact incidence is unknown. Treatment of distal submandibular sialolithiasis has been performed for many years by simple anterior marsupialization (sialodochotomy and/or sialodochoplasty) of Wharton’s duct, which allows the direct removal of the stone. The classic approach to proximal sialolithiasis is antibiotic and anti-inflammatory treatment, in hope of a spontaneous stone extrusion through the papilla. Recurrent episodes of sialadenitis are seen as an indication for open submandibular gland extirpation, although the precise indications remain ill defined. Interestingly, sialolithiasis is the most frequent reason for submandibular gland resection.

Sialendoscopy is a new procedure for visualizing the lumen of the salivary ducts, as well as diagnosing and treating ductal diseases. Because of the required equipment and the complexity, the duration, and the potential complications of the procedure, it appears important to distinguish two different procedures: diagnostic sialendoscopy and interventional sialendoscopy. Diagnostic sialendoscopy is an evaluation procedure, while interventional sialendoscopy must be considered as an operation on the obstructive ductal disease.

We report on the sialendoscopic evaluation and treatment of 129 successive patients with suspected ductal disease, performed by the first author in 2 university centers (Geneva University Hospital, Geneva, Switzerland, and Edouard Herriot Hospital, Claude Bernard University, Lyon, France).
MATERIALS AND METHODS

Patients. Between November 1995 and March 2000, 129 consecutive patients with suspected submandibular duct dysfunction were evaluated. The average (±SD) age was 39 ± 16 years, with a range of 6 to 93 years. Six of the patients had bilateral symptoms, so 135 primary sialendoscopies were attempted.

Most of the patients had preoperative radiologic evaluation studies such as sialography, ultrasonography, magnetic resonance sialography, and, rarely, computed tomography.

The observed disorders of Wharton’s duct were classified as sialolithiasis, stenosis, sialodochitis, and ductal polyps. Each pathological finding in each ductal system was noted. In cases of multiple stones, the largest stone was measured with calipers.

The numbers of diagnostic and interventional procedures per gland were recorded, as well as the type of anesthesia (local versus general), the fragmentation and extraction device used (see below), and the number of open submandibular gland resections performed. “Sialendoscopic success” was recorded when the entire Wharton’s duct and its primary branches were rendered free of disease. “Sialendoscopic failures” were recorded when sialendoscopy was impossible or unsuccessful, or when an open submandibular gland resection was performed.

The pain experienced during the procedure by the last 80 patients of this series was evaluated with a 10-cm visual analog scale.

Statistical analysis for categorical and ordinal variables was performed with the $\chi^2$ test, and continuous variables were analyzed with the t-test (2 groups) or the Kruskal-Wallis test (3 groups) as implemented by SPSS software (version 9.0, Chicago, Illinois).

Endoscopes. The technology of the endoscopes we used evolved in 4 steps: free optical fiber, flexible endoscope, and 2 generations of semirigid endoscopic devices of various diameters.

1. Initial trials were performed with free optical fibers, ranging from 0.5 to 0.8 mm, without a rinsing or a directional system. When visualized, the stones were extracted blindly with commercially available grasping baskets.

2. The second group of endoscopes used were flexible, consisting of a 1.5-mm–outside diameter flexible endoscope and a 0.5-mm working channel. These flexible endoscopes had a directional system, and in 1996, we began to use a rinsing solution.

3. The third system included 2 devices: a semirigid single-lumen device (external diameter of 1.3 mm) for diagnostic endoscopy, and a double-lumen device for interventional procedures. In the latter, one channel was 1.1 mm and the other was 0.8 mm, the total surface being $2.67 \text{ mm}^2$. In both instruments, a 1-mm semirigid endoscope (Karl Storz AG) was secured for visualization in one of the channels. The second channel of the double-lumen device was a working channel used to pass different instruments for sialolith fragmentation and/or retrieval.

4. The final endoscopic system also includes 2 different devices (Karl Storz AG). The Marchal diagnostic sialendoscope resembles the single-lumen diagnostic device described above, with slight modifications (Fig 1A). The outside diameter is 1.3 mm and the cross-sectional area is $1.33 \text{ mm}^2$. The Marchal interventional sialendoscope is a double-lumen device, with one channel of 1.1 mm for the endoscope, and a working channel of 0.8 mm, which is used for custom-made baskets and/or laser fibers (Fig 1B). The overall cross-sectional area is $2.29 \text{ mm}^2$. The tip of the instrument has been beveled and blunted for easier cannulation of the duct. Both instruments were slightly bent to facilitate exploration of the ductal tree.

Other Materials. Other materials include a custom papilla dilator and customized grasping wire baskets (Karl Storz AG; Fig 1C). Fragmentation of larger stones was achieved in this study with the following devices: an electrohydraulic lithotriptor with a 500-µm probe (Calcutrit, Karl Storz AG) and a 400-µm holmium laser probe (Coherent, Versapulse Select, Santa Clara, California).

Surgical Technique. Details of the surgical technique have been previously described. The procedure is done in most cases under local anesthesia. Local anesthesia is achieved by contact anesthesia (10% Xylocaine-soaked pledges) followed by infiltration of the tip of the papilla. In addition, intermittent rinsing through the endoscope is achieved with a local anesthetic solution (1:1 Xylocaine 2% and sodium chloride 0.9%) that provides slight dilation of the duct, cleansing of the endoscope tip, and removal of pus, debris, and occasionally blood.

Dilation of the papilla is performed with customized salivary dilators of increasing diameter, followed by the papilla dilator. Earlier, large sialodochoplasties of the papilla were performed, and the edges of the Wharton’s duct were sutured to the surrounding floor of mouth mucosa. For the past 2 years, simple dilation of the Wharton’s duct papilla is performed in the majority of cases, with rare patients requiring a mini-papillotomy (<5 mm).

The initial procedure is diagnostic sialendoscopy, allowing a minimally invasive but complete explo-
ration of the ductal system. When a stone or another ductal disorder is located, an interventional sialendoscopy is planned. For sialoliths smaller than 5 mm, it is performed during the same stage, with the interventional sialendoscope. The custom wire basket is passed behind the stone and deployed, the stone is trapped (Fig 2), and the whole sialendoscope is removed. For larger stones, fragmentation is required before extraction. As the use of the Calcutript and the use of the holmium laser under local anesthesia have proven to be painful, some sialolithotripsy were performed under general anesthesia. After the last stone is removed, the endoscope is introduced one last time to rinse the duct and verify its integrity.

The intervention is done under antibiotic prophylaxis. Oral antibiotics (amoxicillin–clavulanic acid or clindamycin) and corticosteroids (prednisone 50 mg/d) are administered for 48 hours. Frequent self-massages of the gland are recommended. A follow-up examination is performed 10 days after the procedure.

RESULTS

Diagnostic sialendoscopy was attempted in 135 submandibular glands suspected to have an obstructive process of the Wharton’s duct. Diagnostic sialendoscopy was possible in 131 ducts (97%); the 4 failures were in 2 patients with a complete fibrosis of Wharton’s duct and in 2 patients presenting a spontaneous perforation of the duct in the posterior floor of the mouth after an acute episode of sialadenitis.
The sialendoscopic findings of the 131 Wharton’s ducts explored include 106 cases of sialolithiasis (74.1%), 15 cases of sialodochitis (10.5%), 8 cases of stenosis (5.6%), and 14 normal ducts (9.8%). No Wharton’s duct polyps were identified. In 12 cases (8.4%), a combination of 2 of the above disorders was found. The average duration of diagnostic sialendoscopy was $28 \pm 15$ minutes. No complications were encountered during diagnostic sialendoscopy. The mean value of the pain experienced during the procedure was $2.4 \pm 1.9$ (minimum 0, maximum 10).

Interventional sialendoscopy, as well as diagnostic sialendoscopy, was performed over a 5-year period. The follow-up ranged from 3 months to 5 years; the mean follow-up was 2.75 years.

Submandibular sialendoscopy was attempted in 110 cases of sialolithiasis and stenosis. The average duration of the procedure was $71 \pm 41$ minutes. More than 1 interventional sialendoscopy was necessary in 28 cases (25%), resulting in sialendoscopy failures in 13 cases (47%). In 13 cases (12%), the procedure was performed under general anesthesia. Interventional sialendoscopy was successful in relieving the ductal obstruction in 90 cases, for an overall success rate of 82%. In the remaining 20 cases, failures were due to sialoliths embedded in the ductal wall in 14 cases, unsuccessful dilation of ductal stenosis in 4 cases, and impossibility of retrieving intraparenchymal stones in 2 cases. In 5 of these 20 cases, a submandibular gland resection was performed. The overall rate of submandibular gland resection was 4%.

Considering only sialolithiasis, multiple stones (Fig 3) within Wharton’s duct were found in 31 cases (29%), with 2 sialoliths retrieved in 13 cases, 3 in 10...
cases, 4 in 5 cases, 5 in 2 cases, and 6 in 1 case. The average number of retrieved sialoliths per case was 1.56 ± 1.06. The presence of multiple sialoliths was statistically correlated to long operations, multiple procedures, and sialendoscopy failures (Table 1).

The average size of the submandibular sialoliths was 4.9 ± 2.9 mm. The presence of larger sialoliths was statistically related to procedures performed under general anesthesia, long operations, multiple procedures, use of fragmentation, and sialendoscopy failures (Table 2).

Stones larger than 4 mm were found in 43 cases (41%). Fragmentation prior to fragment extraction was required in 28 cases (26%), while the remaining 15 cases underwent a retrieval of the intact stone with the grasping wire basket. In the fragmentation group, an electrohydraulic device was used in 11 cases, and we were able to achieve complete clearance of the duct in 4 cases (36%). The use of a holmium laser for fragmentation in the other 17 cases was successful in 12 cases (71%; Fig 4).

In the remaining group of 15 patients, extraction of the stone with the grasping basket was successful in 9 cases (60%), although it resulted in canal perforations in all 9 cases, due to canal wall stripping (Fig 5). Two of these patients required hospitalization and intravenously administered corticosteroids and antibiotics because of remarkable submandibular swelling.

Complications of interventional sialendoscopy included ductal wall perforation in 11 cases and 2 wire basket blockages, for an overall complication rate of 10%. Nine of the 11 perforations are discussed above, and 2 perforations occurred with the holmium laser. One resolved with conservative therapy, but the second patient (12-mm stone, partially fragmented) experienced residual Wharton’s duct obstruction followed by an acute blockage that required firm traction under sedation to free the instrument. Fragmentation under general anesthesia was required at a later time, followed by a submandibular gland resection.

Finally, a comparison between flexible endoscopy and semirigid endoscopy (Table 3) shows overall success rates of 54% and 85%, respectively (p < .05).

The mean value of the pain experienced during the sialendoscopy was 2.8 ± 2.1 (minimum 0, maximum 10).

**DISCUSSION**

**Diagnostic Sialendoscopy.** While the anatomic description of the submandibular ducts by Wharton dates to the 17th century, the first attempts to visualize them with an endoscope were reported in the early 1990s.13,14 The instruments used were single optic
fibers similar to the ones we used at the beginning of this study. In our hands, visualization could be achieved only in the main duct, because of the absence of an orientation system and of a technique to dilate the duct. The resulting lack of good visualization makes these simple optic fibers disappointing even for diagnostic sialendoscopy, notwithstanding that any sialendoscopic treatment is performed blindly.8

Major progress was made with the introduction of a rinsing system, which provided for dilation of the duct, defogging, and irrigation of debris. Because rinsing requires a working channel, the principal limitation for diagnostic sialendoscopy becomes the diameter of the instrument. The flexible endoscope we used had a diameter of 1.5 mm, with a working channel of 0.5 mm and an optic fiber of 0.5 mm. This small diameter resulted in poor image quality and a short depth of field. Another problem was that to achieve a small outside diameter, the tip of the endoscope could be oriented only in one direction, requiring frequent 180° twisting of the entire fiberscope in order to advance in the ductal system. To be effective, this torsion had to be applied close to the papilla. Because the overall length of the fiberscope was 40 cm and the resistance of a fiber to torsion is directly proportional to its length, several fiberscopes were damaged. Because of similar experiences, Gundlach et al15 have adopted 2-m-long flexible fiberscopes for salivary duct exploration and treatment.

In light of the advantages of rigid esophagoscopy, which allows straightening and stretching of the

Fig 4. Intracanalar laser lithotripsy and retrieval of fragments with wire basket. A) Laser is positioned against stone. B) Stone is fragmented by laser. C) Fragments are retrieved with basket.
esophagus, we designed a semirigid diagnostic sialendoscope with a 1.3-mm outside diameter. The main advantages of this instrument include a large optic fiber, which provides good illumination and a good image, and a rinsing system. However, as with any rigid endoscopy, special care should be taken to avoid trauma of the ductal walls and perforation. Progression should be done only in the center of the lumen, under clear vision.

Diagnostic sialendoscopy was possible in this study in 131 of the 135 cases (97%) under local anesthesia, with excellent patient tolerance (2.4 on pain scale of 0 to 10), even by children more than 10 years of age. (Younger children required general anesthesia.) The entire Wharton’s duct could be explored and its disorders diagnosed with certainty. Since the endoscope is advanced in the center of the ductal lumen, under clear and direct vision, the associated morbidity remains minimal.

Sialendoscopy may be difficult and challenging for beginners, as any small movements of the sialendoscope against the canal wall result in a blurred image. Another difficulty of Wharton’s duct endoscopy is the presence of a sphincter-like mechanism within the first 3 to 4 cm of the duct. This sphincter is hard to dilate, and its passage renders the beginning of the endoscopy difficult. Our experience differs from that of authors who have described a similar mechanism spanning the entire length of the duct.16

Most of the patient population of this study had an extensive radiologic investigation before sialendoscopy, because it was ordered before their referral, or because of concomitant study protocols (also Marchal et al, unpublished observations). At the present time, we do not require any specific radiologic examination before diagnostic sialendoscopy. We consider sialendoscopy to be the investigation of choice in the suspicion of salivary ductal diseases and any submandibular swelling of unclear origin.

The radiologic evaluation diagnosed sialodochitis in about 15% of patients, half of whom also had sialolithiasis. Endoscopy also revealed zones of the ductal wall that were erythematous, appeared inflamed, and often were associated with a canal narrowing. A frequent association with sialolithiasis was also noted. However, the correlation between radiology and endoscopy is not very good, as has been detailed elsewhere.11 The clinical significance of sialodochitis requires further study. In particular, it is difficult to say whether this inflammation was part of the “initiation process” of calculus formation, or was secondary to an established sialolithiasis.

Interventional Sialendoscopy. The first report of a distal submandibular stone extraction was published in 1990; the procedure was performed blindly with a

TABLE 3. RESULTS OF INTERVENTIONAL SIALENDOSCOPY ACCORDING TO TYPE OF ENDOSCOPE USED

<table>
<thead>
<tr>
<th></th>
<th>Free Optic Fiber</th>
<th>Flexible Endoscope</th>
<th>Modified Feto-</th>
<th>Marchal Sialendoscope</th>
</tr>
</thead>
<tbody>
<tr>
<td>Number</td>
<td>11 (10%)</td>
<td>13 (12%)</td>
<td>27 (24%)</td>
<td>59 (54%)</td>
</tr>
<tr>
<td>General anesthesia</td>
<td>0 (0%)</td>
<td>2 (15%)</td>
<td>2 (7%)</td>
<td>9 (15%)</td>
</tr>
<tr>
<td>Duration of procedure</td>
<td>59 ± 29</td>
<td>92 ± 42</td>
<td>70 ± 36</td>
<td>70 ± 44</td>
</tr>
<tr>
<td>Multiple procedures</td>
<td>1 (9%)</td>
<td>7 (5%)</td>
<td>5 (18%)</td>
<td>15 (25%)</td>
</tr>
<tr>
<td>Sialendoscopy failures</td>
<td>1 (9%)</td>
<td>6 (46%)</td>
<td>4 (15%)</td>
<td>9 (15%)</td>
</tr>
</tbody>
</table>

Data are numbers of cases, except for duration of procedure (minutes).

*Statistically significant difference.
wire basket, during sialography.\(^7\) Our initial endoscope was similar,\(^7\) before technical developments allowed salivary stone extraction under endoscopic control.\(^7,9\) The apparent high success rate with this technique (Table 3) results from case selection, by radiologic studies, of small and unique sialoliths located in the main duct. Although it is possible, we no longer use or recommend the semiblind technique with the grasping wire basket, in contradiction to authors who favor this equipment.\(^13,18\) They present their technique as “endoscopic stone retrieval”; however, it should be clear from our description that during this technique, stone removal remains blind: an exploration of the duct with the optic fiber is followed by the removal of the endoscope and by the blind introduction of the retrieving basket. The potential dangers of this blind technique cannot be overemphasized.

We have also abandoned the use of flexible endoscopes for interventional sialendoscopy, not only because of difficult maneuvering and poor visualization, but also because of difficulty in sterilization of the material, and because the working channel of the fiberscope tends to become stripped by the grasping wire basket. The poor success rate of interventional sialendoscopy (Table 3) with this device might also be due to the particular fiberscope we used, as well as the lack of experience, since these cases were done early on.

Comparison of the 2 semirigid endoscopes does not show any clear advantage in terms of the final success of interventional submandibular sialendoscopy (Table 3). However, the main limitation is a larger diameter, which sometimes causes problems in Wharton’s duct, and becomes a clear limitation for interventional sialendoscopy of the parotid gland.\(^19,20\) Our newly developed sialendoscope was used in 59 patients with Wharton’s duct sialolithiasis, with a success rate of 85%.

Sialoliths can be either directly extracted or fragmented before extraction. Prior publications have focused on fragmentation. The available techniques include use of electrohydraulic devices,\(^21-23\) use of pneumoblastic devices,\(^24\) external lithotripsy,\(^25\) and intraductal laser lithotripsy.\(^14\) A review of the advantages and disadvantages of these techniques can be found elsewhere.\(^10\) In our opinion, the best technique is the fragmentation of sialoliths with a laser fiber, as initially described by Gundlach et al.\(^14\) The laser fiber is introduced in the sialendoscope, laser sialolithotripsy is performed under direct visual control, and retrieval of stone fragments is achieved with grasping wire baskets. A distinct advantage of our technique is the retrieval of sialoliths and their fragments after lithotripsy, contrary to the majority of previously described methods.\(^14,21-25\)

The holmium laser is well known and has proven efficiency for urolithiasis.\(^26,27\) However, one has to be attentive to its potential dangers, because of its absorption characteristics in the surrounding tissues and because of the heat generated from the fragmentation within the narrow salivary ducts. The holmium laser should be used only under clear vision, tangential to the duct, and only in cases of sialolithiasis. The dye laser\(^14\) has proven efficiency and low morbidity, because the high energy delivered is not absorbed by the tissues. Unfortunately, the cost of the device and its specificity may render its acquisition difficult. The combination of dye laser stone fragmentation and endoscopic retrieval is currently being investigated and might decrease the use of general anesthesia, and also further increase the success rate of interventional sialendoscopy.

We initially performed large sialodochoplasties as reported by others.\(^9,16\) For the last 80 patients, we performed only a dilation, with minimal incision only for the stone retrieval. We found no residual stenosis on follow-up. Therefore, we advise against large marsupialization of the ductal papillae, in order to prevent retrograde passage of air and aliment.\(^10,28\)

The results of interventional sialendoscopy were directly related to the size of the stone in the present study. Therefore, we propose the following approach. For stones smaller than 4 mm, interventional sialendoscopy should be performed immediately after the diagnostic procedure, in an ambulatory setting, under local anesthesia, without fragmentation, and with the grasping wire basket used for retrieval of the stone. For sialoliths larger than 4 mm, the shape of the stone is important, and an irregular shape or a very posteriorly located stone would render interventional endoscopy difficult, risking basket blockage or the necessity of using fragmentation procedures. In those cases, the procedure should be performed under general anesthesia.

Despite its apparent simplicity, interventional sialendoscopy is a technically challenging procedure. The maneuvering of the rigid sialendoscopes within the small salivary ducts requires extensive experience. Manipulation is delicate, and progression, which should remain absolutely atraumatic, might be hazardous because of the theoretical risks of perforation and vascular or neural damage. Significant trauma of the ductal wall could result in later stenosis. The necessity of performing the entire procedure under direct visualization cannot be overemphasized.

In 110 submandibular interventional endoscopies, we did not encounter any significant complications,
such as damage to the facial nerve or lingual nerve, gross hemorrhage, or major canal wall perforations. Nevertheless, minor canal wall perforations were found in 11 cases, resulting in 1 submandibular gland resection and 2 admissions for intravenous treatment because of persistent swelling of the submandibular gland. In addition, blockage of the grasping basket is a rather traumatic experience for the patient and the surgeon, potentially resulting in emergency submandibular gland resection.

In conclusion, diagnostic sialendoscopy is a new, minimally invasive technique that may become the investigational procedure of choice for salivary duct disorders. Interventional sialendoscopy is an outpatient procedure performed under local anesthesia that allows the extraction of the majority of sialoliths and thereby prevents salivary gland excision. Further miniaturization of the instruments will allow fragmentation of larger stones and extraction of more deeply located stones.

REFERENCES