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Botulinum toxin injections for chronic sialorrhoea in children are effective regardless of the degree of neurological dysfunction: A single tertiary institution experience

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ABSTRACT

Objective: To determine the effectiveness of submandibular salivary gland Botulinum Toxin Type-A (BTX-A) injection in the treatment of drooling in children with varying degrees of neurological dysfunction.
Methods: A retrospective review of pre- and post-procedure drooling frequency and severity scores of patients receiving BTX-A between January 2008 and January 2013. Stratification to different subgroups of neurological impairment was performed according to Gross Motor Function Classification System (GMFCS) score. Drooling severity was assessed using Thomas-Stonell and Greenberg symptom questionnaires administered at time of initial consultation and 3 months after treatment.
Results: 48 sets of BTX-A injections in 26 patients with an average age of 9.45 years (range 7 months–18 years) were included in the study. Marked improvement in drooling was seen in 60.4% of patients, a marginal or brief improvement was seen in 20.8% and there was no improvement in 18.8%. No adverse events were reported following any of the BTX-A injections. BTX-A was safe and effective in the eight patients with pre-existing swallowing dysfunction. Subsequent drooling surgery was performed in 15 (57.7%) of the cohort, all 15 patients responded to BTX-A injections.
In patients with Cerebral Palsy, there was no correlation between the severity of the neurological dysfunction as measured by the Gross Motor Function Classification System (GMFCS) score and the response to BTX-A treatment.
Conclusions: Injection of BTX-A to the submandibular glands of children with neurological disorders is a safe procedure and results in a reduction in drooling in the majority of patients. Children with severe neurological dysfunction respond to BTX-A injections as effectively as their less impaired peers and the degree of response does not appear to be associated with the severity of neurological disability. BTX-A injection is a good initial procedure when drooling surgery is being considered.

1. Introduction

Drooling, the unintentional loss of saliva from the mouth, is considered abnormal over the age of 4 years and is a common symptom in children with cerebral palsy (CP) and other neurological diseases. Problematic drooling is reported to occur in 10–37% of children with CP [1]. Intrasalivary gland injection of botulinum neurotoxin has emerged as a more targeted, effective and safe option for the treatment of drooling in children with neurological disorders [2–5]. We sought to describe the outcomes of injecting BTX-A to the submandibular glands of children presenting with problematic drooling. Because poor oral motor control and posture also contribute significantly to the aetiology of drooling and are aggravated by neurological impairment, we postulated that the likelihood of achieving a response to BTX-A might be predicted by the level of neurological disability. We used standard questionnaires and classification systems to describe each case. Degree of drooling is assessed using the Thomas-Stonell and

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Greenberg classification [6], which scores frequency of drooling on a scale of 1–4 and severity of drooling on a scale of 1–5 (Table 1); Severity of neurological disability in children with CP can be classified by the Gross Motor Function Classification System (GMFCS) – Expanded and Revised (E&R), 2007 [7]. This classification system is described for use in children up to 18 years of age and is based on functional limitations in daily life. A score of I–V is used, with a score of I indicating only mild limitations and a score of V indicating the most severe.

2. Methods

2.1. Data collection

A retrospective chart review was carried out on all patients at our paediatric tertiary-level institution who had received BTX-A injections to the submandibular glands to treat drooling over the 5-year period from January 2008 until January 2013. The start date was chosen as it corresponded with the point at which our unit adopted a standardised drooling assessment questionnaire.

All patients who were deemed suitable for BTX-A injection from our multidiscipilinary drooling clinic (consensus by patient, caregiver and physician) were included in the study. Cases were identified using a search of the hospital records for ICD-10-AM procedure code 1836000 (Administration of botulinum toxin into soft tissue, not elsewhere classified) and cross-referenced with the operating theatre and departmental procedure lists for BTX-A injection to the submandibular glands. The electronic medical records were then reviewed for each procedure to obtain pre- and post-op drooling frequency and severity scores or other measures of symptom severity, as well as demographic data. Further scrutiny of the medical records up to January 2016 was performed to determine longer term outcomes and whether any patients required surgical intervention.

2.2. Patients and treatment

Patients were seen at a multidisciplinary saliva control clinic comprising ORL surgeons, Dentist and Speech/Language Therapist. The primary caregivers of all patients completed a questionnaire, prior to consultation and again 3 months post-procedure (BTX-A injections). The questionnaire scores were combined to give a total out of 9.

To obtain GMFCS scores, electronic medical records were reviewed and the primary carer(s) of the patient contacted to complete the assessment where the information in the record was insufficient to determine the score.

All procedures were performed under general anesthesia. The patient's head was positioned turned to the opposite side to provide good exposure. A two-person technique was used for the injections whereby the submandibular gland was identified on ultrasound by a radiologist whilst the otolaryngologist injected the BTX-A into the centre of the gland after confirming that the needle had not breached a blood vessel. A standard concentration of 100 U/mL in 0.9% NaCl solution was used via a 1 mL syringe and 22 g needle. All patients had bilateral submandibular gland injections. Other salivary glands were not injected.

The most common dose of BTX-A used in patients weighing over 20 kg was 20 units per gland, with three patients receiving 25 units and one receiving 15 units each side. There was only one patient less than 20 kg, who received 15 units each side.

All patients were followed up for a minimum of 12 months after each procedure.

Table 1
Assessment of drooling.

Frequency
1 = No (or rare) drooling
2 = Occasional drooling (not every day)
3 = Frequent drooling (every day but not all the time)
4 = Constant drooling – always wet
Severity
1 = Dry – never drools
2 = Mild – only the lips are wet
3 = Moderate – wet on the lips and chin
4 = Severe – drools to the extent the clothes get damp and need changing
5 = Profuse – clothing, hands and objects become wet

3. Results

There were 59 procedures performed on 33 patients between January 2008 and January 2013. Records were incomplete for 11 of these procedures therefore they were excluded from the study. A total of 48 sets of injections in 26 patients were considered suitable for analysis.

The average patient age at time of injection was 9.45 years (range 7 months–18 years). Of the 26 patients, 14 (54%) were males, which corresponded to 27 of the 48 total procedures (56%). 17 patients had a diagnosis of CP, the remaining nine patients had neurodevelopmental delays and neurological disabilities due to other conditions.

In our unit, patients are deemed to have had improvement if the total drooling score (obtained by adding the individual frequency and severity scores) is reduced by 2 or more (on the 9 point scale) as this appears to correlate with a parental/carer's subjective impression of improvement.

Improvement was seen in 29/48 cases (60.4%). Of the remaining 19 cases, 10 (20.8%) showed a marginal (improvement of 1 on the 9 point scale) or brief (less than 4 weeks) improvement and 9 (18.8%) showed no improvement (Table 2). Of the 29 cases that improved, duration of effect was recorded in 17 and this showed an average of 3.0 months. There was no statistically significant correlation between the degree of improvement and age of the patient, using Fisher's exact test ($p = 0.43$).

GMFCS scores were reported for the 32 BTX-A injections in patients that had CP. In the remaining 16 procedures, GMFCS scores could not be assigned due to the patient having a neurological diagnosis other than cerebral palsy (Table 2).

No association between GMFCS score and response to BTX-A was seen when the data were examined using Fisher's exact test ($p = 0.52$). Spearman's rank correlation coefficient was calculated as a measure of the relationship between the two groups of data ($r = -0.07$) and this too was not significant ($p = 0.71$).

There were 11 patients who received more than one injection of BTX-A over the study period, with four patients receiving two sets of injections, three receiving three sets of injections and four receiving four sets of injections. This amounted to 33 procedures. Improvement was noted in 21 cases (63.6%), partial response in 8 (24.2%) and no response in 4 cases (12.1%).

Eight patients (30.8%) suffered from swallowing dysfunction prior to BTX-A intervention. This subgroup underwent 19 of 48 procedures (39.6%). Improvement in drooling symptom score was seen following 17 injections. There was no deterioration in swallowing function or need for acute nasogastric tube feeding in the series.

There were 15 of the 26 patients (57.7%) who subsequently proceeded to surgery to manage their drooling, with 14 having bilateral excision of the submandibular glands and one having transposition of the submandibular ducts with sublingual gland excision. All 15 of these cases had some improvement with BTX-A.