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## Case Report

# Botulinum Toxin Type A Treatment for Drooling in Taiwanese Children with Cerebral Palsy: Three Case Reports

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Botulinum toxin is a promising treatment option for the management of drooling in patients with cerebral palsy. However, the effect of botulinum toxin type A in treating drooling of Taiwanese children with cerebral palsy has not been reported thus far. We injected single dose botulinum toxin into bilateral parotid glands of three children with cerebral palsy, and followed the results over a 9-month period. Drooling improved in all three patients without significant adverse effects during follow up. There were individual differences in terms of efficacy and duration of effect. Two patients had longer duration of effect than that reported in previous western studies. The findings for this series of patients suggest that botulinum toxin injection can be effective in reducing saliva secretion and drooling. (Tw J Phys Med Rehabil 2007; 35(3): 165 - 170)

**Key words:** botulinum toxin type A, drooling, cerebral palsy, parotid glands

## INTRODUCTION

Drooling is caused by an upset in the coordinated control of oro-facial and palato-lingual musculature leading to excessive pooling of saliva in the anterior mouth and resultant unintentional loss of saliva from the mouth. Drooling in the awake state is considered to be abnormal beyond the age of 4 years.<sup>[1]</sup> The most common causes of drooling in childhood are cerebral palsy and

mental retardation.<sup>[2]</sup> Drooling has been reported to be a significant problem in 10% to 37% of patients with cerebral palsy.<sup>[3]</sup> Excessive drooling may increase the burden of the caregiver and carry the risk of aspiration and dehydration. Biofeedback techniques, oral and topical medications, surgeries, and radiation have been proposed to treat drooling.<sup>[1,4-6]</sup> Yet none of these is universally successful and many have potential complications.

Nerve endings of the parasympathetic post-ganglionic neurons secrete acetylcholine as the neurotransmitter in

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salivation. Botulinum toxin may be able to inhibit saliva secretion by blocking the release of acetylcholine. A few studies have evaluated the results of botulinum toxin injection to treat drooling in adult and pediatric patients with neurological disorders.<sup>[7-15]</sup> In children, the reported total doses ranged from 10U to 70U within a single treatment. The duration of effect also varied greatly, lasting from 2 weeks to 6 months. Thus, the appropriate dose and duration of effect remain to be established.

The case series was to report the duration, therapeutic effects and side effects of botulinum toxin injection to treat drooling in three children with cerebral palsy.

## CASES REPORT

With the approval of the Ethics Committee, three children with cerebral palsy and severe drooling from our clinic were enrolled. Table 1 lists the characteristics of the three patients. A thorough history taking and physical examination were performed and informed consent was obtained from the parents. Inclusion criteria were: 1) drooling severe enough to bother the children and/or caregiver; 2) no bleeding tendency or other medical problems contra-indicating treatment with botulinum toxin injection; 3) no known allergy to botulinum toxin and the ingredients of toxin formulation; 4) no current use of anti-cholinergic medication, anti-convulsion medication, or drugs that affect salivation; and 5) no previous surgical or radiotherapy procedures to alleviate drooling.

Initial evaluation included body weight, Drooling Severity Scale (1-dry-never drools, 2-mild-only wet lips, 3-moderate-wet lips and chin, 4-severe-damp clothing, 5-profuse-wet clothing, hands and objects),<sup>[16]</sup> and Drooling Frequency Scale (1-never, 2-occasional, 3-frequent, 4-constant).<sup>[16]</sup> With patients sitting erect and awake, at least one hour after a meal, two absorbent dental rolls were placed in the mouth near the orifice of the parotid gland ducts for one minute to collect saliva and were immediately weighed by an electrical scale with a sensitivity of 0.01g. After deducting the dry weight of dental rolls, the net saliva weight was calculated. The saliva weight was recorded pre-injection and once every month during follow up. We diluted 100U botulinum toxin Type A (Botox, Allergan) with 1 ml sterile saline. The total dose injected into the bilateral parotid glands

was weight-dependent (2U per kg body weight).<sup>[9]</sup> We chose to inject the superficial portion of parotid gland lying between the masseter muscle and the mastoid process. First injection was administrated 1 cm behind the ramus of the mandible and the other was at the posterior margin of the masseter muscle 2 cm above the angle of the mandible. The injection was performed manually using a 27-gauge needle and 1-ml syringe 60 minutes after application of EMLA cream (local anesthetic, Astra).

Follow-up evaluation including drooling severity scale and drooling frequency scale was made at monthly intervals. Parents were inquired about the change of drooling manifestations and associated side effects. All three patients were followed at least 9 months after injection, in patient 2 lasting for 15 months. We injected botulinum toxin into the salivary glands and the spastic extremities at the same time in patients 2 and 3, both of them had severe limb spasticity that caused functional disability. The average dose was 1.0U/kg in upper limbs and 1.9U/kg in lower limbs. Botulinum toxin was injected again into the spastic extremities in these two patients four months after the first injection.

Drooling severity and frequency improved in all three patients but the drooling did not stop completely. All parents reported that the drooling improved within one week after injection. Although parents reported an overall improvement, they also described some diurnal and daily variations in drooling.

The weights of the saliva are shown in Figure 1. At the time of first follow-up, all three patients had reduced salivary flow. The saliva weight reached the lowest level at two months after injection in two patients, and at four months in patient 1. In patient 3, the weight of saliva was higher than the baseline seven months after injection. The other two patients showed lower than the baseline throughout the study period of 9 months.

The injection procedures were well tolerated in all three patients. No adverse effects including chewing or swallowing difficulty were found during the study. All three families requested further injection when inquired.

## DISCUSSION

The effect of botulinum toxin for drooling in children with cerebral palsy has not been reported in Taiwan

Table 1. Clinical Characteristics of the Three Patients

Patients	1	2	3
Diagnosis	Spastic Diplegia	Spastic Diplegia	Spastic Quadriplegia
Limbs spasticity by MAS	1	3	3
Drooling severity scale	4	4	4
Drooling frequency scale	3	3	3
Sex	Female	Male	Male
Age (years)	6	5	8
Weight (Kg)	15	13	23

MAS: Modified Ashworth Scale

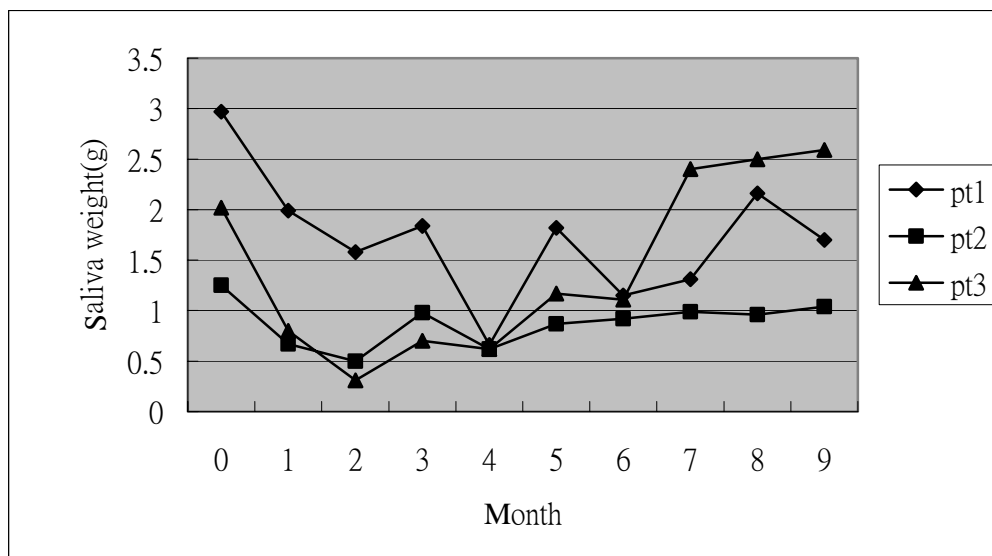


Figure 1. The net saliva weight gathered during a one-minute period before the injection of botulinum toxin and at the following monthly intervals.

yet. We reported the safety, efficacy, and the duration of effect on reducing salivary flow after a single dose botulinum toxin type A injection to bilateral parotid glands in three Taiwanese cerebral palsy children. The results are similar to western studies except that two of our patients had longer duration of effect.

Few studies had reported the long-term follow-up for duration of effect of botulinum toxin in salivary glands. Bhatia et al noted the improvement of drooling lasted 6 to 16 weeks after 20U Dysport injection to each parotid gland in three patients with neurodegeneration.<sup>[7]</sup> Jost reported that the duration of effect ranged from 4 to 7 months in five patients with Parkinsonism.<sup>[8]</sup> Jongerius et al injected the bilateral submandibular glands using a

2U/kg dose of Botox to treat drooling in three children with cerebral palsy and found that the salivary flow rate did not return to the baseline during the following 4 months.<sup>[9]</sup> According to the study by Suskind and Tilton, the duration of effect varied from 2 weeks to 6 months in 17 children with cerebral palsy.<sup>[10]</sup> The effect lasted 6 months in one of our patients and at least 9 months in the other two patients. We chose 9 months after injection as the end point of follow-up because patient 1 dropped out at that time. The duration of effect differed greatly in the two patients who received injection to both salivary glands and spastic extremities. In patient 3, the effect seemed to disappear 6 months after injection and the saliva weight was higher than baseline thereafter, while

the effect lasted for 14 months in patient 2. We do not know whether the drooling will rebound after botulinum toxin type A injection into bilateral parotid glands and it was not reported before. Theoretically, injection of botulinum toxin to the muscles of the upper and lower extremities should have no effect on salivary glands. These results may suggest there is great individual difference in the duration of effect. Similar findings were reported by Ellies et al.<sup>[12]</sup>

It remains unclear why there seems to be a longer duration of botulinum toxin's effect on neuroglandular junction, while the effect on neuromuscular junction is only about three months. Botulinum toxin Type A resulted in anhydrosis for over 6 to 12 months in patients with gustatory sweating.<sup>[17]</sup> Naumann et al postulated two mechanisms affecting the duration of effect.<sup>[18]</sup> First, re-innervation at the neuromuscular junction is related to axonal re-sprouting, and to date it is unknown whether re-sprouting occurs in autonomic nerve endings after administration of botulinum toxin. Second, the rate of SNAP-25 re-synthesis at the neuroglandular junction may be lower. Besides, Garrett pointed out that an absence of nerve stimulation would cause variable degenerative changes in the salivary glandular parenchyma that affect their re-synthetic and secretory capacities.<sup>[19]</sup>

Despite reduction of saliva flow after botulinum toxin injection, the patients retained sufficient secretion to avoid adverse events such as dry mouth. Our goal was to achieve a new balance between the production of saliva and swallowing rather than to completely inhibit saliva production. Previous study by Bhatia showed injecting the parotid glands alone could reduce drooling effectively.<sup>[7]</sup> So we chose to inject the superficially located parotid glands rather than the submandibular glands. We didn't utilize ultrasound to localize parotid glands; however, for more precise localization and to avoid every possible side effect, ultrasound guided injection is strongly recommended.

The families were all satisfied with the reduced drooling throughout the study period although they knew the effect would be temporary only

botulinum toxin injection can be effective in reducing saliva secretion and drooling in these cases of Taiwanese children with cerebral palsy.

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## CONCLUSION

The findings of the present study suggest that

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## 以 A 型肉毒桿菌毒素治療台灣腦性麻痺兒童的流涎問題： 三病例報告

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腦性麻痺患者合併有流涎問題的比例據不同統計有 10-37%，許多治療方法曾被提出且應用過，但沒有任何一項治療方法可以算是完全成功，且許多方法存在著不小的副作用。肉毒桿菌毒素目前被認為是治療腦性麻痺流口水問題的一項新的治療方法，此方法應用在台灣腦性麻痺兒童流口水問題的效果尚未有人報告過。本文報告以 A 型肉毒桿菌素注射於三位腦性麻痺兒童雙側耳下腺治療流口水的效果及可能的副作用。我們發現流口水的問題在三位腦性麻痺兒童接受肉毒桿菌素注射後皆獲得改善，且在追蹤的九個月期間未發現有副作用產生。與國外的研究結果比較，有二位兒童的效果較國外久。由此三例經驗顯示，A 型肉毒桿菌素注射於雙側耳下腺應可以有效且安全地治療腦性麻痺兒童流口水的問題。（台灣復健醫誌 2007；35(3)：165 - 170）

**關鍵詞：**A 型肉毒桿菌毒素(botulinum toxin type A)，流口水(drooling)，腦性麻痺(cerebral palsy)，  
耳下腺(parotid glands)