Objective: To show that sialoendoscopy is both a safe and effective alternative to traditional treatments for juvenile recurrent parotitis and sialolithiasis.

Design: Retrospective medical chart review.

Setting: Two major pediatric tertiary care centers.

Patients: Eighteen pediatric patients.

Interventions: A total of 33 sialendoscopic procedures on 27 glands.

Main Outcome Measures: Indications for surgery, age at onset of symptoms, age at procedure, sex, intraoperative findings, complications, recurrences, need for additional procedures, and follow-up interval.

Results: Juvenile recurrent parotitis was the most common indication for sialendoscopy (12 of 18) followed by sialolithiasis (4 of 18). Ten of 12 patients with juvenile recurrent parotitis were asymptomatic after 1 or 2 sialendoscopies (8 patients and 2 patients, respectively). There were 6 minor complications. Three patients ultimately required gland excision for disease management.

Conclusion: Sialoendscopy is safe and effective as a treatment for pediatric salivary gland disorders.


The treatment of adult salivary gland disorders with sialoendoscopy was popularized in the 1990s by several pioneers in this field, including Francis Marchal, MD, and Oded Nahlieli, DMD.1,2 As the technology has improved, its application has expanded, and there has been a drive to adapt this technique to the management of a variety of pediatric salivary gland disorders, including juvenile recurrent parotitis (JRP) and sialolithiasis.

Juvenile recurrent parotitis is one of the most common salivary diseases of children. It is an inflammatory condition characterized by recurrent and painful swelling of the parotid glands. Although episodes generally cease after puberty, in some cases the disease continues into adulthood.3,4 Surgical treatments have included ligation of the Stensen duct, total parotidectomy or Jacobson neuromectomy.5,6

Salivary stones are uncommon in children; however, when they do occur, it is severe obstructive symptoms that typically bring the child to seek medical attention.7,8 Occasionally, sialolithiasis is not diagnosed until the time of gland exploration because the lower limit of radiologic detection methods approximates 2 mm.9

This study reports the largest series of children treated with sialoendoscopy in the United States and reviews the indications and outcomes of the procedure. Pearls and pitfalls are noted to facilitate appropriate and effective application of this technique in children with salivary gland disease.

Methods

A retrospective medical chart review from September 2009 to June 2011 was conducted at the Children's Hospital of Pittsburgh (Pittsburgh, Pennsylvania) and the University of Iowa (Iowa City) after institutional review board approval was granted at each center. Information was gathered relating to patient demographics, indications, operative technique and interventions, intraoperative findings, complications, outcomes, and need for further interventions.

Patients with obstructive symptoms or with 2 or more episodes of parotitis were referred by the primary care physician or the emergency department to the outpatient otolaryngology clinic for evaluation. No attempt was made to actively recruit patients. Several patients had received imaging prior to referral,
but imaging was not ordered by the otolaryngology clinic. Patients with signs of active infection were treated preoperatively with antibiotics because it is a relative contraindication to sialendoscopy for it to be preformed on an actively infected gland. Patients were offered sialendoscopy after a careful discussion of the risks, benefits, and alternatives to treatment. After informed consent was obtained, the patients were then scheduled for the procedure.

All procedures were performed under general anesthesia with orotracheal intubation. Antibiotic prophylaxis for endocarditis was not indicated for any case. Preoperative medications included the avoidance of anticholinergic medications. Patients were positioned supine with an appropriately sized bite block to access Stensen or Wharton papillae. The selected duct was serially dilated using Marchal dilators until a 1.1-mm or 1.3-mm stent could be introduced (Karl Storz). Saline irrigation, intravenous tubing, was used to assist in advancing the endoscope by opening the duct by gentle irrigant pressure.

A holmium laser (Lumenis) with settings of 0.3 J, pulse rate of 5 per second, and power of 1.5 W was available for management of stones. To use, a 0.4-mm laser fiber was passed through the 0.65-mm working channel of the 1.3-mm endoscope and aimed at the stone under direct visualization.

Patients with JRP had either steroids or a steroid-antibiotic combination instilled into the gland. In addition, 1 stricture was addressed with balloon dilation after a short course of amoxicillin, and patient 4 had an intraoperative complication. She was noted to have transient swelling that resolved without intervention.

Table 1. Summary of Data for Patients With a Diagnosis of Juvenile Recurrent Parotitis

<table>
<thead>
<tr>
<th>Patient/ Sex/Age at Surgery, y</th>
<th>Events Prior to Surgery, No.</th>
<th>Age at Onset, y</th>
<th>Glands</th>
<th>Debris</th>
<th>Pale Mucosa</th>
<th>Stenosis</th>
<th>Complications</th>
<th>Recurrence, mo</th>
<th>Repeated Endoscopies, No.</th>
<th>Follow-up, mo</th>
</tr>
</thead>
<tbody>
<tr>
<td>1/M/10</td>
<td>4</td>
<td>7</td>
<td>B</td>
<td></td>
<td>B</td>
<td></td>
<td></td>
<td>5.0</td>
<td>5.0</td>
<td></td>
</tr>
<tr>
<td>2/M/12</td>
<td>7</td>
<td>2</td>
<td>B</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td>12.0</td>
<td></td>
</tr>
<tr>
<td>3/M/7</td>
<td>6</td>
<td>6</td>
<td>B</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td>4.0</td>
<td></td>
</tr>
<tr>
<td>4/F/16</td>
<td>NA</td>
<td>13</td>
<td>L</td>
<td>B</td>
<td>L</td>
<td>X</td>
<td></td>
<td>1</td>
<td>1</td>
<td>24.0</td>
</tr>
<tr>
<td>5/M/6</td>
<td>9</td>
<td>3</td>
<td>B</td>
<td></td>
<td>B</td>
<td></td>
<td></td>
<td></td>
<td>23.0</td>
<td></td>
</tr>
<tr>
<td>6/F/13</td>
<td>4</td>
<td>13</td>
<td>B</td>
<td></td>
<td>B</td>
<td></td>
<td></td>
<td>6.5</td>
<td>1</td>
<td>15.0</td>
</tr>
<tr>
<td>7/F/8</td>
<td>NA</td>
<td>NA</td>
<td>B</td>
<td></td>
<td>B</td>
<td></td>
<td></td>
<td></td>
<td>7.0</td>
<td></td>
</tr>
<tr>
<td>8/M/9</td>
<td>4</td>
<td>7</td>
<td>R</td>
<td>R</td>
<td>B</td>
<td></td>
<td></td>
<td>4</td>
<td>15.0</td>
<td></td>
</tr>
<tr>
<td>9/M/7</td>
<td>4</td>
<td>6</td>
<td>B</td>
<td></td>
<td>B</td>
<td></td>
<td></td>
<td></td>
<td>6.0</td>
<td></td>
</tr>
<tr>
<td>10/M/7</td>
<td>2</td>
<td>6</td>
<td>R</td>
<td>?</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td>1.0</td>
<td></td>
</tr>
<tr>
<td>11/F/6</td>
<td>3</td>
<td>5</td>
<td>L</td>
<td>L</td>
<td></td>
<td>X</td>
<td></td>
<td></td>
<td>1.5</td>
<td></td>
</tr>
<tr>
<td>12/F/16</td>
<td>NA</td>
<td>16</td>
<td>R</td>
<td>R</td>
<td>X</td>
<td></td>
<td></td>
<td>15 glands</td>
<td>2 glands</td>
<td>6.5</td>
</tr>
</tbody>
</table>

Mean 4.7 7.7 78:2L:3R 9 Pts; 1 Pt; 3 Pts; 3 Pts 15 glands 2 glands 4 glands

Abbreviations: B, bilateral; L, left; NA, not available (data could not be determined from the written medical history and were therefore not available for inclusion); pt, patient; R, right; X, present.

a Seven boys and 5 girls; mean age at surgery, 9.7 years (range, 5-16 years).

Juvenile recurrent parotitis was the most frequent indication for surgery, with 12 children represented and 19 glands involved. The second most common indication was sialolithiasis, which was diagnosed in 3 children preoperatively, although stones were found in a fourth child at sialendoscopy. This child, who had had a hemolymphangiomatous of the right parotid excised via superficial parotidectomy at age 2 years, presented at age 9 years with a 1-year history of recurrent parotitis and was found to have small stones. The remaining 2 patients had submandibular pain and swelling with eating.

INDICATIONS

The findings and outcomes are divided into groups based on diagnosis. Table 1 shows the patients treated for JRP. Patient 5 also had a medical history significant for common variable immunodeficiency; patient 12 had a history of recurrent right parotid swelling and an outside computed tomographic (CT) imaging report noting the presence of a 4-mm stone. Review of the scan suggested that a stone was unlikely, and on sialendoscopy no stone was seen. The medical history and examination were therefore more consistent with the diagnosis of JRP.

Three patients with JRP had complications: patient 11 had transient swelling that resolved without intervention, patient 12 had increased pain at 1 week that resolved with a short course of amoxicillin, and patient 4 had an intraoperative complication. She was noted to have a stricture 1 cm from the papilla that was balloon dilated to 4 mm. Fearing a possible ductal breech owing to the appearance of the duct, a stent fashioned from a 3-Fr feeding tube was sutured in place. The stent was re-
moved in the operating room 5 days later, with sialendoscopy showing no ductal injury. The patient, however, continued to have the same symptoms of recurrent swelling and pain, ultimately requiring a parotidectomy to address her disease. Three patients had recurrent symptoms after the first sialendoscopy. Overall, 8 patients required only 1 procedure to address JRP symptoms, 2 patients required 2 procedures, 1 patient required parotidectomy, and 1 patient was lost to follow-up.

Data for the 4 patients with salivary stones are presented in Table 2. One patient had stones in the parotid, and in 2 of the 3 patients with submandibular gland sialoliths, stones were present in both submandibular ducts. The patient with parotid stones, patient 4, presented 8 years after a right superficial parotidectomy for a hemolymphangioma and was found to have small stones and debris in the parotid duct. Findings from follow-up sialendoscopy were normal, and the child has remained asymptomatic to date (24 months).

There were 2 complications in this group. Patient 3 had a 7-mm stone in the right submandibular gland that could not be removed using a combined approach (ductal cutdown, removing the stone transorally followed by endoscopy to ensure there are no additional sources of proximal or distal obstruction). There was difficulty isolating the stone intraorally owing to stone position, and the procedure was aborted. Therefore, the gland was excised. Patient 2 also required gland excision to address a 3-mm stone when the laser tip became embeded in the stone and broke off on attempted withdrawal of the laser.

Two patients had pain and swelling of a submandibular gland with eating. One child was asymptomatic after stone removal by a community physician but after sialendoscopy and dilation of a strictured papilla, he had been asymptomatic. The other child has also been asymptomatic to date after dilatation of the papillae and sialendoscopy.

### Table 2. Summary of Patient Data for Patients With a Diagnosis of Sialolithiasis

<table>
<thead>
<tr>
<th>Patient/ Sex/Age at Surgery, y</th>
<th>Indication</th>
<th>Age at Onset, y</th>
<th>Glands</th>
<th>Findings</th>
<th>Repeated Endoscopies, No.</th>
<th>Gland Excision</th>
<th>Follow up, mo</th>
</tr>
</thead>
<tbody>
<tr>
<td>1/F/7</td>
<td>Swelling</td>
<td>6</td>
<td>B SMG</td>
<td>Stones: 5-mm L, 2-mm R; Stenosis: X</td>
<td>X</td>
<td>L</td>
<td>31</td>
</tr>
<tr>
<td>2/M/12</td>
<td>Stone</td>
<td>11</td>
<td>L SMG</td>
<td>Stones: 3-mm; Stenosis: L, X</td>
<td>R</td>
<td>R</td>
<td>8</td>
</tr>
<tr>
<td>3/M/8</td>
<td>Recurrent swelling</td>
<td>7</td>
<td>B SMG</td>
<td>Stones: 7-mm R, small L; Stenosis: X</td>
<td>X</td>
<td>X</td>
<td>2</td>
</tr>
<tr>
<td>4/M/10</td>
<td>Recurrent swelling</td>
<td>2</td>
<td>R Parotid</td>
<td>Stones: Small R, Small, 7-mm; Stenosis: 1, 2</td>
<td>1, 2</td>
<td>23</td>
<td>15</td>
</tr>
<tr>
<td>Mean</td>
<td>6.5</td>
<td></td>
<td></td>
<td>Stones: Mean Stenosis: 2</td>
<td>1</td>
<td>2</td>
<td>15</td>
</tr>
</tbody>
</table>

| Abbreviations: B, bilateral; L, left; R, right; SMG, submandibular gland; X, present. | 
| Three boys and 1 girl; mean age at surgery, 9.25 years. |

The minimum requirement for diagnosing JRP is 2 episodes of sialadenitis per year, whereas the severity is determined by the frequency of episodes. Our series was variable in the number of episodes prior to endoscopic management, with an average of 4.7 events and a range of 2 to 9 events. The 1 child who underwent sialendoscopy after only 2 events experienced those events within a single month. Because the natural history of the disease is usually self-limited, a careful discussion with the family must be included in an informed consent. However, it should also be recognized that some children have been noted to have persistent disease into adulthood.3

In aggregate, the literature reports on approximately 100 children with JRP who underwent sialendoscopy. Shacham et al in Israel reported on 70 children treated from 1993 to 2007 for JRP. In 93% of their patients, a single endoscopy was sufficient for symptom resolution. Martins-Carvalho et al in France and Switzerland reported on the second largest group of children with JRP, noting that similar numbers (19 of 23 patients [83%]) required only a single procedure. Konstantinidis et al in Greece and Jabbour et al in Minnesota reported on 7 and 5 patients, respectively, with JRP. One child in the Greek group required a repeated endoscopy, and 2 others had 1 recurrent episode within 1 year (83% improvement with 1 procedure), while in the Minnesota group, 3 children had 1 or 2 recurrent episodes, but their frequency of symptoms was uniformly reduced. In our series, only 4 patients had recurrent symptoms, and 10 of the 11 patients with follow-up (91%) were asymptomatic after 1 or 2 endoscopies. The short-term success rate of sialendoscopy is therefore over 80%, but longer follow-up time is needed to assess long-term efficacy.

Sialendoscopy for JRP is both diagnostic and therapeutic. Common findings include a widened Stensen papilla; a white, avascular duct on endoscopy; and stenosis with debris within the duct. The therapeutic portion of the procedure is the mechanical washout of debris, the hydrostatic pressure dilation of the duct, and the instillation of steroids and/or antibiotics, although evidence-based data supporting the effectiveness of steroid and/or antibiotic infusions directly into the duct are lacking. To our knowledge, a study to validate the efficacy of medical treatments, in addition to dilating the papilla and irrigating the gland, has not been done.

Although small, our series of sialoliths highlights the necessity for diverse treatment options to address stones.
of varying sizes. When endoscopic or hybrid procedures are unsuccessful, gland removal can be completed. We believe, however, that sialendoscopy should be the first approach because it preserves the gland and avoids the small but present complications that can occur with gland excision.

Marchal and Dulguerov\textsuperscript{13} presented an algorithm for management of stones in adults based on the size of the stone. Small stones could be managed entirely endoscopically with forceps or basket devices, whereas 3- to 6-mm stones should first be fragmented with a laser. Stones larger than 6 mm should be managed via a hybrid approach involving ductal cut-down on to the stone and endoscopy to ensure that the remainder of the ductal system is free of stones and debris. This algorithm has not yet been adapted to pediatric sialolithiasis, in part because of the relative rarity of the problem. Nahlieli et al\textsuperscript{9} reported on 15 children with stones successfully managed endoscopically. Martins-Carvalho et al\textsuperscript{10} added an additional 12 patients to this number. Three patients in their series did undergo submandibular gland excision, although they did not give details relating to the decision to proceed with gland excision.

In conclusion, sialendoscopy is a safe and effective technique to use in conjunction with other medical and surgical treatments of salivary gland disorders in pediatric patients. Children most commonly present with JRP, which usually resolves after sialendoscopy. Informed consent should include a discussion of the natural history of JRP, and in patients with obstructive symptoms suggesting a stone, the preoperative discussion should include the possibility of gland excision. Research is needed to determine whether instilling antibiotics and steroids into the gland has an impact on the course of JRP and to improve the endoscopic management of sialoliths in children.

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**Analysis and interpretation of data:** Hackett, Baranano, Smith, and Mehta.

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**Critical revision of the manuscript for important intellectual content:** Hackett, Baranano, Reed, Duvvuri, Smith, and Mehta.

**Study supervision:** Duvvuri and Smith.

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**REFERENCES**