BOTOX-A injection of salivary glands for drooling

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AVAILA

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A B S T R A C T

Background and purpose: Drooling is a challenging entity to manage. Botulinum toxin A (BOTOX-A) infiltration of salivary glands is a promising alternative to surgical treatment. This study aims to assess the outcome of BOTOX-A salivary glands infiltration in children with drooling.

Methods: Patients treated between January 2012 and March 2015 were enrolled. BOTOX-A was injected in the parotid and submandibular glands under ultrasound control and general inhalational anesthesia. The outcome was evaluated through the DSFS: Drooling Severity (1-best to 5-worst) and Frequency (1 to 4) Scale, that was applied before treatment, and 1-, 3-, and 6-month after injection. The inclusion criteria were a DDS ≥4 and/or DFS ≥3. Statistical significance was set at 5%.

Results: There were 17 patients aged 12.1 ± 5.1 [4–19] years, all of them with neurologic impairment. After the first injection, 13 (76.5%) patients had reduction of the severity (S) and 12 (70.6%) of the frequency (F) scale; in 6 (35.5%) patients drooling resolved completely. Pre-treatment S + F score was 8.59 ± 0.71 [7–9]; it decreased significantly to 4.65 ± 2.32 (p = 0.001) at 1-month post-injection evaluation. At 3-month and 6-month the scores were also significantly lower than the pre-treatment one (4.00 ± 1.96, p = 0.002; 5.36 ± 2.20, p = 0.005; respectively), but there was a significant increase between the 3-month and 6-month evaluations (p = 0.01). With a follow-up of 20.1 ± 9.2 [4–38] months, 4 out of the 13 successful injections needed a second one after 7.5 ± 3.1 [3–10] months. The patient with the longest time not requiring re-injection had 28 months of follow-up. One (6%) patient presented mild dysphagia that regressed spontaneously. All but two (88%) parents/caregivers would repeat the treatment.

Conclusions: BOTOX-A seems to be an effective minimal invasive treatment for drooling with few complications. After 6 months the need for re-injection becomes substantial but it may not be necessary for several months. Further studies are needed to establish the most effective dosage and frequency of injections.

Level of evidence: IV

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Drooling can appear in a wide range of diseases being common in the neurologically impaired children. The treatment of these patients is complex and dependent on a multidisciplinary approach. Botulinum toxin A (BOTOX-A) salivary glands infiltration has been used as a promising alternative to surgical approach. The present study aimed to assess the efficacy and safety of BOTOX-A salivary glands infiltration in children with drooling.

1. Material and methods

Pediatric patients that underwent BOTOX-A infiltration in salivary glands between January 2012 and March 2015 were prospectively enrolled. BOTOX-A injections were made by the same radiologist with ultrasound control and under general inhalational anesthesia according to methods published elsewhere [1]. Briefly, BOTOX® (onabotulinumtoxin A – Allergan, Inc., Irvine, CA) was diluted in 5 mL of normal saline; then, 1.5 mL (30 UI) was injected in each parotid gland and 1 mL (20 UI) in each submandibular gland.

The parents/caregivers completed questionnaires before, one, three and six months after injection. Demographic data (age, gender, weight) and associated diseases were collected. The Droslling Severity and Frequency Scales (DSFS), from 1 (best) to 5 (worst) for severity (S) and 1 to 4 for frequency (F), was used to evaluate the efficacy of the treatment, as previously published [2]. The inclusion criteria were a DDS ≥4 and/or a DFS ≥3 in the absence of BOTOX-A contraindications such as myasthenia gravis or previous allergic reaction. S + F score could range from 2 (best) to 9 (worst) points. Treatment was considered a failure when the score did not improve at the 1-month evaluation. A deterioration of 3- or 6-month scores to the pre-treatment ones was considered a recurrence.

Parents/caregivers were questioned whether, after the known experience, they would decide to initiate this kind of treatment.

The variables are described as mean ± standard deviation (SD) and range [minimum–maximum], or frequencies (%). Wilcoxon signed rank test was used for comparisons. Statistical significance was set at 5%.

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test was used to compare Drooling scores (S + F); the statistical significance level was set at 5%.

The study protocol had the approval of institutional ethics committee and parents’ written consent was obtained.

2. Results

Seventeen patients (11 males) with 12.1 ± 5.1 [4–19] years and weighing 31.35 ± 15.7 [14–50] kg were included, all of them with neurologic impairment: 12 hypoxic–ischemic syndromes, and 5 rare syndromes (Dravet, Rett, DiGeorge, Cromossomopathy 13t(9;17), Paquirigia). Seven patients had been previously submitted to transposition of the submandibular ducts and one had had bilateral submandibular excision. Twenty-five injection sessions were performed; all were considered technically successful.

One (6%) patient presented mild dysphagia that regressed spontaneously in short-term.

The distribution of Drooling Severity Scale (DSS) and of Drooling Frequency Scale (DFS) is shown in Figs. 1 and 2, respectively. Before treatment all patients had a DSS of 5 (76%) or 4 (24%), and a DFS of 4 (82%) or 3 (18%). After the first injection, 13 (76.5%) patients had reduction of DSS and 12 (70.6%) of the DFS; in six patients drooling resolved completely. Four cases were failures and underwent re-injection.

At 1- and 6-month evaluations, 8% and 12% of the patients, respectively, presented a score of 4; the most favorable outcome was registered at 3-month evaluation with 85% of patients presenting a DSS of 1 or 2.

Concerning DFS, at 1- and 6-month evaluations, 8% and 27% of the patients, respectively, presented a score of 4; the most favorable outcome was found at 3-month evaluation with 85% of patients presenting a score of 1 or 2.

The drooling S + F score (Fig. 3) before treatment was 8.59 ± 0.71 [7–9]; it decreased significantly (p = 0.001) to 4.65 ± 2.32 at 1-month post-injection evaluation. At 3-month and 6-month evaluations the scores were also significantly lower than the pre-treatment one (4.00 ± 1.96, p = 0.002; and 5.36 ± 2.20, p = 0.005, respectively), but there was a significant increase between the 3-month and 6-month evaluations (4.00 ± 1.96 vs 5.36 ± 2.20; p = 0.01).

The outcome of patients that had previous surgical treatment was similar to the group of patients that did not, concerning S + F scores at 1- (3.67 ± 1.37 vs 4.14 ± 2.34, p = 0.457), 3- (3.33 ± 1.03 vs 3.83 ± 1.60, p = 1.000), and 6-month (5.50 ± 1.38 vs 5.20 ± 3.11, p = 0.357) evaluations.

With a follow-up of 20.1 ± 9.2 [4–38] months, 4 out of the 13 successful injections needed a second one after 7.5 ± 3.1 [3–10] months. The patient with the longest time not requiring re-injection had 28 months of follow-up. From the four patients submitted to re-injection because of failure only one improved. Three out of the four patients submitted to re-injection because of recurrence improved.

All but two (88%) parents/caregivers would repeat the treatment.

3. Discussion

Drooling is the unintentional loss of saliva from the mouth as a result of the lack of coordination of the oral, facial, and neck muscles. It differs from sialorrhea because the quantity and quality of saliva are normal. It is common in children with cerebral palsy (10–37.5%), muscular dystrophy and other rare myopathies [1]. The drooling pathogenesis is multifactorial; bad postural position (e.g., these children tend to keep their mouth open for long periods of time) and impaired swallowing are probably the main mechanisms. As a consequence, the peri-oral area is wet most of the time causing skin alterations, infection and social discrimination.

The treatment of drooling requires a multidisciplinary approach involving pediatricians, neurologists, physiatrist/speech therapist, etc. Drooling treatment can be behavioral, pharmacological or surgical. The behavioral approach is based on physiotherapy and speech therapy, but is associated with a very high relapse rate because it needs high motivation and is very time consuming [3]. Anticholinergic drugs cause many side effects (dry mouth, blurred vision, urinary retention) and do not achieve good results [3]. Surgical approach directly to salivary secretion/excretion may be attained by ducal relocation and/or gland excisions. These are invasive procedures with a quite variable efficacy [4].

BOTOX-A has been used in multiple conditions such as for aesthetic, limb spasticity, bladder dysfunction and anal fissure. The toxin binds permanently with a channel protein responsible for the transport of acetylcholine on the pre-synaptic neuron cell, enabling the synapse to occur. By this mechanism, infiltration of the salivary glands that are controlled by parasympathetic nerves results in the decrease of the saliva secretion and reduction of drooling. The blockade though irreversible is however temporary as new nerves sprout to create new neural connections [5].

The efficacy of BOTOX-A in the current series was in line with others previously published [1,6–8]. We have adopted the dosage advocated by Meece [1], but there are many protocols, such as weight-dependent dose (1.4 U/kg for the parotid and 0.6 U/kg for the submandibular gland [9-11]), or a low dosage and titrate up.

![Fig. 1. Drooling Severity Scale (from 5-worst to 1-best) at: pre-treatment (Spre), 1 month (S1), 3 months (S3) and 6 months (S6) post-injection.](http://dx.doi.org/10.1016/j.jpedsurg.2016.09.074)
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