Pediatric Sialendoscopy

A 5-Year Experience at a Single Institution

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Objectives: To evaluate the outcome of our experience in the treatment of salivary gland disorders in children undergoing sialendoscopy and to assess the evolution of the technique.

Design: Retrospective medical record review.

Setting: Tertiary care university hospital.


Intervention: Diagnostic and interventional sialendoscopy using general anesthesia.

Main Outcome Measures: Demographic, clinical, and surgical variables, including age, sex, date of first symptoms, parotid or submandibular location of disease, preoperative ultrasonographic results, sialendoscopy technique, sialendoscopy observations, and complications.

Results: Pediatric sialendoscopy was performed on the parotid gland in 23 patients (61%) and on the submandibular gland in 15 patients (39%). The most frequent indication for sialendoscopy was recurrent salivary gland swelling. Thirty-two of 38 procedures (84%) were performed endoscopically, whereas a combined intervention was necessary for 3 patients and a submandibular gland excision for another 3 patients. Sialendoscopy allowed the diagnosis of 12 patients with salivary duct lithiasis, 21 with salivary duct stenosis, and 2 with both submandibular lithiasis and stenosis, and findings from 3 sialendoscopies were normal. Preoperative ultrasonographic results were confirmed by sialendoscopy in only 7 patients. Of the 10 patients with lithiasis found using sialendoscopy, only 4 had been detected using preoperative ultrasonography.

Conclusions: Sialendoscopy is a pertinent technique for the diagnosis and treatment of salivary gland disorders in children. It also allows the most conservative treatment of sialolithiasis and juvenile recurrent parotitis.


THE CONCEPT OF SIALENDOSCOPY has been recognized for almost 20 years. The first publication by Katz was in 1990, but sialendoscopy still remains a recent approach to salivary gland obstructive abnormalities in children. Various medical centers have already adopted this technique, and numerous children have been treated by sialendoscopy for salivary gland obstructive swelling. Recently, Shacham et al published the results of an analysis of 70 pediatric patients with juvenile recurrent parotitis (JRP) treated by sialendoscopic lavage of the gland with corticosteroids. There was no recurrence for 93% (n = 65) of the patients during follow-up of 6 to 36 months. Sialendoscopy has also been validated by various researchers as a safe technique in children with salivolithiasis.

During the past 6 years, the sialendoscopic approach to salivary gland disorders has been developed in children at Edouard Herriot University Hospital, Lyon, France, and we evaluated the development of this technique in diagnosis and therapeutics, especially dilatation and introduction of the thulium laser for stone fragmentation. The aims of this study are to show this evolution, to discuss the correlation between ultrasonographic and sialendoscopic results in children, and to define the position of sialendoscopy in the diagnosis and treatment of pediatric salivary gland obstructive abnormalities.

Methods

This study was conducted in a tertiary referral center (the Ear, Nose, and Throat Department of the Edouard Herriot University Hospital). A retrospective analysis was performed of all sialendoscopy procedures in children in the Department of Otorhinolaryngology between January 1, 2003, and November 30, 2008. Demographic, clinical, and surgical variables were assessed, including age, sex, date of first symptoms, parotid or submandibular loca-

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tion of disease, preoperative ultrasonography and magnetic resonance imaging (MRI) sialographic results, sialendoscopy technique, sialendoscopy diagnosis, and complications.

Sialendoscopy was performed using general anesthesia and using 2 different types of sialendoscopes. The first sialendoscope was a semirigid optic endoscopic device (0.9 mm) introduced into a sheath, with an external diameter varying from 1.1 to 1.7 mm. The second sialendoscope was the 1.3-mm “all-in-one” device (reference 11573; Karl Storz GmbH, Tuttingen, Germany) for diagnostic and interventional sialendoscopy.7 Since 2007, we have also used the 1.1-mm all-in-one device, which is more suitable for young children. Diagnostic sialendoscopy was performed to classify ductal lesions such as sialolithiasis or stenosis. Sialendoscopic diagnosis of lithiasis was straightforward via direct visualization of the stone in the duct. Stenosis was diagnosed based on narrowing of the duct area of punctuated high signal in each case, thus confirming JRP. In 1 patient, a generalized stenosis was revealed via MRI sialography.

The MRI sialography was performed for 6 children with recurrent parotid gland swelling episodes; the MRI sialography images showed diffuse sialectasia with a large area of punctuated high signal in each case, thus confirming JRP. In 1 patient, a generalized stenosis was revealed via MRI sialography.

The sialendoscopy procedure allowed the diagnosis of 12 patients with sialolithiasis (1 parotid and 11 submandibular), 21 with stenoses (18 parotid and 3 submandibular), and 2 with both submandibular lithiasis and stenosis. All of the 18 patients with parotid duct stenoses presented with a generalized form of stenosis of the duct system (Figure). For 3 patients (2 parotid and 1 submandibular), diagnostic sialendoscopy did not reveal any abnormalities.

Three patients each underwent 3 sialendoscopic procedures because of recurrence of symptoms (1-3 months). These 3 patients presented with Stensen duct stenoses. Five patients had 2 sialendoscopy procedures, 1 for a Stensen lithiasis, 3 for a Wharton lithiasis, and 1 for parotid swelling without any abnormality noted during the first sialendoscopy.

Of the 18 patients with stenosis of the parotid duct who underwent sialendoscopic dilation with intra-ductal corticosteroid administration, only 4 had a recurrence of symptoms after mean remission of 6 months. The other 14 patients showed no recurrence during mean follow-up of 24 months (range, 4-24 months).

Complications occurred in 3 patients: 1 Stensen duct perforation and 2 airway obstructions. In the child with Stensen duct perforation, the perforation occurred during laser use. After 1 month of follow-up, healing was complete, without fistulas. In the 2 children with airway obstruction, sialendoscopy was performed bilaterally with a high diffusion of lavage solution in the pharyngeal aspect of the parotid gland. Perioperative massage of the pa-

### Table 1. Preoperative Sialendoscopy Indications

<table>
<thead>
<tr>
<th>Indication</th>
<th>Patients, No.</th>
</tr>
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<tbody>
<tr>
<td>Sialolithiasis</td>
<td>1</td>
</tr>
<tr>
<td>Parotid</td>
<td>17</td>
</tr>
<tr>
<td>Submandibular</td>
<td>11</td>
</tr>
<tr>
<td>Sialadenitis</td>
<td>6</td>
</tr>
<tr>
<td>Parotid</td>
<td>6</td>
</tr>
<tr>
<td>Submandibular</td>
<td>5</td>
</tr>
</tbody>
</table>

Among the 420 patients treated by sialendoscopy at Edouard Herriot University Hospital between January 1, 2003, and November 30, 2008, we identified 38 pediatric patients. The mean age of these patients was 9 years (age range, 2-17 years), and the male to female sex ratio was 2:1. Pediatric sialendoscopy was performed on the parotid gland in 23 patients (61%) and on the submandibular gland in 15 patients (39%). Sialendoscopy clinical indications were recurrent salivary gland swelling with inflammatory or me-

chical episodes (Table 1). Mean age at the occurrence of salivary gland symptoms in children was known for 24 patients and was 26 months (range, 2-81 months).

Diagnostic sialendoscopy was possible in all of the patients. For 32 of 38 patients (84%), treatment was performed endoscopically, whereas a combined intervention was necessary for 3 patients (8%) and a submandibular gland excision was necessary for 3 other patients (8%). Nineteen patients underwent ultrasonography before sialendoscopic management. The ultrasonography and sialendoscopy correlation is given in Table 2. For the 5 patients who had normal ultrasonographic findings, sialendoscopy revealed 4 cases of ductal abnormalities, mostly lithiasis. For the 7 patients who had suggested sialolithiasis on ultrasonography, only 4 were confirmed by sialendoscopy.

Each time ultrasonography showed heterogeneous salivary glands (5 patients), sialendoscopy revealed ductal stenoses. And each time ultrasonography showed ductal dilatation (2 patients), sialendoscopy confirmed the diagnosis of stenosis. Of the 10 cases of stenosis revealed by sialendoscopy, only 4 had been detected by ultrasonography. For the 6 cases of stenosis not detected by ultrasonography, the stenosis was large enough to require a combined intervention in 2 cases. Ultrasonographic preoperative results have been confirmed using sialendoscopy in only 7 patients.

The MRI sialography was performed for 6 children with recurrent parotid gland swelling episodes; the MRI sialography images showed diffuse sialectasia with a large area of punctuated high signal in each case, thus confirming JRP. In 1 patient, a generalized stenosis was revealed via MRI sialography.

The sialendoscopy procedure allowed the diagnosis of 12 patients with sialolithiasis (1 parotid and 11 submandibular), 21 with stenoses (18 parotid and 3 submandibular), and 2 with both submandibular lithiasis and stenosis. All of the 18 patients with parotid duct stenoses presented with a generalized form of stenosis of the duct system (Figure). For 3 patients (2 parotid and 1 submandibular), diagnostic sialendoscopy did not reveal any abnormalities.

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Of the 18 patients with stenosis of the parotid duct who underwent sialendoscopic dilation with intra-ductal corticosteroid administration, only 4 had a recurrence of symptoms after mean remission of 6 months. The other 14 patients showed no recurrence during mean follow-up of 24 months (range, 4-24 months).

Complications occurred in 3 patients: 1 Stensen duct perforation and 2 airway obstructions. In the child with Stensen duct perforation, the perforation occurred during laser use. After 1 month of follow-up, healing was complete, without fistulas. In the 2 children with airway obstruction, sialendoscopy was performed bilaterally with a high diffusion of lavage solution in the pharyngeal aspect of the parotid gland. Perioperative massage of the pa-


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rotid gland and perioperative corticosteroid intravenous injection prevented postoperative dyspnea in both patients. No duct avulsion, facial palsy, or bleeding was noted.

COMMENT

Sialendoscopy brings new elements to pediatric salivary gland disease diagnosis and treatment. The main pathologic abnormalities of pediatric sialendoscopy that arise from this study are ductal stenoses and ductal lithiasis. The first element that arises is the importance of sialendoscopic diagnosis of ductal stenoses, especially in the parotid gland. In this series, 21 patients with stenosis were diagnosed by using the sialendoscopic procedure—18 in the parotid and 3 in the submandibular gland. The 18 children with Stensen duct stenosis had recurrent episodes of inflammatory parotitis, and the diagnosis of JRP was established. In the literature, researchers agree to describe the sialendoscopic characteristics of JRP as a “white aspect of the ductal layer without the natural proliferation of blood vessels.” In the present series, we noted the same endoscopic white aspect but always associated it with diffuse ductal stenoses. According to the classification of obstructive salivary abnormalities published in 2009 by Marchal et al, which describes 5 stenosis scores, the aspect found in JRP corresponds to score S4 (generalized ductal stenosis). Unique or multiple intraductal diaphragmatic stenosis is not common in JRP. The stenotic aspect in JRP visible using sialendoscopy has not been reported in other inflammatory salivary gland disorders, such as Gougerot-Sjogren disease or immune deficiency, and seems specific to JRP. A distinction must be made between local stenosis that could be due to inflammation in adults and global stenosis more suggestive of a congenital origin in children. Sialendoscopy allows JRP diagnosis with better sensitivity than ultrasonography even in young children. Results illustrate the difficulty of ultrasonographic interpretation in children with salivary gland disorders.

Concerning salivary duct stenosis, ultrasonography did not contribute to the diagnosis. Most patients with sialendoscopic diagnosis of duct stenosis have no duct dilatation on ultrasonographic analysis. For some of them, ultrasonography shows a heterogeneous gland structure, but this is not a specific radiologic finding. The sonographic features of JRP are described as hypoechoic areas and heterogeneous internal echoes by Shimizu et al., but no duct stenosis was visualized. None of these ultrasonographic variables were specific to JRP because they can be found in all inflammatory salivary gland disorders.

Sialendoscopy allows not only diagnosis but also treatment of JRP. This treatment is based on dilation of the Stensen ductal system by a strong saline solution irrigation or by the sialendoscope itself or sometimes by balloon dilation. Another advantage of the sialendoscopic technique in JRP lies in the possibility of directly irrigating the ductal system of salivary glands with medications such as corticosteroid preparations. This direct administration of drugs into the ductal system provides interesting access for salivary inflammatory disease therapy in adults and children.

Table 2. Correlation Between Preoperative Ultrasonography and Sialendoscopy Findings in Children With Salivary Gland Disorders

<table>
<thead>
<tr>
<th>Sialendoscopy Finding</th>
<th>Normal</th>
<th>Lithiasis</th>
<th>Ductal Dilatation</th>
<th>Heterogeneous Glands</th>
<th>Total</th>
</tr>
</thead>
<tbody>
<tr>
<td>Normal</td>
<td>1</td>
<td>1</td>
<td>0</td>
<td>0</td>
<td>2</td>
</tr>
<tr>
<td>Lithiasis</td>
<td>1</td>
<td>4</td>
<td>0</td>
<td>0</td>
<td>5</td>
</tr>
<tr>
<td>Stenosis</td>
<td>3</td>
<td>2</td>
<td>1</td>
<td>5</td>
<td>11</td>
</tr>
<tr>
<td>Lithiasis + stenosis</td>
<td>0</td>
<td>0</td>
<td>1</td>
<td>0</td>
<td>1</td>
</tr>
<tr>
<td>Total</td>
<td>5</td>
<td>7</td>
<td>2</td>
<td>5</td>
<td>19</td>
</tr>
</tbody>
</table>

a Data are given as number of patients.

Figure. Sialendoscopic views of 3 patients. A, A normal Stensen duct with a normal pink color and visualization of the vessels. B, Juvenile recurrent parotitis with a specific “white aspect” associated with difficulty in introduction and mobilization of the sialendoscope in the duct. C, Diaphragmatic stenosis with the laser probe at the center.
In the literature and in the present series, stenosis was observed more frequently in the parotid gland and sialolithiasis in the submandibular gland in children. Ten children had sialolithiasis—9 in the submandibular gland and 1 in the parotid gland—revealed by recurrent episodes of salivary gland swelling. Infection of the gland, with pain and fever, occurred in 5 children. Because of the tertiary center recruitment, children came to the Ear, Nose, and Throat Department of Edouard Herriot University Hospital, Lyon, France, with radiologic examinations already performed and, specifically, ultrasonography, which is easier to obtain in children without ionizing radiation.

Of the 10 patients with lithiasis diagnosed using sialendoscopy in this study, only 4 had been suspected by previous ultrasonography. Of the 6 undiagnosed patients, 4 had stones measuring 2 mm in diameter. The diagnosis of salivary stones in children is difficult because the lower limit of radiologic detection is 2 mm. Two millimeters is the limit to detect stones using various radiologic methods, such as ultrasonography, computed tomography, and MRI sialography. In children, a stone of 2 mm may be of sufficient size to cause a duct obstruction in the parotid or submandibular gland. Sialendoscopy has better sensitivity in diagnosing salivary stones in children than does ultrasonography. With the present diameter of the sialendoscope, examination of the salivary gland ducts is possible for the primary branches and often for the secondary branches. Sialolithiasis is not a frequent abnormality in children, representing less than 5% of all sialolithiasis in the literature, but sialolithiasis is certainly underestimated using only ultrasonography.

The recent evolution of the sialendoscopic technique is summed in the use of thinner sialendoscopes to access the second-generation branches of the gland and laser probes (250–350 µm) for stone fragmentation. At Edouard Herriot University Hospital, the 2.1-µm continuous YAG-thulium laser is preferred, but in the literature, the use of the 2-µm YAG-holmium laser is more common. The YAG-thulium laser is preferred because of its low residual thermal injury compared with that of the YAG-holmium laser. The variables of laser thulium use are the lower limit of radiologic detection. In children, a stone of 2 mm may be of sufficient size to cause a duct obstruction in the parotid or submandibular gland. Sialendoscopy has better sensitivity in diagnosing salivary stones in children than does ultrasonography. With the present diameter of the sialendoscope, examination of the salivary gland ducts is possible for the primary branches and often for the secondary branches. Sialolithiasis is not a frequent abnormality in children, representing less than 5% of all sialolithiasis in the literature, but sialolithiasis is certainly underestimated using only ultrasonography.

In conclusion, the sialendoscopic approach to pediatric salivary gland obstrucive disease is an effective method for diagnosis and treatment with low morbidity. Diagnostic sialendoscopy must be considered the first step in complementary examinations in children with salivary gland obstructive disease. Based on diagnostic sialendoscopy in this series, we describe a specific stenotic aspect of the ductal system in JRP.

Interventional sialendoscopy is the first conservative treatment to propose in children with salivary disease. In JRP, the sialendoscopy benefit is the dilation of the ductal system under direct vision and the irrigation of the ductal system with corticosteroids. In sialolithiasis, the use of thulium laser fragmentation allows for even larger stones to be extracted endoscopically and decreases the incidence of an external approach with the learning curve.

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Author Contributions: Dr Martins-Carvalho had full access to all the data in the study and takes responsibility for the integrity of the data and the accuracy of the data analysis.

Study concept and design: Martins-Carvalho, Marchal, and Faure. Acquisition of data: Martins-Carvalho, Quenin, and Lesniak. Analysis and interpretation of data: Martins-Carvalho, Plouin-Gaudon, and Froehlich. Drafting of the manuscript: Martins-Carvalho, Quenin, Lesniak, and Faure.

Critical revision of the manuscript for important intellectual content: Plouin-Gaudon, Froehlich, Marchal, and Faure.

Administrative, technical, and material support: Plouin-Gaudon.

Study supervision: Froehlich, Marchal, and Faure.

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REFERENCES