Factors influencing developmental outcome following developmental therapy in children with cerebral palsy

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Abstract

Background: Cerebral palsy (CP) describes a group of motor syndromes resulting from disorders of early brain development. In Bangladesh a single approach therapy has been adopted in many medical institutes as developmental therapy where a developmental therapist assesses the child and provides physiotherapy, occupational therapy, cognitive stimulation, feeding therapy, speech and language therapy to the children according to the need by a holistic approach.

Objective: This study was designed to determine the factors influencing developmental outcome following 6 months developmental therapy in children with cerebral palsy.

Methods: A quasi-experimental study was conducted in the department of Paediatrics, Bangabandhu Sheikh Mujib Medical University on 135 children with CP aged 6 months to 3 years. The motor, mental and behavioral functions of the selected children were initially measured by Bayley Scales of Infant Development II. They were reassessed after 3 months and 6 months following "developmental therapy". Only 62 (45.9%) children have completed the 2 follow-up sessions and hence were evaluated against their baseline motor, mental and behavior score. Data analysis were done by Friedman test, Wilcoxon signed rank test, Chi square test, Fisher's exact test and Unpaired T test. For above tests, significance level α was set at 5% so *p* value <0.05 was considered as significant.

Results: Among 135 children only 62 children completed 2 follow-ups. Mean age of 62 children was 15±8.4 months; male was 66% and female was 44%. After developmental therapy for 6 months 46.8% the children had improved motor skills, 40.3% mental skills and about 69 % behavioral skills; presence of epilepsy, microcephaly and significant illness affected the outcome. Early age, duration and intensity of developmental therapy showed positive impact on the outcome in children with cerebral palsy.

Conclusion: Developmental therapy should be started sooner after the diagnosis of a child with CP and intensity of therapy should as much as possible.

Keywords: Cerebral palsy, Developmental outcome, Developmental therapy.

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Introduction:

Cerebral palsy (CP) describes a group of motor syndromes resulting from disorders of early brain development.[1] The prevalence of cerebral palsy in the world is about 1.6/1000 live birth.[2] In Bangladesh the prevalence is about 3.4/1000 live birth.[3]

Various combination therapies are now practiced in abroad in different names. In Bangladesh a combination therapy named developmental therapy has been advocated for the management of these children. Developmental therapy is the combination of physiotherapy, occupational therapy, cognitive stimulation, and speech and language therapy. It is principally a home based therapy where mother or caregiver plays a role of therapist. She comes to the centre with her children periodically for therapy and learns how to give the therapy to her children at home.[4] It is very much effective where parents are used as therapist. Maximum effectiveness is achieved when parental skills are increased.[5,6] There are some factors that might influence the developmental outcome following developmental therapy. Studies showed that age, epilepsy, intensive therapy, awareness about the condition of CP interfered developmental outcome.[7-11]

There are limited published evidence in this aspect so the present study was conducted on children with CP to find out the associated factors that may influence the developmental outcome following developmental therapy.

Materials & Methods:

It was a quasi-experimental study done in Child Neurology Centre of Bangabandhu Sheikh Mujib Medical University, Dhaka, Bangladesh. A total of 135 children with CP from 6 months to 3 years of age were taken as study population**.** Convenience sampling method was applied. Informed written consent was taken from the caregiver/parents of the patients. Ethical clearance was obtained from the institutional ethical committee of the university. Children with CP who have visual, hearing impairment and who had prior developmental therapy were excluded from the study. Categories of CP and their associated problems like epilepsy, behavioral problem were assessed and recorded. Any illness which occurred in a single or multiple episodes causing interruption of developmental therapy for 15-30 days (total) within 6 months' period were considered as significant illness which included respiratory infection and diarrhoeal diseases. After giving primary management of associated problems by pediatricians, their motor, mental and behavioral status were assessed by the investigators assisted by the psychologist at every visit at three-month interval. After first assessment, counseling of the parents was done. Parents counseling was the major part of the management of children with CP because they would have been the central focus part of the developmental therapy as they had to give the therapy their children at home regularly. Data were entered and analyzed using the software "Statistical Package for Social Science" (SPSS version 18.0 for Windows, IBM, Chicago, IL). Friedman test, Wilcoxon signed rank test, Chi square test, Fisher's exact test and Unpaired T test were done. For above tests significance level α was set at 5% so *p* value <0.05 was considered as significant.

Assessment Tools

Assessment was done by Bayley Scales of Infant Development II (BSID II)[12], administration time for under 14 months was 25-35 minutes and above 15 months up to 60 minutes. The improvement of motor, mental and behavioral functions were assessed as follows;

Firstly, the deviations between the observed scores of the children at baseline and the reference median scores of normal children (obtained from BSID II) of the same age were calculated. The deviations were then converted into percentage. Similarly, the percentage of deviation after 6 months of therapy was calculated with respect of normal children (obtained from BSID II) of the same age. The percentage of deviation after 6 months of therapy was then subtracted from that of the baseline. If the percentage of gap was reduced by >5, the child was considered as "improved" and If the percentage of gap was reduced by ≤5, the child was classified as "not improved".

Follow Up

Parents with each child was asked to come for developmental therapy at 1-month interval. Their motor, mental and behavioral status was assessed by the principal investigator assisted by developmental psychologist in every 3 months' interval and improvement of outcome were assessed. The growth and development and general health measures were monitored by the principal investigator. If any problem or illness was identified in any visit; steps were taken to conduct appropriate investigations (USG, CT or MRI of brain, EEG, Complete blood count etc.) and to provide intervention, advice and specific counseling to the parents. Total 6 month follow up was done.

Results:

A total of 135 children with CP attended the Pediatric Neurology outpatient department but only 62 (45.9%) completed 2 follow up sessions and hence were evaluated against their baseline motor, mental and behavior score.

Table 1: Demographic characteristics of children with CP (n=62)

Characteristic	Frequency	%
Age in months		
06-18	36	58.07
19-36	26	41.93
Mean age in months	15±8.4	
Age range in month	6-36	
Sex		
Male	41	66
Female	21	34
Monthly Income		
< 6000 tk	39	62.9
6000-12000 tk	16	25.8
>12000 tk	7	11.3
Mothers' education		
Illiterate	5	8
Primary education	32	51.6
SSC and HSC	17	27.4
Graduate and above	8	12.9

Baseline characteristics shows most of the children 36(58.07%) in this study were between 6-18 months; mean age was 15.37± 8.4 months. Over two- third 41(66%) were male (male: female ratio 2:1). In terms of income by parents, over two-third 39(62.9%) had earnings of <6000 taka. Mother's education level demonstrated that about 51.6% of the mothers had primary level educated as shown in Table 1.

Table 2: Distribution of children by developmental outcome (n=62)

Outcome	Frequency	Percentage
Motor function improved	29	46.8
Mental function improved	25	40.3
Behavioral function improved	43	69.3

Table 2 shows that nearly half 29(46.8%) the children had improved motor skills, 25(40.3%) improved mental skills and about 43(69 %) improved behavioral skills.

Table 3: Outcome following developmental therapy (n=62)

Outcome variables	Evaluation at			
	Baseline	Month 3	Month 6	*p* value
Motor score	29.0	34.50	48.0	< 0.001
Mental score	60	68	72.50	< 0.001
Behavior score	91	97.50	104	< 0.001

Table 3 shows that as outcome of developmental therapy median motor score of 62 children was increased slowly from 29 at baseline to 34.50 at month 3 and to 48 at month 6 (p<0.001). The mental score was increased from 60 at baseline to over 68 at 3 and 72.50 at 6 months (p<0.001). Significant improvement was also noted at 6 month in behavior score.

Friedman test was done to see if there was any significant difference among the baseline, 3 months and 6 months' score of motor, mental and behavioral function.

Table 4: Association between demographic variables and motor outcome (n-62)

Demographic variables	Outcome		*p*-value
	Improved (n = 29)	Not improved (n = 33)	
Age (months)			
06-18	21 (72.4)	15 (41.7)	0.032
19-36	08 (27.6)	18 (69.3)	
Sex			
Male	18 (62.1)	23 (69.7)	0.527
Female	11 (37.9)	10 (30.3)	
Socioeconomic status			
Poor	19 (65.5)	20 (60.6)	0.589
Middle class & rich	10 (34.5)	13 (39.4)	
Residence			
Urban	15 (51.7)	11 (33.3)	0.143
Rural	14 (48.3)	22 (66.7)	

Data were analyzed using Chi-square Test. Figures in the parentheses denote corresponding percentage.

Association between 4 demographic variables and motor outcome following developmental therapy reveals only age was found to be associated with outcome of developmental therapy (Table 4).

Table 5: Anthropometric variables and developmental outcome (n=62)

Anthropometry	Developmental outcome					
	Motor		Mental		Behavior	
	Improved	Not improved	Improved	Not improved	Improved	Not improved
3rd degree malnutrition* n_1=5	2 (40)	3 (60)	1 (20)	4 (80)	4 (80)	1 (20)
p value	**1.0**		**0.640**		**0.511**	
Microcephaly # n_2=22	5 (22.7)	17 (77.3)	4 (18.1)	18 (81.9)	9 (40.9)	13 (59.1)
p value	**0.005**		**0.008**		**0.001**	

* Data were analyzed using **Fisher's Exact Test**

Data were analyzed using **Chi-square Test**; Figures in the parentheses denote corresponding percentage.

Of the two anthropometric variables, the frequency of microcephaly was 22 and 3rd degree malnutrition was 5 out of 62 children. Microcephaly was less in the children who exhibited significant improvement 5 (22.7 %) in motor function compared to those who did not show any improvement 17(77.3%) where p=0.005. In mental outcome the frequency of microcephaly was much less in the children who exhibited significant improvement 4 (18.1%) compared to those who did not show any improvement 18 (81.9 %) where p=0.008. Regarding behavioral outcome, the frequency of microcephaly was less in the children who exhibited significant improvement 9 (40.9 %) compared to those who did not show any improvement 13(59.1 %) where p = 0.001.

Differences were also compared with nutritional status. As sample size was small for undernourished group, no statistical significant differences were found (Table 5).

Table 6: Influence of epilepsy on developmental outcome (n=62)

Epilepsy	Developmental outcome					
	Motor		Mental		Behavior	
	Improved	Not improved	Improved	Not improved	Improved	Not improved
Present	2 (40)	3 (60)	1 (20)	4 (80)	4 (80)	1 (20)
Absent	5 (22.7)	17 (77.3)	4 (18.1)	18 (81.9)	9 (40.9)	13 (59.1)
p value	**0.001**		**0.041**		**0.025**	

Data were analyzed using Chi-square Test; Figures in the parentheses denote corresponding percentage.

Majority of the children 14 (87.5 %) who had epilepsy did not improve in motor function following "developmental therapy" (p= 0.001). Similar result was evident in mental function where 81.2% did not show any improvement (p=0.04). Regarding behavioral function improvement was seen more of the epileptic children than that of motor and mental function. However, there was significantly more improvement in children without epilepsy (p= 0.02) as shown in Table 6.

Table 7: History of significant illness that interfered with developmental therapy and developmental outcome

History of significant illness	Developmental outcome					
	Motor		Mental		Behavior	
	Improved	Not improved	Improved	Not improved	Improved	Not improved
Present	05 (26.4)	14 (73.6)	4 (21.1)	15 (78.9)	11 (57.7)	08 (42.3)
Absent	24 (55.8)	19 (44.1)	21 (48.8)	22 (51.1)	35 (81.3)	08 (18.6)
p value	**0.032**		**0.040**		**0.002**	

Data were analyzed using Chi-square Test; Figures in the parentheses denote corresponding percentage.

Table 7 shows 19 children had different significant illness that interfered with developmental therapy. Of them only 5 (26.4%) had improved motor function during study period, whereas 43 children did not suffer from any significant illness, amongst them 24 (55.8%) had improved motor function (p= 0.032). In mental outcome 4 (21.1%) children who suffered from significant illness had improved during study period. Whereas 43 children who did not suffer from any significant illness, amongst them 21 (48.8%) had improved in mental function (p = 0.040). In behavioral outcome 11 (57.7%) children had improved during study period suffered from significant illness, whereas 43 children did not suffer from any significant illness, amongst them 35 (81.3) children had improved the behavioral function (p = 0.002).

Table 8: Impact of duration of therapy at home on developmental outcome

Duration of therapy in a week	Developmental outcome					
	Motor		Mental		Behavior	
	Improved	Not improved	Improved	Not improved	Improved	Not improved
Mean hours	8.18	4.97	8.56	5.06	7.45	4.26
SD	3.91	2.62	3.57	2.99	3.64	2.55
p value	**< 0.001**		**< 0.001**		**< 0.001**	

Data were analyzed using unpaired t-Test and were presented as mean ± SD.

The mean duration of developmental therapy in a week was significantly longer as 8.18 hours, 8.56 and 7.45 hours in children who demonstrated improvement in motor, mental and behavioural function ($p < 0.001$) respectively than that in children who did not show any improvement in the above three areas with less duration of developmental therapy as shown in Table 8.

Discussion:

Total 135 cases were included initially in the study. The guardians (particularly mothers) were asked to bring their children at 3 monthly intervals for assessment up to 6 months following therapy. Only 62 (45.9%) completed 2 follow-up sessions and hence were evaluated against their baseline motor, mental and behavior functions.

After giving "developmental therapy", nearly half (46.8%) of the children were improved in motor function, 40.3% in mental function and about 69 % in behavioral functions (Table 03). The behavioral function was improved more in relation to the mental function. This might be explained that the children were neglected before "developmental therapy". When they got extra care from their mother and family members the behavioral function was the first thing that changed in higher proportion.

Similar findings were found in a retrospective study done by Jahan et al. to evaluate the effectiveness of developmental therapy in children with cerebral palsy and other disabilities. Total 100 children were included in the study. The result showed most of the children improved their function slowly in various aspect of development. The maximum improvement was seen in gross motor function (36%), 12% in cognitive function and least in fine motor function (7%). The behavioral function was not assessed in the above study.[13]

In the present study, age group from 6 to 18 months showed more improvement after developmental therapy compared to the age group from 19 to 36 months group (Table 04). Hong shows similar findings in a study which was designed to identify factors influencing the short-term effect of intensive therapy on gross motor function in children with cerebral palsy. The study included 145 cases who received 8 weeks intense therapy. Measurements of gross motor functions were performed using the gross motor function measure-88 (GMFM-88) and gross motor function classification system (GMFCS), and were obtained at the start and end of the course. Result showed that age ≥ 36 months were significantly associated with a poor response after intensive therapy.[8]

Microcephaly was present in 22 (35.4 %) children. Majority of the children who did not show any improvement had microcephaly (Table-5) compared to those who improvement in motor, mental and behavioral function ($p < 0.01$). There are some studies that support these findings. Gordon showed that postnatal etiologies of microcephaly and infants with comorbid epilepsy had worse outcomes.[10] Messerschmidt did a case control study to observe the neurodevelopmental outcome in preterm babies suffering from disrupted cerebellar development. Thirty-one preterm patients with disrupted cerebellar development were taken as cases and a control group of thirty-one gender and gestational age matched premature infants with normal cerebellar development were included in the study. CP was diagnosed in 48% of affected patients later on whereas none of the patients of the control group had CP. Microcephaly was significantly related to disrupted cerebellar development and poor neurodevelopmental outcome.[14] Watemberg et al. did a study to assess the clinical impact of microcephaly among children with developmental disabilities, they reviewed 1393 patients from birth to 5 years of age. Mental retardation was significantly more common among microcephalic patients with cerebral palsy than among normocephalic ones ($p = 0.004$).[15]

One-fourth of the children 16(25.8 %) had history of epilepsy. Majority of the children who had epilepsy did not improve in motor, mental and behavior function following "developmental therapy" compared to children who showed improvement (Table-6). The association between ongoing seizures and cognitive and behavioral regression has been demonstrated convincingly.[9,11,13,16,17,18] Moreover antiepileptic drugs have a detrimental effect on the central nervous system and may affect mood, cognitive and behavior function.[16] Though the epilepsy affect only cognition and behavior function in the above studies but in the present study mental and behavior along with motor outcome was also influenced by epilepsy.This might be explained that epilepsy interfered "developmental therapy" in our children.

In the present study recurrent seizure (epilepsy), recurrent respiratory tract infection and dirrhoea, which were considered as significant illness influenced the developmental outcome. Over one-third of the children who did not improve in motor and mental function and over half of the children who did not improve in behavior function suffered from significant illnesses during study period ($p < 0.05$) shown in Table-7.This may suggest that the significant illness might interfered "developmental therapy" and then developmental outcome.

The present study considered the duration of "developmental theapy" at home as an important associated factor that influenced developmental outcome.The mean duration of "developmental therapy" in a week was significantly longer in children who demonstrated improvement in motor, mental and behavioral function than those who did not show improvement ($\boldsymbol{p} < 0.001$) according to Table-8. There are some studies those suggest the benefit of intensive therapy for CP children. Tsorlakis et al. did a randomized control trial to examine the effect of neurodevelopmental therapy (NDT) and differences in its intensity on gross motor function of 34 children with cerebral palsy. Participants were assigned randomly to two groups, Group A was designed to receive NDT for 16 weeks, twice weekly, for 50 minutes each session. Group B was the intensive group and followed the NDT program for 16 weeks, five times weekly, for 50 minutes each session. Result showed children in group B performed better and showed significantly greater improvement than those in group A ($\boldsymbol{p} < 0.05$).[19] This study underlined the need for intensive application of the therapy.

Stiller did a study to compare the effects of intensive therapy, conductive education and special education services for a group of 19 children diagnosed with cerebral palsy. Results of this study found that physical gains were greatest in children who received intensive physical, occupational, and speech therapy over a five-week period. In addition, children in all groups showed some improvement in physical functioning in spite of the short duration of the study.[20] This also confirm that duration and intensity of developmental therapy affect the outcome significantly.

Conclusion:

The present study observed that presence of epilepsy, microcephaly or significant illness that interfered "developmental therapy" may affect the outcome. Early age, duration and intensity of developmental therapy has positive impact on the outcome. Despite of the high attrition rate (54%), short intervention period for evaluation and variability of response from the parents, it could be concluded that developmental therapy should be started sooner after the diagnosis of a child with CP and intensity of therapy should as much as possible. To lower attrition rate, parents need counseling, regular updating regarding the evaluation report of the children and subsidization of financial charges for service delivery might be other important aspects to be addressed.

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