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The effect of botulinum toxin type A and a variable hip abduction orthosis on gross motor function: a randomized controlled trial

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Hip displacement is the second most common deformity after equinus in children with cerebral palsy (CP), and may result in dislocation, pain, fixed deformity and loss of function. We studied the combined effects of intramuscular injections of botulinum toxin type A (BTX-A) to the adductors and hamstrings and a variable hip abduction orthosis (SWASH), on gross motor function, hip displacement and progression to surgery, in a randomized clinical trial. Thirty-nine children, with bilateral spastic cerebral palsy, and mean age 3 years + 2 months (range 1 year + 7 months–4 years + 10 months) entered the trial. Gross Motor Function Classification System (GMFCS) levels were as follows: one child was level II, 11 were level III, 13 were level IV and 14 were level V. After concealed randomization, 20 were allocated to the control group and 19 to the intervention group. Thirty-five children completed the follow up at 1 year. The novel intervention group received up to 4.0 U BOTOX®/kg/muscle, 16 U/kg/body weight every 6 months plus the use of a SWASH brace. The control group received clinical best practice comprising physiotherapy but no hip abduction bracing. Both groups showed improvements in total Gross Motor Function Measure (GMFM) score [mean 6.0% BTX-A group; 6.1% Control; 95% CI – 6.7, 6.5 (NS)], however, there was no additional treatment effect for the study group. There were similar improvements on GMFM goal scores and GMFM-66 scores, but again no additional treatment effect was observed. Multiple regression of change in total GMFM by GMFCS classification for each group showed greater improvement in the total scores from baseline in the BTX-A/SWASH treated group than the control group. In the first year, nine children (two in the intervention group and seven in the control group) required soft tissue surgery because of progressive hip migration in excess of 40%. A longer-term follow up of a larger cohort may be required to determine the effect of the combined treatment on hip displacement.

Introduction

In children with cerebral palsy (CP), hip displacement is the second most common deformity, after equinus (Cornell, 1995). Progressive hip subluxation and eventually dislocation may occur in these children without intervention. The incidence of hip dislocation varies according to severity of involvement, but 25–30% is the average figure in most large series (Beals, 1966; Samilson *et al.*, 1972; Fujiwara *et al.*, 1976). Hip dislocation in a patient with spastic cerebral palsy may result in significant morbidity because of pain, fixed deformity, and the loss of the ability to sit, stand and walk (Sharrad *et al.*, 1975; Cooperman *et al.*, 1987; Miller *et al.*, 1997).

Attempts to manage hip displacement vary from conservative options such as muscle stretching, postural management (seating and standing frames, static abduction braces) (Nakamura and Ohamu, 1980; Bower, 1990) to intramuscular injections of botulinum toxin type A (BTX-A) (Heinen *et al.*, 1999). Soft tissue and bony reconstructive surgery have also been used extensively (Cornell *et al.*, 1997; Miller *et al.*, 1997). Bracing is perceived as being poorly tolerated, resulting in poor compliance and with no evidence of benefit in the management of spastic hip disorders (Bleck, 1987). Hoffer (1986) questioned the efficacy of braces to prevent hip dislocation although he supported their use in maintaining range of motion after soft tissue

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surgery, without supporting data. There have been no randomized controlled trials investigating the outcome of hip displacement using currently available management options.

There have been no randomized trials of the effect of intramuscular injections of BTX-A on gross motor function in children with moderate to severe cerebral palsy. There have been prospective studies of BTX-A for adductor spasticity (Heinen *et al.*, 1999; Kirschner *et al.*, 1999). There are good quality randomized trials investigating the effects of intramuscular injections of BTX-A on gross motor function in less severely involved children with hemiplegia (Love *et al.*, 2001), as well as in children with diplegia (Flett *et al.*, 1999).

When considering new methods of management in spastic type hip displacement in children with cerebral palsy, the outcome should ideally be determined at skeletal maturity. However, in the intervening period it is important to assess the effect of intervention on gross motor function.

The Gross Motor Function Measure (GMFM) is the most widely used standardized outcome measure to establish baseline profiles and to detect change before and after intervention. It has been shown to have reliability, validity, and sensitivity to detect clinically important change in motor function in children with cerebral palsy (Russell *et al.*, 1989; Rosenbaum *et al.*, 1990; Bower *et al.*, 1996). The interobserver reliability of the GMFM score has been reported as 0.99 and intraobserver reliability 0.99 (Russell *et al.*, 1989). The originators reported a mean change in total GMFM score of 4.0% in 77 children with diplegia and a mean change score of 2.1% in 56 children group spastic quadriplegia over a period of 5.5 months (Russell *et al.*, 1993; Appendix III). The GMFM has been used as a primary outcome measure before and after Selective Dorsal Rhizotomy (SDR) (Steinbok *et al.*, 1997; McLaughlin *et al.*, 1998; Wright *et al.*, 1998) and more recently in a meta-analysis of SDR trials (McLaughlin *et al.*, 2000). In these randomized trials, the increase in GMFM scores in the control groups, who received standardized physiotherapy (PT), varied from 1.9% to 7.2% (Steinbok *et al.*, 1997; McLaughlin *et al.*, 1998). In one of the intervention groups who received SDR and PT, the increase in GMFM was 7.0% (Steinbok *et al.*, 1997).

GMFM has been correlated with muscle performance (Parker *et al.*, 1993) and with the temporospatial characteristics of gait (Damiano and Abel, 1996) in children with CP and mild impairment. A ceiling effect has been reported when measuring outcomes with the GMFM in children with hemiplegia and mild diplegia (Damiano and Abel, 1996) and a floor effect has been proposed in measuring functional change in children with quadriplegia following SDR (Nordmark *et al.*, 2000). Nordmark and colleagues compared assessment of functional outcomes in children undergoing SDR combined with physiotherapy with the GMFM and Pediatric Evaluation of Disability Inventory (PEDI). Both instruments were found to be sensitive to change over time in the series as a whole but there were greater improvements in the group with milder impairment with Gross Motor Function Classification System (GMFCS) levels II & III (Palisano *et al.*, 1997). In the group with more severe impairment (GMFCS levels IV & V), functional changes were detected with the PEDI but not the GMFM.

Intramuscular BTX-A has been shown to be effective in the management of equinus gait (Corry *et al.*, 1998; Flett *et al.*, 1999; Boyd *et al.*, 2000; Koman *et al.*, 2000) and crouch gait (Corry *et al.*, 1999). However, intramuscular injection of the adductor muscles may not be as effective as injection of other lower limb muscle groups. The adductors may not be stretched effectively, during standing and sitting in severely involved children, hence the rationale for combining injections of BTX-A and abduction bracing.

A fixed hip abduction brace does not permit any change in the range of hip abduction in standing and sitting. A new design of variable hip abduction orthosis, the 'Sitting Walking and Standing Hip' (SWASH) orthosis, enables greater stretch of the adductor muscles in sitting with appropriately less abduction in standing and stepping (P. Meyer, pers. comm.) (Figure 1). Following a pilot study of the new brace design, we proposed the combined use of the SWASH orthosis to lengthen the adductor muscles after chemodenervation with intramuscular BTX-A (Boyd *et al.*, 1994). Compliance with brace wear was improved by the combination of improved brace design and BTX-A induced paresis of the adductor and/or hamstring muscles. In addition, the muscle relaxant effects of intramuscular BTX-A injection on the hip adductor muscles might be potentiated by the use of abduction bracing.

Study aims

The short-term aim of this study, was to determine the effectiveness of the combined use of intramuscular injections of botulinum toxin type A and SWASH orthosis (Camp Ltd, Helsinborg, Sweden) on gross motor function in children with moderate to severe spastic type cerebral palsy, compared to current standard of care. The longer-term aim will be to determine if this combined treatment alters the natural history of hip displacement or need for surgery in this

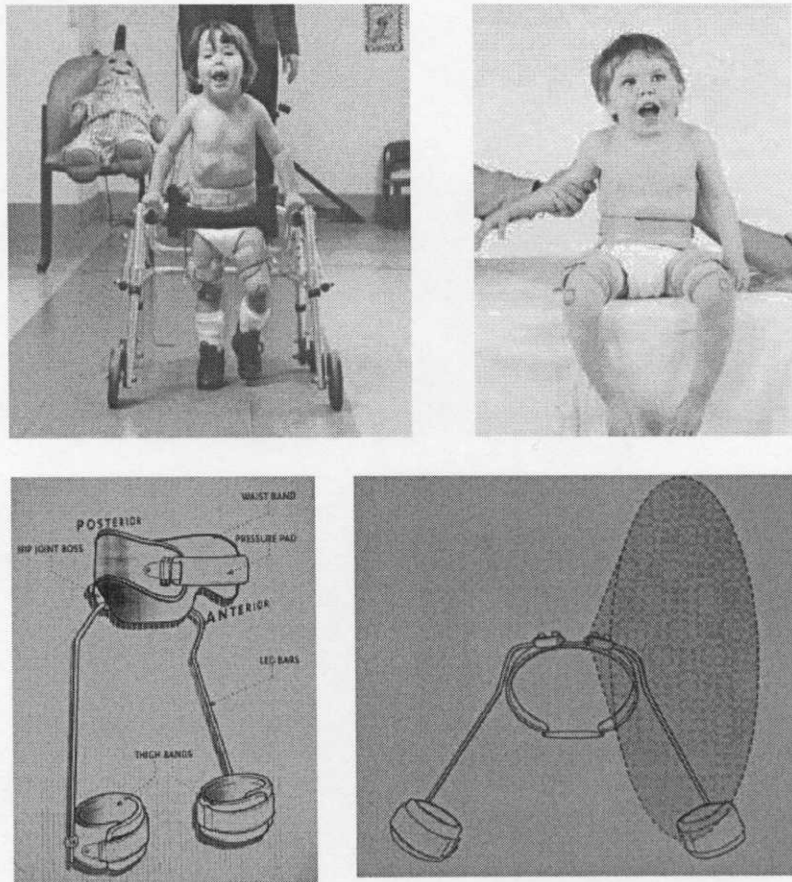

Figure 1 The variable hip abduction orthosis (design and action) (the Sitting Walking and Standing Hip orthoses, SWASH, Camp Ltd, Sweden).

patient group. This study focuses on the changes in gross motor function over 12 months. The study will continue with a larger sample size, to address the second aim.

Methods

Design

The study design was a randomized controlled trial, in which the control group received current clinical practice and the treatment group received current clinical practice with intramuscular injections of BTX-A to the adductors and medial hamstrings, combined with use of the SWASH orthosis. Current clinical practice was defined as the management already in place at recruitment with no alteration or change in medications, the frequency, type of physiotherapy management, seating, standing equipment, or orthoses. Static hip abduction bracing was not permitted. Children who ambulate continued to use their usual walking aids and ankle foot orthoses.

Setting

The study is being conducted in four tertiary referral centres as part of a multicentre trial. The study was approved by the ethics in human research committee of each institution in which the study was conducted (The Royal Children's Hospital, Melbourne; The Royal Hobart Hospital, Hobart; The Princess Margaret Hospital, Perth and the Sydney Children's Hospital, Sydney). Data in this paper are from two of the four sites: The Royal Children's Hospital, Melbourne, and The Royal Hobart Hospital, Hobart.

Subjects

Children with bilateral, spastic CP, aged 1–4 years at enrolment, with clinical and radiological evidence of hip displacement. The children were recruited from special clinics for children with CP at each centre. All children were assessed by an orthopaedic surgeon, paediatrician and research physiotherapist to determine suitability for participation in the study.

Inclusion criteria: all children admitted to the study exhibited features that might benefit from use of a variable hip abduction brace (SWASH). These features included all the following criteria:
• adductor spasticity, as evidenced by a decreased range of dynamic hip abduction;
• scissoring gait (i.e. increased hip adduction during stance or swing); and
• hip displacement, with a Migration Percentage (MP) (Reimers, 1980) between 10 and 40%.

Exclusion criteria: fixed contracture of the hip adductors, with an abduction range of less than 20° in either hip or a combined abduction range of less than 40°;
• previous hip surgery;
• initial MP greater than 40%;
• hip flexion contractures of greater than 30° (Staheli test); and
• scoliosis with a Cobb angle of greater than 20°.

All children were classified according to the Gross Motor Function Classification System (GMFCS) (Palisano *et al.*, 1997) by the research physiotherapist in each centre.

Procedures: 39 children met the inclusion criteria and their parents or carers were invited to enter them into the study. After written informed consent had been obtained from the parent or carer, the children were stratified by:
• motor type: diplegia, quadriplegia (Minear, 1956);
• age: the two age groups were 12 months to 3 years and 3 years to 5 years; and
• migration percentage (MP): 10–20%, 21–30%, 31–40% (Reimers, 1980).

This $2 \times 2 \times 3$ stratification design resulted in 12 subgroups. After central masked randomization by computer generated random number, in blocks by these subgroups, 19 children were allocated to the intervention group and 20 to the control group.

Interventions: subjects in the intervention group received intramuscular injections of BTX-A to the adductors and medial hamstrings up to 4.0 U BOTOX® (Allergan, Irvine, CA, USA) per kg per muscle and 16 U BOTOX® per kg BW as indicated. A standard dilution of BOTOX® was used, by reconstituting 100 U in 1 ml of normal saline. Equinus deformity was managed by plaster of paris casts or ankle foot orthoses in both groups. BTX-A injections to the calf were avoided because of safety considerations (Boyd and Graham, 1997; Graham *et al.*, 1999).

The adductor and hamstring muscles were injected with BTX-A, at 6-monthly intervals as indicated by:
• increased spasticity, modified Ashworth scale (MAS) greater than or equal to grade 1 +; and
• reduced dynamic range of motion, according to the modified Tardieu method (Boyd and Graham, 1999). This was indicated if hip abduction was less than 40° in either or both hips and hamstring range of motion was less than 45° as measured by popliteal θ angle.

BTX-A was injected, according to standard protocol to the hip adductors (Barwood *et al.*, 2000) and to the hamstrings (Cosgrove *et al.*, 1994) using a short-acting general anaesthetic (Cosgrove *et al.*, 1994).

BTX-A was combined with the use of a variable hip abduction orthosis (SWASH, Camp Ltd, Sweden) for 6–8 h/day, based on Tardieu (Tardieu *et al.*, 1988). The SWASH orthosis could be worn during lying, sitting, standing and walking, and enabled increased abduction with hip flexion. It was aimed to be worn in these activities for at least 6–8 h/day and, where appropriate, as a night brace. The period of actual brace wear was monitored by a diary kept by the parents and classroom assistant with additional checks by the child's community physiotherapist.

Subjects in the control group received standard treatment defined as the management already in place at recruitment with no alteration in physiotherapy management, seating, standing equipment, medication or orthoses. Children used their usual walking aids and ankle foot orthoses. The type and intensity of physiotherapy and postural management was recorded at each visit. Parents/carers of children in both groups received an educational session at commencement of the study to explain the project, the importance of hip surveillance and the relevance of the measures undertaken throughout the study to determine their child's progress. All children were followed up at regular 6-monthly intervals in the hip surveillance clinic.

Criteria for exiting the study to progress to surgery were set a priori as:
• MP greater than 40%;
• Acetabular Index (AI) greater than 27° or
• MP increasing by 10% or more in 6 months.

Children could also exit the study if progressive deformity caused loss of function, as defined as a reduction of 6% of the GMFM total score during the previous 12 months (based on McLaughlin *et al.*, 1998).

Measurements

Children were studied at baseline and at 12 months using the GMFM (Russell *et al.*, 1989). The clinical

measures of range of motion and spasticity were undertaken at 6-monthly intervals. In addition, the treatment group had subjective assessment of SWASH brace wear and effectiveness by questionnaire annually.

Gross motor function measure

The GMFM is a standardized observational instrument designed to measure changes in functional tasks (Russell *et al.*, 1989). It is not, however, a qualitative measure of functional skills such as the Gross Motor Performance Measure (Palisano *et al.*, 1997). The subject is scored on five dimensions: 'lying', 'crawling and kneeling', 'sitting', 'standing', and 'walking, running and jumping'. All research physiotherapists underwent GMFM training by the test developers and achieved good inter- and intrarater reliability (0.99–0.87).

Goal areas based on the five dimensions were selected a priori in consultation with the child's treating community-based physiotherapist. All GMFM testing was conducted with the child's parents in attendance and where possible the child's regular treating physiotherapist. All assessments were undertaken initially without aids (including the SWASH orthosis) then with the usual aids (ankle foot orthoses, gait aids but *not* the SWASH orthosis). All GMFM assessments were videotaped for later analysis by two independent examiners reliable in scoring the GMFM who were blinded to the treatment group. Following assessment the total GMFM and goal scores were collated. The raw scores were reanalysed at a later date using the GMFM–66 as it became available from the authors (Russell *et al.*, 2000).

Radiological measures

Standardized antero-posterior hip X-rays were taken every 6 months. Measures taken from these radiographs included:

- hip migration percentage (MP) according to Reimers (1980); and
- acetabular angle (AI) according to Hilgenreiner (1923).

The methodology, as well as interrater and intrarater reliability of the radiological measures has been reported elsewhere (Parrott *et al.*, 2000).

Standardized clinical examination

The clinical examination of static muscle length 'R2' and dynamic muscle length 'R1' was undertaken in the hip adductors, hip abductors, hamstring and calf muscles (Boyd and Graham, 1999). Spasticity was assessed in the muscle groups by the modified Ashworth score (MAS) (Bohannon and Smith, 1987), in the rectus femoris by Duncan Ely test (Gage, 1994), and clinical evaluation of femoral neck anteversion (Ruwe *et al.*, 1992). All these measures were undertaken at baseline and 6-monthly intervals by the research physiotherapist with an experienced assistant.

Questionnaires

Questionnaires were presented to the child's carer and treating physiotherapist at baseline and 12-month follow-up to rate SWASH brace compliance, comfort, durability, cosmesis and perceived function in the child's usual environment. The questionnaires were developed during the pilot study, with open-ended questions and subjective ranking of brace compliance and ease of donning and doffing (Boyd *et al.*, 1994). Parents and the child's local treating physiotherapist completed the questionnaire. Children in all groups were monitored at each follow up as to the intensity and type of physiotherapy intervention; period of use of postural management devices, orthoses and serial casting that the children received during the course of the study.

Statistical analysis

Demographic data were summarized using descriptive statistics. For variables at baseline, two sample Wilcoxon rank sum tests were used to test for differences between the two study groups. The differences in GMFM (total and goal) scores at 12-month follow-up compared to baseline were summarized for the two study groups using means and standard deviations. Two sample *t*-test allowing for unequal variance was used to compare the change in total scores, change in goal scores of GMFM and change in GMFM–66 at 12 months from baseline. In further analysis of changes in total GMFM scores, multiple linear regression was used to adjust for severity (GMFCS) and baseline measure of total GMFM score. An interaction term between study group and baseline GMFCS was also fitted in the model.

Spearman's rank correlation was used to examine the correlation between GMFCS classification (Palisano, 1997) and the change in total GMFM scores. Data analysis was performed in *Stata* 6 (Stata Corporation, Stata Statistical Software, Release 6.0, College Station TX, USA).

Results

There were 39 children who satisfied the entry criteria and whose parents agreed to randomization. The mean

Table 1 Demographics of patient groups at baseline and interventions received over the 12-month follow-up

Study Group	Treatment	Control	Total	*P*
Numbers enrolled	19	20	39	
Numbers completed follow-up	18*	17**	35	
Progression to surgery (% of total cohort)	2 (5%)	7 (18%)	9 (23%)	
Mean age (SD, range) In years + months	3 years + 2 months (SD 10 months)	3 years + 3 months (SD 12 months)	3 years + 2 months (Range 1 y + 7 mo–4 y + 10 mo)	
No with diplegia	9	7	16	
No with quadriplegia	9	14	23	
GMFCS level II	1	0	1 (2.5%)	
Level III	4	7	11 (28%)	
Level IV	7	6	13 (33.5%)	
Level V	7	7	14 (36%)	
Total GMFM at baseline	19.42 (11.2–61.7)	40.4 (8–74.0)	31.0 (8.0–74.0)	NS (median, range)
Total U/BOTOX®/kg/body weight (mean,SD) ($n = 19$)	13.5 U (SD = 2.01)			($n = 39$) $P = 0.16$
Mean U/kg adductors	3.4 U (mean and SD)	(0.92 U)		
Mean dose U/kg/hamstrings (mean, SD)	3.7 U (SD 0.88)			
Brace wear (mean h/day) ($n = 19$)	5.6 (1.5–9)			
Individual PT sessions (median and range, h/month)	4.0 (1–12)	3.5 (1.5–10)	4.0 (1–12)	NS diff $P = 0.8$
Group therapy (median and range, h/month)	8.0 (8–24)	6.0 (2–24)	8.0 (2–24)	NS diff $P = 0.4$
Postural management (median & range h/month)	8.0 (2–20)	4.0 (1–30)	5.0 (1–30)	NS diff $P = 0.08$

** Four of the patients did not complete the 12-month follow-up. Three of the control patients progressed to MP > 40% and underwent soft tissue surgery after 6 months. *One treatment patient did not receive further BTX-A at 6 months due to an adverse event, and moved interstate. Note: All patient demographics (mean age, motor type, GMFCS level) $n = 39$.

age at study entry was 3 years + 2 months (range 1 year + 7 months–4 years + 10 months) with 15 females and 24 males. Twenty-three children had quadriplegia and 16 had diplegia. GMFCS classification ranged from level II (mild impairment) to level V (severe impairment). Patient demographics were compared at baseline for the two groups and were similar except for GMFM scores at enrolment (Table 1). Treatments including BTX-A, SWASH orthoses and amount of physiotherapy are outlined for the two groups in Table 1. Children received a combination of postural management at home or school in the form of use of upright standing frames, modular seating systems and/or corner chairs for long sitting.

Gross motor function

Table 2 gives the mean change and standard deviation (SD) for the GMFM total scores, GMFM goal scores and GMFM–66 scores for the two study groups, respectively. It also shows the difference in the mean change with confidence interval (CI) between the two study groups for the three measurements of gross motor function. The change in GMFM total scores, GMFM goal scores and GMFM–66 scores by group are plotted with group means in Figures 2, 3 and 4, respectively.

There were no significant differences in mean change from baseline in any of the three outcome measures of motor function. There was a moderate correlation between the change in total GMFM score for both groups and the GMFCS ($r = -0.45$, $P < 0.001$).

The estimates for the multiple regression models with the interaction term for change in GMFM total scores and GMFCS are given in Table 3. The model was first fitted to all the available data. The interaction term for model 1 was highly statistically significant ($P = 0.004$) indicating that the gradients of the fitted lines for the two study groups were different. The table shows that for every decrease of one level in baseline GMFCS there was on average a 10-percentage point improvement in GMFM in the group that received the combined treatment. In the control group, there was no statistically significant effect in GMFM total for increasing severity. Residual analysis showed that these results were highly influenced by an outlier (patient 10). This patient received the combined treatment and was the only patient with GMFCS classification level II. This patient showed a very large improvement in the total score at 12 months compared to baseline. The model was refitted excluding this outlier (model 2) resulting in a

Table 2 Summary of mean differences of GMFM total, goal and GMFM–66 scores by group ($n = 35$)

Mean (SD)	Treatment $n = 18$	Control $n = 17$	Mean difference	95% CI	P
GMFM total scores	6.02 (11.8)	6.11 (7.1)	−0.09	(−6.7, 6.5)	0.98
GMFM goal scores	6.58 (11.4)	2.61 (13.8)	+4.0	(−5.0, 13),	0.37
GMFM-66 score	3.01 (4.1)	2.77 (2.8)	+0.24	(−2.2, 2.7)	0.84

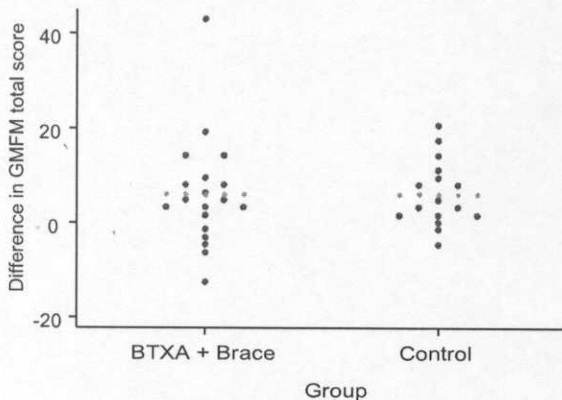

Figure 2 Differences in GMFM total scores between baseline and 12-month follow-up by group ($n = 35$).

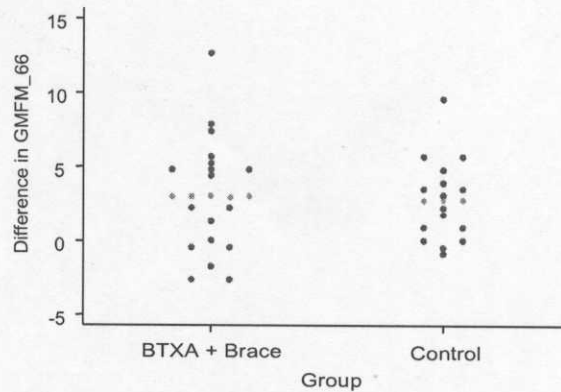

Figure 4 Change in GMFM–66 between baseline and 12-month follow-up by group ($n = 35$).

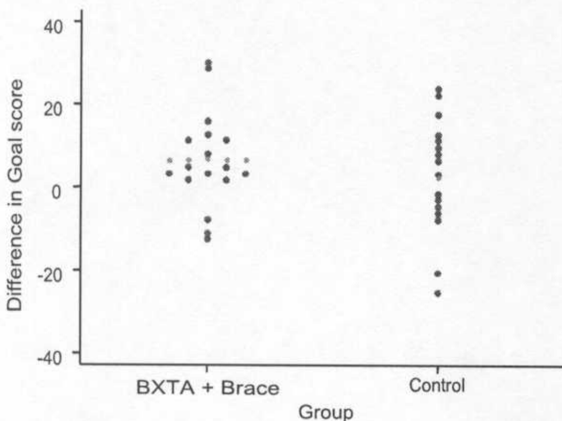

Figure 3 Differences in GMFM goal scores between baseline and 12-month follow-up by group ($n = 35$).

P-value of 0.05 for the interaction term. The coefficient estimate for the controls was unchanged, but the estimated difference in change score per baseline GMFCS category was reduced (Figure 4).

Whether the outlier was included in the analysis or not, the results suggest that if treatment is having any effect, it is reflected primarily in the greater improvement among patients with less severe impairment.

Table 3 Multiple linear regression for change in GMFM total score from baseline, by severity according to the Gross Motor Function Classification System (GMFCS) ($n = 35$)

Variable	Difference	95% CI	P
Model 1 ($n = 39$)			
GMFCS (treatment)	−9.9	(−13, −6.0)	<0.001
GMFCS (control)	−0.47	(−5.1, 4.2)	0.84
Model 2 (excludes patient 10, $n = 34$)			
GMFCS (treatment)	−6.4	(−10, −2.2)	0.004
GMFCS (control)	−0.47	(−4.6, 3.6)	0.82

Adverse events

Two children had chest infections within 2 weeks of BTX-A injections under general anaesthesia. The symptoms were mild, self-limiting and were easily managed by oral antibiotics, without hospital admission. One child received repeat BTX-A injections, without complications. The other child had a pseudobulbar palsy and after appraisal of the risks of aspiration, no further injections of BTX-A were offered.

One SWASH orthosis required a complete pelvic band re-fit as there was a break in the thermoplastic at the posterior section of the brace adjacent to the metal plate holding the thigh bars. This child was large (25 kg) and very active, spending a large proportion of his day crawling and climbing. The brace remained satisfactory with application of

greater thickness of thermoplastic for the pelvic band (3 mm).

Progression to surgery

Two children in the intervention group and seven in the control group progressed to adductor surgery (35%). The influence of the intervention on progression to surgery will require longer follow-up of a larger cohort of children. An external data management committee monitors progression to surgery on a regular basis.

Subjective aspects of SWASH

Parents reported the brace was comfortable, durable and easy to don and doff. The majority reported improved sitting and standing in the brace. Sixty-four per cent of children used the brace in a standard wheelchair. Twenty-one per cent of children in the treatment group required modifications to their seating or wheelchairs to accommodate the brace. The brace was worn for a mean 5.6 h per day (SD 2.4, range = 1.5–9 h). Length of the hip adductors was well maintained in the brace however, all children required additional stretching of the hamstring muscles as these were not adequately stretched in the SWASH orthosis.

Discussion

There was no treatment effect as measured by gross motor function measure in this randomized study of BTX-A and a variable hip abduction orthosis. Children with moderate to severe spasticity improved with and without the intervention. There was a small improvement in gross motor function with the combined use of BTX-A and bracing in the goal areas of gross motor function, but this was not significant. The annual increase in GMFM in this study is comparable to that reported after Selective Dorsal Rhizotomy (McLaughlin *et al.*, 1998). In this study, both groups had a greater improvement in total GMFM than the 4% (Wright *et al.*, 1998) and 2% (Steinbok *et al.*, 1997) reported for control groups in previous studies. In this study, both groups had a highly significant and comparable increase in gross motor function, presumably as the result of the delayed acquisition of motor skills, characteristic of children with more severe CP (Bleck, 1987). Russell reported increases in GMFM of 6% in children with diplegia and 4% in children with quadriplegia, over 6 months, in children not subjected to invasive intervention (Russell *et al.*, 1993). Presumably, this is the natural history of motor development in cerebral palsy. The children in this study in GMFCS groups IV and V had a 6% increase in GMFM, compared to the 4% increase in the cohort with quadriplegia reported by Russell. It may be that our control group who received clinical best practice benefited from being in the trial. Children in the control group received a similar mean number of physiotherapy sessions per month; however, there may be a change in focus of postural management and therapy occurring, knowing that these children have 'hips at risk'. In addition, education on hip surveillance and attendance at measurement sessions on Gross Motor Function by both groups may have covertly influenced practice of tasks undertaken during GMFM testing.

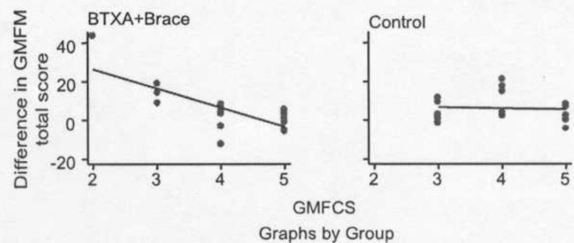

Figure 5 Severity as classified by GMFCS (Palisano *et al.*, 1997) and change in total GMFM scores between baseline and 12-month follow-up ($n = 35$).

However, children at a higher functional level, GMFCS levels II and III achieved a greater improvement in gross motor function in the brace and BTX-A treated group compared to the control group (Figure 5). For the treatment group, it seems that the milder the impairment, the greater the improvement in the total score compared to baseline. In the intervention group children in GMFCS level V showed little or no improvement, compared to the less impaired children in GMFCS levels II and III. In the control group there was no association between the rate of improvement and GMFCS level (Figure 5).

Apart from no genuine treatment, there may be other reasons for the fact that there was no difference on GMFM scores between the groups over time. Despite concealed randomization, the groups had different median GMFM scores at entry to the trial. The difference was not statistically significant, because of the large range of GMFM scores in both groups, but may have been clinically significant. The GMFM measure may not be sensitive for detecting functional change in children with severe motor disability (GMFCS level IV and V), as suggested by Nordmark in their study of children with moderate to severe spasticity following SDR (Nordmark *et al.*, 2000). It may be that our children with a younger mean age range of 3 years + 2 months may not have cooperated

well enough to perform the tasks to the testing criteria. Nordmark and Russell's studies have suggested 5 years and over as the minimum age to assess by the GMFM.

The GMFM is not intended to measure quality of performance. Accordingly, it may be that while there was no change in attainment of new skills during the study there may have been improvements in the quality of those skills. Previous authors have reported the relative insensitivity of the GMFM to detect change in the more demanding dimensions (McLaughlin *et al.*, 1998). It was hoped that use of the Rasch analysis may reduce this possibility in the less severely affected children, but in fact this may have the reverse effect in the more severely affected children (Russell *et al.*, 2000). We did not find any significant difference in our overall results using the GMFM-66 analysis in this group of children with moderate to severe spasticity.

Russell suggested that when testing for GMFM, the child's usual aids and orthoses should be used. However, younger children often require a change in orthoses and assistive devices within a 12-month period. Ideally, these should be consistent at testing and re-testing (Russell *et al.*, 1993). In this study the assessments were performed without ankle foot orthoses and then with the current orthoses and gait aids. The intervention group was not assessed in the SWASH brace. We adopted this assessment protocol because we wished to measure the functional change exhibited by the child, consequent on brace use over time. This may yield a better comparison of motor learning and functional change between the groups. It could be argued that children in the intervention group should have been assessed in the SWASH brace in addition to their usual ankle foot orthoses and gait aids. During testing, the SWASH brace may have enhanced function in certain dimensions (sitting, walking) and hindered it in others (particularly lying and rolling). It was not considered feasible, both with regard to time and compliance, to assess the subjects in all three conditions. This testing protocol would have been too demanding for young children with moderate to severe disability.

Our cohort of younger children with moderate to severe disability may have had less ability to comprehend instructions, comply and complete the lengthy testing procedure during the GMFM. Some children with mild impairment had poor motivation to complete tasks where they could easily achieve them (e.g. rolling and lying in a child who can already stand and walk). The GMFM testing procedure is dependent upon the ability of the child to follow and comprehend instructions and the expertise of the physiotherapist in motivating the child to complete the task (Nordmark *et al.*, 2000). It may be more meaningful to assess children purely in their relevant goal areas. However, goal dimensions may change over time, so analysis between groups may be difficult; change in single subjects over time may be more appropriate.

The SWASH orthosis is well accepted, durable, and sometimes used as an alternative to other postural management systems. It appears to work effectively in combination with chemodenervation with BTX-A to maintain length of the adductors, evidenced by fewer subjects progressing to soft tissue release in this 12-month follow-up. However, parents and physiotherapists require education and help in techniques to maintain hamstring muscle length. These muscles are not stretched appropriately with this combined intervention. A greater proportion of the control group have required soft tissue surgery to the adductor muscles during the first year of this 3-year study. A sample size of 90 subjects, including this initial cohort of 39 subjects, has been recruited and will be followed up for 3 years to assess longer-term radiological outcome. Current trends support the hypothesis that the combined treatment (BTX-A and SWASH) may delay surgery. However, it is not yet known if the intervention delays, reduces or avoids the need for surgery.

Information from this study, both from the control group and from the intervention group, may help define the natural history of hip migration in CP, the effects of nonoperative management, and lead to the development of management algorithms. At the conclusion of the study, it is hoped that the development of management algorithms will indicate the children with spastic hip displacement who may benefit from the combined treatment of BTX-A and bracing, the children who may progress rapidly to surgery regardless of intervention and whether some children maintain hip stability with current best practice.

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