Pediatric Rehabilitation

ISSN: 1363-8491 (Print) (Online) Journal homepage: https://www.tandfonline.com/loi/ipdr19

Botulinum toxin type-A and plaster cast treatment in children with upper brachial plexus palsy

M. Basciani & D. Intiso

To cite this article: M. Basciani & D. Intiso (2006) Botulinum toxin type-A and plaster cast treatment in children with upper brachial plexus palsy, Pediatric Rehabilitation, 9:2, 165-170, DOI: 10.1080/13693780500402229

To link to this article: https://doi.org/10.1080/13693780500402229

Published online: 28 Oct 2009.

Pediatric Rehabilitation, April 2006; 9(2): 165–170

Taylor & Francis
Taylor & Francis Group

Botulinum toxin type-A and plaster cast treatment in children with upper brachial plexus palsy

M. BASCIANI & D. INTISO

Department of Rehabilitation, IRCCS 'Casa Sollievo della Sofferenza', S. Giovanni Rotondo (FG), Italy

(Received 10 May 2005; accepted 11 August 2005)

Abstract

Background and purpose: Electrical stimulation, physical therapy and occupational therapy remain the main treatment for children with upper brachial plexus palsy (UBPP), when surgery has been excluded. A pilot study was undertaken to investigate whether botulinum toxin type A (BoNT-A) and plaster casting, as adjunct to the physical therapy, decreased muscle contracture and improved the position and function of the impaired arm.

Method: Twenty-two children (mean age 5.6 ± 3.4 years) with mild UBPP who previously underwent serial cast treatment, unsuccessfully, were enrolled. Neurological impairment and functional status were quantified using Medical Research Council (MRC) and Mallet scales and the Nine-Hole Peg Test (NHPT). Elbow extension was measured using a goniometer. Biceps brachii, brachialis, pronator teres and pectoralis major muscles were injected with 22 units kg^{-1} BoNT-A (Dysport®, Ipsen). After injection, the treated arm was fixed with a plaster cast and progressively lengthened over 14 days. The cast was maintained for 30 days. Assessments of elbow extension, MRC, Mallet Scale and NHPT were made at baseline, 3, 6 and 12 months.

Results: After BoNT-A injection, children had significant improvement of active elbow extension ($15.5^\circ \pm 17.1$ at 12 months after injection, compared with $42.0^\circ \pm 10.4$ at baseline; $p < 0.001$). NPHT scores improved significantly over the 12 months (51.1 ± 21.8 seconds compared with 56.7 ± 19.3 seconds at baseline, $p < 0.01$). MRC and Mallet scale scores of the paretic muscles were unchanged.

Conclusion: The children showed a reduction in muscular contracture and improvements of the arm position and elbow extension. The data support the use of BoNT-A and plaster casting as an adjunct to physical therapy, in the treatment of children with mild UBPP.

Keywords: *Obstetric brachial plexus palsy, botulinum toxin, rehabilitation, children, plaster cast, BoNT-A (Dysport)*

Introduction

Obstetric brachial plexus palsy can represent a dramatic sequela of dystokia or complicated delivery. Despite improved procedures and diagnosis, the incidence remains unchanged ranging from 0.9–2.6/1000 live births, possibly reflecting an increase in mean birth weight [1,2]. The clinical aspects of the brachial plexus palsy can be very variable and complex, involving several nerve roots and muscle groups. About 80–85% of perinatal brachial plexus injuries concern the upper branch (Erb's type) affecting the C5, C6 and sometimes C7 nerve roots [3,4]. In severe brachial plexus palsies, disabling limitation can result, due to impairment and imbalance of the muscular contraction in the paretic limb. Until now, the strategies used to treat this condition have been either a surgical or a conservative approach, depending on the severity of the brachial plexus lesion. Different surgical techniques have been reported to improve the movement and functional ability in children who had not recovered anti-gravity power of the biceps within the first 3 months of life [5–8]. The hallmarks of conservative management in children with mild brachial plexus palsy remain physical and occupational therapy and electrical stimulation [4,9–12]. In spite of physical therapy, some children continue to experience muscular imbalance, contractures and abnormal posture which hamper complete recovery. Casting is a common therapeutic strategy to treat muscular unbalance and the contractures, even if the main application concerns the reduction of contractures following the spasticity, particularly in cerebral palsy and brain injury [13–15]. Serial casting results in a higher active and passive joints mobility and can enable functional use of affected limb.

Correspondence: D. Intiso, MD, Department of Rehabilitation, IRCCS 'Casa Sollievo della Sofferenza', Viale dei Cappuccini 1, 71013 S. Giovanni Rotondo (FG), Italy. Tel/Fax: 0882-410942. E-mail: fisiatria@operapadrepio.it

ISSN 1363–8491 print/ISSN 1464–5270 online/06/020165–170
DOI: 10.1080/10.80/13693780500402229

The authors undertook a pilot study to investigate whether botulinum toxin type-A (BoNT-A) injection and plaster casting, as adjuncts to physical therapy, reduced muscle contracture and improved the posture and functional status of the impaired limb in children with mild obstetric upper brachial plexus palsy (UBPP).

Method

After gaining parental consent and ethical committee, children were treated with mild UBPP following dystokic delivery. These children had been previously treated by serial casting to obtain the improvement of elbow extension, unsuccessfully, and in spite of physical therapy they presented stable impaired functional ability and range of motion in the paretic arm. Their demographic details are given in Table I. At baseline, the children underwent echography and X-ray scan of the elbow and shoulder joints. Children with severe UBPP and complicated orthopaedic disturbances including dislocation, luxation, sub-luxation and pathological calcification of the elbow and shoulder joint were excluded. Electromyographic (EMG) examination was performed to confirm Erb's type plexus palsy and to identify the innervation status of involved muscles. All patients also underwent a neurological and functional assessment of the impaired arm. Motor function was evaluated using the Medical Research Council (MRC) and Mallet scales [16,17]. MRC is a 6-point graded scale ranging from 0–5 to quantify the power in isolated or muscle groups: 0, no contraction; 1, flicker or trace of contraction; 2, active movement with gravity eliminated; 3, active movement against gravity; 4, active movement against gravity and resistance; and 5, normal power. The Mallet scale includes active abduction, active external rotation and the ability to bring the hand to the mouth, the back and the neck. These five parameter are rated on scale from 1 (absence of function) to 5 (normal function). The Nine-Hole Peg Test (NHPT) was used to evaluate the functional use of the affected arm [18]. Although the MRC scale is widely used to quantify muscular impairment following nerve and muscle injuries, the NHPT is used to evaluate motor dexterity, especially when the functional limitation of the upper limb is not severe. In this test, the subject is seated at a table and is invited to insert nine small pegs into holes located in a 100 mm^2 wooden board. The time taken (in seconds) to perform this task is recorded. The degree of elbow extension was measured using a goniometer. Passive and active goniometric evaluations of the affected limb were performed at baseline and at 3, 6 and 12 months after treatment. The biceps brachii, brachialis, pronator teres and pectoralis major muscles were injected with a total of 200–400 units per single session (mean doses 22 ± 5.1 units per Kg of body weight) BoNT-A (Dysport® Ipsen) [19,20] (Table II). These muscles were selected on the basis that a protracted muscular imbalance favouring the action of flexors, pronators and internal rotators was responsible of characteristic posture and disability in UBPP children. After the first injection, the position of the treated arm was fixed in elbow extension, by a plaster cast, which was lengthened each week for 2 weeks. The plaster cast was removed after 30 days and no further casts were applied. In the study, repeat injections of BoNT-A were given at intervals of 12 weeks for up to 9 months, if the improvements in elbow extension and arm posture were not sustained. During BoNT-A treatment, the children continued with a standard physical therapy rehabilitation programme, including proprioceptive neuromuscular facilitation, stretching and occupational therapy.

Table I. Demographic characteristics of children with UBPP.

	Number	Age (years), M ± SD	Affected side Left	Affected side Right
Male	12	5.5 ± 3.9	6	6
Female	10	5.6 ± 2.8	4	6
Total range	22	5.6 ± 3.4 (3–15)	10	12

UBPP = upper brachial plexus palsy.

Table II. Muscles and BoNT-A doses (mouse unit). Standard deviation is reported in brackets.

Muscles	Pectoralis major	Biceps brachii and brachialis	Pronator teres
Mean doses for single session	88 (17.6)	219 (65.3)	112 (38.8)
Mean doses per kg of body weight (SD)	22 (5.1)		

Subjects

Twenty-two children (12 male and 10 female, mean ages 5.6 ± 3.4 years) were enrolled who have been previously treated by serial casting to obtain the improvement of elbow extension, unsuccessfully. Of these, 10 children did not tolerate the cast. The mean elbow extension resulted in $41.9^\circ \pm 9.6$ and $42.3^\circ \pm 10.6$ before and after casting treatment, respectively, and in spite of physical therapy the children presented a stable range of elbow extension over time. All patients had mild Erb's plexus palsy affecting C5 and C6 nerve roots and anti-gravity function of the biceps muscle. They showed mild impairment of abduction and external rotation of

Table III. Mean scales scores and active angular elbow extension.

	Baseline M ± SD	3 months M ± SD	6 months M ± SD	12 months M ± SD
NHPT (s)	56.7 ± 19.3	54 ± 21.6*	52.7 ± 21.6*	51.1 ± 21.8*
Mallet score***	3.1 ± 0.6; 3	3.2 ± 0.4; 3	3.2 ± 0.4; 3	3.2 ± 0.4; 3
Elbow extension (°)	42° ± 10.4	20.17 ± 16.9**	17 ± 16.2**	15.5 ± 17.1**

NHPT = Nine-hole peg test; SD = standard deviation.
* $p < 0.01$: NHPT scores vs baseline; ** $p < 0.001$: Degree of elbow extension vs baseline; *** mean and median.

the shoulder, as assessed by the Mallet Scale. Almost all children showed a limitation of active elbow extension with a posture of mild internal rotation and adduction of the shoulder, elbow flexion and a pronation contracture of the forearm. Complete passive extension of the elbow with severe pain was possible in 15 children. No child had painless extension of the elbow at baseline.

Statistical analysis

Multiple paired data for the scale scores and goniometric values obtained after casting and BoNT-A treatment at 3, 6 and 12 months of follow-up were compared with the baseline values using ANOVA, adjusted for repeat measurements.

Results

MRC scale scores

The MRC scale scores showed that the deltoid muscle had the greatest impairment. At baseline, the median MRC score of deltoid, biceps brachii and triceps muscles were 3, 3.5 and 3, respectively. The values were unchanged at 3, 6 and 12 months of follow-up. The mean MRC score was 3.1 ± 0.4; 3.4 ± 0.4; and 3 ± 0.2 for the deltoid, biceps and triceps muscles, respectively, at baseline. The values remained fairly stable throughout the observation period: 3.2 ± 0.4; 3.4 ± 0.3; and 3.1 ± 0.3, respectively, at 3 months; 3.2 ± 0.3; 3.4 ± 0.4; and 3.1 ± 0.2, respectively, at 6 months; and 3.2 ± 0.4; 3.4 ± 0.4; and 3.1 ± 0.2, respectively, at 12 months. EMG showed chronic partial denervation of the scapular girdle muscle with impaired motor unit activation involving the deltoid, supra-scapular and infra-spinous muscles. None of the children had biceps and triceps muscle co-contraction in the affected arm.

Angle of elbow extension

After BoNT-A injection and plaster cast treatment, there was a significant improvement in active elbow extension in all but four of the children. At baseline, mean extension was 42° ± 10.4°, which was progressively reduced to 20.1° ± 16.9°, 17° ± 16.26° and 15.5.° ± 17.14°, at 3, 6 and 12 months after treatment, respectively ($p < 0.001$) (Table III). In these patients a reduction of pronation and an improved range of motion of the elbow joint were obtained. This benefit persisted at 12 months, even in the absence of further injections. Children under the age of 5 years with mild impairment of deltoid and biceps muscles had the best outcomes. The four children who did not ameliorate were older (mean age 7.5 ± 1.2) than children responsive to the treatment (mean age 5.25 ± 3.4). They did not show differences in the range of elbow extension (42.2 ± 8.6 vs 41.8 ± 9.5) and arm dexterity compared to the improved children. All four children after the first BoNT-A injection and casting treatment underwent further botulinum A toxin injection sessions, every 12 weeks for up to 9 months, unsuccessfully.

Ten patients achieved complete active extension of the elbow, after a single treatment with BoNT-A injection and the plaster cast treatment (Figure 1). Eight children had two and four children had three BoNT-A injections. The children who did not improve after the second injection did not benefit after further injection. After casting and BoNT-A injections a complete passive painless extension of the elbow was obtained in 14 of the 15 patients who complained severe pain to the elbow extension, at baseline.

NHPT

The NHPT scores decreased significantly after BoNT-A and casting treatment and this functional improvement persisted at 12 months (Table III). The mean time taken to perform this test was reduced from 56.72 ± 19.3 seconds at baseline, 54 ± 21.6 s; 52.72 ± 21.6 s; and 51.1 ± 21.8 s at 3, 6 and 12 months, respectively ($p < 0.01$).

Mallet scale scores

The median Mallet scores changed little throughout the observation period.

Adverse events

Two patients, who had good elbow extension, reported articular pain lasting 5 days after removal of the plaster cast.

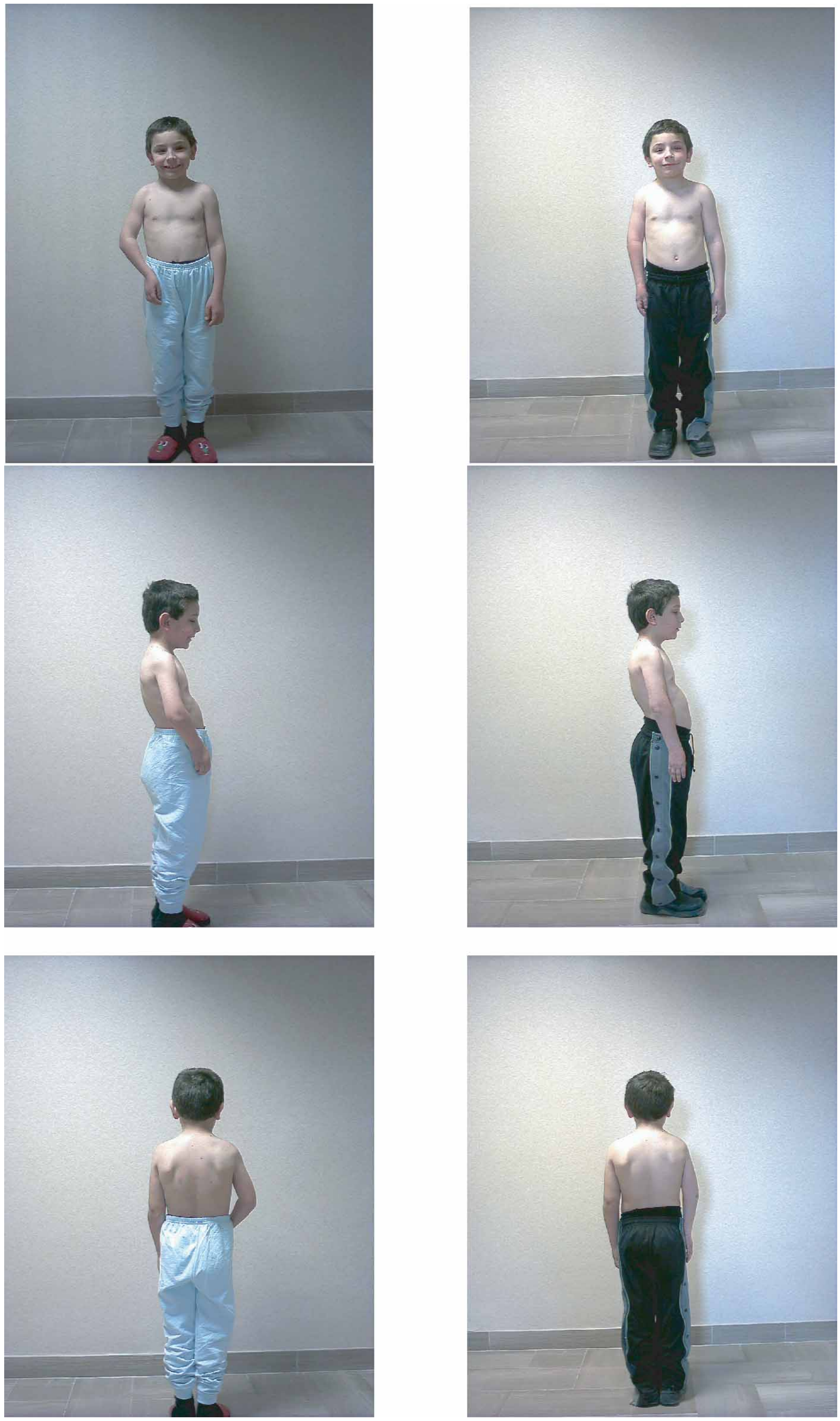

Figure 1. A child with upper brachial plexus palsy before (left) and after (right) BoNT-A and plaster cast treatment.

Discussion

Although there have been previous published reports on the use of botulinum A toxin in the treatment of children with birth-related brachial plexus lesions [19,20], to the best of the authors' knowledge, this is the first report of the combination of BoNT-A with plaster casting in children with mild UBPP. It has been shown that this combination, in conjunction with physical therapy, is a promising new therapeutic strategy in children with this condition.

This treatment increases elbow extension and facilitates improvements in both the posture and function of the affected arm. The prognosis of the brachial plexus palsy varies considerably, depending on the severity of the nerve root injury. Many children with perinatal brachial plexus palsy make a good spontaneous recovery [10–12]; however, others may need aggressive physical therapy to maintain passive range of motion, supple joints and muscle strength if a surgical approach has been excluded. Children with mild UBPP can develop a permanent disability due to the functional limitation and reduced motor skills of the affected arm, despite sufficient re-innervation of the paretic muscles. When supinators and external rotators have been denervated, the unopposed action of the innervated pronators and internal rotators results in a typical pronation contracture at the forearm and posture of the affected arm. This type of muscular unbalance significantly hinders a patient's two-handed grooming activities. The best results were obtained in children under 5 years of age. Several hypotheses could explain this finding, including peripheral and central pathophysiological mechanisms. Early treatment with BoNT-A can prevent irreversible contracture, skeletal deformities and ankylosis. It can also slow myofascial and capsular-ligamentous fibrosis of the joints. The impaired activity and reduced motor skills in children with neonatal brachial plexus palsy might be due to an apraxia arising as a consequence of paralysis at a critical time in infant development [21]. The reduced level or absence of proprioceptive sensory inputs and normal movements early in life could produce abnormal changes in spinal cord and brain development. These, in turn, could lead to abnormal motor neuron recruitment and impaired development of skilled movements [22]. It is possible that treatment with BoNT-A, in conjunction with physical therapy, may modify the muscular imbalance, alleviate problems with posture and motor patterns in the affected arm and could prevent apraxic motor patterns from developing. In this study, since the affected arm was treated with a plaster cast for 30 days after the first BoNT-A injection to promote muscle elongation, it could be argued that the findings resulted from the orthopaedic intervention. However, children were enrolled who previously underwent a traditional plaster cast treatment of paretic arm, but the motion of the elbow and disability of the affected arm were unchanged. It is possible that a greater period of cast application may have produced more positive effects, but the children could not tolerate the orthopaedic orthosis. It was, therefore, believed that the cast reinforces the beneficial effects of BoNT-A injection. Indeed, Desiato and Risina [20] reported good results in children with UBPP, using BoNT-A injections alone. The functionality of the affected arm improved significantly after casting and BoNT treatment as NHPT score (56.72 ± 19.3 and 51.1 ± 21.8 seconds at baseline and at 12 months, respectively) changed, but this finding could simply be explained by maturity as children get older. Although the data from this study are promising, the open design, the small number of patients and the lack of a control group represent limitations to the interpretation of the findings. A proper controlled randomized study of casting alone compared to casting plus botulinum toxin treatment is needed for conclusive remarks. It may be argued that this therapeutic strategy may be helpful in similar pathological conditions such as acquired brachial injuries in adults or in children with UBPP following surgical repair, but according to the findings, one can not support this suggestion. Indeed, the best outcome was obtained in the children under the age of 5 years. Some may consider that the treatment of mild UBPP is unnecessary, since these children are only slightly impaired. However, the sustained results on position of the affected arm, functional improvements, good recovery, with few side effects, would argue against this. The authors hold that it is important to strive for new means to attain the best functional benefits which improve the quality of life in these children.

Conclusion

The complexity and variability of clinical presentations of perinatal brachial plexus palsy requires that different therapeutic strategies be considered at different times, to provide the best outcome for the affected children. The patients showed reduction of muscular contracture and improvements in arm position, elbow extension and dexterity in the paretic limb, even though the neurological impairments remained unchanged. It is proposed that the use of BoNT-A injections together with plaster casting, as an adjunct to physical therapy, be considered for the treatment of children with upper brachial palsy. Further studies including a blinded design and control group are needed before firm conclusions can be drawn.

References

1. Rouse DJ, Owen J, Goldenberg RL, Cliver SP. The effectiveness and costs of elective cesarean delivery for fetal macrosomia diagnosed by ultrasound. Journal of American Medical Association 1996;276:1480–1486.
2. Kay SPJ. Obstetrical brachial palsy. British Journal of Plastic Surgery 1998;51:43–50.
3. Al-Qattan MM, Clarke AM, Curtis CG. Klumpke's birth palsy. Does it really exist? Journal of Hand Surgery 1995;20:19–23.

4. Shenaq SM, Berzin E, Lee R, Laurent JP, Nath R, Nelson MR. Brachial plexus birth injuries and current management. Clinical Plastic Surgery 1998;25:527–536.
5. Clarke HM, Al-Qattan MM, Curtis CG, Zuker RM. Obstetrical brachial plexus palsy: results following neurolisys of conducting neuromas-continuity. Plastic & Reconstructive Surgery 1996;97:974–984.
6. Capek L, Clarke HM, Curtis CG. Neuroma-in-continuity resection: early outcome in obstetrical brachial plexus palsy. Plastic & Reconstructive Surgery 1998;102:1555–1562.
7. Al-Zaharani S. Combined Sever's release of the shoulder and osteotomy of the humerus for Erb's palsy. Journal of Hand Surgery [British] 1997;22:591–593.
8. Chuang DC, Ma HS, Wei FC. A new strategy of muscle transposition for treatment of shoulder deformity caused by obstetric brachial plexus palsy. Plastic & Reconstructive Surgery 1998;101:686–694.
9. Hoffer MM. Brachial plexus palsies in neonates. Western Journal of Medicine 1998;168:126.
10. Greenwald AG, Schute PC, Shiveley JL. Brachial plexus birth palsy: a 10-year report on the incidence and prognosis. Journal of Pediatric Orthopedics 1984;4:689–692.
11. Bager B. Perinatally acquired brachial plexus palsy-a persisting challenge. Acta Paediatrica 1997;86:1214-1219
12. Ramos LE, Zell JP. Rehabilitation program for children with brachial plexus and peripheral nerve injury. Seminars in Pediatric Neurology 2000;7:52–57.
13. Stoeckmann T. Casting for the person with spasticity. Top Stroke Rehabilitation 2001;8:27–35.
14. Phol M, Ruckriem S, Mehrholz J, Ritschel C, Strik H, Pause MR. Effectiveness of serial casting in patients with severe cerebral spasticity: a comparison study. Archives of Physical Medicine Rehabilitation 2002;83:784–790.
15. Singer BJ, Jegasothy GM, Singer KP, Allison GT. Evaluation of serial casting to correct equinovarus deformity of the ankle after acquired brain injury in adults. Archives of Physical Medicine Rehabilitation 2003;84:483–491.
16. Medical Research Council. Aid to the examination of the peripheral nervous system. London: Her Majesty's Stationary Office; 1976.
17. Mallet J. Primautè du traitement de l'èpaule; Methode d'expression des resultats (Part of Symposium: paralysie obstétricale du plexus brachial. Traitement des sequelles. Rev Chir Orthop Reparatrice Appar Mot 1972;58(Suppl):166–168.
18. Mathiowetz V, Wolland G, Kashman N, Weber K. Adult norms for the box and the block test of manual dexterity. American Journal of Occupational Therapy 1985;39:386–391.
19. Rollnik JD, Hierner R, Schubert M, Shen ZL, Johannes S, Troger M, Wohlfarth K, Berger AC, Dengler R. Botulinum toxin treatment of cocontractions after birth-related brachial plexus lesions. Neurology 2000;55:112–114.
20. Desiato MT, Risina B. The role of botulinum toxin in the neuro-rehabilitation of young patients with brachial plexus birth palsy. Pediatric Rehabilitation 2001;4:29–36.
21. Brown T, Cupido C, Scarfone H, Pape K, Galea V, McComas A. Developmental apraxia arising from neonatal brachial plexus palsy. Neurology 2000;55:24–30.
22. Noetzel MJ, Wolpaw JR. Emerging concepts in the pathophysiology of recovery from neonatal brachial plexus injury. Neurology 2000;55:5–6.