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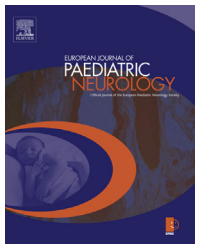

Original article

Effects of botulinum toxin A and/or bimanual task-oriented therapy on upper extremity impairments in unilateral Cerebral Palsy: An explorative study

Lucianne Speth [a,b,*], *Yvonne Janssen-Potten* [b,c], *Pieter Leffers* [d], *Eugene Rameckers* [b,c], *Anke Defesche* [a], *Bjorn Winkens* [e], *Jules Becher* [f], *Rob Smeets* [b,c,g], *J.S.H. Vles* [h,i]

[a] *Adelante, Paediatric Rehabilitation Centre, Onderstestraat 29, 6301 KA Valkenburg, The Netherlands*
[b] *Adelante, Centre of Expertise in Rehabilitation and Audiology, Hoensbroek, The Netherlands*
[c] *Maastricht University, Research School CAPHRI, Department of Rehabilitation Medicine, Maastricht, The Netherlands*
[d] *Maastricht University, Department of Epidemiology, Maastricht, The Netherlands*
[e] *Maastricht University, Department of Methodology and Statistics, Maastricht, The Netherlands*
[f] *Free University Medical Centre, Department of Rehabilitation Medicine, Amsterdam, The Netherlands*
[g] *Maastricht University Medical Centre, Department of Rehabilitation Medicine, Maastricht, The Netherlands*
[h] *Maastricht University Medical Centre, Department of Neurology, Maastricht, The Netherlands*
[i] *Maastricht University, Research School GROW, Department of Neurology, Maastricht, The Netherlands*

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ABSTRACT

Objective: This study reports on the effects of botulinum toxin A (BoNT-A) injections in the upper extremity (UE) of children with unilateral Cerebral Palsy (uCP) combined with bimanual task oriented therapy (BITT) or either treatment modality performed separately on UE range of motion (ROM), spasticity and (functional) strength.

Methods: Thirty-five children, mean age 7.14 years (SD 2.63) of whom 11 had a Manual Ability Classification Score (MACS) I, 15 MACS II and 9 MACS III, participated. The trial started with four study groups: BoNT-A-only (n = 5), BITT-only (n = 11), BoNT-A + BITT (n = 13), and control (n = 6). Twenty-two children were randomized and, due to recruitment problems 13 children received their parents' preferred treatment: BoNT-A + BITT or BITT-only. Three comparisons were analysed: BITT (BoNT-A + BITT and BITT-only; n = 24) versus no BITT (BoNT-A-only and control; n = 11), BoNT-A (BoNT-A-only and BoNT-A + BITT; n = 18) versus no BoNT-A (BITT-only and control; n = 17), and the additional effect of BoNT-A (BoNT-A + BITT versus BITT-only).

Results: BoNT-A significantly decreased key grip strength and finger flexion tone, had a clinically relevant (additional) positive effect on active thumb abduction and supination

* *Corresponding author*. Adelante, Paediatric Rehabilitation Centre, Onderstestraat 29, 6301 KA Valkenburg, The Netherlands. Tel.: +31 45 5282610.
E-mail address: l.speth@adelante-zorggroep.nl (L. Speth).
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and a significantly negative effect on unilateral functional strength. BITT + BoNT-A significantly increased active supination. BITT reduced elbow flexor tone and BITT-only resulted in more improvement than BoNT-A + BITT in functional unimanual and, to a lesser extent, in bimanual grip strength.

Conclusions: In comparison with BoNT-A + BITT, BITT-only gives more improvement on functional grip strength and, therefore, could possibly increase bimanual performance. In this case, the (additional) role of BoNT-A may be an increase in active supination and thumb abduction.

1. Introduction

Grip strength[1,2] and active forearm supination[2] are good predictors for use of the affected arm in bimanual performance of children with predominantly unilateral Cerebral Palsy (uCP) and a Manual Ability Classification Score (MACS) I to III.[3] Muscle weakness and spasticity are assumed to jointly contribute to activity limitations, reflected by the strong relationship between the ability to voluntarily activate a muscle and performance.[4] Children with uCP have been treated with botulinum toxin A (BoNT-A) to reduce impairments in arm and hand function in order to improve use of the affected hand directly or to facilitate the effectiveness of physical and occupational therapy in achieving their individual goals at the activity level of the International Classification of Functioning, Disability and Health for Children and Youth (ICF-CY) of the World Health Organization (WHO) (http://www.who.int/classifications/icf/en/).

In a randomised clinical trial (RCT) on the effect of BoNT-A injections in the UE of children with uCP who received intensive bimanual therapy, we found an increase of active wrist extension of more than 30° and tone reduction of the wrist flexors.[5] These findings were in line with the Cochrane review concluding that BoNT-A injections clearly reduce spasticity and improve range of motion (ROM), but that the effect on performance at activity level is less clearly esthablished.[6] This uncertain effect of BoNT-A on the ICF activity level might be caused by insufficient sensitivity of the measurement instruments used in the trials to detect an existing improvement, or by the fact that BoNT-A might negatively influence muscle strength.

Morphological comparison between wrist flexor and extensor muscles in children with CP shows myopathy of the wrist flexors.[7] The strength in the wrist and finger flexors of the affected hand in children with uCP is only about one third of the strength of their non-affected hand, or that of typically developing children.[8] An RCT on the effect of BoNT-A injections in children with CP receiving functional rehabilitation therapies for the UE showed that increase of muscle strength from therapy is attenuated by BoNT-A injections.[9] Also, in studies on the effect of BoNT-A injections in UE muscles, a transient decrease in grip strength has frequently been reported as a side effect.[6,10]

In a pilot study of 10 children with uCP, Elvrum et al. showed that additional resistance training after the use of BoNT-A resulted in temporary strengthening of the non-injected muscles, reduction of strength loss resulting from BoNT-A without increasing tone, and greater increase of active supination.[11] Although the BoNT-A injections and the resistance training involved only the upper and forearm, and not the hand and wrist muscles, they found an improvement in grip strength in the resistance training group. However, the improvements of these impairments did not result in increased use of the affected hand. The authors questioned whether task-oriented training is a more effective approach than strength training to improve arm and hand use.

Bimanual performance is influenced by grip strength and, to a lesser extent, by active ROM (AROM) of wrist extension and supination.[1,2] As bimanual intensive therapy has proven to have a positive effect on bimanual performance,[12] the objective of the present explorative study is to report effects of BoNT-A injections in the UE in children with uCP combined with bimanual task-oriented therapy (BITT) or either treatment modality separately on UE AROM, spasticity and isometric and functional strength. The latter is used, because it is a better representative for actual strength in daily life.[13]

2. Methods

2.1. Design

This explorative study is part of the BoBiVa (Botuline toxine Bimanuele Vaardigheden) study (http://www.controlled-trials.com/ISRCTN69541857). The BoBiVa study was designed as a multicentre, randomized controlled trial on the effect of BoNT-A injections combined with bimanual task oriented therapy (BITT), or either separately, in children with uCP on UE functions and skills. Medical ethics approval was provided by the METC Atrium-Orbis-Zuyd (ref: 06-p-33) and the Dutch CCMO (ref: NL12005.096.06). The parents gave their informed consent. Initially, besides Adelante/Maastricht University Medical Centre, two other centres participated in this trial. Due to disappointing patient enrolment, other centres joined later. In this paper, the results at the body function level of the ICF-CY are reported. A factorial design was used to determine the effects of BoNT-A and/or BITT (Table 1). As far as possible, the CONSORT guidelines for reporting parallel group randomized trials were followed.[14]