

Narrative Review:

A Conceptual Model of Personal and Environmental Factors Contributing to the Mobility in Children With Cerebral Palsy



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ABSTRACT

Context: The present study aimed at developing a conceptual co-relational model of personal and environmental factors affecting mobility in children with CP.

Evidence Acquisition: This perspective article describes a path model for mobility in children with CP. A conceptual framework for the model was supported by the relevant literature, and published papers on the personal and environmental factors affecting mobility in children with CP were reviewed. A literature search was performed using PubMed, SCOPUS and Google Scholar on papers published from 1970 to 2016.

Results: From the initial 228 articles related to personal and environmental factors for children with cerebral palsy, 72 articles met the inclusion criteria. Different factors, affecting the mobility, directly and indirectly, were analyzed based on available evidence.

Conclusions: If contributing factors that lead to changes in basic motor abilities are identified and subjected to manipulation, the identified determinants that can be optimized through interventions might enhance long-term results of mobility.

1. Context

Cerebral Palsy (CP) is the most common motor disorder in childhood and results in abnormal movement (1). Children with CP have a number of impairments that interfere with their motor function, activities, and participation (2). In addition to the disorders in posture and movement, individuals with CP suffer

from limitations in perception, cognition, communication, and other co-morbidities that further affect their activities and participation (3). Enabling independent functional mobility in children with CP is therefore essential for their daily activities, and reduces their dependency on caregivers and the environment. The ultimate goal of CP treatment is in fact to enhance mobility (4). All therapies focused on decreasing impairments in children with CP are based on the as-

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sumption that reducing motor impairment leads to increased activity and participation in everyday life (5). Improved mobility provides life experiences for children with CP that enable them to later engage in education, work/employment, play/leisure, and social roles (2, 6, 7). Moreover, parents of children with CP are almost always concerned about the possibility that their children will walk independently (8, 9). Hence, improving the motor outcomes in children with CP is an important area of research and practice.

For the successful treatment of CP, it is essential to first determine the factors that affect mobility, and the personal and environmental factors have been shown to be the most influential ones in predicting mobility of children with CP (10). Therefore, a more comprehensive understanding of the underlying mechanisms of these factors is essential. When planning interventions and evaluating the outcomes in CP children, it is necessary for the therapists to know how individual, familial, and environmental factors interact with each other to influence mobility. In addition, the knowledge of environmental obstacles helps develop solutions or strategies that enable participation (11), which is the ultimate aim of intervention as per the International Classification of Functioning, Disability and Health (ICF) (12).

Many comprehensive theoretical and empirical models have been developed to guide clinical decision-making regarding the treatment of CP. A conceptual model was designed by Bartlett and Palisano to present the therapists' perceptions regarding the determinants of motor change in children with CP (13). Gannoti et al. presented a path model for evaluating dosing therapy parameters for treating CP (14). Kim et al. examined the causal relationship between spasticity, muscle weakness, gross motor function, and functional outcome in children with CP in a model, and tested the model by path analysis method (15, 16). Chiarello et al. presented a multivariate model of the determinants of change in gross motor ability and engagement in self-care and play in young children with CP (17).

Despite all these efforts, to the best of our knowledge, no study has collectively analyzed the personal and environmental factors that affect mobility outcome in children with CP in a single model. Therefore, to develop such a model as a preliminary step, the purpose of this study was to design a conceptual correlational model of personal and environmental factors affecting mobility of children with CP. The model can be tested statistically in field research using the path analysis method.

2. Evidence Acquisition

This perspective review article describes a path model for mobility in children with CP. A conceptual framework for the model was supported by the relevant literature. We reviewed published papers on personal and environmental factors affecting mobility in children with CP. A literature search was performed using PubMed, SCOPUS and Google Scholar on papers published from 1970 to 2016.

Our search was based on the following keywords: cerebral palsy, motor outcomes, prognostic model, theoretical models, path model, structural equation modeling, motor development, causal factors, prediction of functional outcome, spasticity, strength, income, balance, quality of life, range of motion, gross motor function, gross motor function classification system, participation, activities of daily living, seizure, intelligence, visual impairment, body mass index, poverty, mobility, function, motor, walking, gait, and environmental factors.

The titles and abstracts of articles identified in the initial search were assessed independently by two researchers for the following inclusion criteria: 1. Children with CP as the study population; 2. Presenting the correlation between personal and environmental factors and mobility; and 3. Study of factors which can be manipulated in clinical practice. When the subject or abstract did not clearly indicate whether an article should be included, the full-text was obtained to determine if the inclusion criteria were met. Finally, the direct or indirect relation of a variable to mobility was illustrated.

3. Results

From the initial selected 228 articles related to personal and environmental factors for children with cerebral palsy, 72 articles met the inclusion criteria. The different factors directly and indirectly affecting mobility are shown in Figure 1 and include the degree of spasticity, pain intensity, Body Mass Index (BMI), and degree of weakness and range of motion. Some other factors that are hypothesized to determine walking abilities are balance status, manual abilities, motivation, daily activities, associated disorders (seizures, visual and auditory problems, behavioral problems, cognitive impairments), medical interventions (surgery, Botox injection), socioeconomic parameters (family income, family education, family social support), environmental factors (physical environment, social support, attitude), and quality of life. We have discussed the relationships between each variable with the other variables based on the available evidence.

3.1. Spasticity

Spasticity is one of the most frequently observed impairments in children with CP and has been operationally expressed as “a velocity-dependent increased resistance to passive movement” (18). It is the main motor impairment related to CP and is inversely related to the activity (19). Studies examining the relationship of spasticity or in a broader sense increased muscle tone with gross motor and self-care abilities in children with CP (4, 19-21) have reported inconsistent findings. A moderate association between quadriceps and hamstring spasticity with motor function has been reported, which was even weaker when muscle power was considered in multivariate analyses (4, 20). Tuzson et al. showed a significant positive correlation between spasticity scores, as measured by the Ashworth Scale, and the Gross Motor Function Measure (GMFM) scores in children with CP (22). Similarly, Østensjø et al. reported that spasticity and GMFM-66 scores were significantly and negatively correlated (23). However, no significant relationship has been found between spasticity and activity limitation (4).

3.2. Selective motor control

Selective Motor Control (SMC) is defined as “the ability to move an individual joint voluntarily and independently of posture and other joints in the same limb” (24). The loss of SMC occurs due to damage in the corticospinal tracts. At the functional level, SMC is one of the most significant impairments affecting gross motor function, such as crawling and walking, in children with CP (23, 25). In addition, SMC is also a significant functional outcome of clinical interventions, such as botulinum toxin treatment (26), selective dorsal rhizotomy (27) and surgery (28). Consequently, it is essential to measure SMC, particularly in lower extremities, by clinical methods during the early stages of CP.

3.3. Range of motion

Limited Range of Motion (ROM) has been documented in children with CP, especially those with spasticity, and usually evolves into contractures constraining function (29). Contracture is caused by a combination of hyperexcitability of the stretch reflex, increased muscle tone, marked loss of sarcomeres, and increased stiffness (30). Furthermore, McDowell et al. (2012) showed a significant association of decreased ROM and the activity limitations as measured by GMFCS (Gross Motor Function Classification System scores) (31).

3.4. Muscle weakness

Muscle strengthening has been shown to be effective in improving functional abilities in patients with CP by a number of recent studies (32). In addition, a moderate correlation between muscle strength and gross motor function has been reported in these children (19). Thompson et al. (2011) observed that patients with CP may see an improvement in their walking ability as a result of muscle strengthening exercises of lower extremities (33). In addition, a systematic review has suggested a reliable enhancement of function and gait after progressive strength training in both the children and adolescents with CP who are ambulatory (34).

3.5. Balance

A good postural control during functional activities is the result of the interactions between the sensory system, Central Nervous System (CNS), and musculoskeletal system (35). Children with CP suffer from disorders in postural control and balance as demonstrated by GMFCS (36). A correlation between postural control and equilibrium has been reported, CP children with GMFCS levels I to III have better functional balance as compared to those with GMFCS levels IV and V (37). Poor balance control is one of the contributing factors to problems with gait and reaching movements, and consequently the functional disabilities associated with CP (37).

The neuromuscular response characteristics contributing to these balance limitations include delayed onset of muscle contractions, abnormal timing of muscle responses, and enhanced co-activation of antagonist muscles with agonists (38). The resulting decreased balance may eventually lead to restricted participation and lower quality of life that is frequently observed in children with CP.

3.6. Manual ability

The description of upper limb function in CP is a considerable challenge to clinicians and researchers (39). Beckung and Hagberg found a strong correlation between functional mobility and bimanual fine motor function (40). However, motor function and manual ability are not analogous, and the cognitive ability and control of voluntary movements are dependent on hand function. In general, a CP child with the ability to walk or sit independently can usually do manual tasks needed in the course of daily living (41). Carnahan et al. reported a poor correlation between motor func-

tion and manual ability, and found that manual ability in hemiplegic CP was more limited than diplegic CP (39).

3.7. Body Mass Index (BMI)

A significant number of children with CP have growth problems due to nutritional deficiencies caused by oral muscle dysfunction and feeding difficulties in early childhood (42). It may be the reason why feeding rehabilitation interventions improve the nutritional status and quality of life in these children (43). Malnutrition also increases the risk of obesity, and may lead to weak respiratory muscles, gastrointestinal disorders, decreased immunity, and slow cognitive development (44). Obesity has been frequently observed in ambulatory children, as well as those with diplegic CP (45).

Some studies have shown that children with better gross motor function had some minor degree of obesity, and most overweight or obese children with CP have the gross motor function level III (46). In contrast, more functionally active children are more likely to be underweight (47). Finally, mobility aids required for many CP children to move can actually be an additional environmental barrier, giving them less incentive to be physically active (32, 47, 48). Taken together, we can surmise that more severe neuromuscular disorders and lower functional levels are associated with greater difficulties with physical activity and mobility, leading to a higher risk of being overweight.

3.8. Pain

Pain is prevalent in children with CP, and is a result of immobility, spasticity, and congenital or acquired deformities (49). Hodgkinson et al. reported a 47% prevalence of hip pain complaints in non-ambulatory young adults with CP (50). According to Bagg et al., dislocated hips may predispose CP patients to degenerative arthritis, pain and limited mobility (51). Significant pain can affect the quality of life in children with CP and their participation during healthcare procedures (52, 53). Finally, studies emphasize disturbed physical function, daily care activities, sleep, mental health, school, participation and quality of life due to pain (54).

3.9. Associated disorders

Sensation, perception, cognition, communication, and behavior disturbances, as well as seizures are categorized as associated disorders with CP. Several studies have shown that children with CP who have visual impairments have more functional constraints and

poorer prognoses for ambulation as compared with those without visual impairments (55, 56). Beckung et al. concluded that acute visual and hearing impairments to be predictive of mobility (57). Nashner et al. suggested different integration of visual, vestibular, and somatosensory information for postural control in various types of CP (58).

Although the contribution of cognitive abilities to motor prognosis is not completely clear, they do not primarily determine ambulation in children with CP. Studies have shown low scores on intelligence tests in children with microcephaly (56). Children with intellectual disabilities labeled as trainable are able to walk before the age of 4, while those with profound intellectual disabilities walked only after the age of 7 (59). The strongest predictive factor for walking ability is intellectual ability (60); for instance, epilepsy is a predictor for poor ambulation in CP children and is associated with intelligence level.

3.10. Motivation

Motivation is an intrinsic psychological force that helps us persist during challenging activities. Thelen and Smith have suggested motivation to be an important determinant of developmental change (61). Children who rigorously explore their surroundings by moving around may acquire motor skills more readily than those who are content to explore only visually (13). In addition, mastery of motivation is thought to be a significant predictor of participation in entertaining activities for children with CP (62).

According to the conceptual model of Bartlett and Palisano (2002), the role of motivation is emphasized because of its intrinsic key role in improving the motor abilities of children with CP (13). It has also been emphasized as an essential element of some contemporary rehabilitation interventions such as virtual reality and constraint-induced movement therapy (63).

3.11. Socioeconomic factors

Children with developmental problems who grow up in poverty are in a "double jeopardy" (63). Poverty affects motor function to a lesser degree than cognitive function among children with a range of developmental problems. Family income may be an index of the dynamics of family function. In other words, families living in poverty seem to suffer from increased stress, diminished social support, and depression (17). Therefore, rather than socioeconomic status directly affecting

the acquisition of motor abilities in the child, the family's resources, social support and functioning level may be the decisive factors that determine the child's motor outcome (13).

Rosenbaum et al. hypothesized that although neuromuscular and possibly cognitive impairments have undeniable impacts on the locomotor development of the children with CP, other factors, including the child's physical and social environment might also determine how movements develop in a child with CP (8).

3.12. Environmental factors

Participation of CP children in daily activities may be restricted by physical, social, and attitudinal environment (64). Environmental factors potentially influencing participation are classified into five chapters in the ICF: 1. Products and technologies; 2. Natural and man-made changes to the environment; 3. Support and relationships; 4. Attitudes; and 5. Services, systems, and policies (65). Products and technologies include facilities adapted or specially designed for the functional improvement of disabled individual. Physical environmental barriers can limit functional mobility and social environment (such as negative attitudes) can reduce participation (66).

3.13. Medical and rehabilitation interventions

Physical therapy, occupational therapy, and community services with the aim of improving motor abilities in young children with CP have not been adequately developed (17). Systematic reviews have shown that a range of treatments or a combination of interventions such as exercise programs focusing on increasing the strength of lower-extremities can help CP children improve their body structures, functions, and motor activities (32, 67). In general, children receiving integrated services have been shown to achieve better rehabilitation results in term of motility as compared to those receiving isolated services (68). In order to successfully address the different motility disorders in CP, a multidisciplinary approach is critical. The aim of such an approach is to adjust movement disorders through oral and injectable medications, physical therapy, occupational therapy, orthoses, and orthopedic surgery (57).

CP children with dystonic movements and gait disorders can be safely and effectively treated with botulinum toxin A injections (57). In addition, selective dorsal rhizotomy, intrathecal baclofen pump placement, as well as deep brain stimulation are other recommended

therapies targeting the central motor disorders. Pathological gait patterns, including abnormal joint motion, muscle timing and spatiotemporal characteristics, such as reduced walking speed and stride length, are frequently reported in spastic CP (69).

Improving body structure or assisting function are the primary objectives of designing or selection of orthoses. In CP children, orthoses are frequently used to achieve both. The objectives of managing lower limb orthotics are correction and prevention of deformity, providing a base of support, facilitating training in skills, and improvement of gait efficiency (70).

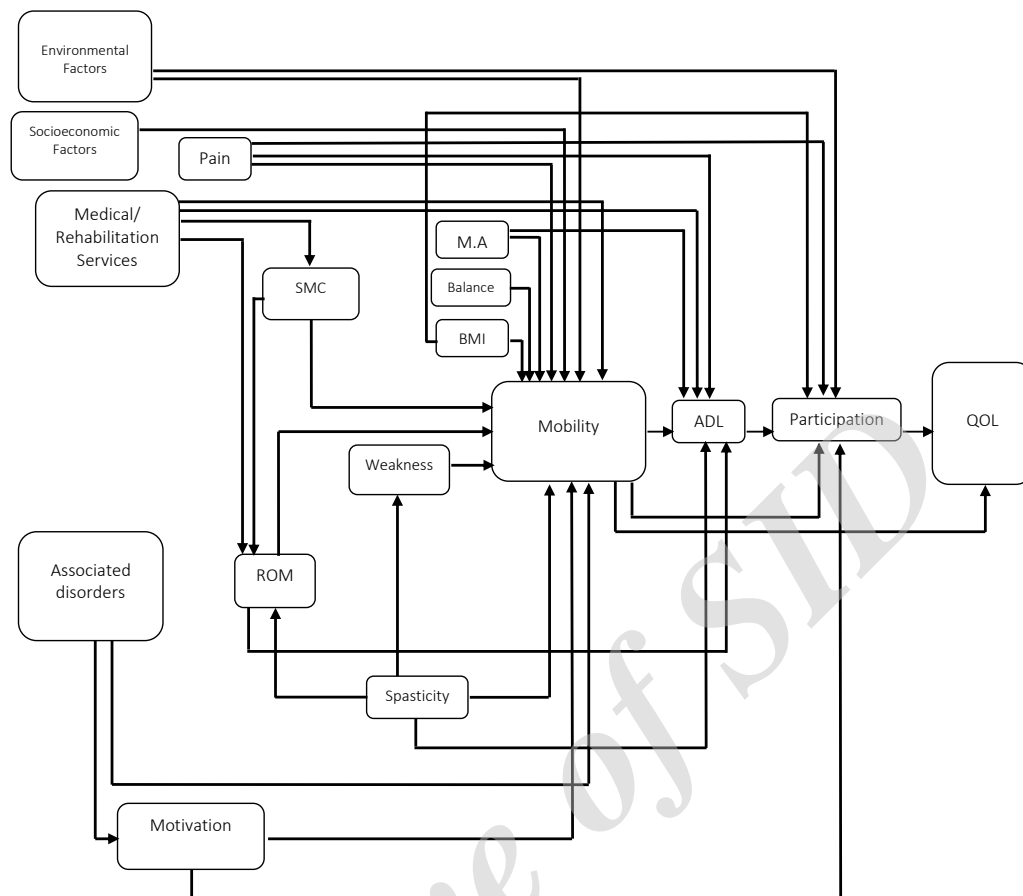
3.14. Activities of daily living

Self-care activities of children with CP improve after adjustment and modification of equipment in their environment (71). CP children with higher functional levels have usually fewer participation limitations and are able to perform better in tasks of daily life (72). In addition, the level of physical abilities in school-aged children with a variety of physical disabilities, including CP are the best predictors of participation during home and community activities (73). CP children with higher gross motor functions have shown to participate in a greater number of activities of daily living and have higher social functioning (2, 74). Furthermore, some studies indicate an association between gross motor function and the degree of self-care independence in CP children aged 4–18 years (75). Finally, GMFCS scores of these children are strongly related to their self-care, daily life activities, and participation (76).

3.15. Participation

Due to their motor disorders, children and adolescents with CP suffer from different forms of limitation in physical activities and participation, or involvement in life situations as defined by ICF (64). Several individual, familial, and environmental factors influence participation (77). Recent studies have highlighted the role of a number of possible predictors of children's participation including environmental factors such as physically accessible and welcoming environments, familial factors like income and family functioning, and individual factors like cognitive ability, preferences, and social skills (78). Moreover, participation has been shown to contribute to the quality of life (79).

Low levels of participation at younger ages can adversely affect the motivation to participate later in life (80). Studies have shown that participation affects qual-



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Figure 1. Full conceptual model of personal and environmental factors contributing to the mobility in children with cerebral palsy

SCM: Selective Motor Control; ROM: Range of Motion; MA: Manual Ability; BMI: Body Mass Index; ADL: Activity Daily of Living; QoL: Quality of Life

ity of life, physical well-being, social support, mood and emotions (79, 81). Motor function is predictive of lower participation in mobility, education, and social relations in children with CP (82). While participation has been considered as the ultimate goal of rehabilitation for children with CP by the ICF, mobility may be the major concern of both parents and therapists.

3.16. Quality of Life

Quality of Life (QoL) is a gold standard for every society, and has been defined by WHO as “an individual’s perception of his position in life in the context of the culture and value systems in which he lives, and in relation to his goals, expectations, standards and concerns” (83). Children with poorer walking ability have demonstrated poorer QoL in the physical wellbeing domain (84). While the presence of pain is associated with poorer QoL, the type and severity of impairments are not significantly correlated with QoL (85). In addition, children with CP

who are more functionally dependent on their parents and caregivers were found to have worse QoL (86).

3.17. The conceptual model

Our proposed model was developed by analyzing the literature on personal and environmental factors, and their correlations with mobility in children with CP. Using the International Classification of Functioning, Disability and Health (ICF) as the basic framework for our model, and considering mobility as the desired outcome, the proposed path model includes factors that potentially cause, mediate and moderate the outcome. The proposed path model is presented in Figure 1, illustrating the process of developing relationships between the constructs. The premise of the model is that the hypothesized personal and environmental factors can influence the mobility of children with CP. We reviewed the evidence to support the constructs in the structural

model and its paths within each factor, and have discussed that below.

The present review evaluates individual, familial, environmental and social factors affecting the mobility in children with CP. The current literature supports the assumption that factors such as degree of spasticity, balance status, manual ability, degree of motivation, participation, daily activities, pain severity, body mass index, biomechanical factors (muscle weakness, joint range of motion), associated disorders (seizure, visual and auditory problems, behavioral problems, cognitive impairments), medical interventions (surgery, Botox injections), socioeconomic parameters (family income, family education, family social support), environmental factors (physical environment, social support, attitude), and finally QoL affect mobility in CP children in the form of a correlational network or path presented in our proposed conceptual model. As shown in [Figure 1](#), variables can either directly or indirectly affect mobility, e.g., spasticity affects mobility in two path, direct and indirect impact on weakness as well as the effect on the ROM.

Park and Kim, in their proposed model, considered only muscular strength, spasticity and gross motor function on mobility, and did not consider the role of other factors ([15](#), [16](#)). The model proposed by Susan et al. included the effect of fatigue and pain on school performance in children with CP ([87](#)). Chiarello et al. presented a multivariate model of factors influencing the change in basic motor abilities in children with CP. They recommended the development and testing of multivariate models for chronic diseases such as CP ([17](#)).

To the best of our knowledge, no study has comprehensively and simultaneously examined the individual and environmental factors affecting the mobility of children with CP. We believe that decision-making regarding intervention options for children with CP should logically be carried out in the context of evidence regarding the relationships between these contributing factors. As a result, the next step in our overall research plan will be to test the proposed model by conducting a field study, collecting the relevant data from the CP children, and finally using standardized regression coefficients to describe the relationship among the different constructs specified by the model. With this method, the respective importance of different causal paths in the context of the whole model can be more accurately interpreted. If contributing factors that lead to changes in basic motor abilities are subjected to manipulation, the

optimized determinants through interventions might accordingly enhance long-term outcomes of mobility.

4. Conclusion

We anticipate that the present model, and ultimately the knowledge gained by testing this model, will help rehabilitation service providers with 1. Consultation with patients and their families regarding the progression of mobility in the future; 2. Setting realistic and achievable outcomes, and 3. Selection of effective interventions to promote motor abilities in CP children. We hope the results will contribute to the planning of more efficient and effective rehabilitation protocols for the CP children at an early stage, with the ultimate goal of optimizing long-term results associated with leisure, education, occupation, social interaction, and quality of life.

Ethical Considerations

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Conflict of interest

The authors declare that they have no conflict of interest.

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