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Surgical management of chronic sialorrhea in pediatric patients: 10-year experience from one tertiary care institution

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ABSTRACT

Background: Chronic sialorrhea is a common problem for pediatric patients with disorders that affect swallowing. While many patients are successfully treated with medical therapies such as Robinul and Scopolamine, a number of such children are not able to tolerate the side effects of these medications. In these cases, surgical treatments can include Botulinum toxin A (Botox) injections into the major salivary glands, sublingual or submandibular gland excision (SMGE), submandibular duct ligation, parotid duct ligation (PDL), or any combination of the above procedures. The purpose of this study is to report on the 10-year experience with the surgical management of chronic sialorrhea at one tertiary care institution, and compare the efficacy of open surgical procedures versus Botox injections for reduction in salivary flow.

Methods: A retrospective chart review identified 27 pediatric patients with chronic sialorrhea; 21 of whom underwent Botox injections and 15 of whom underwent surgical procedures. Preoperative and follow-up clinic notes were reviewed to determine the level and severity of drooling as well as the effectiveness of sialorrhea reduction, as assessed by the Teacher Drooling Scale (TDS).

Results: 42% of those receiving Botox injections reported a reduction in drooling, with the average pre- and post-Botox TDS of 4.3 and 3.9, respectively ($p = 0.02$ by the Wilcoxon signed rank test). Nine of the patients receiving Botox injections (43%) required multiple injections, with an average duration of effect of 3.9 months, and 7 patients (33%) eventually required surgery. All of the children who underwent surgery (7 bilateral SMGE with PDL, 6 SMGE only, and 2 PDL only) experienced a reduction in drooling, with average pre- and post-operative TDS of 4.5 and 2.2, respectively. This reduction was significant by the Wilcoxon signed rank test ($p = 0.001$).

Conclusions: The ten-year experience at our institution demonstrates the safety, efficacy and long-term control of drooling in the patients undergoing surgery for intractable sialorrhea.

1. Introduction

Pediatric patients with neuromuscular and muscular disorders that affect swallowing, such as cerebral palsy (CP), muscular dystrophy and other rarer myopathies commonly experience chronic sialorrhea. Studies have demonstrated that up to 37% of children with CP experience chronic sialorrhea [1]. Swallowing dysfunction in the context of neuromuscular or muscular dyscoordination can lead to long-term, ineffective clearance of saliva produced by the submandibular, parotid, and sublingual glands, which in turn can lead to medical complications such as recurrent aspiration pneumonitis, irritative stomal dermatitis, as well as social and educational impairments secondary to the aesthetic appearance of a child who requires continual clothing changes or frequent mouth wiping [2].

Patients who suffer from chronic drooling are initially managed conservatively, with anticholinergic medications such as Scopolamine patches or Robinul. For those patients with recalcitrant chronic sialorrhea, newer methods of Botulinum A toxin (Botox) injections have curried favor among otolaryngologists due to the less invasive nature of the procedure, its presumed safety, and success rate in reducing drooling. However, results from studies examining the efficacy, duration of effect, and complications of Botox injections vary markedly [3,4].

Reported response rates to Botox injections, as measured by a significant subjective reduction in drooling, range from 51% in a 10 year series of 69 children [3] to 79% in a cohort of 33 pediatric

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patients. When measuring caregiver perceptions, 80% endorsed satisfaction in a series of 45 pediatric patients with neurologic disabilities after Botox injections [1]. This wide range of published response rates is likely due to a combination of different definitions of "success" by investigators, lack of a universally employed quantitative scale for determining amount and changes in drooling, differential sample sizes, and the reporting and sampling bias inherent in retrospective case series. Notwithstanding, Vashishta et al. recently published a meta-analysis of 8 randomized, placebo controlled trials of Botox injection for reduction of saliva, and in spite of the more rigorous experimental designs, the standardized mean differences in saliva pre- and post-Botox injections ranged from −3.03 to −0.51, with an overall mean difference of −1.54 [4]. The magnitude of this difference, which is negative because the scales used to measure saliva quantity decrease if the treatment works, is considered a "large" effect size by Cohen [5]. However, the authors underscore large heterogeneity among the results from studies of Botox treatment for pediatric patients ($I^2 = 61\%$) [4]. From a safety perspective, Botox injections carry a nontrivial risk for both minor complications such as nausea, neck pain, and speech problems, and, though less frequently, for serious complications requiring hospitalization, including aspiration pneumonia, prolonged dysphagia necessitating gastrostomy tube placement, and shortness of breath [1,4]. In the aforementioned study by Chan et al., [3] 28% of the patients experienced complications and 7% experienced complications requiring hospitalization, a rate that was replicated by Khan et al., wherein 26% of the enrolled patients experienced a complication with about one-third of the complications leading to hospitalization [1]. In addition, there is accumulating evidence regarding the transient effects of Botox injections, with maximal reduction in drooling typically noted at 2–4 weeks [6,7] and a steady return to baseline sialorrhea within 2–3 months [6,8].

It has become increasingly clear that open surgical procedures result in higher rates of success, both short- and long-term, in pediatric patients with chronic sialorrhea. In a study involving 93 pediatric patients who received bilateral SMGE and bilateral PDL, 87% reported no further drooling over a follow-up period of 1–10 years [9]. Manrique et al. later recapitulated this finding, observing an 87% response rate in 31 children with neurologic disabilities who underwent bilateral submandibular gland excision and bilateral parotid duct ligation [10]. Similar to studies of Botox injection, the rate of significant saliva reduction varies appreciably according to the sample size and study institution. A recent meta-analysis concluded that most studies were of low-quality and produced varying results, which the authors surmised was attributable to several different surgical techniques and limited sample sizes [2]. However, in the 47 studies they identified, there was an 82% overall success rate with open surgical procedures for control of chronic sialorrhea. Of the surgical approaches, submandibular gland excision with parotid duct ligation has demonstrated the best long-term benefit [2,9], although more conservative techniques such as ligation of the parotid and submandibular ducts have also been reported as effective [11,12]. Surgical correction of sialorrhea can result in xerostomia and subsequent dental problems, in addition to the known risks of general endotracheal anesthesia [2].

Surgical approaches employed at our institution have included both bilateral submandibular gland excision with parotid duct ligation and solely bilateral submandibular gland excision as a more conservative strategy to avoid over-dryness. In addition, our center has extensive experience with Botox injections into the major salivary glands to control sialorrhea. The purpose of this study was to report on the efficacy and safety of both surgery and Botox injections for the management of chronic sialorrhea in pediatric patients. Because many of the patients in this study who received Botox injections underwent subsequent salivary gland surgeries for further management, the natural history of this debilitating and socially isolating condition will be illuminated. Furthermore, this study population permits a comparative analysis of the relative effectiveness and safety considerations in both of these approaches to the management of chronic sialorrhea.

2. Methods

2.1. Patients

After Institutional Review Board approval was granted by the study institution, a retrospective electronic medical record review was performed to identify pediatric patients aged 2–18 years with a diagnosis of chronic sialorrhea who had also received either Botox injection, surgery, or both for management of their drooling during a period from September 2003 through August 2013. Preoperative, operative, and follow-up clinic notes were reviewed to determine the preoperative Teacher Drooling Scale (TDS) [13], the type of operation or injection performed, operative findings and any complications during the surgery or Botox injection, the post-operative course, including length of stay in the hospital and any early complications, such as infection or wound hematomas. The long-term surgical outcome, including recurrence of sialorrhea, the need for additional medical or surgical intervention to control drooling, the post-surgical TDS, and over-dryness or dental problems secondary to xerostomia were determined by reviewing the most recent follow-up visit notes. Postoperative TDS was calculated at a minimum of one month post-surgical procedure and one week post-Botox injection.

2.2. Teacher Drooling Scale

The TDS is a metric for assessing level and frequency of drooling in patients with chronic sialorrhea that ranges from 1 (no drooling) to 5 (profuse drooling all day) [13]. Table 1 outlines the criteria for determining a TDS score. Because a formal TDS calculation was lacking for 6/15 surgical and 14/21 Botox cases due to non-uniformity among surgeon reporting, the TDS for these patients was determined by the authors based on review of the pre-operative notes from the surgeon and clinic notes from at least two other clinicians involved in the patient's care. To circumvent some of the bias inherent in post-hoc ascription of TDS for these cases, an attending otolaryngologist at the institution, who was unassociated with the study or publication and blinded to the intervention type, independently reviewed pre- and post-procedure notes for these 20 unassigned cases, and complete agreement was found for 60% of the cases, with no deviation greater than 1 point on the TDS.

2.3. Surgeries and Procedures

All Botox injections consisted of Botulinum toxin A, delivered in 0.9% sterile saline, at a dose of 8 units to 30 units per major salivary gland (mean 23 units, mode 25 units). Though a strict weight-based algorithm is not used for calculation of Botox dosing at this institution, the average amount of Botox injected for all 21 subjects was 1.1 units per kilogram per gland. Injections were either

Table 1
Teacher Drooling Scale (TDS) characteristics.

Frequency of drooling	Amount of drooling	Score
No drooling	Physiologic	1
Infrequent drooling	Small amount	2
Occasional drooling, intermittent all day	Moderate amount	3
Frequent drooling	Large amount, not profuse	4
Constant drooling	Profuse amount, always wet	5