EUROPEAN JOURNAL OF PHYSIOTHERAPY
https://doi.org/10.1080/21679169.2020.1807602

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ORIGINAL ARTICLE

The effects of using an electrodress (Mollii®) to reduce spasticity and enhance functioning in children with cerebral palsy: a pilot study

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ABSTRACT

Purpose: Spasticity negatively affects the muscles and joints of the extremities and can be harmful to growing children. Children born with cerebral palsy do not have extremity deformities at birth but develop them over time. The goal is to reduce spasticity to avoid deformities. In this pilot study, we tested a non-pharmaceutical non-invasive method using an electrodress for six months.

Material and methods: We included 16 children with cerebral palsy and Gross Motor Function Classification System (GMFCS I-V) age 2–16 y, median age 6.3 y. The intervention lasted 60 min every other day with reciprocal inhibition of the spastic muscle.

Results: Passive range of motion (pROM) improved during treatment with a significant number of improved muscles after one ($p = 0.000$), three ($p = 0.001$) and six ($p = 0.014$) months. The spasticity level measured using the modified Ashworth scale (MAS) significantly decreased at one ($p = 0.007$) and six months ($p = 0.011$) and was almost significant after three months ($p = 0.076$). The modified Tardieu significantly decreased after one month ($p = 0.030$), but not after three ($p = 0.392$) or six months ($p = 0.426$).

Conclusion: The electrodress has effects on spasticity levels and pROM. Further studies are needed to optimise the frequency and intensity of the current with respect to the effects on the level of spasticity.

ARTICLE HISTORY
Received 23 March 2020
Accepted 1 August 2020
Published online 24 August 2020

KEYWORDS
Cerebral Palsy; Children; Electric muscle stimulation; Modified Ashworth; Modified Tardieu; Passive Range of Motion; Spasticity

Introduction

Cerebral palsy (CP) is a complex condition with numerous symptoms and various aetiology. It is characterised by inactive nonprogressive brain damage, where the injury occurs prenatally, perinatally or during the early postnatal period. This may result in many complicated functional impairments, both sensoric and motoric. CP can be characterised by the way it affects a person's movement, the part of the body affected, and the severity of the condition [1,2].

Brain injury leads to impairments of the central nervous system that subsequently affect the muscles when attempting to generate force or movement. The interrelations between the motor cortex in the brain and the lower motor neurons in the spinal cord are affected. This often leads to spasticity in the muscles as well as other phenomena. This is referred to as '"the upper motor neuron syndrome (UMNS)"' [3], defined as a constellation of signs and symptoms including 1) involuntary muscle activity, spasticity, spastic co-contraction, associated movements, and spastic dystonia, referred to as '"positive components"', and 2) impaired voluntary control of movements, such as reduced muscle strength, reduced coordination of movements, and reduced dexterity, referred to as '"negative components"'. Other signs include exaggerated cutaneous withdrawal (flexion and pain) reflexes and the Babinski sign.

One component of UMNS is spasticity characterised by a velocity-dependent increase in the tonic stretch reflex (muscle tone) with exaggerated tendon jerks, resulting from hyperexcitability of the stretch reflexes [4,5]. Other '"positive"' components of UMNS are spastic dystonia referring to abnormal positions, e.g. of the hand or foot. It is caused by chronic involuntary activation of a spastic muscle. These components of UMNS can lead to pain and contracture and cause significant disability [6,7]. Strong spastic muscles cannot be balanced by weak antagonists. This means that there is a risk that the spastic muscle will become shorter and the antagonist will become longer. This muscle imbalance leads to arthrogenic contractures and secondary structural deformities in the growing skeleton. Spasticity causes limitations in movement, involuntary movements, involuntary contraction of the muscles, limited function, and often pain [8].

The aim of treatment of spasticity is reduction of spasticity to minimise the effects and the problems they cause. Current treatment for children with CP include orally or intrathecally administered baclofen [9], injection of botulinum toxin in combination with physiotherapy and sometimes splinting [10], surgery such orthopaedic procedures on muscles, tendons and skeleton, or selective dorsal rhizotomy

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with partial neurectomy designed to allow the spasticity-causing nerves to be destroyed [11]. All current treatments have unwanted adverse effects. Surgical interventions are irreversible. Some treatments are short term in effect, (e.g. botulinum toxin injections). The effect of botulinum toxins lasts 3–4 months and not all necessary muscles can be treated simultaneously. The treatment is painful and requires some kind of sedation; there is a slight risk of immunisation over time. Baclofen may cause unwanted weakness and fatigue.

Among alternative treatments to surgical and pharmacologic interventions is electric stimulation of spastic muscles or their antagonists. Several studies have shown that this reduces spasticity, increases function and increases range of motion [12–15]. Carmick, an American physiotherapist conducted a study in children with CP in which she stimulated spastic muscles or/and their antagonists in both upper and lower extremities. She reported several functional improvements including better stature, balance, fine motor ability and improved ability to perform simple functional tests. A recent systematic review concluded that transcutaneous, electric nerve stimulation might have beneficial effects on spasticity and activity performance after stroke [16].

Mollii is a device developed by Inerventions, a small Swedish med-tech company; it represents an innovative approach for non-invasive electro-stimulation to reduce spasticity and improve motor function. The theoretical background of this treatment method refers to the concept of reciprocal inhibition, in which sensory input from a muscle may inhibit the activation of an antagonistic muscle. By stimulating the antagonist to the spastic muscle, there is reduction of reflex-mediated over-activity in the spastic muscle and strengthening of the weak antagonist to obtain better balance of the muscles. Earlier treatments with electro-stimulation have used separate and patch-based electrodes. The Mollii method uses electrodes embedded in a dress to secure the placement of the electrodes.

This study aims to evaluate the effect of electrostimulation using an electrodress to reduce spasticity and enhance mobility in children with CP.

Material and methods

Equipment

The electrodress (Mollii) is a functional garment that consists of a pair of trousers, a jacket and a detachable control unit that sends electrical signals to the user *via* electrodes on the inside of the garment. A computer control unit activates the chosen electrodes and the intensity of stimulation at each pair. The settings are saved in the control unit, making it simple for the device to be used at home or in school. There are 58 electrodes in the garment. The numbers and locations that are used depend on the child's functional impairment and those muscles that need to be stimulated. Stimulation parameters are pulse width 25–175 μs, frequency 20 Hz, voltage 20 V and pulse shape square wave (Figure 1).

Figure 1. The electrodress (Molii-suit).

Participants and selection criteria

Participants were recruited among children with CP classified according to Gross Motor Function Classification System (GMFCS) 1–5 [17]. All participants were considered to have an immediate need for botulinum toxin injections by the neuropediatric clinic. Through their parents, the children were offered alternative treatment with the electrodress. If they wanted to participate, written informed consent was obtained. Participation was voluntary with the possibility to withdraw from the treatment at any time. The exclusion criteria were as follows: treatment with botulinum or surgery at least three months prior to initiation of the study. They should not have pacemaker, implantable cardioverter defibrillator (ICD), baclofen pump or similar devices. Percutaneous endoscopic gastrotomy (PEG) was an exception.

The number of muscles to be treated was chosen by the child's physiotherapist according to the child's functional impairment. The dress was used for at least 60 min 3–4 times per week, i.e. every other day. The child continued with their usual physiotherapy as prescribed by their physiotherapist who also performed all the measurements in cooperation with the principal physiotherapist in the study, who was one of the co-authors. All measurements were performed within 24 h after using the electrodress.

Sixteen children were included in the study from May 27th, 2016 until February 2^{nd,} 2017. There were eight boys and eight girls, from 2– 16 to sixteen years of age with mean age 6.3 years. Three were classified as GMFCS I, two as GMFCS II, six as GMFCS III, four as GMFCS IV and one as GMFCS V. Twelve had diplegia, one hemiplegia, and three dystonia. Patient demographics are displayed in Table 1.

Eight patients continued the treatment for six months. Three of those continued to one year. Eight patients dropped out at various times and for various reasons (Table 2).

Registration and availability of data

The study was registered and approved by the Regional Review Authority in Uppsala with the registration number Dnr. 2016/119. The platform is now hosted by the Swedish Ethical Review Authority since January 2019

Table 1. Patients demographics.

Patient	Age	Gender	Diagnosis	GMFCS	Treatment time
1	16	girl	Diplegia	IV	3 months
2	4	boy	Diplegia	II	6 months
3	5	boy	Diplegia	III	3 months
4	6	girl	Dystonia	IV	12 month
5	7	boy	Diplegia	II	6 months
6	8	girl	Diplegia	I	12 months
7	4	girl	Diplegia	III	6 months
8	7	boy	Diplegia	I	2 months
9	10	boy	Dystonia	V	12 months
10	2	boy	Diplegia	III	6 months
11	6	girl	Hemiplegia	I	1 month
12	6	boy	Diplegia	III	4 months
13	4	girl	Diplegia	III	6 months
14	4	girl	Dystonia	IV	4 months
15	5	girl	Diplegia	IV	5 months
16	6	boy	Diplegia	III	3 months

Table 2 Reasons for drop-out.

At two weeks	One girl, 6 years, hemiplegia, GMFCS I with a visible improvement in walking when testing the dress. Used it for two weeks, but thought it was too inconvenient and dropped out
At two months	One boy, 7 years, diplegia, GMFCS I. No improvement. Stiff
	Do not get better with increased stimulation. Drop out after 2 months
At three months	One boy, 5 years, diplegia, GMFCS III. Improvement in ROM and spasticity
	His mother had severe dyslexia, and could not fill in the logbook and was not able to manage the dress
	One girl, 16 years, diplegia, GMFCS IV, became more flexible. Had effect on ROM, spasticity, and function Adipositas. Found the dress too inconvenient and dropped out
	One boy, 6 years, diplegia, GMFCS III. Problems with epilepsy and dropped out after three months
At four months	One boy, 6 years old, diplegia, GMFCS II, good function, effect on ROM and spasticity. No improvement after three month. Do not want to use the dress and drops out
	One girl, 4 years, dyskinesia, GMFCS IV, Improvement first, but after 4 month Increasing spasticity in adductors. Drops out
At five months	One girl, 4 years, diplegia, GMFCS IV, Improvement in function, speech, and bowel function. After three month problem with the size
	It takes some time to get a new size and when she gets it, she refuses to go on

The data that support the findings of this study are available from The Centre for Clinical research Region Dalarna, Sweden upon reasonable request provided that the data can be made available in accordance with applicable data protection and privacy regulations.

Measurements

The following measurements were performed for all children belonging to one or the other of the five groups in GMFCS [17]: baseline, one month, three months, six months, and (for some of the participants) after one year.

Joint motion: Hip: extension, abduction, and rotation outwards; knee: extension and flexion; foot: dorsal extension.

Estimation of tonus according to the modified Ashworth scale [18]: adductors, hip flexors, knee flexors, and plantar flexion; knee extension if needed.

A dynamic component according to the modified Tardieu scale [19]: adductors, hip flexors, knee flexors, and plantar flexion; knee extension if needed.

Caregiver/- patient rating

During the treatment with the electrodress, the caregiver/patient registered pain, sleep, and bowel function according to Table 3 for one week prior to the start of the treatment (baseline) and for one week prior to the 1-month, 3-months, 6- months and 1- year follow-up. The caregivers noted in a logbook when and for how long the patient used the dress, which activities they engaged in during the treatment and just afterwards. If possible, they provided a subjective description of the general condition including coldness/discolouration of hands and feet.

Further measurements

Gross Motor Function Measure (GMFM) [22] is a validated assessment tool designed to measure changes in gross

Table 3. Measurements for pain, sleep and bowel function.

Pain	Number of episodes/24 h	context	Intensity/need for medication	FLACC scale if needed (Figure 2)
Sleep	Hours/24 hours		Wakeup episodes	Need for medication
Bowel function	Number/week		Form according to the Bristol scale [20]	Need for laxative

Criteria	Score 0	Score 1	Score 2
Face	No particular expression or smile	Occasional grimace or frown, withdrawn, uninterested	Frequent to constant quivering chin, clenched jaw
Legs	Normal position or relaxed	Uneasy, restless, tense	Kicking, or legs drawn up
Activity	Lying quietly, normal position, moves easily	Squirming, shifting, back and forth, tense	Arched, rigid or jerking
Cry	No cry (awake or asleep)	Moans or whimpers; occasional complaint	Crying steadily, screams or sobs, frequent complaints
Consolability	Content, relaxed	Reassured by occasional touching, hugging or being talked to, distractible	Difficult to console or comfort

Figure 2. FLACC (Face, Legs, Activity, Cry, Consolability) for children < 18 years and for children with multi disabilities [21].

motor function over time or with interventions in children with CP. It was used at baseline and at 6-month follow up. The item sets 66 were used for ambulatory children with level GMFCS levels 1–3. Physical cost index (PCI) [23] was measured at the simultaneously. Caregivers/patients were asked to provide a subjective description of any changes in ability or perhaps new abilities.

The item sets 66 for non-ambulatory children with GMFCS 4–5 were used to assess nursing/practical handling (changing of diapers, dressing, showers, etc.). Caregivers/patients were also asked to describe any changes in ability or perhaps development of new abilities.

The children often had differences between the extremities both in terms of pROM and spasticity. We chose to count the several joints as independent in the measurements. As a result, there were two results for each movement or spasticity, one for the right and one for the left. We considered unchanged or improved as a good result, because we expected spasticity to worsen progressively during the study period. Either the joints were within normal range from the beginning and remained normal or the spasticity would not worsen as expected without treatment with botulinum toxin.

Caregivers were asked for their expectations of the treatment at baseline and were asked during the treatment if their expectations had been fulfilled.

At each follow up the technical equipment was tested to make sure that it was still functioning correctly.

Statistical analysis

Changes in study parameters were expressed as relative percentage changes. Q-Q plots of the collected data were used for analysis and confirmation of normal distribution. Data regarding spasticity and passive range of motion were analysed using the paired t-test. We considered p-values of $\leq$ 0.05 as statistically significant, while *p*-values of $>$ 0.05 to 0.10 were interpreted as non-statistically significant trends. Analyses were performed using IBM SPSS Statistics, Version 22.

Ethical considerations

Electrical stimulation is well known and is used daily in neurological rehabilitation for pain reduction. Side effects and complications are mild and consist mainly of blisters and skin rashes. Using systematic electric stimulation in the electrodress would delay treatment with botulinum toxin injections for 6 months. However, it might be a spasticity-reducing alternative to Botulinum toxin treatment with the advantage of stimulating all warranted muscles at once, unlike the effects of botulinum toxin. The electrodress treatment is non-invasive and can be performed by the child's caregiver or assistant in the normal environment at home or school. There is no need for sedation or treatment in hospital.

The study was approved by the Regional Ethical Review Authority in Uppsala. Diary number 2016/119

Results

Passive range of motion (pROM) improved or remained unchanged for all muscle groups measured after 1 month of treatment in the range of 73.4% – 96.4%. The effect was most pronounced for external rotation of the hip, and knee extension and was somewhat lower for abduction and knee flexors. There was a significantly higher number of muscles with increased or unchanged measurements in the passive range of motion after 1 month ($p = 0.000$). The results decreased slightly after 3 and 6 months, but remained improved for external rotation and abduction of the hip and for extension of the knee. There was a significantly higher number of muscles with increased or unchanged measurements after 3 months ($p = 0.001$) and after six months ($p = 0.014$). The three patients that continued for 1 year maintained their improvements ($p = 0.030$) (Table 4).

Spasticity levels according to the Modified Ashworth scale improved or remained unchanged after 1 month of treatment by 76.8% – 96.7%. There was a significantly higher number of muscles with decreased or unchanged spasticity ($p = 0.007$). The effect was most pronounced in hip adductors and knee flexors and it was somewhat less pronounced for plantar flexors. There was an almost significant number of muscles with decreased or unchanged spasticity after 3 months ($p = 0.076$) with the most pronounced effect for plantar flexors. After 6 months, the number of muscles with decreased or unchanged spasticity was again highly significant ($p = 0.011$). The three patients that continued maintained their improvement for 1 year (Table 5).

Improvements on the Modified Tardieu scale evaluated by the angle of the first catch were categorised as improved or unchanged after 1 month in the range 64.3%– 81.8%. There was a significantly higher number of muscles with improved or unchanged measurements after 1 month ($p = 0.030$). The effect was most pronounced for hip flexors and hip adductors and it was less pronounced for plantar flexors. Even here, there was a slight decrease in the results after 3 and 6 months. The results after 3 and 6 months were not significant ($p = 0.392$ and 0.426, respectively). Only one of the patients that continued for one year had a first catch when evaluated with the Modified Tardieu scale; however, she still had a good result (Table 6).

Caregivers were asked to express their expectations before entering the study and to state whether those expectations had been fulfilled. Some, but not all of the expectations, had been fulfilled concerning motoric function in 13 of 16 patients. Improvement in life quality was seen in four patients (Table 7).

The electrodress did not improve bowel function. Only one child obtained better function as he started to use the toilet instead of diapers. Otherwise, there was no change in toilet visit frequency except for two patients who reported slight increases in frequency. There was also no change according to the Bristol scale (form and consistency). The use of laxative was unchanged during the study. Six of the patients used laxative regularly (Table 8).

Sleep pattern appeared to be unchanged. The frequency of wake-up episodes during the night was recorded. For

Table 4. pROM. Changes at group level.

Time	1 M	%	3 M	%	6 M	%	1 Y
Hip extension							
Number of joints	26/32		20/26		12/16		6/6
Improved or unchanged	22	84.6	13	65	8	66.7	5
Impaired movement	4	15.4	7	35	4	33,3	1
Hip Abduction							
Number of joints	30/32		26/26		16/16		6/6
Improved or unchanged	22	73.4	18	69,2	10	62.5	2
Impaired movement	8	26.7	8	30.8	6	37.5	4
Hip rotation out							
Number of joints	22/32		20/26		12/16		4/6
Improved or unchanged	20	90.9	18	90	11	91.7	4
Impaired movement	2	9.1	2	10	1	8.3	0
Knee extension							
Number of joints	28/32		22/26		14/16		6/6
Improved or unchanged	27	96.4	21	95.5	13	92.9	5
Impaired movement	1	3.6	1	4.5	1	7.1	1
Knee flexion							
Number of joints	30/32		24/26		16/16		6/6
Improved or unchanged	23	76.7	15	62.5	15	93.8	4
Impaired movement	7	23.3	9	37.5	1	6.3	2
Dorsal extension ankle							
Number of joints	30/32		25/26		16/16		6/6
Improved or unchanged	24	80.0	19	76.0	9	56.3	4
Impaired movement	6	20.0	6	24.0	7	43.8	2
Summary							
Number of joints	166/192		137/156		86/96		34/36
Improved or/unchanged	138	83.1	104	75.9	66	76.7	
Impaired movement	2						

Table 5. Modified ashworth. Changes at group Level.

Time	1 M		3 M		6 M		1 Y
Number of joints	132/160	%	116/130	%	68/80	%	30/30
Hip Flexors							
Number of joints	28/32		24/26		12/16		6/6
Decreased or unchanged Spasticity	24	85.7	17	70.8	12	100	5
Increased Spasticity	4	14.3	7	29.2	0	0	1
Hip adductors							
Number of joints	30/32		26/26		16/16		6/6
Decreased or unchanged Spasticity	29	96.7	22	84.6	13	81.2	4
Increased Spasticity	1	3.3	4	15.4	3	18.8	2
Knee flexors							
Number of joints	30/32		26/26		16/16		6/6
Decreased or unchanged Spasticity	28	93.3	22	84.6	16	100	5
Increased Spasticity	2	6.7	4	15.4	0	0	1
Plantar flexors							
Number of joints	28/32		24/36		16/16		6/6
Decreased or unchanged Spasticity	21	75	15	62.5	12	75.0	4
Increased Spasticity	7	25	9	37.5	4	25.0	2
Knee ext							
Number of joints	16/32		16/26		8/16		6/6
Decreased or unchanged Spasticity	15	93.8	14	87.5	8	100	5
Increased Spasticity	1	6.2	2	12.5	0	0	1
Summary		%		%		%	
Decreased or unchanged	119	90.2	90	77.6	61	89.7	24
Increased spasticity	13	9.8	26	22.4	7	10.3	6

eleven of the patients, the pattern was unchanged compared to the baseline. Four children slept better with fewer or no wake-ups. One child reported increased wake-up frequency (Table 8).

Episodes with pain were recorded. Nine of the children had no pain at baseline and no episodes of pain during the study. Four children had reduced frequency of pain, two had unchanged frequency and one child had increased frequency of pain (Table 8).

Two children (patients 5 and 6) managed to perform the test for PCI (physical cost index) at baseline and after 6 months. Both increased their walking speed (m/min), their step length (in cm), and step frequency (step/min), and they lowered their PCI.

The nine children that finished the treatment for six months were evaluated using the GMFM-66 item set (Gross Motor Function Measure) at baseline and after six months. The test was performed by the child's physiotherapist. It measures the child's gross motor function and over time their development. Six children improved their scores. Five were ambulatory and one was non-ambulatory. Three had no improvement in their scores. One was ambulatory and

Table 6. Modified Tardieu. Changes at group level.

Time	1 M	%	3 M	%	6 M	%	1 Y
Hip flexors							
Number of joints	12/32		13/26		6/16		2/6
Improved or unchanged	9	75	9	69.2	2	33.3	2
Increased Spasticity	3	25	4	30.8	4	66.7	0
Hip adductors							
Number of joints	22/32		19/26		14/16		2/6
Improved or unchanged	18	81.8	13	68.4	9	64.3	1
Increased Spasticity	4	18.2	6	31.6	5	35.7	1
Knee flexors							
Number of joints	27/32		24/26		14/16		4/6
Improved or unchanged	18	66.7	10	41.7	10	71.4	2
Increased Spasticity	9	33.3	14	58.3	4	28.6	2
Plantar flexors							
Number of joints	28/32		24/26		16/16		6/6
Improved or unchanged	18	64.3	15	62.5	8	50.0	4
Increased Spasticity	10	35.7	9	37.5	8	50.0	2
Knee extensors							
Number of joints	4/32		2/26		0/16		0/6
Improved or unchanged	2	50.0	2	100	0	0	0
Increased Spasticity	2	50.0	0	0	0	0	0
Summary		%		%		%	
Number of joints	93/160		82/130		50/80		14/30
Improved or unchanged	65	69.9	49	59.8	29	58.0	9
Increased spasticity	28	30.1	33	40.2	21	42.0	5

Table 7. Expectations and fulfilment.

Patient	GMFCS	Expectations	Fulfilment
1	4	Improved motion and easier getting dressed	Could wheel longer in her chair. Able to swim longer
2	2	Improved walking. Be able to sit on the floor.	Easier to stretch. Improved walking. More flexible in moving, Pre-school teacher saw a big difference after 4 weeks leave, Can walk for a longer time without support
3	3	Be suppler in the body. Be able to extend the legs Get the heels down on the floor. Better balance.	Mother had severe dyslexia and was not able to fill in the logbook or put on the dress
4	4	Be more relaxed. Better motion. Better toilet habits	Very pleased. More sweeping gestures with the arms. Better control of the neck and better balance of the body.
5	2	Reduce pain. Improve or preserve motion. An alternative to Botulinum toxin	More flexible. More activity
6	1	More relaxed in the legs especially the right leg. Better balance	Better balance in the right leg. Improved walking pattern. No need of Botulinum toxin
7	3	Improved function. No need of Botulinum toxin	Improved function and improved speech
8	1	Suppler muscles. No need of Botulinum toxin	It is pleasant but with no effect on spasticity or motion. Prefer Botulinum toxin.
9	5	Improved sleep. Improved bowel function, Less tension	Improved sleep, better bowel function, less pain, Think it is pleasant, have better ability to answer questions, improved eating and drinking, easier to get dressed and undressed
10	3	Reduced tension in the thighs, To walk with walking aid without crossing the legs Move the legs side ways	More upright in the trunk. Stronger in holding the legs, No effect on the crossing of the legs. The best effect in the beginning. Better bowel function. Started to use the toilet. Had diapers before
11	1	To walk with less tension. Be able to run better	Visible effect with better walking at the test of the dress. Treated for two weeks. Refuses to use it after that
12	2	Suppler, softer in the calves, less stiff, fever spasms	Runs easier, suppler
13	3	Lower the tension in foot, calf and thigh. Improve circulation in feet, and legs	Still cold feet. Better position even when relaxed. Can walk on the whole foot on the right side and almost on the left side
14	4	Improve walking. Use of walking aids Better bowel function. Improve sleep. Warm feet	Improved bowel function, better sleep, warm hands and feet
15	4	An alternative to Botulinumtoxin.	Improved motion in all joints except hip abduction, Much improved. Less spasticity. Uses the hand.
16	3	Less toe walking. Less pain in the legs. Easier to use the hands (to write, draw, play with Lego, etc.)	Difficulties with using the dress because of epilepsy Slept when using the dress. Besides that no special effect.

two were non-ambulatory. Three children were tested at one year. The one who was ambulatory improved further while the two non-ambulatory children were at the same level as they were at six months.

Patients recruited into the study had previously been treated with botulinum toxin and were considered to be in immediate need of new injections. They were offered treatment with the electrodress as an alternative. None of the participants needed or received botulinum toxin treatment during the test period. However, most needed treatment with botulinum toxin after they finished the study.

There were no reported adverse effects such as skin rash or pain. The children could sometimes feel the stimulation as a tickle, but this was not experienced as uncomfortable.

Table 8. Results for pain, sleep and bowel function.

Pain/times/week or day	Base, 16 children	One Month 14 children	Three months 13 children	Six months 9 children	One year 3 children
No pain	9	10	8	7	2
1–2/day	5	3	2	1	
2–3/day	1		1		
0–1/week			1	1	1
2–3/week	1	1	1		
Sleep. Wake-upp episodes/week	**Base 16 children**	**One month 14 children**	**Three months 13 children**	**Six Months 8 children**	**One year 1 child**
None	6	7	5	2	
0–1	7	2	3	3	1
0–2	1	2	3	1	
2–5	1	2	1	1	
4–10	1	1	1	1	
Bowel function/ times/day	**Base 16 children**	**One month 13 children**	**Three months 13 children**	**Six Months 7 children**	**One year 1 child**
0–1	9	6	8	5	1
0–2	1	2	1		
0–3			2	1	
1–2	5	3		1	
1–3	1	2	2		

Discussion

In this uncontrolled pilot study, we measured the effects of an electrodress with embedded electrodes on spasticity in children with CP using the principle of reciprocal inhibition. We found a significant improvement in pROM and reduced spasticity measured using the modified Ashworth scale (MAS) and the modified Tardieu scale measuring the dynamic component with the angel of the first catch. We found some effect on sleeping patterns and pain but no effect on bowel function. Gross Motor Function (GMFM-66) improved for some of the children mostly for those who were ambulatory. The study of the intervention with the electrodress shows a new way to handle spasticity in children with CP.

Electrical stimulation can be used in various ways using several methods. It can be applied directly to a paretic muscle to improve function or to the antagonist muscles to reduce spasticity of the corresponding agonist muscle, so-called reciprocal inhibition. The mechanism involves enhancement of spinal inhibitory signalling by di-synaptic reciprocal inhibition of Ia afferent fibres and presynaptic Ia inhibition of alpha motor neurons. Electrical stimulation of afferent fibres (sensory) in an extensor muscle activates inhibitory Ia interneurons and reduces excitability of the flexor muscle motor neuron [24]. Reciprocal inhibition plays a fundamental role in the normal performance of movements. This mechanism is used to reduce spasticity, unwanted muscle over-activity and to improve voluntary muscle activation and movement control.

The small portable units used in our study are battery-operated with four small batteries of all together 20 V, which is considered low intensity. The placement and size of the electrodes are essential to secure contact with the skin and to ensure that the correct muscles are stimulated. Earlier studies have used loose patch-on electrodes and use of a gel to contact the skin. This can be ineffective and give unwanted results [25]. In the electrodress, 58 electrodes are embedded in the dress. The dress is tight enough to provide good contact with the skin and the electrodes are small enough so only the proper muscles are stimulated. The device is programmed and tested to stimulate a specific number of muscles. The muscles chosen are the same as would have been chosen for botulinum toxin injections.

The electrodress uses transcutaneous electrical nerve stimulation (TENS) provided through some of the 58 electrodes embedded in the dress. The number of activated electrodes are chosen according to the child's impairment. Electrical stimulation using surface electrodes is a non-invasive therapeutic method. It has been used for many years in patients with upper motor neuron lesions to improve voluntary motor control by increasing muscle strength, reducing spasticity and pain and increasing passive range of motion [24,26]. A systematic review by Mills *et al.* included only randomised controlled trials [16]. None of the studies that met the inclusion criteria were conducted on participants younger than 18 years. In our study TENS, is used through the electrodress; however, because the dress was used in daily activities at home or in school, in serves as a kind of functional electrical stimulation.

We chose to use a low-frequency stimulation of 20 Hz. This is sufficient to provoke both contraction of the antagonist and sensory input to mediate reciprocal inhibition of the spastic agonist. The low frequency might minimise the risk for discomfort or other adverse effects that could jeopardise compliance [26] while at the same time avoiding fatigue of the muscle. The pulse duration in the electrodress is chosen to be 25–175 μs together with the low frequency of 20 Hz. This is sufficient to recruit enough muscle fibres to elicit sensory impulses and perhaps contractions. It is not wide enough to penetrate more deeply and affect secondary tissue layers [27].

The duration of treatment has been discussed. In a review, Thrasher *et al.* found that increasing treatment was not related to more successful outcomes [28]. Positive benefits were seen with short programmes (2.5 h/week) and limited benefits were seen with more extended programmes (21 h/week). In our study, the electrodress was used for 60 min every other day 3–4 times/week. We believed this duration would be sufficient, because the effect of the dress

is expected to last up to 48 h. Children with CP are subjected to demanding programmes with treatments of several kinds; therefor it is crucial to make the interventions as simple as possible. The children were encouraged to carry on with their usual activities while wearing the dress. It was often noticed that the dress had an effect during the treatment. We chose to do the measurements not directly after the treatment but no longer than 24 h after the treatment. The reason for this was that we wanted to see if the treatment effect lasted beyond the effect when the dress was in place. Another study tested the effect of the electrodress for about six weeks [29]. We wanted to see the long-term effects and extended the study time to 6 months (and for three of the children to 1 year).

The children were characterised by all groups of GMFCS I–V and all kinds of CP, including hemiplegia, diplegia and dystonia with an age range of 2–16 years. The diversity of GMFCS levels, types of CP and ages might have influenced the result. We believed that it was important to include all groups in this first study of the garment.

The youngest child was 2 years of age and the oldest was 16. One limitation is the size of the garment. The smallest size is 105 cm. The control unit is the same size for all dresses, and this could be a problem for those smaller children who are non-ambulatory, because the device is placed in front of the stomach (Figure 1).

We used the Gross Motor Function Measure (GMFM) assessment tool designed to measure changes in gross motor function over time for interventions in children with CP. There is an evidence-based possibility of prognostication regarding gross motor progress in children with CP, providing parents and clinicians with means to plan interventions and to judge progress over time [22]. Bakaniene *et al* compared the electrodress to physiotherapy an found that both groups improved their GMFM scores with no significant difference between groups [30]. However, the study time was only three weeks, which would be too short to notice any difference. In our study, nine children were evaluated at baseline and after 6 months. Six of the children improved their overall scores, five of whom were ambulatory. Three showed no improvement, one whom was ambulatory. We consider this study to be exploratory because, with this small a group, it is difficult to determine whether the dress contributed to the improvements or whether improvements resulted from the normal development with age and growth.

Physiological cost index (PCI) has been shown to be a reliable outcome measure of gait efficiency in children with CP. Interventions, that effectively reduce energy consumption, can be identified [31]. Of the eight children who completed 6 months of the study, three were ambulatory. Two of them managed to perform the PCI test. Both improved their index at six months compared to baseline. One girl re-took the test after one year. She had stopped using the dress after 6 months and her results reverted to baseline.

Pain is common in children with CP. In a European multicentre study, self-reported pain in the previous week occurred in 60% and parent-reported pain in the previous 4 weeks occurred in 73% [32]. We wanted to determine whether the electrodress would have any effect on pain. Nine children had no problem with pain at baseline. Of the seven who did have pain, four showed reduced pain, two showed unchanged pain, and one reported increased pain after treatment with the dress, suggesting that the dress may have some pain-reducing effect.

Children with CP often have significant gastrointestinal symptoms. Del Giudice *et al.* found that 74% of these children had chronic constipation [33]. The aetiology is usually multifactorial; however, it is often due to slow motility in the intestines. In a case study by Zollers *et al.* the authors used visceral and neural manipulation to restore the mobility of the organs [34]. They were able to improve the number of bowel movements. We found no improvements or change of bowel function with the use of the electrodress in terms of frequency, consistency, or use of laxatives.

Sleep disturbances are common in children with CP with an incidence between 23% and 46% compared to what is reported for normally developing children (20% to 30%) [35]. Sleep problems include difficulty in initiating and maintaining sleep, sleep-wake transition, sleep breathing disorders, sleep bruxism, excessive daytime sleeping, nightmares and sleep talking [36]. We chose to register wake-up episodes during the study period. Eleven of 16 children had unchanged frequency, four had fewer wake-ups and one child had increased wake-up frequency.

During the study, eight children dropped out at different times for various reasons (Table 2). Some of the children or caregivers chose to drop out despite the fact that their expectations had been fulfilled, at least to some extent. This phenomenon was described in a pilot study by Nordstrom *et al.* [37]. Parental and patient expectations were high and it was important that changes made a difference. It appears that some of the problems with using the dress derive from the fact that the garment is quite tight. It needs to be tight to secure the placement of the electrodes; nevertheless, this problem needs to be resolved. The dress needs to be easier to place on a spastic child. The control unit has to be smaller for the smaller children and for the non-ambulatory children who sit during the treatment.

Our results generate some questions. Why are the results not consistent over time? For pROM, there was a slight decline in the results after three and six months although the improvement remained significant. The overall relative results were almost the same after three and six months (Table 4). The explanation might be that the muscles improved to a certain degree after which there was no room for further improvement. Why are the results better for some muscles? This could be related to the degree of spasticity and differential responses in the muscles to the stimulation. Some muscles may need more stimulation than others. With the dress, it is possible to provide different levels of stimulation for different muscle groups, i.e. the electrodes can be programmed differentially. The improvement of the spasticity as measured using the modified Ashworth Scale was significant after 1 month and six months; however, only almost significant after three months. This might be explained by the fact that some patients dropped out after three months and

the remaining patients were those that derived the most benefit of the device. It is concerning that the modified Tardieu scale only showed a significant improvement after 1 month and not after 3 and 6 months. Not all children have a first catch, and it depends on the degree of spasticity of the muscles. With the improvement of the spasticity, it might be expected that further improvement of the first catch would not be possible.

Conclusion

This pilot study was conducted to test the effect of a non-pharmaceutical and non-invasive intervention using an electrodress to reduce spasticity in children with CP. The treatment lasted 60 min every other day with low frequency, low-intensity stimulation of the antagonist muscle to provide reciprocal inhibition of the agonist (the spastic muscle). The number of muscles with improvement was significantly higher. Passive range of motion was improved after 1, 3, 6 and 12 months. The modified Ashworth scale improved at 1 and 6 months but not after 3 months. The modified Tardieu scale improved after 1 month but not after 3 or 6 months. The children suffering from pain reported less pain after treatment. Some effect was seen on sleeping patterns for a few of the patients. No effect was seen on bowel function except for one child. This study has demonstrated that the electrodress has an effect on spasticity. Further studies are needed to optimise the frequency and intensity of the current with respect to the effect of the level of spasticity as well as to sub-stratify effects on treatment with respect to the level of handicap (GMFCS level). The garment and the control unit would benefit from further improvements in terms of comfort for better compliance. The dress appears to reduce the need for other spasticity reducing treatments such as treatment with botulinum toxin injections and could be a complementary therapy to reduce the need and frequency of injections.

Acknowledgements

We thank all children and their caregivers for participating in this study and thanks to all physiotherapists who performed the measurements.

Disclosure statement

The authors declare no competing interests.

Funding

No funding was received for the study. The company provided the dresses during the study period and the technicians that controlled the control units during the study to ensure that they worked properly.

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