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Original Article

Motor functions, quality of life and maternal anxiety and depression in children with cerebral palsy of different intelligence levels

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ABSTRACT

Background. Cerebral palsy (CP) is the most common motor disability in childhood. In addition to motor impairment, it is frequently accompanied by intellectual disability (ID). We aimed to investigate the associations between motor functions, quality of life (QoL) and maternal psychopathology in children with CP of different intelligence levels.

Methods. In total, 37 children and adolescents (16 females and 21 males) between 4 and 18 years of age diagnosed with CP were recruited from a Pediatric Neurology Outpatient Clinic. Gross Motor Function Classification System (GMFCS) and Bimanual Fine Motor Function (BFMF) were used for the children's motor functions assessment. Quality of life was determined by the caregivers with Pediatric Quality Of Life Inventory-Parent version (PedsQL-P). Maternal anxiety and depression levels were assessed using Beck Anxiety Inventory (BAI) and Beck Depression Inventory (BDI).

Results. Moderate-severe ID (n=19)(13.5%, 37.8%) and normal IQ-mild ID (n=18) (32.4 %,16.2%) groups were evaluated in this study. GMFCS level 2 was more frequent in both groups. The majority of the severe-moderate ID group was at BFMF level 4, while the normal IQ-mild ID group was at BFMF level 2. PedsQL-P scores of children with CP, maternal BAI scores, and maternal BDI scores did not differ between the two groups ($p>0.05$). Psychosocial PedsQL scores had a moderate negative correlation with the maternal BAI scores ($r=-0.41$, $p<0.05$). There was also a moderate positive correlations between the ages of children and maternal BDI scores ($r=0.34$, $p<0.05$).

Conclusions. Our results demonstrated that maternal anxiety was correlated with psychosocial QoL in children with CP. Maternal depression scores increasing with the ages of the children with CP may also indicate the social support needs for mothers with children of chronic diseases. Further studies may reveal the associations with other biopsychosocial factors in children with CP of different intelligence levels by using longitudinal study designs with larger sample sizes.

Key words: anxiety, depression, cerebral palsy, child, quality of life.

Cerebral palsy (CP) is the most common motor disability in childhood.[1] Cerebral palsy (CP) is described as a nonprogressive neuromotor disorder that mainly affects the development of movement, muscle tone, and posture.[2] CP umbrella refers to very heterogeneous clinical presentations in children. In addition to motor impairments, there may be impairments in cognition, communication, hearing, vision, behavior, and epilepsy that may worsen the motor impairment, function, and quality of life (QoL).[3,4]

The most common comorbidities according to the Surveillance of Cerebral Palsy in Europe (SCPE) are speech/language impairments

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(71%), severe intellectual disability (ID) (62%), epilepsy (39%), and visual impairment (22%).[3] CP is accompanied by different levels of ID.[5] Gabis and her colleagues' study[6] reported that 22.5% of children with CP had normal intelligence ,and 41.3% had moderate or severe ID. Moreover, it was suggested that more severe motor function impairments are associated with higher ID.[6] Bertoncelli et al.[7] reported that poor motor skills and epilepsy were associated with severe ID.

Severe motor function impairment in children with CP has been found to be more related to poor QoL in terms of physical health and autonomy. Additionally, children with low intelligence quotient (IQ) were found to have a higher risk of poor QoL in terms of social support.[8] Mobility performances relate to gross motor function levels; however, daily activities mostly depend on children and adolescents' intellectual ability with CP.[9] Caregiving a child who has limited self-mobility is highly psychologically and physically demanding.[10,11] Parents must spend more time caring for children with CP who have limited self-mobility. Greater limited self-mobility in children with CP is associated with greater stress and depressive symptoms among their mothers.[12] Because mothers are mostly primary caregivers, and important team workers of the rehabilitation and care services of children with cerebral palsy, they suffer from many socio-emotional problems and report higher anxiety levels.[13,14] Mothers of children with CP have been found to show depressive symptoms and poor QoL, and it was reported that 30% of mothers having CP children showed symptoms of depression.[15] Also, the prevalence of depression was reported at 20% to 30% in mothers of children with ID.[16,17] The mothers of children with ID reported that they were socially isolated due to caregiving their children and neglected their own social needs. Moreover, caregivers often neglect to receive mental health support for their own self-care needs.[18,19]

Consequently, since about half of children with CP have ID[20], more information is needed about the areas of life for children with CP who also have ID. Quality of life assessment is increasingly used as a mechanism to gain insight into a child's life, identify positive or challenging areas of life, and evaluate interventions. Psychopathology of caregivers may affect the QoL of children by causing inadequate care. Parents with psychiatric problems may subjectively evaluate their children's functionality more negatively. Moreover, it has been reported that caregivers of children with poor QoL experience more psychiatric problems than caregivers of healthy children.[13,21] In this study, we aimed to investigate the parent-proxy QoL of children with CP according to the severity of intelligence levels. In addition to QoL, we examined depression and anxiety in mothers of children with CP to identify challenges in CP children with severe ID.

Material and Methods

Participants and Procedures

In total, 37 children and adolescents (16 females and 21 males) between 4 and 18 years of age diagnosed with CP were recruited from a Pediatric Neurology Outpatient Clinic between April 2018 and September 2018. Also, 37 primary caregivers (all mothers) of children and adolescents with CP were invited to participate in the study. The participants were assessed by a child neurologist for the diagnosis of CP, their mother's literacy status, and abilities to fill out the questionnaires. Participants who reported any genetic disorder, severe visual and auditory impairments, neurological disorders except for epilepsy (e.g., acute cranial trauma, brain tumors) were not included in the study.

Ethical approval was obtained from the Ankara University Medical School Research Ethics Committee (ID-No: 04-215-18). Informed written consent was obtained from parents, and written assent was obtained from children and adolescents.

After the evaluation of the inclusion and exclusion criteria, sociodemographic and clinical assessments were performed. Age, gender, family income levels, gestational age, maternal age, paternal age, birth weight, prematurity, types of birth, and presence of epilepsy, receiving special education were recorded. The cerebral palsy type was determined as spastic (hemiplegic, diplegic, tetraplegic), dyskinetic (athetosis, dystonia), ataxic or mixed types according to the "Surveillance of European Cerebral Palsy Group".[22] Gross Motor Function Classification System (GMFCS) and Bimanual Fine Motor Function (BFMF) were used for motor function assessment of the children by the child neurologist. GMFCS consists of five levels; from Level I (the most independent motor function) to Level V (the most restricted voluntary control of movement and ability to maintain postures, the most dependent motor function).[23] BFMF is designed to evaluate the upper extremity performance in children's daily living activities. Manipulation and gripping ability are evaluated at five levels. Scoring of GMFCS and BFMF are mostly in parallel with each other.[24]

The children's intelligence levels were evaluated by the Turkish version of the Wechsler Intelligence Scale for Children-Revised (WISC-R), Stanford-Binet Intelligence Test, and Ankara Developmental Screening Inventory (ADSI). The children with cerebral palsy were divided into two groups according to their intelligence levels; (i) Normal Intelligence Quotient (IQ)-Mild Intellectual Disability (ID) and (i) Moderate-Severe ID. Quality of life of the sample was determined by their caregivers with the Pediatric QOL Inventory, Parent version (PedsQL-P). Maternal anxiety and depression levels were assessed by using Beck Anxiety Inventory (BAI) and Beck Depression Inventory (BDI).

Questionnaires

Pediatric QOL Inventory-Parent version (PedsQL-P): The PedsQL-P was used to assess health-related quality of life (HRQOL) during childhood. It has two subscales, psychosocial and physical health, and a total score, higher PedsQL-P scores indicate better HRQL. The reliability and validity of the PedsQL-P has been conducted in Turkish children and adolescents.[25,26]

Beck Anxiety Inventory (BAI): The BAI is a self-report scale used to assess the severity of anxiety symptoms. It is a Likert-type scale that is scored between "0" and "3" and comprises 21 items. Higher BAI scores indicate higher anxiety levels. The reliability and validity of the BAI has been conducted in Turkish populations.[27]

Beck Depression Inventory (BDI): The BDI is a self-report inventory used to assess the severity of depression symptoms. The scale is composed of 21 items that are scored between "0" and "3". Higher BDI scores indicate higher depression levels. The reliability and validity of the BDI has been conducted in Turkish populations.[28]

Intelligence Tests

Wechsler Intelligence Scale for Children-Revised (WISC-R) is used for intelligence assessment of children between the ages of 6 and 16 years.[29] The reliability and validity of the Turkish version has been conducted in Turkish children.[30] The intelligence level of the child is classified as normal, borderline, mild, moderate, and severe with WISC-R scores. Stanford-Binet Intelligence Test was used to assess children between the ages of 2 and 16 years and adapted to Turkish by Uğurel-Şemin.[31] The intelligence level is determined according to the standard scores in this test. Ankara Developmental Screening Inventory (ADSI) was used for determining the developmental levels of young children in Turkey.[32] It was administered to caregivers of children who were unable to express themselves, could not communicate, and could not be applied the WISC-R, Stanford-Binet test due to their intellectual levels. The ADSI includes 154 questions which assess 5 domains (language, cognitive, fine motor function, gross motor function, sociality, and self-care abilities) asked to the caregivers and are combined with the clinical observation of the child for determining their intellectual levels. The total scores of these subscales give the general intellectual-developmental levels which are determined according to normative data matched for age.

Statistical analysis

We used the Shapiro-Wilk test to analyze data that was not normally distributed. According to the normal distribution, descriptive statistics were presented as mean ± standard deviation or median (min-max). Count and percentages were used to describe categorical variables; Continuous clinical variables were analyzed using Independent t-tests or the Mann-Whitney U test according to their distribution characteristics in group comparisons. Differences in categorical variables between the groups were examined using Pearson's chi-squared and Fisher exact analysis. Data analysis was performed using Statistical Package for the Social Sciences (SPSS) version 23.0, (IBM SPSS Statistics; New York, USA), and all statistical tests were two-tailed with the significance level set at $\alpha = .05$. Spearman's correlation coefficient was calculated to investigate the association among the clinical measures.

Results

Thirty-seven participants were included in the study; 19 of them were children with moderate-severe ID (moderate: 13.5%, severe: 37.8%), 18 of them were children with normal IQ (32.4 %) or mild ID (16.2%). There were no differences in age, maternal age, paternal age, and family income levels between the two groups (p>0.05). Perinatal factors (gestational age, birth weight, prematurity) were not different in the groups; however, there were differences in types of birth, gender, and presence of epilepsy (p= 0.012, p= 0.012, p= 0.012, respectively). Children with CP received special education for learning needs (Normal IQ-mild ID group: 77.8%; Moderate-severe ID group: 73.7%), and for their motor disabilities (Normal IQ-mild ID group: 88.9%; Moderate-severe ID group: 84.2%).

The CP types examinations indicated that the frequency of the spastic type was highest in both groups, and most of these children were spastic bilateral hemiplegia (Normal IQ-mild ID group: 55.6%; Moderate-severe ID group: 47.4%). GMFCS level 2 was more frequent in both groups according to gross motor functions examination of the children. There were no differences in severity of gross motor function (level 1-3=non-severe; level 4-5=severe) between the two groups (p=0.197). The majority of the severe-moderate ID group was at BFMF level 4, while the normal IQ-mild ID group was at BFMF level 2. There was a significant difference in severity of bimanual fine motor function (level 1-3= non-severe; level 4-5= severe) between the two groups ($X2$ (1, N = 37) = 17.79, p <0.001). Sociodemographic and clinical characteristics of the sample are shown in Table I.

Total PedsQL-P scores, Psychosocial PedsQL-P scores, Physical PedsQL-P scores of children with CP did not differ between normal IQ-mild ID group and moderate-severe ID group (t(35) = -.809, p = .424; t(35) = .098, p = 0.922; t(35) = -.088, p = .930, respectively). There were no differences in maternal BAI scores (U = 151.00, z = –0.610, p = .558) and maternal BDI scores (U = 156.50, z = –0.7443, p = .663) between two groups. . Results of maternal psychopathology and quality of life scores between the two groups are shown in Table II.

The correlations among the perinatal factors, gross and fine motor functions, intelligence levels, maternal anxiety scores, maternal depression scores, and PedsQL scores were calculated. Psychosocial PedsQL scores had a moderate negative correlation with the maternal BAI scores. PedsQL scores did not correlate with the other variables included in the correlational analyses. There were also moderate positive correlations between the children's ages and maternal BDI scores. There were also moderate positive correlations between epilepsy and female gender, GMFCS levels, and high negative correlations among the intelligence levels and BFMF level scores. All of the bivariate correlations between the variables are presented in Table III.

Table I. Sociodemographic and clinical characteristics of the sample.

	Normal IQ-mild ID (n=18)	Moderate-severe ID (n=19)	*p*
Age (mean ±SD) (year)	9.88 ± 3.68	11.65± 4.92	0.226
Gender			0.012*
Male	14 (77.8%)	7 (36.8%)	
Female	4 (22.2%)	12 (63.2%)	
Maternal age (mean± SD)	37.74±7.58	39.68±6.33	0.404
Paternal age (mean± SD)	41.72±7.61	44.15±7.41	0.331
Family income			0.638
Low	8 (44.4%)	7 (36.8%)	
Middle-high	10 (55.6%)	12 (63.2%)	
Gestational age (weeks)	34.66±5.50	35.84±4.89	0.496
Birth weight (grams)	2435.55±1115.90	2644.73 ±952.26	0.543
Prematurity	10 (55.6%)	8 (42.1%)	0.413
Types of birth			0.012*
Vaginal birth	6 (33.3%)	9 (47.4%)	
C/S	12 (66.7%)	10 (52.6%)	
Epilepsy	4 (22.2%)	12 (63.2%)	0.012*
Special education for cognitive ability			0.538
Yes	14 (77.8%)	14 (73.7%)	
No	4 (22.2%)	5 (26.3%)	
Special education for motor ability			0.527
Yes	16 (88.9%)	16 (84.2%)	
No	2 (11.1%)	3 (15.8%)	
CP type			N/A
Spastic bilateral hemiplegia	10 (55.6%)	9 (47.4%)	
Spastic unilateral hemiplegia	5 (27.8%)	4 (21.1%)	
Dystonic	-	1 (5.3%)	
Ataxic	2 (11.1%)	2 (10.5%)	
Mixed	1 (5.6%)	3 (15.8%)	
GMFCS			N/A
Level 1	6 (33.3%)	2 (10.5%)	
Level 2	7 (38.9%)	8 (42.1%)	
Level 3	1 (5.6%)	1 (5.3%)	
Level 4	1 (5.6%)	3 (15.8%)	
Level 5	3 (16.7%)	5 (26.3%)	
BFMF			N/A
Level 1	8 (44.4%)	1 (5.3%)	
Level 2	3 (16.7%)	2 (10.5%)	
Level 3	6 (33.3%)	2 (10.5%)	
Level 4	1 (5.6%)	9 (47.4%)	
Level 5	-	5 (26.3%)	

CP: cerebral palsy, C/S: cesarian section, GMFM: gross motor function measure, BFMF: bimanual fine motor function, IQ: intelligence quotient, ID: intellectual disability, SD: standard deviations. N/A: not applicable. * p<0. 05.

Table II. Quality of life scores and maternal psychopathology scores of the sample.

	Normal IQ-mild ID (n=18)	Moderate-severe ID (n=19)	p
Psychosocial PedsQL-P (mean± SD)	58.17±21.12	57.51±19.30	0.922
Physical PedsQL-P (mean± SD)	56.67±22.20	57.36±24.78	0.930
Total PedsQL-P (mean± SD)	55.48±16.09	59.97±17.56	0.424
BAI (mother) (median)(range)	11.50 (1-38)	13.50 (0-47)	0.558
BDI (mother) (median)(range)	11.36 (2-35)	11.72 (0-25)	0.663

PedsQL-P: the pediatric quality of life inventory parent version, BAI: beck anxiety inventory, BDI: beck depression inventory, IQ: intelligence quotient, ID: intellectual disability, SD: standard deviations.

Table III. Intercorrelations among study variables.

	2	3	4	5	6	7	8	9	10	11	12	13	14
1.Age	-0.19	0.03	-0.08	-0.01	-0.06	0.07	-0.07	-0.08	0.29	0.34*	-0.18	0.12	-0.04
2.Gender		-0.07	-0.23	0.13	0.28	0.37*	-0.45**	0.33*	0.03	-0.14	0.20	0.23	0.25
3.Gestational Age			0.78**	-0.87**	-0.21	-0.00	0.05	-0.03	-0.11	0.03	0.25	0.21	0.19
4.Birth Weight				-0.74**	-0.27	0.04	-0.01	-0.18	-0.01	-0.01	0.09	-0.01	0.01
5.Prematurity					0.17	0.05	0.003	-0.08	0.03	0.003	-0.19	-0.05	-0.07
6.GMFCS						0.64**	-0.31	0.38*	0.11	0.13	-0.03	-0.02	0.04
7.BFMF							-0.72**	0.28	0.18	0.12	-0.13	-0.01	-0.05
8.Intelligence level								-0.38*	-0.30	-11	0.14	0.01	0.01
9.Epilepsy									-0.01	-0.09	0.00	0.05	0.16
10.BAI (mother)										0.65**	-0.41*	-0.03	-0.20
11.BDI (mother)											-0.32	0.11	-0.13
12.Psychosocial PedsQL-P												0.49**	0.72**
13.Physical PedsQL-P													0.81**
14.Total PedsQL-P													

GMFM: gross motor function measure, BFMF: bimanual fine motor function, BAI: beck anxiety inventory, BDI: beck depression inventory, PedsQL-P: the pediatric quality of life inventory. Parent version.* p<0. 05; ** p<0. 01.

Discussion

We compared parent-proxy quality of life, maternal anxiety, and depression between the normal IQ-mild ID group and moderate-severe ID group in children with CP. We hypothesized that parent-proxy QoL, maternal psychopathology would be significantly different between the two groups, and our findings did not support this. However, there were differences in gender, types of birth, and presence of epilepsy between the two groups. There was a significant difference in BFMF severity between the two groups; however, GMFCS severity was not different between the two groups. Children with CP at different intelligence levels have contradictory results of QoL studies. Our findings were in line with studies showing that QoL scores were similar for children with and without cognitive impairment.[33,34] However, another large study found that CP children with cognitive impairment have lower QoL scores in the social support domain, compared to CP children with normal intelligence.[8]

Four large studies reported that children with CP have poor QoL, compared to the general population in all domains. The scores in the physical, but not the psychosocial domains, correlated with the level of motor impairment.[8,33,35,36] In our study, QoL scores did not correlate with the level of gross and bimanual fine motor impairment. Furthermore, there was no correlation between maternal depression and parent proxy-reported QoL. In previous studies examining the relationship between parental depression and proxy reports of QoL, parental depression was negatively correlated with parent proxy-reported QoL, contrary to our results.[37,38] In our study, maternal anxiety scores correlated with the psychosocial domain of PedsQL-P, this result is consistent with previous studies which showed a negative correlation between BAI scores of mothers and the PedsQL P scores of their CP children.[39,40] Physical domains of QoL scores correlate well with the severity of motor impairment, but the psychosocial scores are low regardless of the impairment severity.[41] This finding underlines the importance of factors other than the severity of CP that may affect QoL in these children. According to our study results, maternal anxiety may be one of the factors affecting the psychosocial domain of QoL. In some studies, family variables such as parenting style and family functioning were found to be important factors affecting the psychosocial aspects of QOL of children with CP.[42,43] In future studies, parenting style, and family functioning can be examined along with maternal psychological status. In our study, the ages of children positively correlated with maternal BDI scores. It may be related to the increase in the mother's caregiving burden and the decrease in coping capacity as the child grows with chronic diseases.[44]

Presence of epilepsy correlated with a lower level of intelligence in our study. In Gabis and her colleagues' study[6], epilepsy was found in disabled individuals (33%), and both presence of epilepsy and GMFCS levels predicted 29.9% of variance in the IQ score. In our study, intelligence was associated strongly with BFMF but not GMFCS. CP with epilepsy is more often accompanied by ID than CP without epilepsy. Correspondingly, CP with ID is associated with a high risk of developing epilepsy.[45,46]

Our study has some limitations. It is a cross-sectional study, and had a small sample size, which precludes the determination of a causal relationship between clinical factors. Only the mother was evaluated as a caregiver. Familial characteristics such as family dynamics, parenting roles, and coping strategies were not evaluated. Emotional and behavioral assessment of children with CP were not applied, and structured psychiatric interviews for mothers were not implemented by researchers. Additionally, epilepsy rates are generally higher in the moderate-severe ID group as expected, and epilepsy could be an important factor that may affect the QoL of children with CP.

In conclusion, several biopsychosocial factors may be related to OoL in children with CP. Our results demonstrate that maternal anxiety was related to psychosocial QoL in children with CP. Maternal depression scores increased with the age of children with CP which may also indicate the social support needs of mothers with children of chronic diseases. We could not find an association with intelligence levels and QoL of children with CP, maternal anxiety, maternal depression in our study sample. Further studies may reveal the associations with other biopsychosocial factors in children with cerebral palsy of different intelligence levels by longitudinal study design with larger sample sizes.

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Author contribution

The authors confirm contribution to the paper as follows: study conception and design: EA, BGK; data collection: EA, YT, STT; analysis and interpretation of results: BGK, EA, YT, STT; draft manuscript preparation: EA, BGK. All authors reviewed the results and approved the final version of the manuscript.

Ethical approval

Ethical approval was obtained from the Ankara University Medical School Research Ethics Committee (ID-No: 04-215-18).

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Conflict of interest

The author(s) declare no competing interests.

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