RESEARCH ARTICLE

Effectiveness of Standing Frame on Constipation in Children with Cerebral Palsy: A Single-Subject Study

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Abstract

Children with cerebral palsy (CP) and quadriplegia or severe diplegia suffer from highly reduced mobility and consequent constipation. Clinicians recommend standing frames to exercise the support reaction in this population, sharing the opinion that the upright position may facilitate intestinal transit, although no evidence supports this assumption. We conducted this study to determine the effects of the standing frame on spontaneous evacuation in children with CP. Moreover, we studied its effects on the frequency of induction of evacuation, the characteristics of the stool and the pain suffered by the child due to constipation and/or evacuation.

We implemented a single-subject research design in one chronically constipated child with CP and quadriplegia, Gross Motor Function Classification System Level V. To monitor the effects of the standing frame, we measured the outcome of interest throughout the study using a daily diary and the Bristol Stool Scale. This study was approved by the local Ethics Committee.

This study has several limitation; primarily, the use of a single-subject research design only makes possible the visual analysis of data obtained from a unique patient. So, by themselves, data obtained do not allow us any generalization for the target population. Future research should verify our results collecting more data and also investigating the effect of the standing-frame on respiratory functions.

Although the standing frame did not affect the frequency of evacuations or the characteristics of the stool, its employment reduced the inductions of evacuation and the related pain suffered by the child. However, this study has several limitations, such as the lack of generalization due to the fact that we studied a unique patient and the overall brevity of the study due to external circumstances. Therefore, we suggest future research to verify our results, also investigating the effect of the standing frame on respiratory functions.

Relevance to clinical practice. The standing frame may positively influence the management of constipation of these children, possibly improving their quality of life. Copyright © 2014 John Wiley & Sons, Ltd.

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Keywords

cerebral palsy; chronic constipation; standing frame; severe diplegia; paediatric occupational therapy

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Cerebral Palsy (CP) has been recently defined as "a group of disorders of the development of movement and posture, causing activity limitation, that are attributed to non-progressive disturbances that occurred in the developing foetal or infant brain. The motor disorders of Cerebral Palsy are often accompanied by disturbances of sensation, cognition, communication, perception and/or behaviour and/or by a seizure disorder" (Bax et al., 2005). This definition emphasizes the importance of assessing activity limitations to be able to obtain a comprehensive picture of the disease, according to the approach proposed by the International Classification of Functioning.

Almost all children with quadriplegia or severe diplegia show internistic problems that limit their functions and quality of life; as a matter of fact, constipation is widespread in this population (Veugelers et al., 2010). Constipation is probably caused by neurological abnormalities of the digestive system control, and it might also be due to the limited mobility that characterizes these children (Araújo et al., 2012). Sometimes this problem can lead to serious complications, such as intestinal obstruction and bleeding. In addition, constipation involves a strong commitment and effort in its management by caregivers.

Children with CP in the quadriplegic or severe diplegic forms are frequently prescribed a standing frame in order to exercise the support reaction and passively maintain the upright position. This activity may be indicated for several reasons; the most important are the improvement of the social interaction and the participation in meaningful occupations (Wright, 2011). Other common and important indications are the improvement of the postural alignment, facilitating the extension of the head and the trunk, the increase of the load over the lower limbs, reducing the risk for osteoporosis and the lengthening of the muscular chains.

In the literature, there are studies suggesting that in children with CP, the upright position facilitates the intestinal transit, improves cardiovascular system functionality, maintains the muscle trophism and helps the resolution of urinary tract infections and respiratory problems (Wright, 2011). Additionally, it is common opinion among experts that the standing frame can be beneficial in a number of areas of internal medicine, because it probably has an effect on gastrointestinal and respiratory functions (Mogul-Rotman, 2008; Labandz, 2010; Rosen, 2010). Health care

professionals share the opinion that the upright position contributes to relieving constipation, so clinicians frequently indicate the standing frame with this clinical purpose. In searching the literature, we found one systematic review (Pin, 2007) and eight clinical trials dealing with the use of the standing frame in children with CP (Ivey et al., 1981; Motloch and Brearley, 1983; Herman et al., 2007; Kecskemethy et al., 2008; Eisenberg et al., 2009; Gibson et al., 2009; Gudjonsdottir and Stemmons, 2009; Salem et al., 2010) (Figure I). However, none of these studies investigated the effectiveness of the standing frame on constipation.

Therefore, the principal aim of this single-subject research study was to assess the effectiveness of the standing frame on constipation, measuring the frequency of spontaneous evacuations, which means without the use of inductions, in a child with CP and consequent severe diplegia or quadriplegia. As secondary outcomes, we chose to measure the effects of the standing frame on the frequency of inductions of evacuation, the type of stool and the pain caused by constipation and/or evacuation.

Methods

Participant

The eligibility criteria for this study were CP in the manifestation of severe diplegia or quadriplegia, constipation problems as defined by guidelines (Clinical Practice Guideline, 2006) and age ranging from 3 up to 17 years. We excluded children currently using a standing frame and those with medical conditions who, according to clinical judgment, advise against the use of the standing frame.

According to the criteria we set for our study, we identified one child who met all the requirements, whose parents gave the informed consent to let him participate to this study.

A.H. is a five-year-old child, who has CP in the clinical manifestation of quadriplegia, Gross Motor Function Classification System Level V. He has dystonia in rotation, particularly located in the head. The child is in the charge of the Child Neuropsychiatry Unit of Carpi (Modena, Emilia-Romagna, Italy), and he is periodically examined by physicians and physical therapists at the Regional Unit for Severe Disability in the Developmental Age of Reggio Emilia.

His first observation took place in 2009 at the age of three when the child presented a severe motor impairment, poor head control and sciallorea.

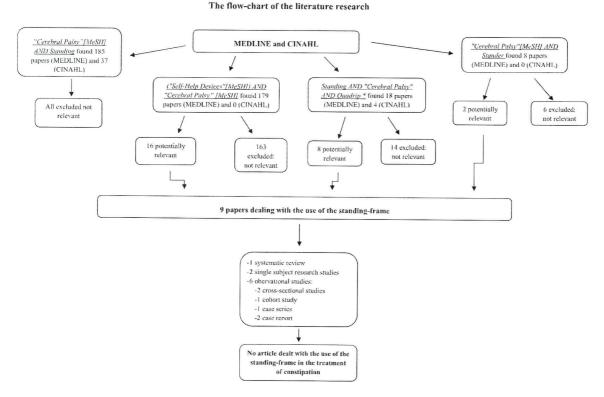


Figure 1 The flow chart of the literature research

At the present time, the child presents the automatic walking reflex, he has not reached the upright posture yet, and because of the important axial hypotonia, he manages to maintain the sitting position only by using a system for postural control in the wheelchair. This postural aid includes a pelvic belt at 45°, a butterfly harness at the level of the trunk and a head restraint that envelops his upper body up to the sternal level, in order to maintain the head in the correct position while accompanying it during dystonic movements. The motor repertoire of A.H. is very poor and primitive: He does not show much freedom in movement combinations because of the predominance of pathological dystonic patterns.

During a consultation in 2012 in Reggio Emilia, clinicians observed a variation in the tone of the neck muscles that increased his head control. The startle reactions had stopped. Therefore, the use of the standing frame was recommended to further stimulate postural control.

Because of the low degree of mobility, which limits his gastro-intestinal function, the child suffers from chronic constipation that his parents report to be the cause of significant abdominal pain. For this reason, the child needs to use enemas to induce bowel movements.

In addition to constipation, A.H. also has a large amount of mucus in the lungs that causes breathing problems and frequent pneumonias. For this reason, the child uses the Pep Mask, and occasionally, the mucus must be mechanically extracted.

Therefore, we though that in this child, the regular use of the standing frame might be ideal to improve postural control and to prevent secondary damages, such as constipation, potentially caused by immobility and the lack of standing posture.

Design

The population included in this study is rare and presents a wide variety of clinical manifestations. Therefore, we chose to implement a single-subject research design to study this topic (Backman et al., 1997). We chose to implement the $A_1B_1A_2B_2$ design, because we judged it appropriate to the context. This research design provides an alternation of phases of control (A_1 and A_2), without the use of the standing frame, and phases of intervention (B_1 and B_2), with the daily use of the standing frame in the living environment of the child, in accordance with the usual clinical practice (Herman et al., 2007; Kecskemethy et al., 2008; Eisenberg et al., 2009; Gudjonsdottir and Stemmons, 2009).

Our study had an estimated duration of 14 weeks divided into four periods. The first 2 weeks constituted the baseline phase, during which the initial assessment took place, without the use of the standing frame (A₁). After the baseline phase, there were 4 weeks of daily use of the standing frame (B₁) followed by an additional 4 weeks of suspension of the use of the device (A₂). During the final further period (B₂), the standing frame was reintroduced. To monitor the effects of the standing frame, we measured the outcome of interest throughout all phases using the daily diary and the Bristol Stool Scale (Heaton and Lewis, 1997).

Outcomes

The daily diary

The outcomes of interest were measured using a daily diary completed by the child's parents and the assistant teacher assigned to the child at school.

The daily diary looks like a simple grid that has to be filled out daily by choosing one of the options provided for any item investigated (Annex 1 in the Supporting Information).

The diary has a specific space where the caregiver could write notes regarding any change in the daily habits that could be considered useful for this study.

The Bristol Stool Scale

The Bristol Stool Scale (Heaton and Lewis, 1997) is a diagnostic tool commonly used in clinical practice by paediatricians and gastroenterologists. This scale categorizes the different types of stool into seven classes, on the basis of their shape and texture. The first two classes are symptoms of constipation, the third and the fourth classes indicate an ideal consistency of the stool, whereas the last three classes approach progressively to diarrhoea. Each class is represented in the scale by a graphical depiction of the stool type, accompanied by a quick qualitative description and a numerical correspondence (e.g. type 1: separate hard lumps, like nuts, hard to pass).

In our study, the Bristol Stool Scale was attached to the daily diary, and the numerical data were registered by the caregiver on it.

Questions on burden of care

We created a questionnaire to collect data regarding the burden on caregivers because of the evacuation management (Annex 2 in the Supporting Information) and tested it on three caregivers of children with CP before the beginning of this study. Using a four-category rating scale, we investigated the daily load due to the management of child's evacuation. By way of the same scale, we investigated the financial cost of inductions, the time spent in inducing and/or managing the evacuation and the uneasiness caused to caregivers by these manoeuvres, due to the fact that they cause discomfort and/or pain to their child. We also collected data regarding the physical fatigue due to the management of the evacuation and the diaper change, by way of a visual rating scale. We decided to collect data regarding the physical fatigue on a six-point scale, in order to catch even the smallest variation in this area.

Procedure

Because of the influence of lifestyle and diet on the frequency and type of evacuation, at the very beginning of the study, just before the baseline started, we asked parents to complete a questionnaire regarding the eating and the evacuation habits and modalities, the duration of the constipation and the drug regimen of their child (Annex 3 in the Supporting Information). During the study, we controlled all these potential sources of variability by asking caregivers to report any variation that might occur in these parameters in the daily diary.

The principal investigator of this study, a senior student graduating in occupational therapy, met the assistant teacher and the parents to teach them the correct mode of filling out the daily diary. The diary had to be filled out every day for the entire study duration. In order to collect complete data, we gave one copy of the daily diary to the assistant teacher and another copy to the parents. We chose to delegate the filling out of the diary at home to the parent that was more frequently involved in the induction and evacuation manoeuvres (principal caregiver), in order to achieve the best reliability of the responses. The principal investigator was responsible for the monitoring of the data collection in the diary; so she called the caregivers on a daily basis during phase A1 of the study and visited the child in his environment during all the following

A professional team composed of a physical therapist, the occupational therapy student and an orthopaedic technician chose the appropriate standing frame. The standing frame is a position prone model, which allows a gradual verticalization; this characteristic was

considered essential for this type of pathology, which is frequently associated to perceptual disorder that makes any postural changes less tolerable without continuous restraint or visual control. Besides, the position prone standing frame stimulates the muscular activity of the extensors of the trunk and the head, allowing for a better postural control over these anatomical areas. This characteristic was considered to be appropriate for this child, whose axial skeletal muscles were particularly hypotonic.

The occupational therapy student and the physical therapist, together with the parents, discussed the best location where the standing frame could be continuously utilized in total security, in order to reach the best results: They chose to use the device at school where the amplitude of the environment facilitated its management. The assistant teacher took on the task of managing the device on every school day, for a minimum of 5 days a week.

The very first trial of the standing frame took place in the Child Neuropsychiatry Unit of Carpi. After that trial, we chose the most appropriate standing frame and planned a second trial in the school of the child, in the presence of A.H. and his parents, his assistant teacher and the professional team. We made all the necessary adjustments to adapt the device to the child, and the occupational therapist student trained the assistant teacher on how to use the device correctly. From the beginning, the child liked the vertical position and was able to hold it for 30 minutes. However, as could be expected, he required being involved in games or other occupations and needed some encouragement because of his exhaustibility in the upright position.

Then, we began the single-subject research design. We started the baseline phase A_1 on 23 May 2012. The first intervention phase B_1 was supposed to start on 6 June, but, on 29 May, the second and biggest earthquake in the area of Modena occurred, with its epicentre in Novi di Modena; the earthquake brought great inconvenience and considerable damage to the surrounding area. This event caused serious damage both to the school attended by A.H. and his family house, making both of them temporarily inaccessible.

The family of A.H. decided to temporarily move into another Italian Region, thus interrupting the study.

At the end of the summer, both the school and the house of the child had been declared fit for use and the family came back home. Together with the parents and the school personnel, we decided to start the study

again from the beginning; however, because the occupational therapist's thesis to graduate had to be completed by November, we decided to undertake only the first two phases of the study $(A_1 \text{ and } B_1)$ and reduce the duration of phase B_1 , which lasted 3 weeks, instead of 4, as initially programmed.

Then, baseline phase A_1 began again on 13 September and ended on 26 September 2012. The first intervention phase, B_1 , began on 27 September and ended on 17 October 2012.

The programme of use of the standing frame consisted of a minimum of 30 minutes of verticalization per day for 5 days a week. If well tolerated, the duration of verticalization could be prolonged up to 1 hour. In order to allow the child to maintain the upright position for a prolonged time, the assistant teacher involved him in games or activities also including other pupils.

Data analysis

Because this is a single-subject research design, data analyses consisted of graphical representation of the parameters monitored and the visual comparison of the trends shown in the phases of the study, looking for any noticeable differences (Backman et al., 1997). We searched for a change in the level, or a change in the trend and/or a reduction in the variability of the responses.

Results

Data collected from the initial questionnaire told us that the child usually eats mashed food and drinks liquids in regular amounts; his habitual evacuation frequency was about twice a week, and it was always necessary to induce the evacuation by stimulation, an enema associated to a facilitative posture, because he could not evacuate spontaneously. The type of stool evacuated usually falls into the first class of the Bristol Stool Scale.

Regarding the burden on his caregivers (Table I), the principal caregiver, A.H.'s father, reported that the daily load due to the management of the evacuation and the financial cost of the stimulation were minimal in both phases A_1 and B_1 ; however, the impact on the caregiver due to the time spent doing the stimulation and the discomfort caused by these manoeuvres decreased by one and two points, respectively, on a four-point scale. Notably, the discomfort was totally

Table I. The results of the questionnaire on burden of care

	Phase A ₁	Phase B ₁
Question no. 1. How much is the daily load due to the management of your child's evacuation?	1	1
Question no. 1. How much is the daily load due to the management of your data. Question no. 2.1. How much is the impact on you about: the financial cost of the stimulations?	2	2
Question no. 2.1. How much is the impact on you about the impact of the stimulations? Question no. 2.2. How much is the impact on you about the time spent doing the stimulations?	2	1
Question no. 2.2. How much is the impact on you about: the uneasiness caused by the discomfort	3	1
of your child during the stimulations? Question no. 3. How much is your physical fatigue due to the management of the evacuation?		
Question no. 4. How much is your physical fatigue due to the management of the diaper change? (if your child uses diapers)		

1, not at all; 2, minimal; 3, moderate; 4, a lot.

eliminated when the child started to use the standing frame. The physical fatigue due to the management of the evacuation decreased by one point, and the physical fatigue due to the diaper change decreased by two points, on a six-point scale.

The results of the data collected by the daily diary showed that the standing frame was used every school day, but one, when the child was absent from school because of a medical examination. The duration of use of the standing frame was from 30 to 45 minutes a day in both phases, depending on the tolerance of the child.

During phase A₁, the child evacuated five times in 14 days (28%), and during phase B₁, the child evacuated six times in 21 days (28%): Thus, the evacuation frequency was exactly the same in both phases (Figure II). However, the item relating to the presence of inductions to stimulate the evacuation showed us a relevant difference between the two study phases: In fact, in the baseline phase, the principal caregiver reported two inductions in 14 days, whereas

in the intervention phase, he reported no induction in 21 days; this result means that in B_1 phase, all the evacuations occurred spontaneously (Figure III). Thus, regarding the principal aim of this study, the frequency of spontaneous evacuation, we observed that during the regular use of the standing frame, A.H. always evacuated spontaneously.

According to the item relating to the time of the evacuation, we noticed that during the baseline phase, when evacuation occurred, it always occurred at the same time of the day, after its induction. In contrast, during the intervention phase, the evacuations occurred at different times of the day.

The stools were of the class two of the Bristol Stool Scale in both phases, with the exception of a single episode, during the intervention phase, when it became of class six. According to the written notes regarding the changes in the daily habits, the day before that episode, the child was administered an antibiotic to cure an infection of the upper respiratory airways and associated high fever.

Frequency of evacuation

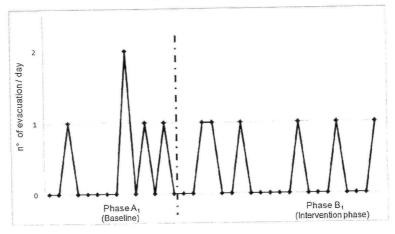


Figure 2 Frequency of evacuation

Frequency of inductions of evacuation

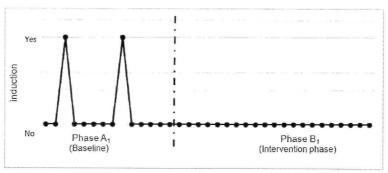


Figure 3 Frequency of inductions of evacuation

The modality of evacuation did not present any differences between the two phases because the child constantly used a diaper.

A notable improvement regards the item "pain caused by constipation and/or evacuation". In fact, during the 2 weeks of baseline phase, the principal caregiver reported four episodes of pain due to constipation or evacuation (28%), whereas in the 3 weeks of intervention phase, he reported no experience of pain related to constipation or evacuation (Figure IV).

Finally, looking at any possible changes to the daily habits and/or any unusual manifestations (e.g. behaviour, clinical signs and symptoms) that could have happened during the study, in phase B₁, the child presented two days with high fever; furthermore, in phase B₁, the assistant teacher reported that the child had frequent episodes of productive coughing during the use of the standing frame, episodes that subsided at the end of the verticalization session.

Discussion

The results of this study support the hypothesis that the use of the standing frame in children with CP and quadriplegia or severe diplegia has beneficial effects on constipation. The daily use of this device improves the frequency of spontaneous evacuations that in the intervention phase, always happened without any kind of induction. On the basis of this data, we can presume a positive correlation between the habitual use of the standing frame and the improvement in the intestinal transit. However, we cannot postulate the timely effect of verticalization because spontaneous evacuation occurred at various times of the day, whereas the device was always used during the morning.

During the intervention phase, not to be underestimated was a noticeable reduction in the pain felt by the child due to constipation and/or evacuation, as reported by the principal caregiver (spontaneous moaning, crying and facial grimaces). In fact, pain was completely absent in the intervention phase, whereas during the baseline phase, the child was affected on different days. Although we did not measure the participation in activities or the quality of life of this child, our data strongly suggest that the use of the standing frame might have increased both these important outcomes, as well as the level of participation in meaningful activities.

Pain due to constipation or evacuation

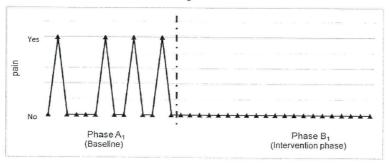


Figure 4 Pain due to constipation or evacuation

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We hypothesized that the use of the standing frame would induce a noticeable change of the type of stool, because the improvement of the intestinal transit would allow a reduction of the intestinal stasis. Our data did not verify this hypothesis, but these results are not surprising if we take into account that the evacuation frequency did not increase during the intervention phase.

Finally, on the basis of the questionnaire that the principal caregiver filled out, we noticed that the burden on the parents decreased with the introduction of the standing frame, and the reported discomfort was totally eliminated.

Even though measuring the effects of the standing frame on bronchial stasis was not among our outcomes, the assistant teacher reported that the child had excessive coughing episodes during verticalization, which was promptly relieved at the end of the session. Thus, the use of the device would probably affect the respiratory functions.

This study has several important weaknesses. Primarily, the use of a single-subject research design, although justified by the heterogeneity of the clinical manifestations of CP, only makes possible the visual analysis of the data obtained (Backman et al., 1997). Another typical limitation of this study design is the shortage of data due to the fact that we studied a unique patient. So, by themselves, data obtained do not allow us any generalization for the target population. Secondly, the earthquake that took place on 29 May had dramatic effects, and we were forced to lessen the overall duration of this study, reducing the number of weeks of phase B₁ and totally eliminating phases A₂ and B2. The implementation of these last two phases would have allowed us to verify with greater certainty the effects of the device on the outcomes of interest. Another important limitation that might have affected our study is the lack of original research in literature regarding this area: This fact has prevented us from comparing different study designs and maybe from implementing a more feasible and valid research. Certainly, it has resulted in having no data available to match up and discuss our findings. Nonetheless, we should keep in mind that we undertook this study because we could not find any evidence to support the shared opinion that the use of this device in children with CP is beneficial in a number of areas of internal medicine.

Future research should verify our results by way of longer periods of study and a multiple baseline design that includes more subjects (Backman et al., 1997). Given the encouraging data we registered on coughing, we also suggest investigating the effect of the standing frame on the bronchial stasis, assessing the amount of sputum, the cough efficacy and the upper respiratory airways infection rate.

In conclusion, this study was the first research design conducted in this very specific field. It brought out the first, although slender, piece of evidence to support the use of the standing frame for the treatment of constipation in quadriplegic or severe diplegic children with CP. Our single-subject research design demonstrated that the regular use of the standing frame did facilitate spontaneous evacuation and reduced the pain suffered by this child to zero, also thanks to the cessation of the need of inductions.

Conflict of interest

The author declares no conflict of interest.

Ethical approval

This study was approved by the Ethics Committee of the Province of Modena (Italy).

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Supporting information

Additional supporting information may be found in the online version of this article:

Annex 1

Annex 2

Annex 3