

Efficacy of whole-body vibration on balance and motor function in children with osteogenesis imperfecta: A randomized controlled trial

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ABSTRACT

The current study aimed to examine the influence of whole-body vibration (WBV) on balance and motor function in children with osteogenesis imperfecta (OI). This randomized controlled trial (single-blinded) included forty children with OI, aged 8 to 13 years. Two groups with equal numbers were randomly selected; the control group received 40 min. standard program of physical therapy while the WBV group received the identical program as the control group for 20 min. plus 20 min. of WBV training. The program was carried out for all children twice weekly for three successive months. Biodex Balance System was utilized to evaluate the dynamic balance by measuring the stability indices and Gross Motor Function Measure was employed to evaluate the motor function for two groups, pre and post intervention. Following intervention, results showed statistically significant improvements in all evaluated outcomes of both groups, with the favor of WBV group ($p < 0.05$).

Incorporating WBV training into a standard program of physical therapy has a greater effect on dynamic balance and motor function than a standard program of physical therapy only among children with OI.

KEYWORDS

Balance; Motor Function; Osteogenesis Imperfecta; Whole-Body Vibration

1. INTRODUCTION

Osteogenesis Imperfecta (OI) is an infrequent hereditary condition that affects the connective tissues and bones. It is manifested by bone fragility that are prone to fracture, blue sclera, dentinogenesis imperfecta, and other skeletal deformities (Mueller et al., 2018). Its incidence is approximately 1 in 15000 to 20000 worldwide. It was brought about by mutations in the type I collagen-producing genes, which is a basic protein element of bone and connective tissue providing their strength and structure (Forlino & Marini 2000). There are several types of OI, each caused by a different genetic mutation. The severity of OI can vary greatly from person to person, ranging from mild to severe forms. These children are characterized by reduced level of activity and present with delayed motor development due to immobilization periods after fractures that lead to loss of muscle bulk which is followed by reduction in bone mass and increase risk of fractures and prolonged immobility (Mc Donald et al., 2023). Treatment options for OI include physical therapy, medications, surgery, and lifestyle modifications (Rauch & Schoenau 2001).

Whole-body vibration (WBV) is becoming a more popular way as a substitute method of enhancing neuromuscular function in the field of physical therapy (Yang et al., 2015). It has been demonstrated to have favorable effects on every organ in the human body, including the reduction of oxygen intake, enhancement of strength, balance, functional walking, and life quality, as well as an increase in blood circulation, mineral density of bone, heart and lung functions, and blood vessels function (Alvarez-Barbosa et al., 2020; Mohammad Rahimi et al., 2020; Van Dijk & Sillence 2014).

Generally, there is a scarcity of research on optimal or best management intervention exercise strategies for children with OI. Even though researchers have discussed the influence of WBV training on bone mass in individuals with OI, the underlying mechanism for this conclusion is still unclear. Various studies utilized different guidelines and reported that WBV provides minimal impact on enhancing motor function or mobility in OI population (Högler et al., 2017), but it is still a desirable method to promote bone strength that can affect balance control in people with OI (Kang et

al., 2014). Therefore, the trial's aim was to determine the effectiveness of WBV on balance, and motor function in Osteogenesis Imperfecta children. So, we hypothesized that WBV could provide an impact on balance and motor function in Osteogenesis Imperfecta children.

2. METHODS

2.1. Study design and participants

This randomized controlled trial (single-blinded) was conducted at the Faculty of Physical Therapy outpatient clinic, Benha University, and the Hospitals of the General Authority for Health Insurance in Zagazig between April 2021 and December 2023. Forty children with osteogenesis imperfecta (OI) participated in the study, with ages ranging from eight to thirteen years. The diagnosis of children with OI was dependent on meticulous clinical evaluation by a pediatrician. Following screening, children of both genders with OI (type I) were picked according to these criteria; (i) they were between the ages of eight and thirteen, (ii) they had normal BMI, (iii) their score of gross motor function (GMFM-88) were ≥ 45 , (iv) they had stable medical and psychological status. On the other hand, we excluded; (i) children who walk with any assisted device, (ii) children with auditory or visual problems, (iii) children who have recently broken their upper limb and are still in plaster, (iv) children who have broken their lower limbs within the last three months, and (v) children who are unable to follow verbal commands or instructions. Each child's parents provided informed consent. The procedures that followed were authorized by the Cairo University Faculty of Physical Therapy's Institutional Ethical Committee under the protocol No. P.T. REC/012/002574.

2.2. Study randomization

A total of forty-five boys with OI assessed for eligibility. Three children failed to convene the study criteria, and two children refused to enroll. Forty children with OI were divided into two equal-sized groups at random. The control group underwent to a standard program of physical therapy, while WBV group got the similar standard program of physical therapy in addition to WBV. Reducing selection bias by using random allocation software (Saghaei 2004). A diagram of the study's randomization and retention of children is shown in Figure 1.

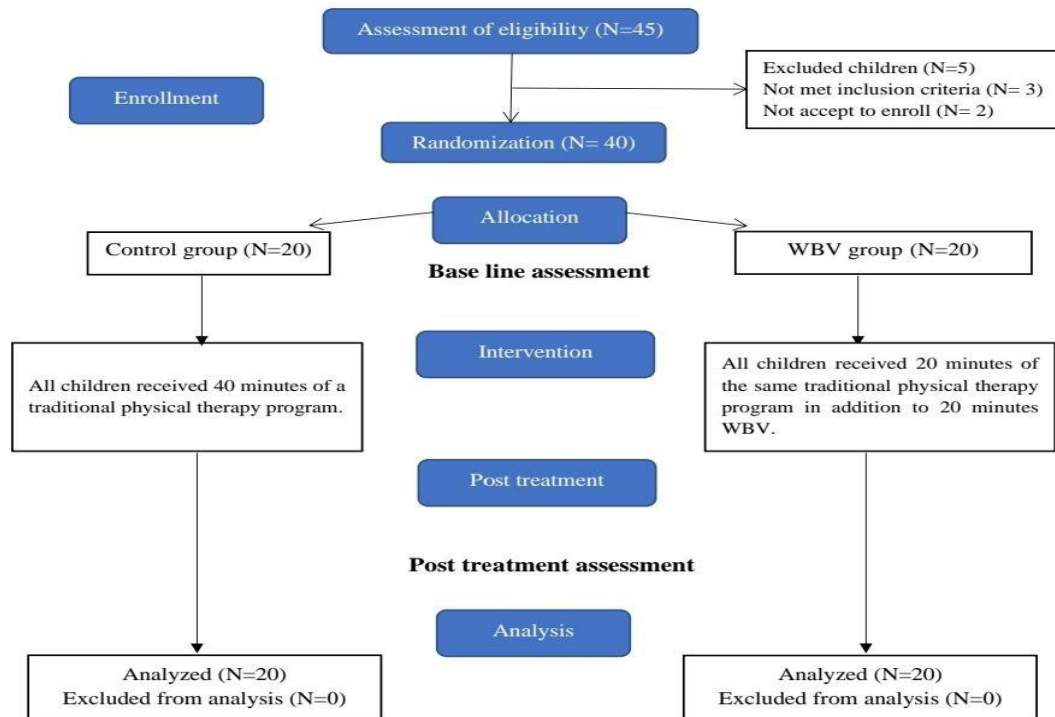


Fig. (1): Flowchart of Children Randomization

2.3. Sample size

Sample size calculation using version 3.1 program of G power, Heinrich-Heine-University, Düsseldorf, Germany according to F tests (MANOVA: Specific interactions and effects), Type I error (α) = 0.05, power (1- α error probability) = 0.80, Pillai V = 0.420, and effect size f^2 (V) = 0.1627 with 2 independent groups comparing the 4 main variables outcomes. A minimum sample size of twenty children in each group was appropriate.

2.4. Measurement procedures

2.4.1. Dynamic balance

The Biodex Balance System (BBS) is an essential therapy device for evaluating or treating people with balance problems (Abo-zaid et al., 2020). The Biodex Balance System SDTM (Model 950-441) is a trustworthy and valid apparatus employed for assessing the child's capacity to preserve the unstable tilting platform for both static and dynamic balance (Shousha et al., 2021). Previous pilot research stated that a dynamic balance test was conducted on stability level eight. The assessment was done pre and post three successive months of intervention.

Before the procedures of evaluation, all children received an explanatory session to help them understand the various test steps. Every child in the two groups was instructed to take a two-leg stance while standing in the middle of the "locked" platform with bare feet. The weight, height, and age of the child were presented to the apparatus. Then the child was instructed to center himself on an unsteady platform by changing his feet placement until it became simple to maintain the platform's center, or the cursor, on the screen panel while maintaining a straight posture. The child was instructed to keep his feet in that posture until the platform stabilized after centering had been reached and the cursor was centered within the display target. The next step was to record the platform's heel coordinates and foot angles. The platform was labelled with degree angles ranging from 0° to 45°. The coordinate of platform X was indicated with numerals, while the coordinate platform Y was indicated with letters. The foot angle of each child was ascertained by locating a parallel line on the platform that connected to the center line of the foot, and his heel coordinates were calculated starting at the heel's posterior center. The test started once heels and foot angles were entered into the BBS. Each child was asked to stand with both arms at his sides without gripping the handrails, face the visually feedback screen squarely in front of him, and try to maintain the pointer in the bullseye at the middle of the screen as the platform moved into an unstable state. For every child, the process was performed three times for a total of thirty seconds, and the mean was computed. Every test run concluded with the acquisition of a printout report. The antero-posterior stability index (APSI) represents the child's capacity in controlling balance from front to rear, the medio-lateral stability index (MLSI), represents a child's capacity in controlling balance from side to side, and the overall stability index (OASI), represents a child's capacity in controlling balance in all directions. High scores indicate that the child struggled with balance (Shousha et al., 2021).

2.4.2. Motor function

Gross Motor Function Measure (GMFM) is a semi-quantitative, validated, reliable, and standardized evaluated tool for motor function that was used for children with OI (Lundkvist et al., 2009; Hoyer-Kuhn et al., 2014). We assess the sector standing, walking, running and jumping in the current study. The scoring key is intended to serve as a broad reference. For every score, most of the sub-items have descriptors. It is essential that each item be scored according to the manual's instructions (scoring key 0 = does not initiate, 1 = initiates, 2 = partially completes, 3 = completes) (Ruck-Gibis et al., (2001). Each child was evaluated for motor function pre and post three successive months of intervention.

2.5. Interventions

The control group received 40 minutes of standard program of physical therapy, twice a week for 3 months according to Powell et al., 2023; Marr et. al. 2017. The program was established based on neurodevelopmental basis, and including sit-to-stand, single-legged, sit-up, heel-rise, strengthening exercises for abdominal and spinal muscles, weight shifting exercises from stride standing position, standing on balance board, and balancing exercises in the form of righting and equilibrium from standing position.

On the other hand, children in WBV group received 20 minutes of WBV plus 20 minutes of the same standard program of physical therapy. A motorized board on the WBV device generates alternating vertical sinusoidal (rotational) vibrations from side to side around a fulcrum located in the plate's center. The child could choose the frequency of vibration while standing on the board with two feet. The farther the feet are from the vibrating board's center line, the greater the peak-to-peak movement to which they are subjected. Children were instructed to stand straight on the WBV device's platform (Bestpro Gymform Vibro Max 20 speed), with bending knees (10 degrees, semi-squat position). For the first two weeks, amplitude 1 mm was utilized; after that, the amplitude was increased up to 2 mm, then it was raised up to 3 mm, if individually possible, the frequencies employed were between 20 and 25 Hz. Children were also instructed to rotate their trunks while changing their weight from side to side. Each WBV training session consisted of two sets, each set consisted of 3 min of WBV for two positions, 2 min breaks between every position, and 4 min breaks between sets, two times per week for 3 months (Högler et al., 2017).

2.6. Statistical analysis

Descriptive statistics using version 21 in SPSS software for analysis of data were utilized to determine each variable's mean and standard deviation. Comparing both group's number and percentage of sex distribution using Chi-square test. Comparing all measured variables within every group before and after treatment using Paired t-test. Comparing all measured variables among both groups pre and post treatment using an unpaired t-test. P-value ≤ 0.05 was considered significant. The relevant test variables were compared at various tested groups and measuring times using multivariate analysis of variance (2 x 2 mixed design MANOVA). Every statistical analysis was significant when ($p \leq 0.05$).

3. RESULTS

Children`s demographic data is demonstrated in Table 1. Between the control and WBV groups, no statistically significant variations were found in age, weight, height, as well as BMI ($p > 0.05$).

Table 1. Children demographic data among both groups

Variables	Control Group (n=20)	WBV Group (n=20)	<i>p</i>
Age (year)	10.05±1.432	9.85±1.182	0.6328
Weight (kg)	35.1±4.734	33.3± 4.105	0.2067
Height (cm)	129.8±8.069	128.6±7.81	0.6355
BMI (kg/m ²)	20.76±1.342	20.12±1.589	0.1739

Note. BMI: Body Mass Index, SD: Standard Deviation, P: Probability, Non-significant = $P > 0.05$.

Mixed design MANOVA 2 x 2 for measurement of outcomes showed a statistically non-significant consequence in tested groups (p -values =0.085). On the other hand, there was a statistically substantial consequence in measuring time and interaction effect (p -values =0.0001, and 0.01) respectively, as presented in Table 2.

Table 2. Main effects of independent variables by MANOVA test for all dependent measured outcomes

Source of variation	Wilk's Lambada	Partial Eta ² (η^2)	F	<i>p</i>
Tested groups effect	0.797	0.203	2.234	0.085
Measuring period effect	0.298	0.702	20.588	0.0001*
Interaction effect	0.690	0.373	3.10	0.01*

Note. *p*: probability value, * Significant = P -value < 0.05

Within each group, the statistical analysis revealed a significant reduction in all stability indices ($P < 0.05$) and a significant enhancement in GMFM score ($p < 0.05$) post-intervention compared to pre-intervention inside the control and WBV groups (Table 3). Between groups, pre-intervention there was no statistically notable difference in all assessed outcomes ($P > 0.05$). Post-intervention, there was a statistically considerable variation among two groups in all assessed outcomes in favor of WBV group (p -value < 0.05) as presented in Table 3.

Table 3. The design MANOVA test for all dependent measured outcomes within and between groups

Outcomes	Intervention	Groups (Mean \pm SD)		F	p	
		Control group (n=20)	WBV group (n=20)			
Stability Indices	OSI	Pre-intervention	5.215 \pm 0.737	5.055 \pm 1.061	0.333	0.571
		Post-intervention	4.72 \pm 0.705	4.00 \pm 0.748	10.319	0.005
		F-value	10.831	12.305		
		P-value	0.005	0.003		
	APSI	Pre-intervention	4.34 \pm 0.901	4.09 \pm 0.885	0.783	0.387
		Post-intervention	3.9 \pm 0.803	3.28 \pm 0.626	7.393	0.014
		F-value	15.403	12.495		
		P-value	0.009	0.007		
	MLSI	Pre-intervention	3.55 \pm 0.611	3.21 \pm 0.637	3.172	0.091
		Post-intervention	3.22 \pm 0.499	2.58 \pm 0.679	10.626	0.004
		F-value	12.459	10.598		
		P-value	0.021	0.0018		
Motor Function	GMFM	Pre-intervention	54.75 \pm 3.16	54.5 \pm 3.395	0.068	0.797
		Post-intervention	56.3 \pm 2.45	59.05 \pm 2.31	16.167	0.001
		F-value	10.516	57.133		
		P-value	0.008	0.0001		

Note. SD: Standard Deviation, p: Probability Value, * Significant = P-value < 0.05

4. DISCUSSION

Reduced stability of the musculoskeletal system, muscle hypotonia, defective bone modelling, and resorption are the main characteristics in children with OI resulting in a decrease of bone mechanical properties (Rauch & Glorieux 2004). So, our trial aimed to evaluate the efficiency of adding WBV to standard program of physical therapy on balance and motor function in children with OI. The findings of our trial revealed a significant reduction in all stability indices as well as a significant enhancement in GMFM within two groups in favor of the WBV group.

Decreasing balance and motor function in each participant in the pre-intervention was directly related to OI when compared to typically developing (TD) individuals. This fact was supported by Pouliot-Laforte et al. (2017) who reported that a proprioceptive deficit could justify poor postural balance in type I children with OI, and Caudill et al. 2010 who found that restriction in every-day tasks might be secondary to OI which affects dynamic postural stability in individuals with OI. However, Takken et al. (2004); Semler et al. (2008) in their studies on children with osteogenesis imperfecta documented a considerable reduction in muscle strength that in turn had a strong adverse impact on motor performance and standard of living in those children.

Our findings revealed that the control group's post-intervention mean values of all assessed variables (stability indices and motor function) showed a statistically significant improvement. This might be due to the efficiency of the standard program of physical therapy and can be explained by Marr et al. (2017), who declared that abdominal and trunk muscles strength is crucial exercise to enhance controlling posture and balance in OI children. Van Brussel et al. 2008 demonstrated that twelve weeks of physical training program on thirty-four children OI (type I) improved both muscular force and aerobic capacity and reduced fatigue levels in a safe and effective manner. This in turn reflects an improvement in gross motor function.

Besides, our results represented a significant reduction in post-intervention mean values of every stability index in the WBV group. It might be justified by Rittweger et al. (2003); Semler et al. (2008) who concluded that WBV could increase mineral density of bone and muscle strength in children with OI, which makes an enhancement in postural balance.

Additionally, Melnyk et al. (2008); Pollock et al. (2010) confirmed that vibration-induced alterations in joint stiffness make an enhancement in ankle and knee joint stabilization, stimulates neuromuscular activation which leads to co-activation of lower extremity extensor and flexor muscles, and consequently could improve the postural stability.

Furthermore, our results indicated that there was a substantial increase of GMFM scores following intervention contrasted with pre-intervention in the WBV group. These findings correspond with Hoyer-Kuhn et al., 2014 who performed a 12-month of WBV training program for physically limited children with OI, and recorded an improvement in GMFM score that reflects a positive effect on the child's mobility.

Contrary to the current findings, Höglér et al. (2017) noted that WBV has no statistically notable changes in motor function after five months of application in twenty-four children with OI. The disagreement may be related to the selected sample which included OI (type IV) with limited mobility.

5. LIMITATIONS

Some restrictions to this study should be considered. First, total number of sessions per week were limited to twice a week in order to prevent children from becoming fatigued owing to muscle weakness. Second, our study was limited to OI children with normal BMI. Further work is needed to investigate the impact of WBV over a lengthy period of time, with an adequate follow-up period to guarantee findings last more than a year.

6. CONCLUSIONS

The results of our investigation revealed that WBV training in conjunction with a standard program of physical therapy has an obvious therapeutic benefit on improving balance and motor function in OI children.

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CONFLICTS OF INTEREST

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