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Walking Ability, Participation, and Quality of Life in Children with Spastic Diplegic Cerebral Palsy: A Path Analysis Study

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Abstract

Objectives

This study aims to design a conceptual model for the effect of various factors on walking ability, participation, and quality of life in children with spastic diplegic cerebral palsy (SDCP) and test it based on field data using path analysis.

Materials & Methods

This cross-sectional study was performed on 181 children with SDCP. The following were used to measure each of the variables: the Modified Ashworth Scale, the Micro Manual Muscle Tester, the Timed Up and Go Test (TUG), the Boyd and Graham test, the goniometer, weight and height, the Gross Motor Function Classification System, the Life Habits Questionnaire, and cerebral palsy Quality of Life Questionnaire for Children. The structural model was tested in Amos 17.

Results

All paths of the proposed model were significant ($P < 0.05$). Among evaluated variables, muscle strength ($B = -0.466$), balance ($B = 0.326$), and spasticity ($B = 0.143$) affected walking ability. Moreover, as an intermediate factor, walking ability affected the subjects' participation ($B = -0.819$) and quality of life ($B = -0.183$).

Conclusion

Muscle strength, balance, and spasticity are the most influential factors in the walking ability of children with SDCP. Furthermore, walking ability and participation are two critical factors in promoting the quality of life of these children.

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Introduction

Cerebral palsy (CP) is the most common cause of motor disability in childhood (1), resulting from a chronic, non-progressive lesion in the developing brain. It is associated with impaired movement, posture, and function (2). According to the International Classification of Functioning, Disability, and Health (ICF), CP is associated with defects in body structure and function (muscle tone, muscle strength, balance, body mass index (BMI), selective movement, and range of motion (ROM)), significant limitations in activities (e.g., motor function), and limitations in social participation and communication (3). Spastic diplegic cerebral palsy (SDCP) is the most common clinical type of CP (4). Previous studies have reported a prevalence of 62.5% for this condition among different types of CP (5).

The main goal of all rehabilitation treatments in this context is to enable children with CP to walk independently (6). One of the most common concerns among these children's parents is whether their child can start walking and when it will take place (7). Meanwhile, therapists find it difficult to answer this question because several factors affect the child's walking ability.

Numerous environmental and individual factors directly or indirectly influence motor performance in children with CP (8). Identifying these factors and discovering their relationship with each other and with walking help researchers and clinical specialists to further consider all these factors in treatment planning. As a result, they can develop interventions that could improve the walking ability of these children by simultaneously acting on different factors.

Although previous studies have examined the correlation between factors such as spasticity (9-12), muscle strength (13-16), balance (17, 18), BMI (19, 20), selective movement (21, 22), and ROM (10, 23, 24) in children with CP, they have often addressed children's walking ability concerning only one or two factors.

If a child with CP does not participate enough, certain complications, such as shortness of limbs, deformities, and obesity, will occur (25). Nowadays, the two variables of social participation and quality of life have become appropriate and acceptable criteria for evaluating the success of therapeutic and rehabilitation interventions in many chronic disorders (26, 27), including developmental disorders such as CP. Therefore, enhancing the ability to walk in children with CP to effectively raise their participation and, ultimately, their quality of life is expected (28).

Most models proposed for CP are theoretical (8, 29-31), and only three models have been tested (14, 23, 32). Moreover, previous studies have dealt with a limited number of variables and have included children with any degree of involvement. This study aims to design a theoretical or conceptual model for the effect of different factors such as ROM, muscle strength, spasticity, selective movement, balance, and BMI on walking ability, participation, and quality of life in children with SDCP and evaluate the relationship between these factors and test the proposed model based on field data and using path analysis.

Materials & Methods

The draft of the initial conceptual model was prepared by browsing scholarly databases and reviewing scientific texts and expert opinions. The relationship between children's walking ability and various potentially influential factors was evaluated. Worth mentioning that only those relationships for which good sources were found were incorporated into the model.

Three pediatric specialists, three experts with Master's degrees, and two with a Ph.D. in occupational therapy were selected by purposive sampling and an interview subsequently. These experts have been involved in rehabilitation programs for children with CP for more than five years. The experts were first given the initial model to prepare for the interview. After a while, in a face-to-face meeting, their opinions were recorded. Finally, in another meeting, the research team made decisions regarding the need to change the model.

To design the initial model according to the model proposed by the World Health Organization, the authors studied muscle weakness, balance, ROM, BMI, selective movement, and spasticity as factors of body function and structure. On the other hand, walking ability (based on the gross motor function classification system) was analyzed as part of activities, and social participation was regarded as a participation component. Eventually, these variables' impact on quality of life was assessed.

This cross-sectional path analysis was carried out on children aged six to 12 diagnosed with SDCP, and the diagnosis had been stated in the children's medical records. The subjects were chosen using convenience sampling from several rehabilitation centers in Tehran, Mazandaran, and Alborz provinces. The inclusion criteria for these children were passing at least six months after surgery or possible Botox injection, and the exclusion criteria were disorders such as hydrocephalus, blindness, and deafness.

Necessary explanations were given to children's parents about the objectives and methods of the study. After obtaining parents' or legal guardians' written informed consent, the authors began measuring the variables proposed in the initial model. The ethical committee of the University of Social Welfare and Rehabilitation Sciences has approved the study protocol.

An occupational therapist collected data with ten years of experience in clinical and educational activities in the area of CP. Importantly, parents answered questions on the two variables of participation and quality of life.

The study was approved by the Ethics Committee of Mazandaran University of Medical Sciences.

Measurement tools and methods

Muscle strength was measured using the Micro Manual Muscle Tester (MMMT). Seven muscle groups (hip extensor, hip flexor, hip abductor, knee flexor, knee extensor, ankle plantar flexor, and ankle dorsiflexor) of the lower extremities were examined. The child was asked to perform specific movements strongly, and the number indicated by the MMMT was considered the subject's muscle strength (33).

The Timed Up and Go test (TUG) measured balance. Thus, the child sitting in a chair was asked to stand up, move three meters forward, then turn and sit back. The time the child spent doing these tasks was recorded (34).

Passive ROM in the pelvis, knee, and ankle flexor and extensor muscle groups in both lower extremities were measured by a goniometer using the Kendall technique at the sagittal level (35). The authors used the full joint angles to evaluate the ROM in both lower extremities to determine the disorder's extent.

BMI was calculated by dividing weight by the square of height. A digital scale was used to measure weight, and a tape measure was used to determine the height. First, several stretching movements were performed to make the joints flexible. Then the child laid down with his/her back on the ground, and the distance between the highest point on the child's head and the end of the heel was measured using the tape.

The authors used the Boyd and Graham test (35) to measure selective movement, which evaluates selective voluntary motor control in the ankle joint when the subject performs dorsiflexion. This test has four scores: zero indicates no independent movement, and four means the child can independently raise the ankle without moving the knee.

The modified Ashworth scale (36) was used to assess spasticity in the flexors and extensors of the pelvis, knees, ankles, and hip abductors at a moderate velocity in both lower extremities. In this scale, zero represents the absence of spasticity, 1 represents a slight increase in tone manifested by a catch and release, 1^+ presents a slight increase in tone manifested by a catch flowing by minimal resistance, 2 represents a more marked increase in muscle tone but affected part can easily be moved, 3 shows a considerable increase in muscle tone, and 4 indicates rigidity. In assessing spasticity in both limbs, the total spasticity score was used to determine the magnitude of the disorder.

The ability to walk was assessed by the gross motor function classification system (GMFCS) (37). It has five levels of classification. Level I applies to children who can do all their peers' activities, though with limited speed, balance, and coordination. Level II applies to children with limitations in walking long distances outside the house and using the railing to go up and down the stairs. In Level III, children need physical assistance to walk around the house and a wheeled device to go over long distances. In Level IV, children have limited mobility and move using a manual or electric wheelchair. In Level V (38), children have more severe limitations in keeping antigravity head and trunk postures and rely on someone else's help or more substantial support technologies, but they can move on their own if they learn to use an electric wheelchair.

To assess participation, the authors used a short form of the Life Habits questionnaire (LIFE-H) (39) to measure social participation in people with disabilities. In this questionnaire, completed by children's parents, LIFE-H is classified into two general categories: daily activities and social roles. The LIFE-H is scored from 0 to 9, which suggests the level of difficulty and the type of assistance; zero indicates a high level of limitation, and nine indicates desirable participation (40).

The subjects' quality of life was assessed using the CP Quality of Life Questionnaire for Children (CP QOL-Child), which parents completed. The questions are answered using a 9-point Likert scale (41).

All participating children cooperated well. However, in cases where the child was tired or did not have the necessary cooperation, the examiners re-evaluated them after ten minutes.

SPSS version 16 was used to analyze data, and values below 0.05 were considered statistically significant. The Pearson correlation test was used to evaluate any correlation between variables. The initial (conceptual) model considered the variables with significant correlation. Goodness-of-fit statistics were used to assess the fitness of the conceptual model. The following conditions have to be met to fit the model perfectly: P> 0.05; the ratio of Chi-square to the degree of freedom (Chi-square / DF) should be 1 to 3; the root mean square error of approximation (RMSEA) should be <0.10; the goodness of fit index (GFI) should be > 0.9; the adjusted goodness of fit index (AGFI) should be > 0.8; and the standard fit index (NFI), comparative fit index (CFI), and the incremental fit index (IFI) should be > 0.9 (42). Structural equations modeling (SEM) was performed using Amos version 17.

Results

This research was conducted on 181 children with SDCP (109 boys and seventy-two girls) with a mean age of 135.81±25.97 months. Based on the results, forty-six (25.4%) children were at level one, fifty-three (29.3%) were at level two, fifty-seven (31.5%) were at level three, and twenty-five (13.8%) were at level four according to GMFCS (Table 1).

After reviewing scientific texts and expert opinions, the initial conceptual model was designed in Amos software (Figure 1). Pearson coefficient between variables showed that the relationships proposed for the variables in the initial model were significant (p <0.05) (Table 2).

As shown in Table 3, path analysis confirmed that all proposed paths between variables were acceptable (P <0.05). However, even though the goodness of fit index (GFI = .904) and incremental fit index (IFI > 0.9) confirmed the fitness of the model, the Chi-square (x2 = 100.81, P <0.001) and the ratio of Chi-square to the degree of freedom (CMIN / DF = 6.30) indicated that the conceptual model was not fit.

Due to the lack of fitness in the initial model, many modified relationships and paths were incorporated into the initial conceptual model based on scientific texts. Accordingly, new paths between mobility and participation, quality of life, and muscle strength, as well as BMI and participation, were added to the initial model (Figure 2).

As can be seen in Table 4, all one-way and two-way paths were acceptable in the modified model (p <0.05). Furthermore, the multiple regression square (R^2), which indicates how much variation of a dependent variable is explained by the independent variable(s), was 0.94 for quality of life, 0.89 for mobility, and 0.63 for participation.

As given in Table 5, the modified model showed an excellent fit in terms of the absolute fit indices, including the Chi-square value (X^2 = 33.863, P = .001), the ratio of Chi-square to the degree of freedom (X^2/DF = 2.605), the root mean square error of approximation (RMSEA = .094), the goodness of fit index (GFI = 963), and the adjusted goodness of fit index (AGFI = .871), as well as Normal Fit Index (NFI = .984), Comparative Fit Index (CFI = .990), and Incremental Fit Index (IFI = .990).

Table1. **Characteristics** of participants

Characteristic	Number	Percentage
Gender		
Girl	72	39.8
Boy	109	60.2
GMFCS level		
I	46	25.4
II	53	29.2
III	57	31.4
IV	25	13.8

Table 2. Inter-correlation of variables

	Balance	BMI	ROM	Strength	Spasticity	Selective movement	GMFCS	Participation	Quality of life
Balance		.155*	-.800**	-.746**	.733**	-.837**	.837**		
BMI			-.213**	-.60*	278**.	-.194**	.209**		
ROM				.810**	-.755**	.875**	-.811**		
Strength					-.755**	.875**	-.888**		
Spasticity		.				-.781**	.817**		
Selective movement							-.879**		
GMFCS								-.774**	-.804**
Participation									.867**
Quality of life									

** $p < 0.01$.
** $p < 0.05$.

Table 3. Path coefficients of the conceptual model

Path			B^a	Standard error	Critical ratio	P
POW.LOWER	<---	SPAS.LOWER	-.755	.243	-15.463	***
BALANCE	<---	POW.LOWER	-.447	.178	-6.405	***
BALANCE	<---	SPAS.LOWER	.396	.888	5.670	***
BMI	<---	POW.LOWER	.349	.038	3.294	***
BMI	<---	SPAS.LOWER	.542	.188	5.110	***
SELE.MOV.AN	<---	BALANCE	-.402	.001	-9.464	***
P.ROM.LOWER	<---	BALANCE	-.423	.053	-7.752	***
SELE.MOV.AN	<---	BMI	-.120	.007	-4.228	***
P.ROM.LOWER	<---	BMI	-.142	.251	-3.899	***
P.ROM.LOWER	<---	POW.LOWER	.484	.134	8.908	***
SELE.MOV.AN	<---	POW.LOWER	.566	.004	13.381	***
GMFCS	<---	BALANCE	.315	.002	6.155	***
GMFCS	<---	BMI	-.101	.007	-3.552	***
GMFCS	<---	P.ROM.LOWER	.053	.002	1.009	.048
GMFCS	<---	POW.LOWER	-.508	.005	-8.613	***
GMFCS	<---	SPAS.LOWER	.159	.019	3.514	***
GMFCS	<---	SELE.MOV.AN	-.071	.067	-1.047	.049
PARTICIPATION	<---	GMFCS	-.775	5.495	-16.405	***
Q.O.L	<---	PARTICIPATION	.860	.024	30.611	***
Q.O.L	<---	GMFCS	-.138	2.804	-4.923	***

*** $p<0.001$, B^a Standard regression weight

Table 4. The fit indices of the [initial] conceptual model

IFI	**CFI**	**NFI**	**AGFI**	**GFI**	**RMSEA**	**CMIN/ DF**	p	x^2	**Fit index**
.960	**.960**	**.953**	**.729**	**.904**	**.172**	**6.30**	**0.001**	**100.81**	

Table 5. Path coefficients of the conceptual model

Path			B^a	Standard error	Critical ratio	P	R^2
POW.LOWER	<---	SPAS.LOWER	-.753	.240	-15.542	***	
BALANCE	<---	POW.LOWER	-.446	.178	-6.405	***	
BALANCE	<---	SPAS.LOWER	.396	.888	5.670	***	
BMI	<---	POW.LOWER	.348	.038	3.294	***	
BMI	<---	SPAS.LOWER	.542	.188	5.110	***	
SELE.MOV.AN	<---	BALANCE	-.402	.001	-9.464	***	
P.ROM.LOWER	<---	BALANCE	-.423	.053	-7.752	***	
SELE.MOV.AN	<---	BMI	-.120	.007	-4.228	***	
P.ROM.LOWER	<---	BMI	-.142	.251	-3.899	***	
P.ROM.LOWER	<---	POW.LOWER	.484	.134	8.908	***	
SELE.MOV.AN	<---	POW.LOWER	.566	.004	13.381	***	
GMFCS	<---	BALANCE	.326	.002	6.913	***	
GMFCS	<---	BMI	-.099	.007	-3.516	***	
GMFCS	<---	P.ROM.LOWER	.122	.002	3.475	.048	0.89
GMFCS	<---	POW.LOWER	-.466	.005	-8.440	***	
GMFCS	<---	SPAS.LOWER	.143	.018	3.411	***	
GMFCS	<---	SELE.MOV.AN	-.133	.063	-2.113	.035	
PARTICIPATION	<---	GMFCS	-.819	5.798	-16.486	***	0.63
PARTICIPATION	<---	BMI	-.173	1.302	-3.708	***	
Q.O.L	<---	PARTICIPATION	.853	.023	32.373	***	0.94
Q.O.L	<---	GMFCS	-.183	2.838	-6.447	***	
e8	<-->	e7	9.737	2.112	4.611	***	
e9	<-->	e4	-72.065	16.301	-4.421	***	

*** $p<0.001$, B^a Standard regression weight

Table 6. The fit indices of the modified model

Fit index	x^2	p	**CMIN/DF**	**RMSEA**	**GFI**	**AGFI**	**NFI**	**CFI**	**IFI**
	33.863	**.001**	**2.605**	**.094**	**.963**	**.871**	**.984**	**.990**	**.990**

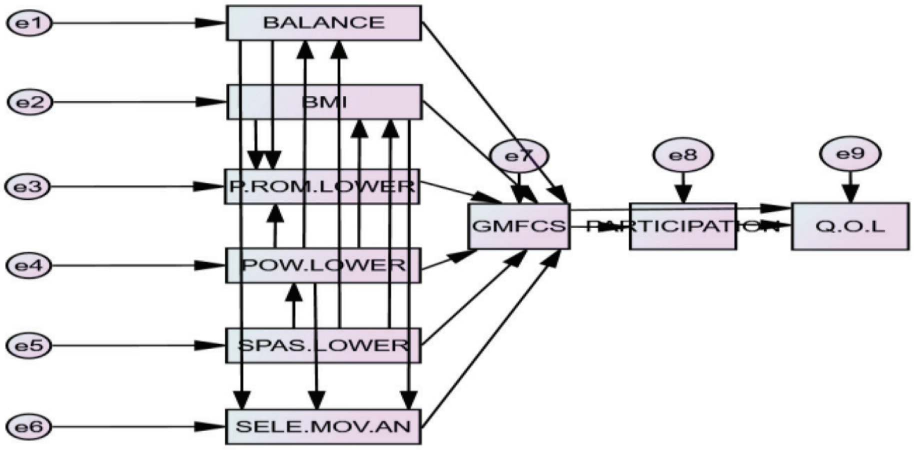

Figure 1. The [proposed] conceptual SEM model

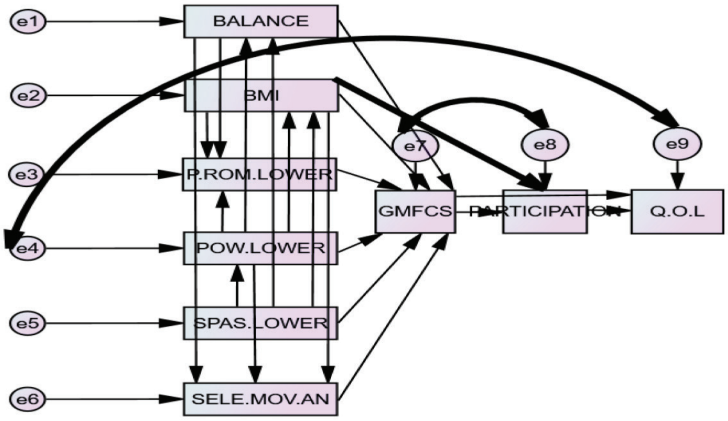

***Figure 2**. The proposed SEM model*

Discussion

The present study introduced an excellent model for the walking ability, participation, and quality of life of SDCP children, which indicates ROM, muscle strength, spasticity, selective movement, balance, and BMI directly or indirectly affect the walking ability in SDCP children.

The present study's findings illustrated that muscle strength was the most important factor directly affecting the walking ability of SDCP children. Additionally, the authors found that the muscle strength variable indirectly influences walking ability through its impact on balance, ROM, and BMI variables. In line with the current study's results, previous studies confirm that muscle strength is adequate for walking ability (43-45). Moreover, a strong relationship between lower limb muscle weakness and activity limitation in CP patients has been reported. Reportedly, a one standard deviation increase in muscle strength leads to a 0.466 standard deviation increase in walking ability. (14, 46). Kim et al. concluded that a one standard deviation increase in muscle strength results in a 0.447 standard deviation increase in the ability of children with CP to stand and walk (14). In another study in 2013, this value was 0.733 (23). The results of the present study suggest that muscle strength directly impacts the quality of life, which is in line with the results reported by Park, who reported a direct relationship between muscle strength and quality of life (32). Seemingly, better muscle strength, as well as enhanced walking ability positively affect other activities of the child, such as sitting and standing without assistance, and even daily life activities, such as going to the bathroom. Consequently, the quality of life of the child improves as well.

Balance was the second important factor directly affecting walking ability. It both directly influenced the walking ability of children and indirectly affected this variable through selective movement and BMI. Previous studies support the relationship between balance and walking ability (47, 48). Balance is affected by the delayed onset of muscle contractions, the reaction time of muscles, and increased abnormal contractions in agonist and antagonist muscles. Children with CP at levels 4 and 5 of the GMFCS have significant problems in balance and postural control, but those identified with levels 1 to 3 of this system show an acceptable functional balance (49). In line with previous reports, this study's results demonstrate that a one standard deviation increase in balance could lead to a 0.326 standard deviation increase in walking ability.

Spasticity was the third important factor that directly affected the walking ability of subjects. It also indirectly affected the walking ability of the children through muscle strength, BMI, and balance. The following spasticity includedmuscle flexibility, the number of sarcomeres decreased, muscle growth impaired, and muscle movement diminished (12).

Spasticity plays a significant role in motor dysfunction in children with CP (50). Kim and Park found that by increasing spasticity by one unit, the ability of children with CP to stand and walk declined by 0.67 and 0.39 (14, 32), respectively. Other studies similarly discussed the link between spasticity and the ability to stand and walk. While some authors have observed an intermediate relationship between motor function and spasticity in the quadriceps and hamstring muscles (51), others have found no link between spasticity and mobility limitation (52). A negative relationship between spasticity and gross motor function is proposed (11). However, according to this study's results, one standard deviation increase in spasticity leads to a 0.143 standard deviation decrease in the ability to walk. The controversy between the results of different studies could be attributed to the difference in the population studied. The authors studied just SDCP cases, while in previous studies, different subtypes of CP were evaluated.

Similar to previous reports, the present study's findings supported that selective movement affects the walking ability of children with SDCP. Muscle strength impairment, spasticity, and decreased ROM are essential factors in reducing or losing voluntary selective movement (21). In a study by Kim and Park (2013), it was observed that a one standard deviation increase in selective movement would cause the standing and walking ability of children with CP to rise by 0.663 (23). The results of several other studies substantiate that the lack of the selective movement negatively affects motor function and walking ability. The better the selective movement of the lower limb joints, the better the motor level of the child with CP (53-55).

As expected, in addition to the direct impact that BMI exerts on the walking ability of CP children, the fitted model revealed its indirect role on the walking ability of these individuals through the variables of ROM and selective movement. Moreover, BMI acted as a mediator, indirectly affecting walking ability through muscle strength and spasticity. This variable had a direct impact on children's participation as well. Edward et al. concluded that children with CP at levels I, II, and III were at risk of obesity and overeating. A review study by Pascoe et al. (2016) concluded that 19.4% of children with CP who could walk were obese and overweight, and children who showed a higher level of mobility had a more normal BMI (20). Colver et al. (56) also confirmed the relationship between BMI and participation.

As expected, this study's results confirmed that reduced ROM leads to the decreased walking ability of SDCP patients. According to the literature, reduced ROM is one of the most prevalent disorders in children with CP. Bell et al. reported that reduced ROM in hip abductors, knee extensors, and dorsiflexors was significantly linked with reduced mobility between the ages of eight and 12 years (57). Anette et al. reported that the knee and ankle ROM plays a crucial role in the ability of children with CP to move independently or use assistive devices like a wheelchair (58).

According to the findings of this study and the final model, the ability to walk affects participation, and participation, in turn, affects the quality of life. Indeed, walking ability significantly influences participation, such that a one standard deviation increase in the ability to walk leads to an 0.81 increase in social participation among children with SDCP. Besides its direct relationship with walking ability, participation is an intermediate factor between the ability to walk and quality of life. In line with previous reports (59-66), the impact of participation on quality of life was significant, such that a one standard deviation increase in participation is associated with an 0.85 standard deviation increase in the quality of life of these children. All the remaining variables in this study's proposed model indirectly affected the subjects' quality of life through the mediating role of walking ability and participation. Park et al. showed that a one standard deviation reduction in the walking ability of children with CP causes a 0.61 standard deviation reduction in their quality of life (32). The relationship between participation and quality of life in people with CP has been confirmed in multiple studies (56, 67, 68).

Overall, this study's results indicated that the final fitted model could explain 89% of the variance in the walking ability of children with SDCP. Therefore, only 11% of the variance in the walking ability is due to other variables that are not predicted in the study's model. On the other hand, the ability to walk and BMI accounted for 63% of the variance of participation.

In addition, walking ability and participation together explained 94% of the variance in quality of life, which indicates the high significance of these two variables in raising the quality of life of children with SDCP.

This study was the first to use path analysis to introduce a model for walking ability, participation, and quality of life in the SDCP subtype. However, this study has some limitations. First, the quality of life questionnaire was completed by parents, which could affect the accuracy of the scores obtained for the quality of life of their children. Meanwhile, due to children's cognitive and communicative limitations, the authors could not use instruments that children themselves might fill out. Another limitation of this study is that the authors did not investigate other factors such as intelligence, history of seizures, motivation, behavioral disorders, history of surgery, history of Botox injections, and environmental considerations that could have a bearing on the walking ability, participation, and quality of life of CP children. Deploying SEM in future studies to address the role of other variables in the walking ability, participation, and quality of life of other subtypes of CP children is recommended.

In Conclusion

Overall, the final fitted model, which proved consistent with the ICT model, demonstrated the direct effects of balance, BMI, passive ROM, selective movement, muscle strength, and spasticity on the walking ability of children with SDCP. In this regard, muscle strength, followed by balance and spasticity exerted the highest impact on children's walking ability. According to the findings of this study, concentrating rehabilitation programs designed for these children on three factors is necessary: muscle strength, balance, and spasticity. Finally, walking ability and participation were two crucial factors in raising the quality of life of these children.

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Authors declare any sources of potential conflicting interest. The authors are grateful for all children and parents who participated in this study.

Author's Contribution

All authors contributed to the design and implementation of the research, to the analysis of the results and to the writing of the manuscript.

Conflict of interest

The authors declare that there is no conflict of interest.

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