Effect of a new physiotherapy concept on bone mineral density, muscle force and gross motor function in children with bilateral cerebral palsy

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Abstract

Objective: The purpose of this study was to determine the effect of a new physiotherapy concept on bone density, muscle force and motor function in bilateral spastic cerebral palsy children. Methods: In a retrospective data analysis 78 children were analysed. The concept included whole body vibration, physiotherapy, resistance training and treadmill training. The concept is structured in two in-patient stays and two periods of three months home-based vibration training. Outcome measures were dual-energy x-ray absorption (DXA), Leonardo Tilt Table and a modified Gross Motor Function Measure before and after six months of training. Results: Percent changes were highly significant for bone mineral density, -content, muscle mass and significant for angle of verticalisation, muscle force and modified Gross Motor Function Measure after six months training. Conclusions: The new physiotherapy concept had a significant effect on bone mineral density, muscle force and gross motor function in bilateral spastic cerebral palsy children. This implicates an amelioration in all International Classification of Functioning, Disability and Health levels. The study serves as a basis for future research on evidence based paediatric physiotherapy taking into account developmental implications.

Keywords: Cerebral Palsy, Physiotherapy, Bone density, Motor function, Whole Body Vibration

Introduction

Cerebral Palsy (CP) is the most common cause of physical disability in childhood1. 57% of CP children in Europe are bilateral spastic (BSCP). They perform on a severely reduced activity level and suffer from reduced mobility or immobility. Immobilisation of the musculoskeletal system causes muscle mass loss which is followed by reduced bone mass which results in higher risk of low energy fractures and further immobilisation. Bone mineral density (BMD) is shown to be reduced in children with CP and correlated to immobility and non-weight-bearing. However more good quality research is needed. Research showed that muscular function correlates with skeletal morphology. Consequently, activation of the musculoskeletal system seems to be a promising approach to harness BMD and improve gross motor function in CP children.

There is a lack of evidence supporting effective strategies for musculoskeletal activation in CP children. The most common approaches used are Bobath/NDT and Conductive Education. More modern concepts arrogate a task-specific repetitive approach. Recently new approaches such as resistance training (RT), body weight supported treadmill training (BWSTT) and whole body vibration (WBV) have been described for musculoskeletal activation in CP patients.

WBV seems to be beneficial to improve BMD in disabled children, but functional improvements have not been investigated. RT has been shown to be beneficial to improve muscle strength and muscle volume and better strength can be translated into functional improvements. However improvements in BMD changes have not been investigated and the effect of RT in CP children is still controversially discussed. BWSTT seems to improve walking parameters in CP children, however more research is needed to confirm the effect and no BMD change is evaluated in the literature.
The new physiotherapy concept *On your feet* combines the bone-mass-harnessing approach of WBV, RT, BWSTT and physiotherapy. The CP child might benefit from an amelioration in all International Classification of Functioning, Disability and Health (ICF) levels (body structure and function and activity and participation) by restored bone mass, increased muscle force, better motor function, and better participation in all day living activities. According to our knowledge a concept like *On your feet* has never been described before and will provide valuable information that may be utilised to guide future physiotherapy clinical practice and research. Primary objective was to determine the effect of *On your feet* on BMD in BSCP children. Secondary objectives were to evaluate the effect of *On your feet* on bone mineral content (BMC), muscle mass, muscle force and gross motor function in BSCP children and the difference between spastic diplegia, quadriplegia and above and below age ten.

**Materials and Methods**

**Design.** *On your feet* is not a clinical trial but a routine procedure and component of the basic health care system in Germany. It is carried out in a rehabilitation centre (medifitreha for children) equipped for the requirements of the concept and affiliated to the Children’s University Hospital Cologne. The analysed data is part of patient observations, therefore sample sizes may vary due to incomplete data sets. Ethical approval was obtained from the Research Ethics Committee of the University of Cologne. Data was analysed retrospectively including all BSCP children attending *On your feet* since it was opened in 2006. Children were excluded if they did not participate in the concept, did not complete 6 months of training or were not diagnosed BSCP. Informed consent was obtained from legal guardians prior to participation.

**Interventions and Procedure.** The concept *On your feet* is...
designed to apply the full diversity of treatment approaches shown in Figure 1 as agreed on a legal basis with the health insurance providers in Germany. However if the child was too weak it could be altered individually according to the child’s goal setting. Therapy components were determined by utilising WBV as a reflective (neuro-muscular) possibility to activate muscular contraction beyond the conscious ability of the child; complemented by RT for selective muscle activation, BWSTT for implementing WBV and RT results in a more functional lower extremity training accompanied by pool therapy (utilising buoyancy) and rounded down by extensive physiotherapy sessions to work on individual goal settings. Bobath/NDT and Vojta were selected because they are the two most popular physiotherapy techniques for children in Germany. WBV settings were set individually according to the child’s progress and comfort. A selection of five different training apparatus is available at the medifitreha for children (abductor/outer thigh, leg press, cable machines, dips and extension leg curl). A selection of four different apparatus was used to target isolated muscle activity of the lower extremity; selected by the child’s goal setting and capabilities. Dips were also included to train the upper extremity for using walking aids if adequate. The training apparatus was adapted to children’s body size by Stolzenberg GmbH-Dynamed (Erftstadt, Germany).

The first in-patient stay contained of 13 consecutive days with one day break from therapy. The participating children were in-patients because they were taking part in the concept only and for no other medical reasons. Therapies were applied as listed in Figure 1. During this stay the baseline assessment (M0) was taken. After the first in-patient stay the children exercised with the Galileo® system (Galileo® or Galileo TT®) at home twice daily (3x3 minutes) for three months. After these three months the second in-patient stay (six consecutive days) took place also including the therapy components shown in Figure 1. Therapy and goals were adjusted to the progress of the child. Again, three months home-based Galileo® training followed. Six months after M0, the children were assessed in an out-patient examination (M6) and the Galileo system was returned to the clinic.

**Outcome Measures** were taken at baseline (M0, first in-patient stay) and after 6 months of training (M6, out-patient examination). BMD, bone mineral content (BMC) and muscle mass were assessed at ICF body structures level by dual-energy x-ray absorption (DXA) with the GE Lunar PRODIGY device (GE Ultraschall GmbH, Solingen, Germany). DXA is the current standard for measuring BMD in children. Muscle force (F) was assessed at the ICF body functions level and analysed by a ground reaction force plate combined with a tilt table (TT) (Leonardo TT®, Novotec Medical GmbH, Pforzheim, Germany). Ground reaction force (Test B) was calculated using the formula \[ F = \sin(\text{tilt angle}) \times (\text{body mass (kg)} \times 9.81 \text{ kg/ms}^2) \]. Test A assessed the angle of verticalisation. Commonly isokinetic testing devices or hand held dynamometry are utilized to assess muscle force. However, a limitation of isokinetic testing is the time required to test multiple joints and the assessment of individual muscle groups in contrast to the more functional interaction to maintain body weight against gravity. In Leonardo TT Test B ground reaction force increases with augmented verticalisation, because more muscular power is needed to maintain verticalisation. Therefore, the tilt angle (Test A) determines muscle force of the lower extremity. We found the TT a more functional assessment method for lower extremity muscle force quantification. Gross motor function was assessed at ICF activity level by the Gross Motor Function Measure (GMFM)²⁸. For practical reasons for the first two years of the concept On your feet an abbreviated version of the GMFM (modified GMFM, mGMFM) including only 30 items instead of 66 was used. The physiotherapeutic

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### Abbreviations

<table>
<thead>
<tr>
<th>Abbreviation</th>
<th>Description</th>
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<tbody>
<tr>
<td>BMC</td>
<td>Bone Mineral Content</td>
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<tr>
<td>BMD</td>
<td>Bone Mineral Density</td>
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<td>BSCP</td>
<td>Bilateral Spastic Cerebral Palsy</td>
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<td>BWSTT</td>
<td>Body Weight Supported Treadmill Training</td>
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<tr>
<td>CP</td>
<td>Cerebral Palsy</td>
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<tr>
<td>GMFCS</td>
<td>Gross Motor Function Classification System</td>
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<tr>
<td>GMFM</td>
<td>Gross Motor Function Measure</td>
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<tr>
<td>ICF</td>
<td>International Classification of Function</td>
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<tr>
<td>DT</td>
<td>Disability and Health</td>
</tr>
<tr>
<td>RT</td>
<td>Resistance Training</td>
</tr>
<tr>
<td>TT</td>
<td>Tilt Table</td>
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<tr>
<td>WBV</td>
<td>Whole Body Vibration</td>
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### Table 1. Participant details, diagnosis and age in M0 before participation in On your feet.

<table>
<thead>
<tr>
<th>Age</th>
<th>n</th>
<th>%</th>
<th>Mean</th>
<th>SD</th>
<th>Range</th>
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<tbody>
<tr>
<td>Total BSCP</td>
<td>78</td>
<td>100</td>
<td>9,76</td>
<td>4,03</td>
<td>2,27-24,62</td>
</tr>
<tr>
<td>Spastic quadriplegia &gt;10 years</td>
<td>47</td>
<td>60</td>
<td>10,2</td>
<td>4,28</td>
<td>2,95-24,62</td>
</tr>
<tr>
<td>Spastic diplegia &lt;10 years</td>
<td>24</td>
<td>31</td>
<td>13,38</td>
<td>3,34</td>
<td>10,30-24,62</td>
</tr>
<tr>
<td>Spastic diplegia &gt;10 years</td>
<td>23</td>
<td>29</td>
<td>6,89</td>
<td>2,05</td>
<td>2,95-9,62</td>
</tr>
<tr>
<td>Spastic quadriplegia &lt;10 years</td>
<td>12</td>
<td>16</td>
<td>12,72</td>
<td>2,5</td>
<td>10,01-16,22</td>
</tr>
<tr>
<td>Spastic diplegia &gt;10 years</td>
<td>19</td>
<td>24</td>
<td>6,8</td>
<td>1,79</td>
<td>2,27-9,84</td>
</tr>
</tbody>
</table>

### Table 2. Total height and weight before and after participation at On your feet with z-scores and p-value.

<table>
<thead>
<tr>
<th>Total (n 78)</th>
<th>M0</th>
<th>M6</th>
<th>p</th>
</tr>
</thead>
<tbody>
<tr>
<td>Height (cm)</td>
<td>128,3</td>
<td>130,9</td>
<td>&lt;0,0001</td>
</tr>
<tr>
<td>z-score</td>
<td>-1,76</td>
<td>-1,79</td>
<td>0,42</td>
</tr>
<tr>
<td>Weight (kg)</td>
<td>29,62</td>
<td>31,16</td>
<td>&lt;0,0001</td>
</tr>
<tr>
<td>z-score</td>
<td>-0,86</td>
<td>-0,86</td>
<td>0,71</td>
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team at the medifitreh in Cologne was not appropriately staffed in the beginning to conduct the full version of the GMFFM. It would have taken too much time to conduct the full version additionally to the running rehabilitation clinic.

Data Analysis. Data was analysed using PC Statistics version 4.0 (Hoffmann-Software, Giessen, Germany). The effect of On your feet on BMD, BMC, muscle mass, muscle force and gross motor function was tested using the dependent Wilcoxon test for pair differences. The difference between spastic diplegic and quadriplegic respectively above and below age ten was tested with the independent samples Wilcoxon test. Results for before and after On your feet are presented as percent change and box and whisker plots. Group differences are presented in percent change and mean±SD with 95% confidence intervals (CI). BMD is calculated as whole body without head in g/cm². Means±SD for BMC and muscle mass are adjusted for height (BMC/cm and muscle mass/cm). The mGMFM is presented in points. The significance level was set at 0.05. A power calculation was conducted for BMD (whole body without head) and found 29 patients the minimum sample for a study power of 99.999%.

Results

Since the concept On your feet started in 2006, 78 subjects diagnosed with BSCP participated (56% male, 44% female) with a mean age of 9.76 years in M0 (median 9.33 years). Two children were Gross Motor Function Classification System (GMFCS) level I (3%), nine were level II (12%), 32 level III (41%), 28 level IV (36%) and seven level V (9%). 47 children were diagnosed with spastic quadriplegia and 31 with spastic diplegia (Table 1). There was no significant difference in age between the spastic quadriplegia and the spastic diplegia group (p=0.2224). The two groups were subdivided into children above age ten and below age ten, whereas the younger group was less than ten years and the older group ten years and above (Table 1).

BMD ranks showed a significant difference (p<0.0001) in total of 2.3% after completion of On your feet (Figure 2). With a marginal significant difference between the improvement of the spastic quadriplegic and diplegic children (2.00% vs. 2.77%, p=0.0575) and no significant difference between above and below age ten (1.98% vs. 2.64%, p=0.1264). Children above age ten started (M0) with a significantly higher mean BMD than the children below age ten (0.41±0.14kN, CI 0.20-0.73 vs. 0.24±0.08kN, CI 0.12-0.42, p<0.0001). At M6 the difference was still apparent (0.44±0.13 kN, CI 0.20-0.69 vs. 0.26±0.09 kN, CI 0.14-0.39, p=0.0001).

The mGMFM ranks showed a significant difference (p<0.0001) in total of 12.67% (Figure 2). mGMFM Sitting ranks showed a significant difference (p=0.0012) in total of 9.78%. Spastic quadriplegic and children below age ten improved significantly (12.41% and 12.4% respectively, p<0.05), whereas spastic diplegic and children above age ten did not improve significantly (4.89% and 6.35% respectively). Crawling and Kneeling ranks showed a significant difference (p<0.0001) in total of 13.75%. Spastic diplegic children did not improve significantly, whereas all other groups improved significantly (>10: 24.26%, <10: 8.65% and spastic quadriplegic: 22.86%, p<0.05). Standing ranks showed a significant difference (p=0.0101) in total of 35.11%. Spastic quadriplegic children improved by 45.29% and the group above age ten by 14.29%, but not significantly. The group below age ten (45.47%) and the spastic diplegic group (29.48%) improved significantly (p<0.05).

Standing, Running and jumping ranks showed a significant difference (p=0.0497) in total of 8.42%, but none of the subdivided groups improved significantly.

Height and weight. Participating children gained height and weight while training and z-scores stayed stable (Table 2).

Discussion

After participating in On your feet for six months the 78 BSCP children improved significantly in BMD, BMC/cm,
muscle mass/cm, angle of verticalisation, maximal force in extension and mGMFM dimensions sitting, crawling and kneeling, standing and walking, running and jumping. Therefore the new physiotherapy concept On your feet seems to be feasible to improve BMD, BMC, muscle force and gross motor function in BSCP children.

The BSCP group included spastic quadriplegic and diplegic children. According to Kulak et al. significantly more children diagnosed spastic diplegia are able to walk than quadriplegic children. Perhaps because of the better use of the upper extremity for walking aids in diplegic children. Therefore it is to be assumed that diplegic children perform on a better mobility level, have better muscle force and higher BMD. Contradicting this, after participating in the concept On your feet there was no significant difference in per cent change between these groups and no significant difference in M0 parameters; only a significantly lower angle of verticalisation was observed in the quadriplegic group, which reflects the diminished ability to stand. Children diagnosed with spastic quadriplegia improved significantly in mGMFM dimensions sitting and crawling and kneeling and diplegic children in standing. Which also reflects the better ability to stand in children diagnosed with spastic diplegia. In dimension walking, running and jumping neither of the groups improved significantly. Inconsistencies in
mGMFM and TT results could most likely be due to the modification of the GMFM, varying sample sizes and large standard deviations. Neither mGMFM nor the original GMFM were tested for intra-rater reliability among the physiotherapists at the setting. Apparently On your feet improves gross motor function in children diagnosed spastic quadriplegia predominantly in lower GMFM dimensions and in spastic diplegic children in standing. This supports Kulak et al.’s results and clinical experience that quadriplegic children perform better on lower mobility levels. Accordingly children diagnosed spastic diplegia should be supported on higher gross motor function levels than quadriplegic children. However this can only act as a suggestion and should always be considered under the individual goal setting.

According to the motor development curves created by Rosenbaum et al. main motor development in CP children is to be observed below age ten and then remains more or less static. On the basis of these curves the BSCP group in this study was divided in two groups above and below age ten. BMD, BMC and muscle mass are obviously age dependent parameters due to adolescence. Although BMC and muscle mass were adjusted for height, a difference in baseline values was apparent which leads to the assumption of other developmental factors influencing results than height. No significant difference in per cent change could be observed for muscle mass/cm and BMD, but a strong difference for BMC/cm. BMD is calculated by dividing BMC by bone area. The lack of difference between groups for BMD is reasonable because the bone area increases with development and qualifies BMD results. Therefore BMC seems to be the more reliable value in developing systems.

Highly significant differences for groups above and below age ten could be observed in maximal force in extension in M0 and M6. Children above age ten showed almost twice as much ground reaction force than below age ten in M0 (0.41±0.14 vs. 0.24±0.08) and M6 (0.44±0.13 vs. 0.26±0.09). Unfortunately we had no reference data for these children, so interpretation of these results is difficult, but most likely due to adolescence.

In mGMFM the group above age ten improved significantly in crawling and kneeling and children below age ten in sitting, crawling and kneeling and standing; supporting Rosenbaum et al. in stating less motor development above age ten for CP children. This observation is not unique in CP children, but is evident in normal development. Again, more reliable data would have been provided by using the full, validated original version of the GMFM. When using the full GMFM data also should have been correlated to the now existing development curves. As well as BMD data should have been correlated to references like Wilmshurst et al.

As in other studies on children the affiliation of adolescence has an high impact on outcomes. Nevertheless, this study provides solid data of a large, heterogeneous sample. On your feet is a routine procedure in the German health system, therefore it is not a controlled trial and flawed by circumstances resulting from this setting: no control group, varying sample sizes due to missing data, large variability in execution of assessments due to a variety of therapists and no control whether the children trained at home or not. The power calculation was only conducted for BMD (whole body without head). Therefore ideal sample sizes for outcomes like GMFM, muscle force etc. are unfortunately unknown. The sustainability of the positive effects has not been investigated in this study but is in progress.

According to the mechanostat theory intensive exercise evolves strain signals from bone tissue during loading and therefore enhances bone density. Henderson et al. found that BMD of the distal femur is reduced (z-score lower than -2) in 77% (95% CI 65.0-87.1) of CP children. Unfortunately we did not calculate BMD z-scores in our results in order to compare data. We calculated “whole body without head” BMD data which we are not able to compare with lumbar spine, femur or hip BMD data of other studies. We used a DXA measurement, which we are not able to compare to quantitative computed tomography (QCT) data of other studies. Unfortunately there is high inconsistency in BMD measurements so comparisons are very difficult. To our knowledge no studies are available on evolved strain signals from bone tissue during WBV, RT, WBV or “physiotherapy”. Comparisons are very difficult due to high inconsistency and more good quality research is needed. Recently a pilot study by Rauch (2009) showed a positive effect of a twice-a-week, six months WBV training on GMFM (dimension D), lumbar spine BMD and bone mass in four CP children. This supports Ward et al. and Semler et al. that WBV is beneficial to improve BMD in CP children. However, to our knowledge, there is no proof of how isolated RT, BWSTT, “physiotherapy” or pool therapy would have influenced BMD.

RT has been shown to improve muscle strength, muscle volume and induce functional improvements. However the effect of RT in CP children is still controversially discussed. BWSTT seems to improve walking parameters in CP children, however again, more research is needed to confirm the effect.

We decided to combine the available, though weak evidence for the best functional improvement of the CP child in order to benefit from an amelioration in all International Classification of Functioning, Disability and Health (ICF) levels (body structure and function and activity and participation) by restored bone mass, increased muscle force, better motor function, and better participation. Therefore, the effect of On your feet can not be assigned to a single intervention but is the result of the combination of the different interventions orchestrated in the concept.

A general problem is the definition of “physiotherapy”. In On your feet physiotherapy sessions set a stage for implementing results of RT, BWSTT and WBV in all day living activities. The methods for achieving the individual goals are variable and dependent on the physiotherapist’s clinical decision which causes problems in reproducibility. RT is reasonably precise in terms of definition of action; however repetitions, intensities and frequencies depend on the individual capability of the child and therefore can not be standardised. In BWSTT again parameters can not be generalised for the whole sample. On your feet should treat and evaluate children on all ICF levels (body functions and structures,
activities, and participation). The concept was successful in treating and assessing on ICF body functions and structures and activities levels. Participation level was influenced in therapy but unfortunately not assessed and therefore could not be presented as a result. This will be solved in future by evaluating participation within the concept On your feet27. However a trend towards an ICF based treatment concept is clearly visible.

With this study a concept like On your feet is described to our knowledge for the first time. In the field of paediatric physiotherapy there is very little evidence for successful treatment methods for children with disabilities. To our knowledge WBV is not part of the standard care for CP rehabilitation in Germany nor other countries and should perhaps be considered to be included in the rehabilitative procedure. With this study a stage is set for evidence based, effective treatment for children with BSCP. This study serves as a basis for future research on evidence based paediatric physiotherapy taking into account developmental implications. Further studies need to be conducted on the effectiveness of the single interventions like RT and BWSTT on BMD and functional abilities of the CP child. BMD measurements should be standardised for comparability.

Conclusion

On your feet is effective for children with BSCP as a physical fitness intervention as Fowler et al. arrogate27. There was no difference in improvement between children diagnosed spastic quadriplegia or diplegia; however the results show a trend that diplegic children should be supported in higher gross motor function levels and quadriplegic children in lower according to their predicted motor development. BMC/cm, muscle mass and gross motor function can be influenced better below age ten. However, it has to be taken into account that the motor development of children below age ten is in progress and that these children might improve naturally. Results showed less motor development above age ten than below age ten which is consistent with the motor development curves for CP by Rosenbaum et al.30. The new physiotherapy concept On your feet had a significant effect on all dimensions investigated and seems to be feasible to improve BMC, muscle force and gross motor function in children with BSCP.

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References