Major Salivary Duct Clipping for Control Problems in Developmentally Challenged Children

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Objectives: To introduce a technical modification for interruption of the parotid and submandibular salivary ducts and to demonstrate the technique's effectiveness and effect on quality of life.

Design: Retrospective, uncontrolled, consecutive case series.

Setting: A tertiary care pediatric otolaryngology practice.

Patients: Eighteen drooling and aspirating children.

Intervention: Transoral interruption of parotid and submandibular ducts using vascular clips.

Main Outcome Measures: Manifestations of poor saliva control (visible drooling, number of shirts and bibs used, choking episodes, embarrassment, and incidence of salivary aspiration), Glasgow Children's Benefit Inventory scores (possible score range, −100 to +100), and complications.

Results: Eighteen patients (10 boys and 8 girls) were treated in 14 months. Patient age ranged from 2 to 14 years. Follow-up ranged from 3 to 18 months. No complications occurred. Nine patients had no drooling at all after surgery. There was a significant reduction in the number of bibs and shirts used ($P < .001$). Regarding measures indicating circumoral skin problems, embarrassment, choking episodes, and aspiration pneumonia, all the patients had significant improvements after surgery. The mean Glasgow Children's Benefit Inventory score was 33.2.

Conclusions: Salivary duct clipping is an efficient and safe method of controlling saliva in neurologically challenged children. The operation positively affects the children's quality of life.


Efficient saliva control in children is a complex management problem. Almost two-thirds of all patients with cerebral palsy drool, and in one-third the drooling is severe. Treatment with systemic anticholinergic drugs is decreasing in popularity because of their frequent adverse effects and contraindications. Intraglandular injection of botulinum toxin type A has recently received a great deal of interest, although the effect is temporary and variable in many cases, and patients will, therefore, require subsequent treatments. Across many years, numerous surgical solutions have been reported. Mostly, they are complex procedures with considerable morbidity.

In 1999, Klem and Mair described a technique for ligating submandibular and parotid ducts. Shirley et al subsequently reported a case series using 4-duct ligation with good results in 21 patients. This technique, however, has not been reported in wider use since. Herein, we report on a modification of the procedure in an attempt to simplify it technically, prevent subsequent canalization, and reduce postoperative morbidity. We also assess the impact of surgery on the quality of life of the children as judged by their caregivers and discuss the rationale of this treatment.

METHODS

This is a retrospective medical record review conducted in a tertiary care pediatric hospital (The Stollery Children's Hospital). The main source of identification of patients was a prospectively documented surgical database (Microsoft Access 2000; Microsoft Corp, Redmond, Washington) kept by 1 of us (H.E.-H.) beginning June 1, 2002. This database documents demographic information, surgeries involved, mode of anesthesia, the urgency of each procedure, diagnoses, all procedures per-
formed, special instrumentation, and complications. All children (<17 years of age) who were treated by means of submandibular with or without parotid duct clipping for saliva control problems were included. These were patients who experienced either excessive drooling and/or recurrent salivary aspiration pneumonia. The patients were required to have persistent symptoms or inadequate control despite a trial of nonsurgical therapy (unless nonsurgical therapy was not suitable or contraindicated). At this stage, the families or other caregivers were offered interventions directed at the major salivary glands or their ducts (botulinum toxin type A or ductal surgery). According to the choice, informed consent was obtained. Although initially a 4-duct clipping procedure was offered, this was modified, and the consenting caregivers were presented with the option of staging the clipping procedure (starting with the submandibular ducts and reserving clipping of the parotid ducts as a second stage if required). The rationale was based on parental satisfaction (in response to favorable initial results, the short duration of surgery, and the low morbidity) and readiness to stage the procedure, coupled with the desire to avoid xerostomia and its consequences. The information documented included age, sex, diagnoses, previous treatment modalities, procedures, and complications. Patients and caregivers were either followed up in the outpatient clinic or contacted by telephone. Outcome measures included comparison of preoperative and postoperative symptoms that related to saliva control and a measure of quality of life in addition to a record of perioperative complications.

Preoperatively, the patients' caregivers were asked a minimum set of questions related to symptoms of deficient saliva control. These questions were readministered after the intervention by a pediatric otolaryngology student clinical nurse specialist (S.R.) (Figure 1). These questions included the maximum number of shirts or bibs used in 24 hours, circumoral skin problems (redness, breakdown, and scaling), social embarrassment (by the children or their caregivers), choking episodes, and aspiration pneumonia (the latter 4 variables as binomial scores [0 or 1]).

The Glasgow Children's Benefit Inventory (GCBI) was also administered. The GCBI is a health-related quality-of-life tool assessing the child's condition after surgery. The GCBI comprises 24 questions on various aspects of the child's day-to-day life. The questionnaire evaluates general symptoms and shows 4 dimensions related to emotion, physical health, learning, and vitality. The GCBI measurements range from −100 to 100, with a 0 score indicating no change and 100 indicating maximum benefit. It is the only instrument designed specifically for measuring postsurgical changes in pediatric quality of life. Any postoperative complications were also noted (bleeding, swelling, pain, infection, delayed dietary intake, ranula formation, and airway obstruction).

The results were analyzed statistically on an intention-to-treat basis. Data were collected using a spreadsheet (Microsoft Excel 2000; Microsoft Corp). Descriptive statistics, the McNemar test (comparison of proportions before and after the intervention), and the paired t test (parametric data) were used (SigmaStat and SigmaPlot version 3; Systat Software Inc, San Jose, California). The Ethics Review Board approval was provided.

**SUBMANDIBULAR DUCT CLIPPING**

General anesthesia is induced, and nasotracheal intubation proceeds in the usual manner. With the patient in the tonsillectomy position, the oral cavity is kept open by means of dental blocks. Bupivacaine, 0.25%, with 1:200 000 epinephrine is used for infiltration of the floor of the mouth. Cotton-tipped applicators are used for displacing the tongue and, on occasion, are moistened with 1:1000 epinephrine for hemostasis.

Using magnifying surgical loops (×2.5), the opening of Wharton duct and the plica sublingualis are identified. Midway between the 2 landmarks, a mucosal incision is made using a size 15 blade. This incision must be superficial to prevent injury to the underlying duct. Blunt dissection is performed anterior and posterior to the mediolateral course of the duct. Once the duct is identified, further skeletonization proceeds using a Jake forceps. Once 0.5 cm is cleared from the surrounding connective tissue, 2 vascular clips (Ligaclips, No. 100; Ethicon, Somerville, New Jersey) are applied. The duct is not divided. Minimal manipulation of the duct is observed. No attempt is made to grasp or cannulate the duct to avoid false passage and fistula formation. The mucosa over the duct is closed using 3/0 chromic catgut, again ensuring that the duct is not included or traumatized.

**PAROTID DUCT CLIPPING**

The patient is positioned as previously described. Once again, magnifying surgical loops (×2.5) are used. The Stensen duct orifice is identified and cannulated using a lacrimal probe, and a 4/0 polypropylene (Prolene) suture is threaded around it to act as a stay suture. The area around it is infiltrated with bupivacaine, 0.25%, with 1:200 000 epinephrine. Then, the probe duct, with a small amount of mucosa, is grasped using a small Allis forceps. An elliptical incision, including 2 mm of the buccal mucosa and the mouth of the duct, is fashioned; 0.5 cm of the duct is identified and skeletonized. Two vascular clips (Ligaclips) are applied to the duct. The duct opening is buried, and the wound is closed using 3/0 chromic catgut. Patients are not routinely given antibiotic agents. Recomencement of dietary intake proceeds directly when the patient is fully recovered from anesthesia.

**RESULTS**

Nineteen procedures were performed on 18 patients in 14 months (September 1, 2005, to October 31, 2006). Medical therapy was attempted and failed to work in 12 patients before surgery. The other patients were deemed unsuitable for or their parents had refused other methods. Twelve patients were fed by gastrostomy tube. There was a slight male preponderance (10:8), with an age range of 2 to 14 years (mean, 5.7 years). The underlying diagnosis varied (Table). Five patients underwent 4-duct clipping, and 13 underwent submandibular duct clipping.
also including cardiac and limb anomalies.

tracheoesophageal fistula, and radial and renal dysplasia) association but anomalies similar to VATER (vertebral defects, imperforate anus, and ear anomalies; VACTERL, a nonrandom association of congenital coloboma of the eye, heart anomaly, choanal atresia, retardation, and genital and renal dysplasia) association but also including cardiac and limb anomalies.

only. Two patients who underwent submandibular duct clipping had a poor response. One patient subsequently had the procedure revised and had both parotid ducts clipped. The other patients who underwent submandibular duct clipping had good results. The duration of follow-up was 2 to 18 months.

Figure 2 displays results pertaining to manifestations of deficient saliva control. Eight patients had no drooling at all after surgery (P = .004). Ten patients complained of circumoral skin problems before surgery, with only 2 still symptomatic after surgery. The difference was significant (P = .01). The caregivers of 13 patients reported embarrassment before surgery. This number decreased to 5 after surgery (P = .008). All the patients experienced choking spells, but only 8 continued to have episodes after surgery (P = .008). Finally, of 13 patients who sustained aspiration pneumonias before surgery, only 1 had an episode during follow-up (P = .001).

The mean number of shirts changed each day (n=11) was 3.73 at baseline and 0.32 after the intervention (P = .001). The mean number of bibs changed daily (n=12) was 5.25 at baseline and 1.42 after the intervention (P = .001; 95% confidence interval, 2.38-3.83; power, 0.9). Shirt and bib use was not universal in the study group. The mean (SD) GCBI score was 33.2 (22.36). The highest value was 81.2, and the lowest was 0, with the latter being scored by 2 patients (Figure 3). The mean score for the questions weighted toward physical health (questions 1, 6, 14, and 21-24) was 44. There were no negative scores representing a decrease in quality of life after surgery.

There were 2 patients in whom surgical treatment failed, both with a GCBI score of 0. One patient underwent revision and subsequently has had no further drooling. The GCBI score for this patient after revision was 37.5, with no symptoms of poor saliva control. There were no complications in this series, and most patients were admitted to the hospital for a single postoperative night. There was no gland swelling, transient or otherwise. Postoperative analgesia requirements were minimal and were limited to acetaminophen. There have been no cases of ranula formation in the study group.

To our knowledge, this is the third study in the English-language literature on major salivary duct interruption surgery for deficient saliva control. The first study furnished the anatomical basis and described the operative procedure. The clinical application was on 4 children with chronic aspiration, and the response was excellent, with no significant complications. Subsequently, the procedure was successful in 81% of a 21-patient series. All the patients, however, experienced transient postoperative neck swelling and, in addition, 1 had a ranula and 1 had bilateral sialadenitis that required surgical intervention. According to the authors, several of the families thought that the postoperative morbidity was too high and would not consider subsequent surgery. In the present study, we demonstrated significant improvements in all the outcome measures, albeit within a short follow-up. We used, broadly speaking, 2 main measures: quality of life and traditional

| Table. Underlying Diagnosis in 18 Patients Undergoing Salivary Duct Clipping |
|---------------------------------|------------------|
| Diagnosis                      | No. of Patients  |
| Hypotonia                      | 1                |
| Angelman syndrome              | 1                |
| CHARGE                         | 2                |
| VACTERL                        | 1                |
| Cri du chat syndrome           | 1                |
| Cerebral palsy                 | 3                |
| Vocal cord paralysis           | 2                |
| Aicardi syndrome               | 1                |
| Secondary neuronal migration disorder | 1 |
| Global developmental delay     | 3                |
| DiGeorge syndrome              | 1                |
| West syndrome                  | 1                |

Abbreviations: CHARGE, a syndrome of associated defects, including coloboma of the eye, heart anomaly, choanal atresia, retardation, and genital and ear anomalies; VACTERL, a nonrandom association of congenital anomalies similar to VATER (vertebral defects, imperforate anus, tracheoesophageal fistula, and radial and renal dysplasia) association but also including cardiac and limb anomalies.
manipulations of deficient saliva control. Regarding the first measure, to our knowledge, only 1 previous study has measured changes in quality of life after surgery using a purpose-designed tool. This study uses a tool specifically designed and validated for pediatric surgery. The mean GCBI score is favorably comparable with unpublished results documented for bone-anchored hearing aid implantation for young children in the Birmingham program (GCBI score, 19.5; n=37) (M.S.T., M. D. Siemers, FRCS, A. P. Reid, FRCS, and D. W. Proops, FRCS, unpublished data, 2006). It also increases when the questions related to physical health are considered in their own right. This is a reflection of the original standardization on otherwise healthy children, whose baseline social activities and interaction would be considerably different from those of the present patient population. It is specifically reassuring that there was no perceived decrease in the quality of life of any of the patients studied, and only 2 parents reported no positive change in quality of life.

This result is particularly important. Recent trends in the literature emphasize that improvement in the increased daily care needs of patients and increased demands on caregivers, and the adverse effect on social interaction and self, should be important benchmarks to evaluate interventions. There is also interest in monitoring quality of life in these patients using specifically designed tools. The second aim of realizing a difference in manifestations of uncontrolled salivary flow has been achieved. The improvements in recurrent aspiration pneumonia were particularly encouraging because this group of patients may require more drastic measures involving laryngeal or tracheal surgery and few researchers have reported on salivary gland interventions to eliminate their problems. The other measured variables improved as well. Bibs used and shirt changing were reduced significantly, reducing the load of care. Regarding the remaining variables, we used ordinal scales to detect total abolishment of problems rather than encounter the ambiguity of reporting on graded responses. Therefore, we know, for example, that the success in complete elimination of drooling, which occurred in approximately half of the patients, is comparable with that in other studies. We encountered no complications in this series, and the mean duration of the procedure was 42 minutes, as collected from the records of the operating room. No patients required additional pain control measures other than acetaminophen treatment. The minimal morbidity is one of the most attractive aspects of this procedure. These results compare favorably with others because according to a recent review, complication rates of up to 69% have been reported.

Many different surgical approaches have been described. Submandibular duct relocation seems to be the most widely reported procedure. It is, however, associated with substantial postoperative pain, and 88% of patients who were dissatisfied cited postoperative pain as the main factor. Ranula formation, seen in all series, may be as high as 9%, and transient edema of the floor of the mouth resulting in feeding difficulty is near universal. Other complications include secondary hemorrhage, lower lip droop, and submandibular gland swelling requiring excision. In the context of morbidity, bilateral simultaneous submandibular gland excision with single parotid duct ligation is also described. In addition to an external scar, marginal mandibular nerve weakness postoperatively, and the theoretical risk of traumatizing both nerves in a patient with preexisting poor oromotor control make this option less attractive. Similarly, combined parotid and submandibular duct relocation is undesirable in this population with multiple comorbidities, taking more than 4 hours to accomplish and followed by persistent submandibular gland swelling and xerostomia.

To combine effectiveness with less morbidity, we applied basic principles to modify the procedures. We believed that the use of magnification would help identify the ducts, reduce manipulation, and avoid probing the submandibular ducts to minimize the risk of fistula formation. Following the information from the original description, we interrupt the duct no further than 1 cm lateral to its opening to avoid disruption of the sublingual ductules that enter the duct proximal to this point. This is a key step in avoiding ranula formation, although the sublingual glands are left intact. We also chose to use vascular titanium clips instead of silk ties to help minimize the traction that might occur during the tying of ligatures, and they are technically easier to use in the restricted space of a child’s mouth. The clips also elicit less tissue reaction than silk ligatures and are less likely to slip.

Finally, as the experience evolved, it became apparent that the families’ impression was favorable regarding the procedure and its postoperative course. The parents are informed of the major contribution of the submandibular glands to resting salivary flow rate compared with the parotids and that the position of the orifices of their ducts is strategically placed to enhance pooling and escape of secretions in the anterior floor of the mouth. When informed of the potential deleterious effects of dryness on oral and dental hygiene, most parents are in favor of staggering the surgery in 2 steps. This is a stronger case if the child is orally fed rather than using a permanent alternate route. There is some evidence that effective control of the submandibular glands even by botulinum toxin is comparable with the effect of systemic anticholinergic agents. Suskind and Tilton and Crysdale et al commented on the notable increase in the thickness of secretions on addressing the parotids as well as the submandibular glands. Although there were 2 initial surgical failures in the submandibular duct clipping group, the probable reason was loose and improper application of the clips. The remaining patients responded well and avoided the risk of xerostomia. When the 2- vs 4-duct clipping procedures are examined in relation to drooling control and GCBI scores, a weak correlation (Pearson correlation coefficient, 0.4) is found using these data. However, this of its own right and with small numbers in each arm cannot make a strong case for a 4-duct clipping procedure. When the primary complaint is aspiration, we recommend 4-duct clipping in the first instance, but we concede to the wants of the parents to some degree as well.

Anecdotally, physicians believe that the technique of duct ligation fails because the remaining glands compensate. There is some literature indicates that total obstruction of the ducts causes ipsilateral atrophy of the glandular and ductular cellular elements and contralateral hyperplasia of...
those on the control side. However, submandibular gland excision combined with parotid duct ligation is associated with high success rates and there is a limit to extrapolation from animal experiments to human applications. Botulinum toxin type A, in comparison, has a reproducible good success rate, which may be as high as 80%, but this varies among studies, as does the duration of action, which may be as short as 8 weeks. Thus, this approach will lead to successive treatments, which are costly (owing to off-label use), and may require a general anesthetic in many patients. Unless there is an overriding parental choice, a contraindication, or a temporary cause for the deficient saliva control, we perceive its role as diminishing compared with that of duct clipping.

In conclusion, major salivary gland clipping offers a simple, quick, effective, and safe technique for dealing with deficient saliva control. We recommend the consideration of this technique before more complex surgical procedures. Optional staggering of the 4-duct clipping and starting by the submandibular glands is based on the overriding contribution to resting salivary flow, the comparable benefit of including the parotid, and the low morbidity overall. In this series, this method has been shown to significantly improve the care needs, physical health, and quality of life of these children.

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