

Functional profiles of children with cerebral palsy in a series of Tunisian cases

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ABSTRACT

Cerebral palsy (CP) is the most frequent cause of childhood impairment. Several factors including age and clinical status have been found to be influential on functional outcome of these children with CP. The aim of this study was to assess Tunisian children with CP in a population-based sample by characterizing their capabilities in numerous domains and scenarios in order to acquire a solid understanding of their abilities based on their clinical classifications and using common international functional measures. In addition, to identify the factors linked with functional status.

Methods : This is a cross sectional study that included children with CP. The clinical evaluation questionnaire includes clinical data and functional scales: the MACS (Manual Ability classification system), the FMS (Functional Mobility Scale), the CFCS (Communication Function Classification System) and the WeeFIM (Functional Independence Measure for children).

Results : A total of 80 Children with CP had been included. Fifty eight percent were boys, and had a mean age of 3.49 ± 6.55 years. CP was especially spastic (%75) and the spastic forms were bilateral in the majority of cases (36 cases) followed by hemiplegic forms in %30 of cases. Children were mainly classified as MACS level IV in %27.5 of cases, FMS level 5 in %37.5 of cases and CFCS level IV in %30 of cases. Total independence was achieved in %23 of cases according to the WeeFIM score. Age and clinical status, communication (CFCS) and mobility (FMS) were identified in multivariate analysis as predictors of functional outcome in children with CP ($p=0.000$).

Conclusion : Functional status was altered in Tunisian children with different subtypes of CP. Hemiplegic group members performed higher on both mobility and manual ability scores. The spastic bilateral group had more difficulty with mobility and hand function activities. Age, language and mobility were predictors of functional status.

Key words: Cerebral palsy, Manual Ability classification system, Functional Mobility Scale, Communication Function Classification System, Clinical status, Functional outcome, Tunisian children.

RÉSUMÉ

Introduction : La paralysie cérébrale (PC) est la cause la plus fréquente de handicap chez l'enfant. Plusieurs facteurs, y compris l'âge et la forme clinique, se sont avérés influencer l'état fonctionnel. L'objectif de cette étude était d'évaluer un échantillon de population Tunisienne incluant des enfants atteints de PC en utilisant des mesures fonctionnelles internationales communes afin de déterminer les facteurs liés à l'état fonctionnel.

Méthodes : Il s'agit d'une étude transversale incluant des enfants atteints de PC. Le questionnaire de l'évaluation clinique comprend des données cliniques et des échelles fonctionnelles : le MACS (Manual Ability classification system), le FMS (Functional Mobility Scale), le CFCS (Communication Function Classification System) et le WeeFIM (Functional Independence Measure for children).

Résultats : Quarante-vingt enfants atteints de PC ont été inclus. Cinquante-huit pour cent étaient des garçons et avaient un âge moyen de $3,49 \pm 6,5$ ans. La PC était surtout spastique (%75) et cette forme était bilatérale dans la majorité des cas (36 cas) suivie de forme hémiplegique dans %30 des cas. Les enfants étaient principalement classés niveau IV selon MACS dans %27,5 des cas, niveau 5 selon FMS dans %37,5 des cas et niveau IV selon CFCS dans %30 des cas. L'indépendance était totale dans %23 des cas.

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cas selon le score WeeFIM. L'âge, la forme clinique, la communication (CFCS) et la mobilité (FMS) ont été identifiés dans l'analyse multivariée comme des facteurs prédictifs de l'état fonctionnel chez les enfants atteints de PC ($p = 0,000$).

Conclusion : L'état fonctionnel était altéré chez les enfants atteints de différents types de PC. Les hémiplégiques avaient des meilleurs résultats pour les scores de mobilité et de capacité manuelle. La forme spastique bilatérale avait plus de difficulté pour la mobilité et les activités fonctionnelles de la main. L'âge, la communication et la mobilité étaient des facteurs prédictifs de l'état fonctionnel.

Mots clés : Paralyse cérébrale, Manual Ability classification system, Functional Mobility Scale, Communication Function Classification System, Forme clinique, Etat fonctionnel, Enfants Tunisiens.

INTRODUCTION

Cerebral palsy (CP) is the most prevalent cause of neurological disability in children (1). Variety of clinical presentations can be observed in children with CP ranging from children who can ambulate and handle objects independently to children who have severe limitations in mobility and manual abilities. Categorizing children with CP into cohesive groups based on functional abilities is beneficial. The use of functional profiles in clinical settings is intended to offer a comprehensive description of children with CP's skills, perhaps improving service planning and research (2). Several factors including age and clinical status have been found to be influential on functional outcome of these children with CP. In Tunisia, CP is an issue with uncertain scope, causes, and remedies. In reality, in Tunisia, there are no guidelines or research on the subject of CP children and outcome measures using numerous functional measures.

As a consequence, the objective of our study was to assess Tunisian children with CP in a population-based sample by characterizing their capabilities in numerous domains and scenarios in order to acquire a solid understanding of their abilities based on their clinical classifications and using common international functional measures. In addition, to identify the factors linked with functional status.

METHODS

This was a cross sectional study on children with CP followed in the Physical Medicine and Rehabilitation Department (PMR) at the University Hospital Fattouma Bourguiba of Monastir (Tunisia) from January 2019 to June 2020.

The parents of the children were informed about the study and provided written consents.

The inclusion criteria were as follows:

- Patients aged from 2 to 18 years
- Previously diagnosed with CP by their pediatric/physician or their Physiotherapist and have a medical file in the PMR department

Exclusion criteria were as follows:

- The following causes: head trauma sequelae, children treated for brain tumor
- Abnormalities caused by a single mental defect
- Spina bifida
- Prematurity-related transient motor disturbance
- Motor problems caused by a gradual degenerative illness (neuromuscular disease, metabolic disease)

The collection of socio-demographic and clinical data was carried out on pre-established forms including: gender, age, description of the CP classification, etiology of CP, weight at birth, term of pregnancy at birth, type of delivery, age of diagnosis of CP, number of siblings, marital status of parents, associated disorder, treatment team, schooling, physiotherapy, ergo-tracking.

Clinical and functional evaluations included:

- Manual ability classification system (MACS): The MACS reflects children's self-initiated manual capabilities to handle goods as well as their need for help or modification throughout everyday manual tasks (3). The MACS stresses effectiveness at household, school, and in the environment rather than capacity in a controlled situation. This scale categorizes youngsters from Level I (handles objects easily and successfully) to Level V (Has very limited ability to conduct simple activities).

- The Communication Function Classification System (CFCS): To define appropriate communication performance, the CFCS contains five levels (CFCS I, II, III, IV, V).

The CFCS offers a scalable and accurate categorization of communication performance and activity constraints for use in scientific and therapeutic settings (4).

- Functional Mobility Scale (FMS): The FMS was developed to classify functional mobility in children while accounting for the variety of adaptive equipment needed. The FMS is used to assess walking abilities across three distinct ranges: 50, 5, and 500 meters (5).

- The Functional Independence Measure For children (Wee-FIM): The Wee-FIM aims at evaluating disability and determining functional independence rates. The Wee-FIM assesses 6 functional aspects, which are separated into two categories: personal hygiene, sphincter awareness, movement / transfer, locomotion, communicating, and social interaction (Motor and Cognitive) (6).

Statistical analysis: The Statistical Package for Social Sciences (SPSS) version 21.0 has been used for data gathering.

The univariate analyses were used to determine whether characteristics were risk factor indicators for the Wee-FIM score. The logistic regression analysis was used in multivariate analysis to find factors that were significantly linked with Wee-FIM score. The univariate analysis factors with $p < 0.25$ would be included in the multiple regressions calculation. The adjusted R^2 , which represents the proportion of variable that is explained by the explanatory va-

riables, was used to assess the model's predictive potential. The odds ratio was estimated using a %95 confidence range. PO.05 was considered important unless otherwise specified.

The Wee-FIM was used as the dependent variable. CP disability categories, gender, age, related comorbidities and CFS-FMS-MACS were the independent factors in the regression model.

RESULTS

The demographic and clinical characteristics of patients are given in Table I.

The mean age was 3.49 ± 6.55 years and the sex ratio was 1.16. Seventy percent came from urban areas, while %30 came from rural areas. In %62.5 of cases, medical care was virtually evenly split between both the physiatrist and the paediatrician. Fifty percent of children were unable to attend school. The CP was especially spastic (%75) and the spastic forms were bilateral in the majority of cases (36cases) followed by hemiplegic forms (24 cases).

Of all the children with spastic bilateral form, diplegia was dominant in 20 % of cases, followed by triplegia in %15 of cases.

CP etiology varies essentially between prematurity (%26.3), perinatal asphyxia (%26.3), and other prenatal and postnatal causes (Table I).

Table I: Demographic and clinical of the study population (N=80)

Variable	Categories	frequency	Percentages
(Age (year mean \pm SD	± 6.55		
Age groups	2-4	26	32,5
	4-6	20	25
	6<	34	42,5
Gender	Female	37	46,3
	Male	43	53,8
Social Security	Yes	79	98,75
	No	1	1,25
Origin	Rural	24	30
	Urban	56	70
Family care	Parents live together	75	93,8
	Parents live together	1	1,3
	Orphan	4	5
Medical care	Pediatric	61	76,25
	Neurologist	3	3,8
	Psychiatrist	64	80
	General practitioner	1	1,3
	Pediatric and physiotherapist	50	62,5
Schooling	Ordinary class	25	31,3
	Specialized institution	15	18,8
	Schooling impossible	40	50
CP Etiology	Prenatal causes	7	8,75
	Postnatal causes	15	18,75
	Prematurity	21	26,3
	Perinatal asphyxia	21	26,3
	Unspecified	16	20
CP type	Spastic bilateral	36	45
	Hemiplegic	24	30
	Dyskinetic	11	13,8
	Ataxic	9	11,3
Spastic bilateral form	Diplegia	16	20
	quadriplegia	8	10
	Triplegia	12	15

SD: standard deviation, CP: cerebral palsy

The MAC describes children's self-motivated manual capacity to handle goods as well as the need

for help or modification throughout regular manual tasks. According to the figure %27,5 ,1 were classified as level IV. Level II was achieved in %25 of instances.

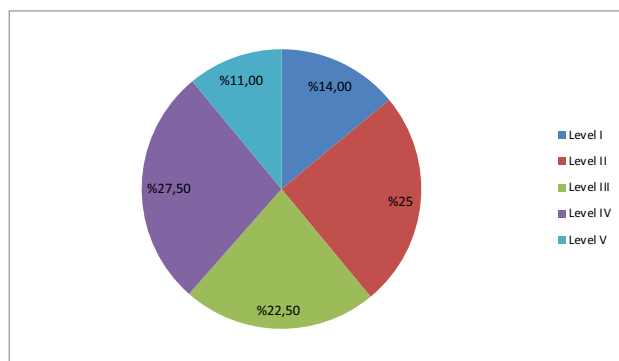


Figure 1: Scores of Manual Ability Classification System (N=80)

According to CSFS, level II was allocated to %30 of participants, whereas Level I was attributed to %26. Sixty percent were classified as verbal speakers, whereas %40 were classified as nonverbal communicators (Figure 2).

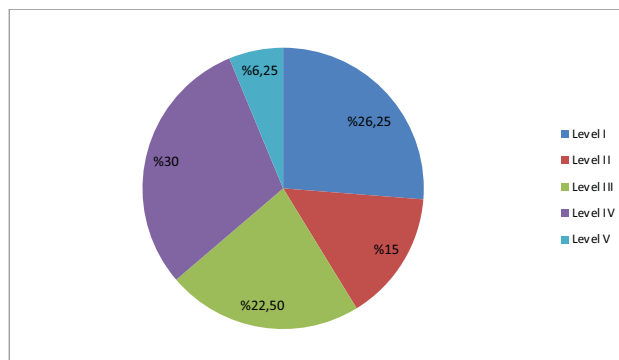


Figure 2: Scores of the communication Function Classification System (N = 80)

According to the FMS 500, the participants were categorised as level 5 in the majority of cases (%36) and level 1 in %32 of cases.

The Wee-FIM was used in our study for 80 children with CP. Our youngsters with CP had a total score of 64.81 on the CP scale.

As shown in table II, Self-care, mobility, and cognition sub-quotients were %52.7 ,%45, and %57, respectively. Only %11 were entirely independent (scoring 7-6), and %72.5 needed support with self-care skills (score 4-1).

Sixty percent needed assistance with mobility, whereas only %20 needed no assistance with daily mobility tasks.

Although nearly half of the children required assistance with speech and social interaction, 13 children required monitoring, and 23 children were entirely independent in cognitive capacity activities.

Table II: Average Functional Independence Measure scores for three domains for CP children in the research sample (N=80):

Domains	Mean score	SD	Quotient (%)	Need assistance (4-1 score) (%) n	Need supervision (5 Score) (%)n	No help needed (7-6 Score) (%)n
Self - care (/56)	25.27	16.9	45	(72.5) 58	(13.8) 11	(13.8) 11
Mobility (/35)	18.46	11	52.7	(60)48	(15)12	(25)20
Cognition (/35)	19.6	10.1	57	(55)44	(16.25)13	(28.75)23
Total (/126)	64.81	36.11	54	(62.5) 50	(15)12	(22.5)18

Table III shows the dispersion of youngsters across Wee-FIM, MACS, CFCS, and FMS levels by CP clinical topography.

Table III: Distribution of participant children's functional scale levels by CP subtypes (N=80):

Level/Quoting	Spastic bilateral	Hemiplegic	Dyskinetic	Ataxic	Total	Percent
MACS level (V)	4	0	1	4	9	11,25
MACS level (IV)	11	6	3	2	11	27,5
MACS level III	9	7	1	1	18	22,5
MACS level II	6	9	4	1	20	25
MACS level I	6	2	2	1	11	13,75
FMS 500 level (5-6)	6	16	6	2	30	37,5
FMS 500 LEVEL (3-4)	9	2	1	2	14	17,5
FMS 500 Level (1-2)	21	6	4	5	36	45
CFCS level (V)	1	1	0	3	5	6,25
CFCS level (IV)	14	3	3	4	24	30
CFCS level (III)	8	7	3	0	18	22,5
CFCS level (II)	4	6	2	0	12	15
CFCS level (I)	9	7	3	2	21	26,25
FIM Self-care level (4-1)	29	14	7	8	58	72,5
FIM Self-care level (5)	3	5	2	1	11	13,75
FIM Self-care level (7-6)	4	5	2	0	11	13,75
FIM mobility level (4-1)	26	7	7	8	48	60
FIM mobility level (5)	7	8	1	0	16	20
FIM mobility level (7-6)	2	3	1	0	6	7,5
FIM cognition Level (4-1)	20	10	7	7	44	55
FIM cognition level (5)	10	9	0	0	19	23,75
FIM cognition level (7-6)	6	5	4	2	17	21,25
Total	36	24	11	9	80	100

MACS: Manual Ability classification system, FMS: Functional Mobility Scale, CFCS: Communication Function Classification System, FIM: Functional Independence Measure

We found it useful to regroup CP subtypes into more homogeneous subgroups by classifying the resulted scores based on the level of independence and by 3 main domains: manual capacity, motor capacity and communication capacity. For the Wee-FIM self-care domain all four CP subtypes groups faced an important difficulty to perform in self-care tasks as %72 of all CP children were classified as level 1 to 4 of Wee-FIM. In the Wee-FIM Cognitive domain, %5 of spastic bilateral group (36/20) were classified as level 4-1. The hemiplegic group performed as level (4-1) in 24/10 of cases and level 5 in 24/9 of cases. The majority of children belonging to the spastic bilateral group needed help with mobility and transfer in 36/26 of cases as well as for the dyskinetic and ataxic group while the hemiplegic

group scored on the majority of cases as level 5 of the Wee-FIM.

The most important determinants of functional status were determined by studying the following factors: The Wee-FIM as the dependent variable, age, sex, CP subtypes, associated impairment, and presence of epilepsy, MACS score, CFCS score and FMS score as factors.

Simple linear regression was used in a univariate statistical analysis to investigate the factors that influence functional outcome of CP patients.

At the result of this chapter, a multiple linear regression model was used to assess the key predictors of functional outcome in children with CP. The factors added in the first model were those that were significant at the %25 level in the univariate analysis, and those that were kept in the final model were those that were significant at the %5 criterion. The adjusted R2, which represents the proportion of variation explained by the explanatory variables, was used to assess the model's predictive potential. The odds ratio was estimated using a %95 confidence range. If not mentioned differently, P 0.05 was considered significant.

The findings of a linear regression analysis with the functional independence measure (FIM) as the dependent variable are shown in Table VI.

Table IV: Factors linked with FIM score in cerebral palsy children (univariate analysis) (N=80):

Variables	β	p
Clinical status	-0.127	0.015
Age	0.242	0,00
Gender	-0.055	0.267
Concomitant impairments yes or no	-0.04	0.530
Epilepsy yes or no	-0.064	0.320
MACS score	-0.088	0.213
CFCS score	-0.381	0,0
FMS 500 score	0.493	0,0

MACS: Manual Ability classification system, CFCS: Communication Function Classification System, FMS: Functional Mobility Scale,

CFCS and FMS 500 scores were also shown to be independently related to the functional independence measure (FIM) (p=0.00).

These variables were found as dependent factors for Wee-FIM classification for children with severe

CP Clinical status ($p=0.015$), age ($p=0.00$ (Multiple linear regression analysis/ stepwise linear regression analysis) (Table V). Gender, concurrent disability, epilepsy, and MACS, FMS and CFCS were therefore unrelated to Wee-FIM scores.

Table V: WeeFIM statistically dependent variables in children with cerebral palsy in Tunisia (multivariate regression analysis) (N=80):

Domain	factors	Adjusted R2	β	CI %95	P
Wee-FIM	CFCS	%66.6	0.000	[2.864 ; 2.488]	1.000
	FMS 500	%61.7	0.001	[3.521 ; 2.979]	0.999
	Clinical status	%10.9	0.328	[2.014 ; 1.535]	0.735
	age	%6.5	0.402	[7.365 ; 5.685]	0.694

CI: confidence interval, CFCS: Communication Function Classification System, FMS: Functional Mobility Scale,

DISCUSSION

The CP was particularly spastic, and the majority of spastic forms were bilateral, followed by hemiplegic forms and dyskinetic forms. Diplegia predominated in %20 of the children with spastic bilateral form, followed by triplegia in %15 of the cases. Indeed, many studies have shown that the spastic form was the most common form, on which functional profiles and functional ability classifications were heavily influenced and improved over time (7).

Our study focused on categorising CP children in more homogeneous groups based on CP subtypes, by describing the results of functional outcome in each of the following scales: FMS, Wee-FIM, CFCS and MACS.

Children with spastic bilateral CP, the most common CP subtype, had more difficulty and less independence as their FMS level increased. None of the members of this group were able to perform at a level 6 in FMS 500, to which the majority of our young people belong. The hemiplegic group performed consistently from 5 to 500 meters with no significant change, as did the ataxic and dyskinetic groups.

The latest result could be explained by the change of movement tactics utilized by children with CP at changing ranges. As a result, although the changing need of assistance differ from one range to another does not have a heavy impact on the nature of disability, it obviously contributes to a changing score in every condition reflecting the FMS's primer objective.

As previously indicated, scientific studies in the literature (9,8) support these conclusions.

According to Wee-FIM, results vary from CP subtypes as these children do not have the same capacities in the three domains. We observed that spastic bilateral youngsters face more serious challenge in self-hygiene and care tasks. This can be explained that upper limbs are more functionally limited. The hemiplegic youngsters on the other hand had bet-

ter results. That also can be explained by having an alternative upper limb that can perform efficiently. Ataxic and dyskinetic children struggled with self-care tasks and mobility but scored better in cognition domains. Similar results were found by Adrienne R. and al by performing a study on 73 young patients with CP by using Wee-FIM. These researchers have conducted that more than %50 of young patients could accomplish functional autonomy; even %62 of them could achieve everyday tasks either by themselves or with supervision of their care givers (10). In terms of CP subtypes of our study, the spastic bilateral group performed mainly as MACS level IV than as level III while the hemiplegic and dyskinetic groups performed best as level II, Nonetheless, children with ataxic form performed at MACS level V. These results are similar to the research done by Ann-Christin Eliasson and al others, a neuropsychiatric practitioner at Astrid Lindgren Children's Hospital Sweden. Their research conducted hemiplegic young patients have mostly scored as level II and III whereas youngsters with diplegia performed mainly as Levels I to IV. Furthermore, dyskinetic group members ranked mostly as they scored as level IV and V (11).

The spastic bilateral group performed mostly at level IV of the CFCS scale, whereas the hemiplegic group performed equally at levels I, II, and III. The same observation was made for the dyskinetic group, despite the fact that the ataxic group was predominantly classified as level IV.

A similarity between our results and other studies like the research done by Kate Himmelmanna and al that have evaluated 68 youngsters belonging to the unilateral group, 35 were classified as unilateral spastic members and the rest (19 cases) were in the dyskinetic group (12).

In Our study, in the multivariate analysis, there is no significant association between MACS and Wee-FIM ($p=0.1$) despite the fact that our population had spastic had in %55 of cases a spastic upper limb impairment and %13.8 were dyskinetic. Manual ability has no influence on functional outcome of children with CP. This could be explained by the fact that the majority of our patients are children with diplegic CP. However, other studies have found a strong correlation between Wee-FIM and MACS score. In the study by Mintaze Kerem Gunel et al has found a correlation between the MACS and the Wee-FIM subscales according to CP subtypes ($p < 0.01$). In this study, authors have used the chi-square test, %60 of children have spastic upper limb dysfunction and %40 were classified in level 4 and 5 (poorly handles objects) (13).

Moreover, a study by Byoung-Hee Lee, performed on 77 children with spastic CP demonstrated a strong correlation between MACS score and Wee-FIM scores ($r = 0.828$, $p < 0.001$).

Our study demonstrated an important correlation between Wee-FIM total score and CFCS scores by using multivariate analysis ($p= 0.00$). These results

were similar to a study by Kavlak Erdogan and al (2019) and Gokcen Akyurek and al (2018) as they found a significant correlation between CFCS levels and Wee-FIM total score (16 ,15).By using multivariate analysis ,we found a strong correlation between Wee-FIM and FMS score ($p=0.000$) .A study by Corinne Ammann-Reiffer (2019) performed on 64 children with CP showed a positive correlations between changes in FMS scores and changes in outcomes of other measures of functional activity assessed with Wee-FIM score ($r=0.83-0.75$; $p<0.005$) (17) . Our study found that there was a significant amelioration in Wee-FIM scores by more increasing age ($p=0.000$). This may be explained by the amelioration of adaptation capacities by more advanced age and the effect of physical rehabilitation management. Many studies support these findings: Sheila S. N.Wong and al (18) (2007), Meigenliu and al (1998) (19), Michael E. Msall and al (20) (1994), So Yean Park (21) (2010) and Virginia Wong and al (22) (2015). As recommendation, it is essential to continue the rehabilitation management (physiotherapy, equipment, botulinum toxin injection) throughout growth of these children to improve their functional capacity.

CONCLUSION

Functional status was altered in Tunisian children with different subtypes of CP. Hemiplegic group members performed higher on both mobility and manual ability scores. The spastic bilateral group had more difficulty with mobility and hand function activities. Age, clinical status, communication and mobility were identified as predictors of functional status in this population. Therefore, it is essential to continue the rehabilitation management throughout growth of these children to improve both motor and cognitive outcome.

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