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Effects of Robotic Rehabilitation on Motor Functions in Children with Cerebral Palsy

Serebral Palsili Çocuklarda Robotik Rehabilitasyonun Motor Fonksiyon Üzerine Etkileri

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Abstract

Objective: The aim of this study was to investigate the effects of robotic rehabilitation (RR) on spasticity and motor functions of children with varying types and functional levels of cerebral palsy (CP).
Materials and Methods: A total of 28 children were evaluated aged 6-16 years, with level 2-4 CP according to the Gross Motor Function Classification System (GMFCS) who were planned to undergo 30 sessions of RR. Motor functions were evaluated before and after RR using the Gross Motor Function scale-66 (GMFS-66) B, C, D and E dimensions, gastrosoleus spasticity with the Modified Ashworth scale (MAS) and a target was defined for each patient with a Goal Attainment scale (GAS). Following the RR treatment, the efficacy was evaluated by grouping the patients according to the GMFCS level and the type of CP.
Results: 11% of the patients were at level 2, 36% were at level 3, and 54% were at level 4.61% of them were identified as bilateral spastic, 21% unilateral spastic and 14% mixed type. The patients comprised 50% male and 50% female children with a mean age of 10.8±2.7 years. Mean participation in the RR program was 23±9.6 sessions. A statistically significant improvement was determined in the D dimension of the GMFS in the children at level 2 and 3 of GMFCS ($p<0.05$), and there were no differences in respect of the MAS and GAS ($p>0.05$). No differences were determined between the type of CP groups in respect of GMFS, MAS and GAS.
Conclusion: It was concluded that the application of RR was of benefit for the children with CP at the level 2 and 3 of GMFCS in respect of the development of standing activities.

Öz

Amaç: Bu çalışmanın amacı, farklı fonksiyon düzey ve tipteki serebral palsili (SP) çocuklarda robotik rehabilitasyonun (RR), spastisite ve motor fonksiyonlara etkisini araştırmaktır.
Gereç ve Yöntemler: Otuz seans RR planlanan, 6-16 yaş arasında, Kaba Motor Fonksiyon Sınıflama Sistemi (KMFSS) 2-4 düzeyinde olan 28 SP'li çocuğun sonuçları değerlendirildi. RR öncesi ve sonrasında fonksiyon Kaba Motor Fonksiyon ölçüm-66

(KMFÖ-66) B, C, D, E boyutları ile, gastrosoleus spastisitesi Modifiye Ashwort skalası (MAS) testi ile değerlendirildi ve her hasta için Hedefe Ulaşma skalası (HUS) ile hedef belirlendi. RR tedavisi sonrası, KMFSS düzeyi ve SP tipine göre gruplama yapılarak etkinlik değerlendirildi.
Bulgular: Çocukların %11'i evre 2, %36'sı evre 3, %54'ü evre 4'de, %61'i bilateral spastik, %21'i unilateral spastik, %14'ü miks tipteydi. Hastaların %50'si erkek ve ortalama yaş 10,8±2,7 yıldı. RR programına ortalama katılım 23±9,6 seanstı. RR sonrası KMFSS evre 2 ve 3 çocuklarda KMFÖ, D boyutunda gelişme olduğu saptandı (p<0,05), MAS ve HUS yönünden fark yoktu (p>0,05). SP tipine göre gruplar arasında KMFÖ, MAS ve HUS yönünden anlamlı fark saptanmadı.
Sonuç: RR uygulamasının, KMFSS 2 ve 3 evredeki çocuklarda ayakta durma aktivitelerinin gelişmesinde yararlı olduğu kararına varıldı.

Introduction

One of the targets in cerebral palsy (CP) rehabilitation is to provide mobility according to the expectations of the patient, the clinical examination findings and patient characteristics. Therefore, in rehabilitation programs there has been increasing widespread use of robotic technology that has been developed with the aim of facilitating or correcting the gait, thereby providing functional walking (1-3). Robot-assisted gait training is based on the principles of sensorimotor learning. Through the intensive and repeated visual and auditory stimuli given in the different phases of walking, it is intended to regain functional ambulation (4).

There is not yet considered to be sufficient information to be able to form recommendations for robot-assisted walking in CP childen (5,6). Some studies have reported that robot-assisted walking has provided an increase in gross motor skills, walking speed and resistance (4,7-13), and an increase in participation and performance of daily living activities (9,14). However, it has been claimed that there is a need for stronger evidence of the role and efficacy of robot-assisted walking in the clinical treatment of CP children (10,15).

There have been seen to be conflicting results related to the efficacy of robot-assisted walking training in CP children at different motor functional levels (7,13,16). van Hedel et al. (7) reported developments in mobility and gross motor function in children of level 3 and 4 according to the Gross Motor Function Classification System (GMFCS), while there was no development in level 2 children. Borggraefe et al. (13) determined a greater development in gross motor function in level 1 and 2 children compared to those of level 3 and 4. In a study by Willoughby et al. (16) it was reported that gait training on a walking band was of more benefit to children who were more severely affected functionally. However, to the best of our knowledge, there has been no previous study that has investigated the efficacy of robotic rehabilitation (RR) in different types of CP. Therefore, the aim of this study was to evaluate the response to RR in terms of motor function development in children with different types and motor functional level of CP, who were planned to undergo robot-assisted walking training.

Materials and Methods

The data of children with CP, aged 6-16 years, who applied to the outpatient clinic of physical therapy (PT) and rehabilitation department and who were eligible for RR were analysed retrospectively. The Ethics Committee of the University of Health Sciences, Antalya Training and Research Hospital approved the study (approvel number: 2015-029). A total of 30 sessions of RR was scheduled for the children with CP who were at GMFCS level 2-4, had received no botulinum toxin injections or surgical treatment within the previous 3 months, had sufficient mental function and did not have treatment-resistant epilepsy, any sight or hearing problem, contracture of the lower extremity, fracture or joint instability, hip dislocation or any circulation problems. Motor functions were evaluated using the Gross Motor Function scale (GMFS), and gastrosoleus spasticity with the Modified Ashworth scale (MAS). Taking the GMFS results into consideration and using a Goal Attainment scale (GAS), a target was defined for each patient. All evaluations were performed twice, before and after RR, in all patients.

The GMFCS is a standardised method that classifies gross motor function in CP Children into 5 levels: level 1, walking without restriction; level 2, walking with restriction; level 3, walking with a hand-held assistive device; level 4, limited independent movement and can use a motorised mobility device; level 5, moved by wheelchair (17).

Classification according to the type of CP was made according to the Surveillance of Cerebral Palsy in Europe (SCPE) and was defined as bilateral spastic, unilateral spastic and mixed (18). Although there is no mixed type in the SCPE classification, the presence of spasticity and ataxia together was defined as mixed type.

To measure the gross motor functions, the standard points of the GMFS were calculated. This scale comprises 66 items in 5 dimensions in terms of A, lying and turning (4 items); B, sitting (15 items); C, crawling-kneeling (10 items); D, standing (13 items); and E, walking, running, jumping (24 items) (19). In the study, lying and turning functions were not evaluated, so dimensions B, C, D and E were calculated for evaluation.

The MAS was applied to classify gastrosoleus spasticity. Resistance to passive motion was measured on the following 5-point scale: 0) no increased resistance; 1) slightly increased reisitance; 2) clear resistance throughout most of the range of motion; 3) strong resistance, movement is difficult; 4) rigit flexion or extension (20).

GAS is an individualized, goal-oriented measurement tool rated by the physician and patient to track functional improvement; scores ranged from –3 (worse than start) to +2 (much more than expected, improvements clearly exceeded the defined therapeutic goal) based on a 6-point scale. Active and/or passive goals were set for each child by the patient and the investigator at screening based on the GMFS results and the performance of the child (21) (Table 1).

The RR application as performed 5 days per week, 40 min per session, 30 sessions in total with the RoboGait (BAMA Technology, Turkey) device, which is a robot-aided gait training system that helps the patients improve their walking abilities. The system was composed of a suspension system which could move the patient up and down, backwards and forwards, a screen providing feedback on the interaction of the patient and formed as a robotic walking orthosis providing active movements of the legs and the low-speed walking band on which the patient is walking. The robotic orthosis has motors providing active movement in the hip and knee joints and power sensors measuring the torque on the joints.

Statistical Analysis

All statistical analyses were applied using SPSS version 18.0 software. The differences in the GMFS, MAS and GAS data before and after treatment were calculated and values recorded as median, minimum and maximum. In the comparison of the gender distribution in the groups, the chi-square test was used, and for intra-group evaluations, the Wilcoxon test. The differences before and after RR were compared between groups using the Kruskal-Wallis test. For the comparison of significant data between groups, the Mann-Whitney U test was applied. A value of $p<0.05$ was accepted as statistically significant.

Results

The study included 28 children with CP; 14 males and 14 females. The GMFCS levels were 2 in 11%, 3 in 36% and 4 in 54%. All the GMFCS level 2 children were unilateral spastic, those at level 3 were 20% unilateral spastic, 70% bilateral spastic and 10% mixed type. Those at level 4 were 7% unilateral spastic, 7% dyskinetic, 67% bilateral spastic and 20% mixed type. The 1 child who was dyskinetic was not included in the statistical evaluation.

The age and duration of each session were smilar in all patients when they were stratified according to the GMFCS or CP type ($p>0.05$). The number of RR sessions in the bilateral spastic group was greater than in the mixed group ($p=0.031$) (Tables 2, 3).

At baseline, there were no differences between the GMFCS groups in respect of GMFS-B and MAS ($p>0.05$). A statistically sinificant difference was determined between the groups in respect of GMFS-C, (chi-square=11.770, $p=0.003$), GMFS-D, (chi-square=15.332, $p<0.001$), and GMFS-E (chi-square=17.138, $p<0.001$). The points of the level 2 children were higher than those of the level 3 and 4 children, and the points of the level 3 children were higher than those of level 4 ($p<0.05$) (Table 2).

At baseline, there were no differences between the CP type groups in respect of GMFS-B, C and E points ($p>0.05$). The GMFS-D points were determined to be different between the groups (chi-square=7.805, $p=0.020$). No difference was determined between the bilateral spastic and the mixed group in respect of the GMFS-D points ($p>0.05$). The GMFS-D (standing) points of the unilateral spastic group were higher than those of the bilateral spastic group ($p<0.05$).

In intra-group comparisons according to the GMFCS level, of before and after treatment, no significant differences were observed in the GMFCS level 2 children in respect of all the GMFS, MAS and GAS (p>0.05). In the GMFCS grade 3 children, no difference was determined in the GMFS-C points (p>0.05), however there were significant improvements in the GMFS-B, D and E values (z=-2.214, p=0.027; z=-2.812, p=0.005; z=-2.201, p=0.028, respectively) and in MAS and GAS (z=-2.530, p=0.011; z=-2.558, p=0.011, respectively). In the GMFCS level 4 children, GMFS-D and E dimensions (z=-2.032, p=0.042; z=-2.032, p=0.042, respectively) and MAS and GAS (z=-2.460, p=0.014; z=-2.714, p=0.007, respectively) improved significantly when compared to baseline values, but no significant difference was observed in values of GMFS-B and C dimensions (p<0.05) (Table 2).

When intra-group comparisons were done according to the CP type, no significant change was observed after treatment in the bilateral spastic children in respect of the GMFS-B and C points (p>0.05), however there were significant improvements in D and E points (z=-2.821, p=0.005; z=-2.403, p=0.016, respectively), and MAS and GAS measurements (z=-3.217, p=0.001; z=-3.100, p=0.002, respectively). No significant change was observed in the unilateral spastic children in

Table 1. The targets of the patients according to the Goal Attainment scale

	Patient	Target
1	A.M.Y.	To sit on the mat for at least 3 secs with the arms free and independent
2	B.E.Ş.	To sit on a chair for at least 10 secs with the arms free and independent
3	B.K.	To stand for at least 20 secs with the arms free and independent
4	C.B.	To sit on a chair for at least 10 secs with the arms free and independent
5	D.D.	To stand supported by 1 hand
6	E.D.	To walk 10 steps forward and turn 180°
7	E.C.İ.	In a standing position supported by 1 hand, to raise 1 foot from the floor and hold this position for at least 3 secs
8	E.M.Ç.	In a full kneeeling position, to advance 4 steps with the arms free
9	E.A.	To come to a half-kneeling position from a full kneeling position and hold this position for at least 10 secs
10	E.B.	To sit on the mat for at least 3 secs with the arms free and independent
11	E.H.U.	To stand for at least 20 secs with the arms free and independent
12	E.K.	To extend one arm forward while in a crawling position
13	E.K.	To come to a half-kneeling position from a full kneeling position and hold this position for at least 10 secs
14	F.A.K.	To come to a half-kneeling position from a full kneeling position and hold this position for at least 10 secs
15	H.İ.	To sit on the mat for at least 3 secs with the arms free and independent
16	H.G.	To stand independently and raising 1 leg, hold this position for at least 10 secs
17	H.C.U.	To step over an obstruction at ankle level
18	M.K.	To stand for at least 20 secs with the arms free and independent
19	M.C.K.	To stand for at least 20 secs with the arms free and independent
20	R.Ö.	To stand independently and raising 1 leg, hold this position for at least 10 secs
21	S.A.	In a crawling position, to make reciprocal forward crawling
22	S.G.E.	To walk 10 steps forward
23	S.E.	To walk 10 steps forward and turn 180°
24	S.K.	To walk 10 steps forward
25	Ş.K.	To sit on the mat for at least 3 secs with the arms free and independent
26	T.T.	To sit on the mat for at least 3 secs with the arms free and independent
27	T.Ş.	In a crawling position, to make reciprocal forward crawling
28	V.Y.	To step over an obstruction at ankle level

respect of the GMFS points, MAS and GAS (p>0.05). In the mixed type spastic children, no difference was determined in the GMFS-B, C and E points and MAS (p>0.05). A significant increase was determined in the GMFS-D points and in GAS (z=-2.207, p=0.027; z=-2.121, p=0.034, respectively) (Table 3).

When inter-group comparisons were performed after the RR, no difference was determined between the GMFCS groups in respect of GAS, MAS and GMFS-B, C and E points (p>0.05), however, GMFS-D values were significantly different (chi-square=16.564, p<0.001). The increase in the GMFS-D points in the level 2 and level 3 children was greater than the increase in those at grade 4 (z=-2.214, p=0.027; z=-3.915, p<0.001, respectively) (Table 2). No statistically significant difference was determined between the CP type groups in respect of all the GMFS points, MAS and GAS (p>0.05) (Table 3).

Discussion

The aim of this study was to evaluate the response to RR in respect of motor function development in children with CP of different types and with different levels of motor function. At the end of treatment, while no change was observed in the GMFS points of the level 2 children, an increase was determined in the GMFS-B, D and E points of the level 3 children, and in the GMFS-D and E points of the level 4 children. No change was seen in MAS and GAS in level 2 children, whereas a decrease in MAS and an increase in GAS were seen in level 3 and level 4 children. In the group comparisons, the increase in the GMFS-D points of the level 2 and 3 children was greater than in the grade 4 children.

There have been conflicting reports related to the efficacy of RR applied to children with CP at different gross motor function levels (7,13). van Hedel et al. (7) applied a mean of 20 RR sessions in addition to a regular physiotherapy program in a group of children with CP aged 4-20 years, comprising 15% level 2, 23% level 3, and 29% level 4. They reported that while there was no development in level 2 children, significant improvements were observed in GMFS-D and E points of level 3 and 4 children, without significant differences between the groups. Borggraefe et al. (13) applied 12 sessions of RR and determined a greater development in GMFS-D in level 1 and 2 children compared to those of level 3 and 4. In a study by Willoughby et al. (16), it

Table 2. Improvement in spasticity, Gross Motor Function Measure, and Goal Attainment scale in different levels of Gross Motor Funtion Classification System

		Gross Motor Function Classification System								
		Level 2 n=3 median (min-max)			Level 3 n=10 median (min-max)			Level 4 n=15 median (min-max)		
Age, year		12 (10-13)			11 (8-14)			11 (6-16)		
Number of sessions		26 (26-31)			31.5 (25-32)			28 (20-32)		
Duration per session, min		37 (33-40)			39.5 (33-54)			34 (26-44)		
		BT	AT	D	BT	AT	D	BT	AT	D
MAS		2 (1-2)	1 (1)	-1 (-1-0)	2 (1-3)	1 (1-3)§	-1 (-1-0)	2 (1-3)	1 (1-3)§	0 (-1-0)
GAS		-2 (-2)	-1 (-2-1)	1 (0-1)	-2 (-2)	-1 (-2-1)§	1 (0-2)	-2 (-2)	-1 (-2-1)§	1 (0-2)
GMFM (%)	B	88 (77-100)	100 (93-100)	16 (0-16)	96 (75-100)	100 (76-100)§	1 (0-13)	84 (24-100)	86 (24-100)	0 (-5-7)
	C	100 (70-100)*	100 (80-100)	4 (0-10)	96 (66-100)*	100 (68-100)	0 (0-2)	52 (0-96)*†‡	51 (0-96)	0 (-11-2)
	D	64 (23-74)*	76 (27-100)	4 (2-36)†	37 (2-56)*	56 (10-71)§	7 (4-58)‡	2 (0-20)*†‡	2 (0-22)§	0 (0-5)*†
	E	66 (17-73)*	67 (18-73)	1 (0-1)	21 (2-54)*	23 (5-57)§	2 (-2-19)	0 (0-20)*†‡	0 (0-24)§	0 (0-11)

*Kruskal Wallis, †Mann-Whitney U, level 2-4, ‡Mann-Whitney U, level 3-4, §Wilcoxon, p<0.05

BT: Before treatment, AT: After treatment, D: Difference, MAS: Modifiye Ashwort scale, GAS: Goal Attainment scale, GMFS: Gross Motor Function scale, B: Sitting dimension, C: Crawling- kneeling dimension, D: Standing dimension, E: Walking, running, jumping dimension, min: Minimum, max: Maximum was reported that the children with more functional disability were more likely to benefit from RR.

In studies that have compared RR with conventional PT, different results have been reported in respect of efficacy (4,10,11,22,23). In a study which compared 20 sessions of RR and PT in bilateral spastic, level 2 children aged 8-10 years, improvement was recorded in the RR group in GMFS-D and E points (4). Another study found no difference between a PT group and an RR group in kinematics and step length, step width and walking speed measurement in level 2-3 CP children aged 6-13 years with spastic diplegia (10). Level 2-4, bilateral spastic CP children aged 4-12 years were compared as a hospitalised group applied with 20x45-min sessions of PT and an outpatient group applied with 12x60-min sessions of RR, and no difference was determined between the groups, with a similar increase in walking speed and GMFS-D points in both groups (11). In level 1-3, bilateral spastic CP children aged 4-17 years, treatment was applied for 4-10 weeks at a frequency of 4-10 sessions per week and no difference was determined between the RR, PT and PT+RR protocols. Similar results were determined to have been obtained from the robotic application and the physiotherapy, and it was concluded that a single application was more beneficial than combined applications, with reported improvements in GMFS-D and E points in all groups (22). In a study by Romei et al. (23), 20 sessions of RR and PT were compared with RR alone in bilateral spastic CP children, and it was reported that both groups improved and there was no difference between the groups in respect of the improvement in gross motor function.

van Hedel et al. (7) determined a strong relationship between the GMFS-E dimension and the total number of treatment sessions in level 2 CP children. In that study, although not significant, the total number of RR sessions was higher, and the duration of the sessions was longer in grade 2 and 3 children. Despite the lack of studies directly comparing RR treatment doses, that children who were more affected in respect of motor function were likely to benefit more from the application of a greater total number of sessions and longer session duration (7). Researchers have emphasised that more severely impaired children benefit more from a greater number of sessions, and longer walking distance and duration of the sessions, while lesser affected children benefit from high

Table 3. Improvement in spasticity, Gross Motor Function Measure, and Goal Attainment scale according to cerebral palsy type

		Bilateral spastic n=17 median (min-max)			Unilateral spastic n=6 median (min-max)			Mixed n=4 median (min-max)		
Age, year		12 (6-16)			10 (8-13)			11 (7-16)		
Number of sessions		30 (20-32)†			26 (21-32)			22.5 (21-27)*†		
Duration per session, min		37 (29-54)			36.5 (32-44)			32 (26-39)		
		BT	AT	D	BT	AT	D	BT	AT	D
MAS		2 (1-3)	1 (1-3)	-1 (-1-0)‡	2 (1-2)	1 (1-2)	-0.5 (-1-0)	2.5 (2-3)	2.5 (1-3)	0 (-1-0)
GAS		-2 (-2)	-1 (-2-1)	1 (0-2)‡	-2 -2 (-2)	-1 (-2-1)	1 (0-1)‡	-2 (-2)	-1.5 (-2-0)	0.5 (0-2)
GMFS (%)	B	86 (24-100)	87 (24-100)	0 (-5-13)	92,5 (77-100)	100 (90-100)	2.5 (0-16)	94 (44-97)	94.5 (44-100)	0.5 (0-3)
	C	76 (0-100)	75 (0-100)	0 (-11-2)	91 (46-100)	91 (46-100)	0.5 (0-10)	55 (0-100)	56 (0-100)	0 (0-2)
	D	10 (0-56)*†	15 (0-71)	4 (0-58)‡	58 (15-100)*†	58 (15-100)	4 (1-36)‡	10 (0-43)*	11 (0-48)	1 (0-5)
	E	11 (0-47)	11 (0-57)	1 (-2-19)‡	29.5 (0-73)	32.5 (0-73)	1 (0-4)	6 (0-54)	6.5 (0-56)	0.5 (0-2)

*Kruskal-Wallis; †Mann-Whitney U, ‡Wilcoxon, $p<0.05$

BT: Before treatment, AT: After treatment, D: Difference, MAS: Modifiye Ashwort scale, GAS: Goal Attainment scale, GMFS: Gross Motor Function scale, B: Sitting dimension, C: Crawling- kneeling dimension, D: Standing dimension, E: Walking, running, jumping dimension, min: Minimum, max: Maximum intensity programs of shorter duration (24,25).

In the current study, while no change was determined in GMFS points, MAS and GAS in unilateral spastic children, an increase was determined in GMFS-D and E and an improvement in MAS and GAS measurements in bilateral spastic children. In the mixed type children, an increase was determined in GMFS-D points and GAS. No difference was determined in the GMFS, MAS and GAS between the CP types. As there is no previous study in literature that has evaluated the efficacy of RR in different CP types, no comparison could be made. Although there was no difference between the CP types in respect of MAS, the MAS median value was found to be higher in the bilateral spastic and mixed type groups. The improvement in motor function in the bilateral spastic and mixed type groups can be considered to be due to a reduction in partial spasticity, associated with the negative effect of spasticity on gross motor function (26).

It has been emphasised that for there to be benefits of robotic walking training in respect of improvements in walking speed, resistance and gross motor functions in children with CP, the program should be applied at a frequency of 4 days per week with sessions of at least 30 mins (30) and it has been stated that strong evidence is required to determine the role of robot-assisted walking in CP children (10,15).

The primary limitation of the current study was the low sample size of different motor function grades and different CP types, and that there was no control group. Second is that the anthropometric details of the children and energy consumption were not recorded as it has been previously reported that anthropometric differences affect gait kinetics and motor behaviour (3). It has been emphasised that a reduction in energy consumption is related to a reduction in contractions of spastic muscles and a more effective gait pattern (3). A third limitation was that details could not be obtained of the reliable application of the RR as it was implemented at a different centre.

Conclusion

In conclusion, although several methodological limitations prevent definite conclusions, a number of potential clinical implications emerge from this study. Our results showed that RR application is beneficial for improving standing activities in bilateral spastic children with CP at GMFCS level 2 and 3. Further prospective studies are needed to confirm these results in larger groups of patients to improve the evidence-based application of RR.

Ethics

Ethics Committee Approval: The Ethics Committee of the University of Health Sciences, Antalya Training and Research Hospital approved the study (approvel number: 2015-029).

Informed Consent: As this was a resrospective study, there is no informed consent.

Peer-review: Externally and internally peer-reviewed.

Authorship Contributions

Surgical and Medical Practices: M.B.F., N.F.T., T.Ç., H.A., Concept: N.F.T., C.M.A.Ç., Ş.K.D., Design: N.F.T., C.M.A.Ç., H.A., T.Ç., Data Collection or Processing: C.M.A.Ç., H.A., Ş.K.D., Analysis or Interpretation: M.B.F., N.F.T., Literature Search: T.Ç., Ş.K.D., Writing: M.B.F., N.F.T.

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