IMPORTANCE  The 4-duct ligation procedure has appeal for its surgical simplicity and limited invasiveness in the management of pediatric sialorrhea. However, more information is required to understand the benefits, risks, success rates, and long-term effects of this intervention.

OBJECTIVES  To report the clinical outcomes of the 4-duct ligation procedure in pediatric patients diagnosed as having sialorrhea and the associated complication rates and to characterize patient and caregiver satisfaction in a consecutive series.

DESIGN, SETTING, AND PARTICIPANTS  This investigation was a retrospective cohort study at an academic tertiary pediatric center and pediatric rehabilitation hospital. Patients included 38 children with neurological impairment who underwent a 4-duct salivary gland ligation (parotid and submandibular glands) between January 1, 2004, and July 31, 2012. The dates of the analysis were August 2013 through February 2015.

MAIN OUTCOMES AND MEASURES  Posttreatment assessments included duration of effect, severity and frequency of drooling before and after the procedure, patient complications, caregiver satisfaction, caregiver recommendation of the procedure, and caregiver overall assessment of the child's quality of life. Clinical and outcome measures were collected before the procedure, 1 month after the procedure, 1 year after the procedure, and at the most recent follow-up (range, 3-8 years).

RESULTS  The study cohort comprised 38 participants. Their median age was 11 years (age range, 5-17 years), and 37% (14 of 38) were female. The mean (SD) duration of effect was 52.6 (20.4) months. Patients with previous sialorrhea management were more likely to demonstrate an improvement in their drooling frequency score at 1 year. Thirteen complications were documented in 12 patients. The most common complications were persistent facial swelling and aspiration pneumonia. Eighty percent (28 of 35) of caregivers reported an improvement in their child's drooling at 1 month, while 69% (25 of 36) and 71% (24 of 34) stated that there was an improvement at the 1-year follow-up and the most recent follow-up, respectively.

CONCLUSIONS AND RELEVANCE  The 4-duct ligation procedure offers a simple, effective, and minimally invasive approach to the management of sialorrhea in children.
Sialorrhea is defined as spillage of saliva due to decreased frequency of swallowing (secondary sialorrhea) or excessive saliva production (primary sialorrhea). It can be further characterized as either poor anterior or posterior saliva control. Anterior sialorrhea (or drooling) occurs when saliva leaves the oral cavity and spills from the mouth. Posterior sialorrhea results in the pooling of secretions in the hypopharynx, which increases the risk of aspiration pneumonia.3

Saliva is produced by 6 major salivary glands (2 parotid, 2 submandibular, and 2 sublingual) working in conjunction with several hundred minor salivary glands.1,2 Patients with neurological disorders often experience sialorrhea. In children with cerebral palsy (CP), excessive drooling occurs in an estimated 10% of cases.1,4 Other health complications that may cause or contribute to drooling include dental-related problems, poor head control and posture, nasal obstruction, and facial paralysis.2,5

The physical and psychosocial implications of sialorrhea are a significant source of distress for patients and their caregivers. Drooling can cause physical morbidity (eg, impaired masticatory function, interference with speech, and increased likelihood of perioral infections) and result in the loss of fluid, electrolytes, and proteins.2,5,6 Beyond the physical implications of sialorrhea, caregivers are most concerned about the individual's socialization and risk of social isolation, embarrassment, and stigmatization.2,5 Overall, it is not uncommon for patients with excessive salivation to experience both a physical and an emotional barrier to the outside world.6

The management of sialorrhea in a pediatric population requires a multidisciplinary team-based approach to understand the wide range of contributing factors and patient and family goals and to determine the most appropriate therapeutic approach. A series of evaluations is necessary for each patient and includes a review of the medical history, medications, cognitive abilities, speech, language, and level of alertness.5,7 When conservative treatment measures, such as behavior modifications, physical therapy, and pharmacotherapy fail, to manage drooling, surgical options are recommended. Surgical therapy includes sublingual gland excision, parotid duct ligation, parotid duct rerouting, and submandibular gland excision, rerouting, or ligation. A combination of these procedures is often performed.1,5

Concurrent ligation of the parotid and submandibular gland ducts (4-duct ligation) has been described in the literature as a viable first-line surgical option to manage sialorrhea because of its simplicity and limited invasiveness.6,7 However, the reported success rate has been variable.1,8,9 Authors of a 2009 meta-analysis on the surgical management of drooling have strongly suggested that more studies focused on long-term safety and efficacy are required. The objectives of this retrospective cohort study were to report the clinical outcomes of the 4-duct ligation procedure and the associated complication rates and to characterize patient and caregiver satisfaction in a consecutive series of individuals treated by a single surgeon (P.C.) at The Hospital for Sick Children (SickKids), Toronto, Ontario, Canada.

Methods

Participants

This study was a retrospective medical record review of consecutive pediatric patients who underwent a 4-duct salivary gland ligation (parotid and submandibular glands) between January 1, 2004, and July 31, 2012. The dates of the analysis were August 2013 through February 2015. The cohort included patients treated with 4-duct ligation exclusively and those who had previous behavioral, physical, botulinum toxin A, or pharmacotherapy treatment for drooling. No patients were excluded from the review, and none received concurrent or ongoing medical therapy for their sialorrhea after 4-duct ligation.

The patients were evaluated before the procedure at a multidisciplinary Saliva Management Clinic at Holland Bloorview Kids Rehabilitation Hospital in Toronto. The multidisciplinary team included an otolaryngologist (P.C.), an outside dentist, speech-language pathologists (C.A.M. and R.P.), and a social worker (M.S.J.). The 4-duct ligation procedure was performed at The Hospital for Sick Children (SickKids). Posttreatment assessments were completed at the multidisciplinary clinic or by telephone. All of the patients were assessed in clinic at least once. The caregivers of 7 of the patients contacted the clinic by telephone for one of their assessments. The Holland Bloorview Research Ethics Board and the Hospital for Sick Children Research Ethics Board approved this study.

Operative Technique

Surgery was performed using general anesthesia with nasotracheal intubation. At the start of the procedure, patients received prophylactic antibiotics intravenously, including clindamycin hydrochloride (10 mg/kg of body weight) and dexamethasone sodium phosphate (2-4 mg). The neck was placed in extension with a shoulder roll.

The oral cavity was exposed using a Boyle-Davis mouth gag. A rectal retractor was used to retract the cheek. A size 0 lacrimal probe was inserted into the parotid duct opening and advanced toward the gland. Lidocaine hydrochloride was then applied after cannulation. Infusion of lidocaine before cannulation may obscure and prevent cannulation of the duct, which is a critical step. One milliliter of lidocaine hydrochloride, 1%, with epinephrine 1:100,000 was injected around the ductal orifice. The mucosa anterior to the duct opening was transected using bipolar cautery approximately 5 mm from the duct. The incision was made from the 6 o’clock to 12 o’clock positions. Guided by the lacrimal probe, dissection was performed along the duct submuscosally for a length of approximately 0.5 cm. Isolation of the duct required transection of the buccal fascia and surrounding muscles. A 3-0 polyester fiber suture was then applied around the isolated duct as the lacrimal probe was withdrawn. The suture knot was firmly applied to the duct. The mucosa and fascia were then sutured with interrupted, overlying 4-0 chromic sutures.

To ligate the submandibular ducts, the Boyle-Davis mouth gag was removed and replaced with a 2-prong, self-retaining retractor. The tip of the tongue was then temporarily sutured to the base of the uvula to expose the anatomy of the floor of
mouth. Lidocaine hydrochloride (1-2 mL) was then infiltrated submucosally immediately posterior to the submandibular duct papillae. A submucosal incision was made around the papilla using curved sharp scissors approximately 1 cm posterior to the duct openings. Submucosal dissection was then performed to expose the distal ends of both submandibular ducts through the same incision. The submandibular ducts were then ligated with 3-0 polyester fiber sutures, and the overlying mucosa was closed with interrupted 4-0 chromic sutures.

Patients recovered in a postanesthetic care unit and were admitted to a step-down unit for typically a 2-day observation period unless other medical conditions or complications were observed. They were given parenteral antibiotics for 2 days and were maintained on oral antibiotics (clindamycin) for 1 week after surgery.

Outcome Measures
Outcome measures were collected before the procedure, 1 month after the procedure, 1 year after the procedure, and over a follow-up period of 3 to 8 years. Severity and frequency of drooling before and after the procedure were graded using the scale by Thomas-Stonell and Greenberg. Severity of drooling is scored as dry, mild (wet lips only), moderate (wet lips and chin), severe (clothing damp), or profuse (clothing, hands, and objects wet). Frequency of drooling is scored as never drools, occasionally drools, frequently drools, or constantly drools. This scale is validated, and a 1-point change in severity, frequency, or both indicates a clinically significant and detectable change. Duration of effect was defined as the period during which there was a subjective decrease in the amount of saliva spillage observed by the caregiver. The period extends to the last follow-up but may persist beyond that time. Other outcome measures recorded were caregiver satisfaction and caregiver overall assessment of the child’s quality of life, which were measured as the percentage of caregivers who responded yes, no, or not applicable. Any symptom or problem reported by the patient or caregiver was labeled as a complication. Postprocedural assessments were conducted through outpatient clinic visits and follow-up telephone interviews with a primary caregiver.

Statistical Analysis
Data were analyzed using a statistical software package (SAS for Windows, version 3.1; SAS Institute Inc). Given that this investigation was a retrospective study, there was no sample size calculation performed. Rather, all available data from patients who underwent a 4-duct ligation procedure for sialorrhea management were included in the final analysis. The results presented herein are descriptive (eg, observed procedural outcomes, safety, and efficacy) and analytical (eg, hypothesis-generating regression models). Summary statistics were used to describe the patient cohort, while \( \chi^2 \) test or Fisher exact test and logistic regression were used for inferences. For linear regression analysis, any response variable that was not normally distributed was log transformed before analysis. \( P \leq .05 \) was considered statistically significant.

Results

Patient Characteristics

Between January 1, 2004, and July 31, 2012, a total of 38 patients underwent a 4-duct ligation procedure for the treatment of sialorrhea. Of the 38 patients, 34 had postoperative data collected over a follow-up period of 3 to 8 years. The data collection period for the remaining 4 patients varied between 1 month and 1 year after the procedure. Patient demographics and clinical characteristics are summarized in Table 1. The me-
Four-Duct Ligation for the Treatment of Sialorrhea in Children

Original Investigation | Research

Table 2. Drooling Severity Scores and Drooling Frequency Scores at Various Time Points

<table>
<thead>
<tr>
<th>Variable</th>
<th>Drooling Severity Score</th>
<th></th>
<th></th>
<th></th>
<th>Drooling Frequency Score</th>
<th></th>
<th></th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Before the Procedure</td>
<td>1 Year After the Procedure</td>
<td>At the Most Recent Follow-up</td>
<td>Before the Procedure</td>
<td>1 Year After the Procedure</td>
<td>At the Most Recent Follow-up</td>
<td></td>
</tr>
<tr>
<td></td>
<td>(n = 37)</td>
<td>(n = 33)</td>
<td>(n = 33)</td>
<td>(n = 37)</td>
<td>(n = 33)</td>
<td>(n = 33)</td>
<td>(n = 33)</td>
</tr>
<tr>
<td>Mean (SD)</td>
<td>4.6 (0.4)</td>
<td>3.0 (1.9)</td>
<td>3.9 (1.2)</td>
<td>3.9 (0.3)</td>
<td>2.5 (1.3)</td>
<td>3.2 (0.9)</td>
<td></td>
</tr>
<tr>
<td>P value across groupsa</td>
<td>&lt;.001</td>
<td>&lt;.001</td>
<td>&lt;.02</td>
<td>&lt;.001</td>
<td>&lt;.001</td>
<td>.01</td>
<td></td>
</tr>
</tbody>
</table>

a Groups were compared using the independent samples t test.

Table 3. Complications Experienced by Patients After the 4-Duct Ligation Procedure

<table>
<thead>
<tr>
<th>Complication</th>
<th>Total No. of Occurrences</th>
<th>No. of Occurrences at Each Follow-up</th>
<th>Total No. of Occurrences</th>
<th>No. of Occurrences at Each Follow-up</th>
</tr>
</thead>
<tbody>
<tr>
<td>Aspiration pneumonia</td>
<td>3</td>
<td>3 at 1 mo</td>
<td>3</td>
<td>3 at 1 mo</td>
</tr>
<tr>
<td>Persistent facial swelling &gt;1 wk</td>
<td>3</td>
<td>3 at 1 mo</td>
<td>3</td>
<td>3 at 1 mo</td>
</tr>
<tr>
<td>Oxygen desaturation</td>
<td>3</td>
<td>3 at 1 mo</td>
<td>3</td>
<td>3 at 1 mo</td>
</tr>
<tr>
<td>Vomiting</td>
<td>2</td>
<td>2 at 1 mo</td>
<td>2</td>
<td>2 at 1 mo</td>
</tr>
<tr>
<td>Delayed reinitiation of oral feeding</td>
<td>1</td>
<td>1 at 1 mo</td>
<td>1</td>
<td>1 at 1 mo</td>
</tr>
<tr>
<td>Recurrent parotid gland swelling</td>
<td>1</td>
<td>1 at 1 y</td>
<td>1</td>
<td>1 at 1 y</td>
</tr>
</tbody>
</table>

Median age of the patients was 11 years (range, 5–17 years), and the mean (SD) duration of hospitalization after the procedure was 2.7 (1.3) days (range, 1–6 days).

Sialorrhea Outcome
At the most recent follow-up, the mean (SD) duration of effect was 52.6 (20.4) months (range, 1–96 months) among 34 patients. Univariable linear regression analyses revealed no significant association between duration of effect (log transformed) and age, sex, previous sialorrhea management, mobility (ability to ambulate unassisted), head control, oral motor impairment, drooling frequency, and drooling severity at 1 month, 1 year, and the most recent follow-up (P > .05 for all). However, patients with an underlying diagnosis other than CP were more likely to have no improvement in drooling at 1 month after the procedure (P = .001). The difference was no longer significant at 1 year and the most recent follow-up (P > .05 for both).

Using a paired t test, a significant difference was observed when comparing the mean drooling frequency and drooling severity scores before the procedure, 1 year after the procedure, and at the most recent follow-up (Table 2). Univariable linear regression analyses revealed no significant association between drooling frequency and drooling severity scores (log transformed) and age, sex, underlying diagnosis, mobility, head control, and oral motor impairment at 1 month and 1 year after the procedure (P > .05 for all). However, patients with previous sialorrhea management were more likely to demonstrate an improvement in their drooling frequency score at 1 year (P = .03).

Caregivers reported that their child demonstrated mean (SD) percentage improvements in drooling of 62% (32%) at 1 month after the procedure (n = 35) and 47% (35%) at the most recent follow-up (n = 34) (range, 0%-100% at both intervals). The difference in the percentage improvements in drooling between these intervals was not significant (P = .05). Of the 34 patients whose follow-up extended beyond 1 year, 26 patients (76%) had no return of their drooling. Seven patients (21%) experienced a return of their drooling, and the response to surgery was unknown in 1 patient (3%). The mean (SD) duration of drooling improvement was 3.5 (4.0) months for the 7 patients who experienced a return of their drooling.

Complications
Patient complications were captured at 1 month and 1 year after the procedure. In total, 13 complications were documented in 32% (12 of 38) of the patients (Table 3). The most common complications were persistent facial swelling (23% [3 of 13]) and aspiration pneumonia (23% [3 of 13]). Other observed complications included oxygen desaturation (23% [3 of 13]) and vomiting (15% [2 of 13]). One patient required placement of a nasogastric tube for feeding because of delayed reinitiation of oral intake. Another patient experienced recurrent parotid gland swelling at 1 year after treatment, with no improvement in the sialorrhea. Univariable linear regression analyses found no significant association between the reporting of a complication (log transformed) and sex, previous sialorrhea management, mobility, head control, drooling frequency score, and drooling severity score at 1 month and 1 year after the procedure (P > .05 for all). No deaths occurred.

Caregiver Satisfaction and Overall Assessment of the Child’s Quality of Life
After the procedure, 28 of 35 caregivers (80%) reported an improvement in their child’s drooling at 1 month, while 25 of 36 (69%) and 24 of 34 (71%) caregivers stated that there was an improvement at the 1-year and most recent follow-ups, respectively. Caregiver overall satisfaction with the procedure at 1 month was 63% (17 of 27) and decreased to 53% (18 of 34) at the most recent follow-up.

Caregivers had mixed opinions on whether they would recommend this procedure to others. When asked at 1 month after the procedure, 11 of 20 caregivers (55%) stated that they would suggest the procedure, while 18 of 34 caregivers (53%) expressed the same sentiment at their most recent follow-up. Variables significantly associated with caregiver overall satisfaction in univariable logistic regression models included underlying diagnosis and improvement in drooling. Caregivers of patients with an underlying diagnosis other than CP were less likely to be satisfied with the procedure at 1 month (P = .001).
Table 4. Pediatric Studies Examining the Treatment of Sialorrhea With the 4-Duct Ligation Procedure

<table>
<thead>
<tr>
<th>Source</th>
<th>Design</th>
<th>No. of Patients</th>
<th>Follow-up, mo</th>
<th>Success Rate, %</th>
<th>Recurrence Rate, %</th>
<th>Timing of Recurrence After the Procedure, mo</th>
</tr>
</thead>
<tbody>
<tr>
<td>Present study</td>
<td>Retrospective</td>
<td>38</td>
<td>≤102</td>
<td>77</td>
<td>24</td>
<td>3.5</td>
</tr>
<tr>
<td>Chanu et al., 11</td>
<td>Prospective</td>
<td>30</td>
<td>12</td>
<td>93</td>
<td>NA</td>
<td>NA</td>
</tr>
<tr>
<td>El-Hakim et al., 12</td>
<td>Retrospective</td>
<td>5</td>
<td>≤18</td>
<td>100</td>
<td>0</td>
<td>0</td>
</tr>
<tr>
<td>Stamataki et al., 2</td>
<td>Retrospective</td>
<td>13</td>
<td>≤36</td>
<td>30</td>
<td>61</td>
<td>9</td>
</tr>
<tr>
<td>Martin and Conley, 7</td>
<td>Retrospective</td>
<td>16</td>
<td>≤48</td>
<td>31</td>
<td>69</td>
<td>6</td>
</tr>
<tr>
<td>Shirley et al., 8</td>
<td>Retrospective</td>
<td>21</td>
<td>≤14</td>
<td>81</td>
<td>19</td>
<td>NA</td>
</tr>
<tr>
<td>Klem and Mair, 9</td>
<td>Retrospective</td>
<td>5</td>
<td>≤15</td>
<td>100</td>
<td>0</td>
<td>0</td>
</tr>
</tbody>
</table>

Abbreviation: NA, not available.

Discussion

There are numerous surgical treatments used in the management of sialorrhea, including submandibular gland excision, submandibular gland duct rerouting, sublingual gland excision, submandibular gland duct ligation, parotid duct rerouting, parotid duct ligation, and any combination of these treatment modalities. However, botulinum toxin A administration to the parotid and submandibular glands remains the most commonly used first-line treatment modality at The Hospital for Sick Children (SickKids). There are several scenarios in which a surgical approach, such as the 4-duct ligation procedure (ligation of the parotid and submandibular gland ducts), is considered or preferred. In addition to patient or caregiver preference, these situations include the following: botulinum toxin A therapy fails to manage the sialorrhea, the patient is at high risk of developing aspiration pneumonia, the patient is at high risk of anesthetic complications, the patient lives a long distance from the treating institution that makes successive treatments onerous, or the patient has very severe sialorrhea. In these instances, the 4-duct ligation procedure offers a minimally invasive technique that has been reported to be efficacious, is easy to perform, and causes few complications.13-15 The procedure immediately decreases the volume of saliva entering the oral cavity. Over time, it is believed that the ligated glands eventually atrophy and cease to produce saliva. In contrast, the sublingual and minor salivary glands continue to produce saliva and may upregulate saliva production to maintain dental health and prevent xerostomia.10

After the first report on 4-duct ligation by Klem and Mair9 in 1999, few studies3,7,8,11,12 have assessed the safety and efficacy of the 4-duct ligation procedure (Table 4).

To our knowledge, the present study represents the largest cohort of children treated with the 4-duct ligation procedure and observed for a significant period. Moreover, it is the only report to date to examine patients at several follow-up time points and assess patients who underwent previous sialorrhea treatments. One of the primary outcomes measured was the efficacy of the technique. The results herein demonstrate that most patients (26 of 34) with follow-up beyond 1 year experienced an improvement in their sialorrhea. However, 7 of 34 children had a recurrence of drooling approximately 3.5 months after the procedure. This result is comparable to the findings in other studies3,7,8 assessing the 4-duct ligation technique. One possibility for the recurrence of sialorrhea is the recanalization of the ligated ducts. Another possible explanation is the formation of accessory openings along the ducts that develop spontaneously or because of intraoperative trauma.7,12

Similar to previous findings, most of our patient caregivers reported a subjective reduction in the amount of saliva after 4-duct ligation.6,9,11,12 The mean duration of effect herein was 52.6 months at the most recent follow-up, which is much longer than what has been reported for 4-duct ligation in the literature.3,7,9,11,12 As expected, the observed duration of effect is significantly longer than what has been observed for botulinum toxin A injections, which is approximately 4.5 months.12-15 Patients herein with a diagnosis other than CP were less likely to have an improvement at the 1-month follow-up. However, this observation diminished after 1 year, suggesting either a change in caregiver perception or a delay in observable improvement.

Within the cohort studied, a notable fluctuation was observed in the mean drooling frequency scores and the mean drooling severity scores. Scores were high before the procedure, decreased at 1 year after the procedure, and increased again at the most recent follow-up. However, scores remained significantly below the baseline measurement. Children with progressive neurological conditions, the improvement in sialorrhea may gradually decrease because of deterioration in the patient’s posture, neck muscle control, or cognitive level.16

To assess caregiver satisfaction, a posttreatment questionnaire was administered. Sixty-three percent (17 of 27) of caregivers were satisfied with the results of this technique at 1 month, while only 53% (18 of 34) of caregivers maintained this sentiment at the most recent follow-up. Overall improvement in sialorrhea was higher than the satisfaction scores, with caregivers stating improvement rates of 80% (28 of 35), 69% (25 of 36), and 71% (24 of 34) at 1 month, 1 year, and the most recent follow-up, respectively. This discrepancy may be explained by changes in caregiver expectations over time and by a confounding effect caused by other patient comorbidities.

Approximately one-third (12 of 38) of our patient cohort experienced at least 1 complication. This complication rate is consistent with other studies3,7,9,11,12 describing the efficacy of the 4-duct ligation procedure. Although we cannot defini-
tively state which complications were the result of the procedure or the anesthesia, we intuitively recognize persistent facial swelling, delayed reinitiation of oral feeding, and recurrent parotid swelling as being more related to the procedure. The most serious complication observed was aspiration pneumonia in 3 patients within the first month after treatment. Aspiration pneumonia, oxygen desaturation, and vomiting are common complications in this patient population, especially if the patient has compromised neurological function. As such, postoperative admission to a monitored setting is recommended. Only 1 patient in the cohort experienced recurrent unilateral parotid gland swelling at 1 year after treatment. These episodes were managed conservatively with warm compresses, facial massage, and oral antibiotics. None of the patients developed xerostomia.

There are several limitations inherent in this study, including the retrospective nature of our data collection, a possible overstatement of complications, and the use of subjective questionnaires to measure outcomes. The wide range in follow-up periods also demonstrates the challenge in establishing an assessment at consistent time points. In addition, the study groups were small, and the patients had complicated medical conditions that may have affected the outcome measurements.

Conclusions

The 4-duct ligation procedure offers a simple, effective, and minimally invasive approach to the management of sialorrhea in children. Nonetheless, further studies (including randomized clinical trials) are required to address specific questions relating to patient selection, optimal timing of effect, and the risks of this procedure. Continued research will help standardize treatment protocols and thereby improve the quality of life of affected children and their caregivers.

REFERENCES