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Case Report

Immediate Postural Change in a Child with Choreic Movement Disorder Using Elasto-Compressive Bodysuit

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Abstract

Background/Objectives: Dynamic movement orthoses (DMO) are elasto-compressive bodysuits used in the rehabilitation of children with motor disabilities and mainly in children with cerebral palsy. Among the DMO, the FLEXA® represents one of the most frequently used orthoses in clinical practice due to its adaptability and flexibility. The purpose of the present case study is to describe the application of FLEXA® in a female child of 18 months of correct age with a choreic form of cerebral palsy. **Methods:** To evaluate the effect of the dynamic movement orthosis's (FLEXA®), the *Movement Disorder-Childhood Rating Scale* (MD-CRS) 0-3 was administered. The child was evaluated before the use of the FLEXA® bodysuit and with the bodysuit donned at approximately 30 minutes after its application. **Results:** The results showed an important change in the severity of the movements according to the MD-CRS; mainly the child's movement disorder severity changed from a grade 5 severity (profoundly affected) performed without the bodysuit to grade 3 (moderately affected) with the use of the bodysuit. The evaluation also shows better trunk posture with use of FLEXA®. **Conclusions:** This case report highlights the potential benefits of dynamic movement orthosis like the FLEXA® in managing movement disorders in children with choreic form of cerebral palsy. A follow up evaluation is necessary to confirm the beneficial effects of continuous use of the DMO in a short and a long period of time.

Keywords: cerebral palsy; movement disorders; dynamic movement orthoses; dyskinetic; rehabilitation

1. Introduction

Cerebral palsy (CP) is one of the most common physical disorders of childhood, defined as a group of permanent disorders of the development of movement and posture, causing activity limitation, that are attributed to non-progressive disturbances that occurred in the developing fetal or child brain. [1].

The scientific literature of the last ten years has increasingly focused on the importance of early diagnosis of children with cerebral palsy to provide early rehabilitation care that can promote and optimize child neuroplasticity, prevent complications, and enhance parent and caregiver well-being. [2]

Depending the type of movement disorder, cerebral palsy is classified in spastic, dyskinetic including dystonia and athetosis, ataxic and hypotonic. About 10% of cerebral palsy is represented by the dyskinetic forms, characterized by abnormal movements, abrupt changes in tone and the persistence of primitive reflexes that limit the ability to organize and execute voluntary movements and the ability to maintain posture. The dyskinetic form of CP represents one of the most frequent

forms of movement disorder in childhood [3] and are frequently associated with disorder of cerebral cortex, basal ganglia, cerebellum and other motor pathways which are the cause of a brain injury.[4]

According to the recent literature, the most evident therapeutic approaches for the rehabilitation of dyskinetic forms of child cerebral palsy are taping, mobility, strength trainings and environmental enrichment to promote task performance. [5–7]

A pharmacological approach using baclofen, trihexyphenidyl, and benzodiazepines is often necessary to accompany rehabilitation treatment. Tetrabenazine also acts as a weak postsynaptic dopamine-2 receptor antagonist. The efficacy of this treatment is limited by the lack of clinical trials on long-term effects in this patient population and potential dose-related side-effects including drowsiness, nausea, parkinsonism, depression, and insomnia. [6]

Dynamic movement orthoses (DMO) are elasto-compressive tools used in the rehabilitation of children with motor disabilities and in children with cerebral palsy. [7] The literature review on preventive and rehabilitative interventions in child with cerebral palsy identifies the use of DMO as an intervention that is likely to benefit these children. [5] The use of bodysuit is part of the interventions that can benefit the motor function [5]. The aim of these orthoses is to improve postural stability, posture symmetry, and movement, without restricting functional movement. The elastic fabric consists of individual panels that are sewn together so that pressure is applied to the trunk and limbs, combined with zips to allow for ease of application. The use of DMOs, to support the rehabilitation of children with motor disabilities, has been extensively used in clinical practice and in various scientific works to promote normalization of muscle tone, increased perception of body position in space, symmetry, posture and movement, stability and proximal control, reduction of pain [8] [9].

Many types of DMO are used in clinical practice associated with standard therapy or specific treatment programs. These include Theratogs suit, Adeli suit treatment, Lycra, FLEXA®, SPIO, Spinecor, SelLi suit. [7] These suits are dynamic orthoses available in different designs from full body suits to smaller garments such as sleeves/gloves and leggings. Although there is no consensus regarding the advantages and disadvantages of wearing these suits, they are used in clinical practice with specific rehabilitation programme and monitoring by therapists. [7]

FLEXA®

Among the DMO, the FLEXA® represents one of the most frequently used in our clinical practice due to its adaptability and flexibility; it can be applied as a bodysuit, glove, or tight-fitting legging that completely covers the trunk, arms, and legs. FLEXA® is less invasive than traditional orthoses, is breathable and has a wide weave. It has seven lines of force, and it can be reinforced to exert specific directional forces without restricting functional movement. The pressure applied by body improved stability and a decreasing effect of external force vectors. It can contain and reduce involuntary movements, favouring realignment and giving at the muscles a biomechanical advantage. The FLEXA® is a specific DMO that has been widely used in clinical practice in different type of children with motor and orthopaedic disorders. This type of DMO is distinguished from others using innovative materials and attention to packaging, elements that contribute to comfort and resistance. The base of the bodysuit is made of a natural-synthetic fibre. Cotton is the component in direct contact with the skin. A significant aspect of the FLEXA® is the use of elasticised velcro, which allows prolonged use of the brace, adapting to the patient's growing and changing needs. The materials used for the FLEXA® are supplied by European manufacturers (Germany and Italy).

A further advantage offered by the FLEXA® is the possibility of customization, responding to the requests of physician and therapists and providing a personalised solution for each patient.

Although its large use in clinical practice, no specific study has been published in literature about the use in both adult and paediatric patients. The specific use of FLEXA is reported exclusively in a case report that explains the use of a FLEXA glove in a boy with cerebral palsy as an alternative to the use of neuromuscular tape to increase autonomy and independence. [10].

The purpose of the present case study is to describe the application of FLEXA® in a female child of 18 months of correct age with a choreic form of cerebral palsy.

2. Materials and Methods

This case report details about the application of FLEXA® in a female child, corrected age (CA) 18 months, with cerebral palsy, specifically with a choreic movement disorder. The child was born prematurely at 32 weeks and 6 days due to premature rupture of membranes and was urgently delivered via caesarean section. At term age, a MRI indicated extensive gliotic-malacic changes, cortical necrosis, and hemosiderin deposits primarily in the posterior periventricular white matter and basal nuclei. By 4 months CA, a neurological structured evaluation [2] identifying motor delays with a predominant extensor tone at the axial level and a significant visual deficit. The child used Valproic acid from the 9 months CA due to the presence of electroclinical episodes characterized by focal onset epileptic spasms. During follow-up, a psychomotor development using the Bayley-III scale at 12 months CA revealed delays in cognitive and language development, as well as in socio-emotional behaviour and adaptive functioning. Genetic analyses for isolated congenital heart defects and through array- Comparative Genomic Hybridization (array-CGH) were conducted, with normal results. At 12 months CA, significant axial hypotonia, lack of trunk control, and poor, chaotic motility were observed. At the 15-month CA, the child shows a level V at the Gross Motor function classification system E & R (12% at Gross Motor Function system-66) [1,11,12] and the motility evolved into a choreiform movement disorder affecting the trunk, limbs, and peribuccal area. Due to the presence of movement disorders that impacted the motor performance, the family was introduced to the use of a DMO (FLEXA®) from 18 months CA, to improve axial control, alignment, and stabilization. An occupational therapist performed the measures and fitted the FLEXA® before ordering it and educated the family for use. At the time of introduction of the DMO the child was not taking medication for chorea.

Outcomes

The subsequent follow-up noted good compliance with the bodysuit and improved postural stability and reduced abrupt limb movements. The (MD-CRS) 0-3 was used to evaluate the effect of the DMO (FLEXA®). This scale is age-specific and identify the presence of the movement disorder by pointing out its prevalent pattern; it also evaluates how much the movement disorder affects motor functions and activities of daily living, and rate the intensity of the disorder in different body regions. The MD-CRS allow monitoring of the disorder over time and the effectiveness of pharmacological, surgical or rehabilitative treatment. [13] The scale is composed of two sections: General assessment (part I) and Movement Disorder (MD) assessment (part II). Part I is to allow a global assessment of the child from observing how much the movement disorder affects motor function, oral/verbal function, level of responsiveness to the environment, and in older children, personal autonomies. Part II assesses the severity of movement disorder in different parts of the body. Scoring the scale to generate an Index I related to Part I (General assessment), an Index II related to Part II (Movement-disorder severity), and a Global Index. Each Index can range from 0 to 1 and it is divided into five classes, according to the severity of movement disorder. Class 1 included a 0–0.2 Index (healthy), class 2 a 0.2–0.4 Index (mildly affected), class 3 a 0.4–0.6 Index (moderately affected), class 4 a 0.6–0.8 Index (severely affected), and class 5 a 0.8–1 Index (profoundly affected). The MD-CRS scale is validated and extensively used in clinical practice for the evaluation of rehabilitative and pharmacological treatments due to its easy reproducibility and it's inter and intra-rater reliability. [13–16]

2. Results

The Movement Disorder-Childhood Rating Scale (MD-CRS) 0-3 was administered to the child twice (T0 and T1) on the same day: once without the bodysuit (T0) and then with the bodysuit on after wearing it for approximately 30 minutes after its application (T1). The assessment was administered by one paediatric therapist (CV) trained in the use of Movement Disorder-Childhood Rating Scale (MD-CRS). Administration of the scale with and without the bodysuit showed an

important change in the severity of movement disorder (Index II) (Table1, Video). The severity of the child's movement disorder (part II) with the use of the bodysuit was placed in class 3 (moderately affected by MD) in comparison with the assessment without the use of the bodysuit, which showed a class 5 for severity (profoundly affected by MD) (Table 1). In details, the improvements were observed in the trunk, upper limb, and lower limb items. However, no differences were found in Part I (General Assessment) and Global Index with and without the bodysuit. The child enjoyed wearing the body with no specific adverse effect. There were no reports of difficulties on managing toileting by parent.

Table 1. Movement disorder child rating scale results

	Index I	Class I	Index II	Class II	Global Index	Globale Class
Without Bodysuit (T0)	0,675	4 (Severly affected)	0,857143	5 Profoundly affected)	0,75	4 (Severly affected)
With Bodysuit (T1)	0,675	4 (Severly affected)	0,571429	3 (Moderately Affected)	0,632353	4 (Severly affected)

2. Discussion

This case report is the first study presenting the use of a FLEXA® orthosis in a pediatric choreiform movement disorder. The effectiveness of DMO orthoses in improving posture, dynamic movement, function and quality of movement appears to be confirmed by the use of instruments largely used in scientific research and clinical practice, such as gait and motion analysis, the Gross Motor Function Measure (GMFM) the Pediatric Evaluation of Disability Inventory (PEDI), the Quality of Upper Extremity Skills test (QUEST), the Ten meter walking test, the Visual Analogue Scale (VAS) performed before and after the use of DMO aids, also in studies with control groups using traditional rehabilitation tools. [17,18] Previous studies have measured the effects of DMO on gait and motor skills with tools such as motion analysis, GMFM, PEDI, Quest, ten meter walk test and visual analogue scale.

In the present case study, the FLEXA®, a specific type of DMO, has been used for the first time to improve the movement disorder. Mainly, a 18 month old female child with a choreic movement disorder showed a significant improvement in clinical assessment with the MD-CRS after the use of a FLEXA ® bodysuit for 30 minutes, compared to the evaluation without it. The greatest clinical change was observed in the assessment of the severity of the presence of the movement disorder (Index II) with a change of two classes, especially in the trunk, upper limbs and lower limbs.

No change in General assessment (Index I) and in the Global Score were observed. However, the interval between the two assessments with and without body suit is too short to observe changes in functional competence. A modification in competence should be evaluated with the follow up at least six months later, as it requires the combined use of orthoses and rehabilitation treatment. The efficacy of the FLEXA® on the movement disorders, represented by the reduction of movement in the examined districts, may be related to the proprioceptive stimulus given by the orthosis. The FLEXA® acts like a second skin, providing a feeling of containment and a perceptive margin without restricting movement. Pressure from skin contact and increased internal soft tissue pressure leads to enhanced proprioceptive feedback to improve positional limb and body awareness, improving muscle activation and movement control.

The literature review in children with dyskinetic disorders shows the importance of a dynamic orthosis to be better tolerated due to the presence of fluctuations in muscle tone. A dynamic orthosis it also provides a proprioceptive stimulation that supports the movement [19,20]

Moreover, the mechanism of action of these orthoses can modify the impulses that reach the central nervous system, consequently modifying the motor response.[21] Furthermore, the use of DMO would better provide cutaneous and proprioceptive sensory feedback by improving the execution of accurate movements and inhibiting motor cortex responses by reducing pathological reflexes and improving muscle synergies through the normalization of vestibulo-proprioceptive inputs. [18,21]

An intensive therapy with the suit on can enables re-education of the brain to recognize and form the correct movement of the muscles. The more correct proprioceptive input result in the more proper alignment. [21]

2. Conclusions

In conclusion, this case report highlights the potential benefits of dynamic movement orthosis like FLEXA® in managing movement disorders in children with choreic form of cerebral palsy. It allows a modification of afferent stimuli to the central nervous system and a consequent modification of the tone and motor pattern [21]. Due to a reduction of movement disorder affecting the 4 limbs and the beneficial effects on trunk posture in a timeframe of 30 minutes, the authors proposed the prolonged use of this DMO to analyze the persistence of these achievements to promote rehabilitative goals. Therefore, a follow up is mandatory to confirm the beneficial effects of continuous use of the DMO in a short and a long period of time. It is also a necessary to increase knowledge about the use of these orthoses in children with movement disorders by expanding the study sample.

The: authors report there are no competing interests to declare.

Disclosure of interest: The authors report no conflict of interest.

Abbreviations

The following abbreviations are used in this manuscript:

DMO	Dynamic Movement orthoses
MD-CRS	Movement Disorder-Childhood Rating Scale
CP	Cerebral Palsy
CA	Correct Age
Array- CGH	Array Comparative Genomic Hybridization
MD	Movement Disorder
GMFM	Gross Motor Function Measure
PEDI	Pediatric Evaluation of Dysability Inventory
QUEST	Quality of upper limb skills test
VAS	Visual Analogue Scale

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