

# Treatment of cerebral palsy with botulinum toxin A: functional benefit and reduction of disability. Three case reports.

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## Summary

Three patients with cerebral palsy are described suffering, respectively, of pes equinus, spasm of the m. teres major and flexion spasm of the hand, who were treated with botulinum toxin A. These patients demonstrate not only the local reduction of the muscular hyperactivity following treatment with botulinum toxin A but also the potential functional benefit resulting from such a treatment. Thus, local intramuscular injection of botulinum toxin A in children with cerebral palsy should be considered as part of a multidisciplinary treatment concept, since reduction of the disability and the functional improvements could have high impact on daily living activities.

## Introduction

One of the leading symptoms of cerebral palsy is spastic muscular hyperactivity which is part of the severe movement disorders with major impact on the patients' disability. The aim of most current treatment strategies in these patients is to reduce the spastic muscular hyperactivity. One possible way to achieve this is by weakening selected muscles through chemical denervation using local injections of botulinum toxin A. Although botulinum toxin A acts by irreversible inhibition of acetylcholine release at the pre-synaptic neuromuscular junctions [1-3], subsequent growth of new neuromuscular junctions leads to reinnervation of the muscle within 3-6 months.

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In adults, botulinum toxin A is the therapy of choice in blepharospasm, hemifacial spasm and spasmodic torticollis and is effective and safe in the treatment of spasticity [4-6]. Several studies have shown that children with cerebral palsy, who were treated with botulinum toxin A, achieved reduction of local spastic muscular hyperactivity and improved their joint mobility [7-19]. This form of therapy in patients with cerebral palsy represents a local treatment of a generalized movement disorder. Thus, it is important to evaluate the impact of the local reduction of muscular hyperactivity on the patients' overall condition and daily activities.

In this report we present three children with cerebral palsy whose disabilities were reduced and their quality of every-day life improved following local intramuscular injections of botulinum toxin A.

## Subjects and methods

The following parameters were assessed before and at six months after the start of the treatment: spastic muscular hyperactivity using the modified Ashworth Scale [20] and joint mobility (range of motion). Using a standardized questionnaire — adapted from the German multicentre study on behavioural aspects in torticollis spasmodicus — an interview was conducted with the patient and the parents after 6 months of treatment [21-25]. During the interview the impact of the treatment with botulinum toxin A on disability and daily activities were discussed and evaluated.

## PATIENT 1

This patient is a 13-year-old boy with severe tetraspasticity with dystonic features due to perinatal asphyctic brain damage. He suffered of painful shoulder luxation caused mainly by muscular hyperactivity of the m. teres

major. This was considered the main disability factor both by the parents and the physicians. The spastic hyperactive muscle caused bending of the arm towards the back into a very uncomfortable position. During the night, the patient woke up 2–4 times requiring his parents to move him into a more comfortable position. Conservative treatment including physiotherapy and oral medication failed. Local intramuscular injection of botulinum toxin A into the m. teres major was performed and well tolerated.

Following treatment the arm was not bent towards the back, allowing him and his parents uninterrupted sleep for the first time in 12 years. Moreover, the patient was able to lift himself unaided into a sitting position; getting dressed, with the help of his parents, became easier and less painful. One treatment with botulinum toxin A was sufficient for up to seven months, after which the symptoms reappeared requiring re-injection. Over the last three years the patient was treated nine times with the intramuscular injection, maintaining the described functional benefits.

PATIENT 2

A 16-year-old girl with tetraspasticity presented with flexion contracture of the left hand. EMG-controlled injections of botulinum toxin A into the m. flexor carpi radialis and m. flexor digiti profundus were performed, resulting in decreased muscular hyperactivity and increasing her ability to extend the whole hand (Table 1). The extension splint was better tolerated during the night; physiotherapy and nursing of the open hand could be performed without causing pain. Maceration of the palmar surface disappeared.

As a result of treatment with botulinum toxin A the patient experienced another functional benefit which

strikingly improved her mobility. She was now able to properly hold onto a handlebar, thus enabling her for the first time to steer a tricycle.

PATIENT 3

This patient is a 15-year-old-boy with spastic diplegia and pes equinus. The patient had to be supported during standing and walking. Local injection of botulinum toxin A into the m. gastrocnemius on both sides led to a significant reduction of their muscle tone and increased joint mobility (Table 1). After the intramuscular injection of botulinum toxin A a significant improvement of the gait pattern was observed after two weeks, three weeks later the patient was able to walk unaided for about 50 metres for the first time in his life.

Discussion

Local weakening of muscles caused by botulinum toxin A reduces spastic muscular hyperactivity and increases joint mobility. While in adults this treatment is well accepted for various hypertonic muscle disorders, experience in children is limited. In recent years however, encouraging results have been reported following treatment of children with botulinum toxin A. This was illustrated by 13 separate studies with a total of 327 children with cerebral palsy, treated with botulinum toxin A [7–19].

Systemic side effects such as dysphagia and ptosis of the eyelid, reported in adults during the treatment of spasmodic torticollis [14,26,27] were not observed in these studies. Similarly, long-term side effects were not reported either during the clinical use of botulinum toxin A nor after food intoxication accidents, when the concentration reached was several thousand times

**Table 1** Dose regime, injected muscle, and local effect of botulinum toxin A

	Muscle	Mean dose* per		Ashworth scale		Range of motion				
		Muscle	Kg body weight	Pre	Post	Joint	Function	Pre	Post	Normal
Patient I	m. teres major	500 U	29 U	4	2	shoulder	abd/add rotation	40° 70°	160° 110°	225° 160°
Patient II	m. flexor carpi ulnaris m. flexor dig. prof. m. add. pollicis brevis	200 U 200 U 100 U	8,3 U	4	2	wrist	ext/flex	10°	40°	130°–150°
Patient III	m. gastrocnemius caput lat., caput med.	500 U	25 U	4	3	foot ankle	ext/flex	30°	40°	65°

\*Dysport®.

higher than the applied therapeutic doses [28–30]. Nevertheless, because of the limited experience and lack of long term observation periods with botulinum toxin A, the possibility of side effects should not be underestimated.

The three case reports described here demonstrate that treatment with botulinum toxin A leads not only to local muscle weakening and thereby reduction of muscular hyperactivity, it also provides functional improvement which has additional positive impact on the patients' overall amelioration of disabilities. Thus, local injection of the m. teres major led to a more comfortable sleeping position and reduced the nightly interruptions of the patient's and his parents' sleep. Local weakening of the m. flexor carpi ulnaris and the m. flexor digiti profundus made it possible to use the affected hand for support. Reduction of pes equinus permitted standing and walking without assistance, enabling the patient to perform daily personal activities more independently.

In these patients it could be demonstrated that local injections of botulinum toxin A reduce the patients' disability and thus alleviate the need of assistance. In order to achieve more than local muscle weakening, careful evaluation of realistic treatment goals with the patient, parents, physiotherapists and physicians is an important part of the procedure.

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