Juvenile Recurrent Parotitis

Sialendoscopic Approach

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Objective: To assess the relevance of sialendoscopy as a diagnostic and interventional procedure in juvenile recurrent parotitis (JRP).

Design: Prospective case series study.

Setting: Tertiary care teaching hospital.

Patients: Sialendoscopy was used to examine 10 children (age range, 1.8-13.0 years) with symptomatic JRP for recurrent swelling of the parotid glands between January 2003 and January 2005. Diagnostic sialendoscopy allowed classification of ductal lesions, and interventional sialendoscopy was used to treat the lesions. Initial data analyzed included the type of endoscope used as well as the size and form of the main duct of the parotid gland. Outcome variables were resolution of symptoms and endoscopic enlargement of the ductal tree.

Results: Initial ultrasound evaluation of the diseased gland revealed a white Stensen duct without the natural proliferation of blood vessels in all 10 cases. This finding was associated with a true stenosis of the Stensen duct. Two cases of suspected stones according to ultrasonography were subsequently diagnosed as localized stenoses. The sialendoscope was used to dilate the duct with pressurized saline solution in all cases as well as to dilate the 2 cases of stenoses. There were no major complications. The average length of follow-up was 11 months (range, 2-24 months). Seventeen parotid glands were dilated in all 10 patients, with a success rate of 89%. One patient needed repeated sialendoscopies for recurrent symptoms. Two patients presented with a second episode of JRP contralateral to the side initially treated.

Conclusions: Diagnostic sialendoscopy is a new procedure that can be used in children for reliable evaluation of salivary ductal disorders, with low morbidity. Sialendoscopic dilation of the main parotid ducts appears to be a safe and effective method for treating JRP.


Juvenile recurrent parotitis (JRP) is defined as recurrent episodes of inflammatory parotitis in children. Although JRP is an uncommon disorder, it is the second most common salivary disease in children, after mumps. It is characterized by recurrent episodes of swelling and/or pain associated with fever and malaise and is usually unilateral. In some cases, it can occur bilaterally, usually with a more predominant side. The first episode typically occurs between the ages of 3 and 6 years. The interval between 2 episodes is variable. The number of attacks also varies individually. The main criterion for establishing the severity of JRP is the frequency of the recurrences. The pathogenesis of JRP remains unclear.

The treatment of acute episodes of JRP aims to prevent damage to the gland parenchyma and to alleviate the symptoms (eg, with antibiotic agents and pain medication). Because of the supposed bacterial origin, most authors classically recommend the continuous administration of low doses of antibiotics or standard doses early in an attack. The antibiotic treatment is also believed to prevent additional damage to the glandular parenchyma. To our knowledge, no preventive therapy for JRP is available as yet.

Following our previous study, which included 150 adults and children, we investigated diagnostic and interventional sialendoscopy as an alternative to the classic approach to the treatment of JRP. We present a preliminary report involving 17 pediatric sialendoscopies in 10 cases of JRP.
The first episode usually occurred at approximately 2 1/2 years (age range, 2.0-12.0). The sex ratio of 60% favored a female preponderance. The average age of the 6 girls was 6.6 years (age range, 3.5-13.0 years). The sex ratio of 60% favored a female preponderance. The average age of the boys was 3.6 years (age range, 1.8-3.8 years). Between January 2003 and January 2005, 10 children (4 boys and 6 girls; mean age, 5.0 years; age range, 1.8-13.0 years) with symptomatic JRP were treated in our institutions (Table). The average age of the boys was 3.6 years (age range, 1.8-3.8 years). The average age of the 6 girls was 6.6 years (age range, 3.5-13.0 years). The sex ratio of 60% favored a female preponderance. The first episode usually occurred at approximately 2 1/2 years in boys and 4 years in girls. The average number of episodes was 4.8 (range, 2.0-12.0).

One child had a mother with sicca syndrome. Therefore, without evidence of another specific cause, a diagnosis of sicca syndrome could not be totally ruled out in this particular child. All 10 children had a predominantly affected gland, although 7 presented with intermittent bilateral swelling. Therefore, in these 7 cases, both sides were explored. All patients underwent ultrasonography, and 2 underwent magnetic resonance sialography. In 8 patients, ultrasonography revealed inflammatory parotid enlargement with hypoechoic areas corresponding to punctate sialectasia. Two children were initially diagnosed as having sialolithiasis by ultrasonography. The decision to perform endoscopic intervention was made when 2 episodes occurred within a 6-month period.

We used 2 last-generation sialendoscopes (Karl Storz, Tuttlingen, Germany). The first system uses a semirigid 0.9-mm optic endoscopic device, which can be introduced in 2 types of sheaths and gives the endoscope some flexibility. The first type of sheath is used for pure diagnostic purposes and has an external diameter of 1.1 mm. The second type is used for interventional purposes and has 1 working channel of 1.15 mm and a telescope channel of 0.9 mm. The telescope itself has an internal diameter of 0.75 mm. The second system uses an all-in-1 instrument (Marchal Sialendoscope; Karl Storz) (Figure 1), which has an external diameter of 1.3 mm, a fiberoptic scope, an operating channel (0.65 mm in diameter), and an irrigation channel (0.25 mm in diameter). This unit was used either as a diagnostic or as an interventional tool. Papilla dilation was achieved with a set of customized dilators (Nos. 745910 and 745845-744856; Karl Storz). Dilation of ductal stenoses was performed either with the interventional endoscope itself or with balloon catheters in localized cases (No. 28635P; Karl Storz).

The endoscopic procedure was performed with the patient under general anesthesia. The first step involved dilation of the papilla with salivary probes of increasing diameter, followed by the use of the papilla dilator. The second step consisted of the diagnostic sialendoscopy. The cleansing of the endoscope tip and the slight dilation of the duct to obtain good visualization were achieved by intermittently rinsing a local anesthetic solution (50% xylocaine [2%] and 50% sodium chloride saline solution [0.9%]) through the endoscope. Diagnostic sialendoscopy allowed a minimally invasive exploration of the ductal system. When a stenosis was located, a balloon catheter was introduced and inflated at the level of the stenosis under visual control. In most cases, the sialendoscope itself was also used to dilate the duct. Finally, the parotid gland was rinsed with saline solution and steroids. The patients remained hospitalized for 24 hours. Antibiotics (amoxicillin–clavulanic acid) and corticosteroids (prednisolone, 50 mg) were administered for 48 hours. Massages of the parotid gland were recommended in the 24 first hours to decrease swelling. All children were reexamined 1 month after the procedure. Control sialendoscopy was never performed.

The average time of the endoscopic procedure was 57 minutes. The Stensen duct was examined in all 10 cases, and in 4 cases, the duct was large enough to explore the second-generation branches (Table). A white Stensen duct without the natural proliferation of blood vessels (Figure 2) was seen in all cases and was

<table>
<thead>
<tr>
<th>Patient No./Age, y</th>
<th>Sex</th>
<th>No. of Episodes (Presentation)</th>
<th>Ultrasound Findings</th>
<th>Relapse</th>
<th>Follow-up, mo</th>
<th>Sialendoscopy</th>
</tr>
</thead>
<tbody>
<tr>
<td>1/F/13.0</td>
<td>7.0</td>
<td>6 (bilateral)</td>
<td>Sialectasia</td>
<td>No</td>
<td>6</td>
<td>Bilateral, overall stenosis on right side</td>
</tr>
<tr>
<td>2/F/7.0</td>
<td>5.0</td>
<td>4 (unilateral)</td>
<td>Sialectasia</td>
<td>No</td>
<td>4</td>
<td>Unilateral, first-generation branches and overall stenosis</td>
</tr>
<tr>
<td>3/F/6.5</td>
<td>3.5</td>
<td>6 (bilateral)</td>
<td>Sialectasia</td>
<td>No</td>
<td>24</td>
<td>Bilateral, second-generation branches and overall stenosis</td>
</tr>
<tr>
<td>4/M/5.5</td>
<td>3.0</td>
<td>5 (bilateral)</td>
<td>Lithiiasis</td>
<td>No</td>
<td>2</td>
<td>Second-generation branches, sludge, and overall stenosis</td>
</tr>
<tr>
<td>5/F/5.0</td>
<td>3.5</td>
<td>12 (bilateral)</td>
<td>Sialectasia</td>
<td>Yes</td>
<td>16</td>
<td>Second-generation branches, sludge, and overall stenosis</td>
</tr>
<tr>
<td>6/F/4.5</td>
<td>4.0</td>
<td>3 (unilateral)</td>
<td>Lithiiasis</td>
<td>No</td>
<td>8</td>
<td>Unilateral, localized stenosis and papillotomy of Stensen duct</td>
</tr>
<tr>
<td>7/M/3.8</td>
<td>3.2</td>
<td>2 (unilateral)</td>
<td>Sialectasia</td>
<td>No, but 4 contralateral episodes</td>
<td>18</td>
<td>Unilateral, overall stenosis and dilation of the pharyngeal part of parotid gland</td>
</tr>
<tr>
<td>8/F/3.5</td>
<td>3.5</td>
<td>2 (bilateral)</td>
<td>Sialectasia</td>
<td>Yes, right side</td>
<td>8</td>
<td>Bilateral, second generation branches on left side; impossible on right side because of duct perforation</td>
</tr>
<tr>
<td>9/M/3.3</td>
<td>2.0</td>
<td>7 (bilateral)</td>
<td>Sialectasia</td>
<td>No</td>
<td>3</td>
<td>Bilateral, overall stenosis</td>
</tr>
<tr>
<td>10/M/1.8</td>
<td>1.7</td>
<td>2 (bilateral)</td>
<td>Sialectasia</td>
<td>No</td>
<td>18</td>
<td>Bilateral, overall stenosis</td>
</tr>
</tbody>
</table>

![Figure 1. The all-in-1 sialendoscope (1.3 mm in diameter).](image-url)
associated with true stenosis of the Stensen duct. Stenoses of the entire Stensen duct were diagnosed according to 2 criteria: (1) global narrowing of the duct under endoscopic vision control and (2) difficulty in introducing the sialendoscope. Local stenosis was recognized in 1 case as a diaphragm and correlated with the area of the parotid swelling.

Sialendoscopy was performed for bilateral swelling in 7 patients. Endoscopic examination was indicated on 1 side in 3 patients. Contralateral sialendoscopy was not possible in 2 of the 7 patients. In these 2 cases, swelling of the pharyngeal part of the parotid gland, with partial upper airway obstruction (Figure 3), was observed. Therefore, in both cases, the second parotid gland was not explored in order to avoid major airway obstruction due to bilateral enlargement of the pharyngeal extension of the parotid gland. Papillotomy of the Stensen duct was necessary in 1 case because dilation of the papilla was impossible. Sludge within the ductal system was seen in 2 cases. Swelling of the parotid gland after sialendoscopy can result from high-pressure injection of the rinsing solution in the gland during the procedure (Figure 4). Such episodes of swelling resolved in all cases within 24 hours. There were no major complications such as facial palsy or abscess after sialendoscopy in any of our patients.

The average length of follow-up was 11 months (range, 2-24 months). Only 1 patient underwent repeated sialendoscopy for recurrent symptoms. Her recurrent swelling had diminished, and she was free of symptoms for 10 months in the right side and for 2 months in the left side. A localized stenosis, which was observed during the second endoscopy, was dilated with the endoscope, forcing irrigation. Two patients presented with second episodes of JRP contralateral to the side that had initially been treated.

**COMMENT**

Several studies report a JRP sex distribution favoring males. In our study, we found a predominance of female patients, with a ratio of 6:4. Other authors found the same sex distribution, with a higher proportion of females in cases in which the symptoms appeared later in life. As in the literature, JRP occurred on average between the ages of 3 and 6 years in our study. Ericson et al describe 20 patients with JRP, with an age at onset ranging from 3 months to 16 years.

According to Nozaki et al, ultrasonography may be useful for diagnosis and follow-up of JRP. In our group of patients, sialendoscopy appeared to be of better diagnostic value than ultrasonography. Indeed, preoperative ultrasonography misdiagnosed 2 cases of stenosis as sialolithiasis. In fact, no stone was found at the time of the endoscopic procedure. In these 2 cases, sialendoscopy provided further evidence for the diagnosis of JRP. Moreover, sialendoscopy is a minimally invasive technique with greater sensibility and specificity than imaging. The technique is safe and without major complications.

Sialendoscopy is a diagnostic tool. However, the actual diameter of the endoscope allows dilation and exploration of the Stensen duct, but only up to the first-generation branches. In our series, it was not possible to explore the secondary branches in 7 cases. In accordance with the study by Nahlieli et al, we recognized the “white aspect of the ductal layer without the natural proliferation of blood vessels.” This aspect is believed to be characteristic of JRP. In our pediatric series, the
diameter of the duct was found to be smaller than in adults. There is no normative value of the diameter of the Stensen duct according to age, but it is believed that the duct grows and enlarges during childhood. This theory is difficult to confirm, because the duct is expandable. During our measurements, we could only evaluate the maximal diameter according to the progressive resistance during dilatation of the papilla.

A review of the relevant literature found no study on maturation of the diameter of the Stensen duct. The study by Zenk et al. involving patients aged 49 to 89 years found that the diameter of the Stensen duct ranged from 0.9 to 2.3 mm. These measurements are larger than those we found with ductal probes. Such findings represent an argument for secondary development of the parotid gland ductal system in childhood.

The pathogenesis of JRP remains unclear. Some factors that have been suggested to contribute to the development of JRP include ductal congenital malformations, hereditary genetic factors, viral or bacterial infection, allergy, and local manifestation of an autoimmune disease. The theory of allergy as a causative factor in JRP was not validated by Ericson et al. Fazekas et al. found a correlation between JRP and a selective IgA deficiency. For Chitre and Premchandra, the most probable hypothesis for pathogenesis of JRP involved a congenital abnormality of the salivary gland ducts. Congenital malformation was believed to be responsible for retrograde infection, which was further worsened by dehydration. Reid et al. proposed an explanation that involved hereditary genetic factors in a model of autosomal inheritance with incomplete penetrance. Taking into consideration the various theories and endoscopic observations of parotid glands at different ages, we suggest that there is an inadequacy between the excretion and the secretion capacities of parotid glands in children with symptomatic JRP. This inadequacy may cause symptoms as the secretion of the gland increases. Also, we make a distinction between local and global stenoses. Local stenoses are usually found in adults and could be due to inflammation. Global stenosis of the main duct and secondary branches of the ductal tree seems more suggestive of a congenital origin.

Until now, therapeutic abstention was a traditional and recognized attitude based on the disappearance of the signs in 95% of the cases before puberty. However, recurrent swelling over a long period can often cause repercussions in a patient’s social life and school activities. Furthermore, a few patients have developed sequelae such as chronic recurrent pain with chronic recurrent parotid swelling.

Galili and Marmar were the first to use lavage to treat JRP with sialography, with good results in 13 of the 15 children with unilateral swellings. Symptoms subsided in 5 of 7 children with bilateral episodes. Nahlieli et al. in a series of 26 cases of JRP treated by dilatation and abundant washing under endoscopic control, achieved a resolution of symptoms in 92% of the cases with 36 months of follow-up. Yet, preliminary sialography had been performed in all of their patients.

Our study was limited by a short follow-up period in patients with a disease that has a variable recurrence rate. It is unknown whether the lack of recurrence within this short follow-up period was the result of the natural history of the disease or the procedure itself. Moreover, the lack of a control group of nonsymptomatic children makes it difficult to generalize about the endoscopic findings. Obviously, it is unethical to perform this procedure on healthy children. However, in a previous pediatric series of sialolithiasis treated by sialendoscopy, we did not observe such narrow ducts or the white aspect of the Stensen ducts. Our findings confirm this preliminary therapeutic result, with 9 of 10 cases of JRP (89%) successfully treated by complete dilatation.

According to Galili and Marmar, the severity of JRP is established by the frequency of recurrence. We decided to perform sialendoscopy when patients reported a minimum of 2 episodes of swelling within 6 months. We undertook dilatation only on the side of the JRP attacks. In our series, 2 patients developed episodes of JRP secondarily, contralateral to the treated side. This observation should encourage bilateral systematic sialendoscopy, even in cases in which the JRP episodes are unilateral.

The rate of good results seems to be higher with sialendoscopy than with simple dilatation of the duct with a probe. We suggest that the forced high-pressure injection of water through the endoscope is more efficient than single-probe dilation of the Stensen duct. We hope that the diameter of the endoscope will decrease in the future so that exploration of the distal ducts of salivary glands and treatment of ductal disorders will be easier and more effective. Our hypothesis needs to be confirmed in a larger series, with a longer follow-up period. With the evolution of endoscopy, our knowledge of the anatomy of the salivary ductal tree will gradually continue to increase.

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Author Contributions: All authors had full access to all the data in the study and take responsibility for the integrity of the data and the accuracy of the data analysis. Study concept and design: Quenin, Marchal, Disant, and Faure. Acquisition of data: Quenin, Marchal, Disant, and Faure. Analysis and interpretation of data: Quenin, Plouin-Gaudon, Froehlich, and Faure. Drafting of the manuscript: Quenin and Faure. Critical revision of the manuscript for important intellectual content: Plouin-Gaudon, Froehlich, Marchal, Disant, and Faure. Statistical analysis: Quenin and Faure. Obtained funding: Faure. Administrative, technical, and material support: Quenin, Disant, and Faure. Study supervision: Plouin-Gaudon, Froehlich, Marchal, Disant, and Faure.

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