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Bimanual hand use in children and adolescents with unilateral spastic cerebral palsy: an exploratory study

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Abstract

Background: Individuals with unilateral spastic cerebral palsy (USCP) often show difficulties using their hands during activities of daily living.

Objective: To investigate the factors that interfere with hand use during bimanual activities in children and adolescents with USCP.

Methods: We conducted a cross-sectional study with 102 children and adolescents with USCP, aged 6 to 18 years. We collected information with the caregivers about the classification of the child's manual ability, according to the Manual Ability Classification System (MACS); child's age; side of the involvement; Children's Hand-Use Experience Questionnaire- CHEQ2.0. Cluster analysis identified groups of children and adolescents who performed CHEQ activities with or without assistance. Multiple linear regression analyses identified the contribution of the factors: age, sex, MACS level, side of hemiparesis, and clusters of assistance, on the outcomes of efficacy, time, and feeling bothered.

Results: MACS and clusters of assistance explained the variance in efficacy ($p<0.05$; R^2=0.31) and time ($p<0.05$; R^2=0.37). MACS explained 22% of the variance in feeling bothered. Children and adolescents with increased difficulty to perform activities that involve hand use (i.e., MACS III) and who receive assistance during most bimanual activities showed less efficacy of use, were slower in their performance, and presented greater feeling of being bothered.

Conclusion: Assistance in bimanual activities and MACS level contributed to explain the efficacy of use, time, and feeling bothered in performing bimanual activities. Intervention strategies aimed at promoting the performance of bimanual activities in the daily routine of children with USCP should consider these outcomes.

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Introduction

Activities of daily living involve actions that require the combined use of both hands.[1,2] Such activities can be challenging for children and adolescents with unilateral spastic cerebral palsy (USCP).[2,3] They often show difficulties in reaching, grasping, manipulating, and releasing objects with the affected extremity, compromising their bimanual performance.[2-8] However, these children and adolescents can use different strategies to perform bimanual tasks in different contexts.[2,3,9-12] They can opt whether or not to use the affected hand, use other parts of the body (e.g., elbow, chin) to stabilize, and/or seek parental assistance to perform bimanual activities.[3,8] These strategies may interfere, both positively (e.g., task completion) or negatively (e.g., frustration, need for extra time), in their daily routine.

Limitations for using the upper affected extremity by children and adolescents with USCP during bimanual performance may result from deficits in motor and sensory components.[4,9,13] Klingels et al.[11] analyzed the impact of motor and sensory factors in the performance of bimanual activities of children with USCP. In this study, grip strength explained 76% of bimanual performance.[11] Similarly, Sakzewski et al.[5] showed that stereognosis explained 75.1% of the bimanual performance of children with USCP. The integrity of interhemispheric connectivity (i.e., corpus callosum) is also predictive of bimanual coordination.[12] The bimanual performance of children and adolescents with USCP in daily activities can also be influenced by contextual factors (i.e., environmental and individual factors).[4] Family structure (single-parent families), low socioeconomic level, and high level of caregivers' stress interfere with the performance of activities of daily living of children with USCP.[4]

Previous studies analyzed children's and adolescents' perceptions regarding their performance in bimanual activities. Skold et al.[4] conducted a qualitative study with adolescents with USCP, who reported that hand use is influenced by environmental (e.g., expectations of people who integrate the contexts) and personal factors (e.g., concentration and choice of individualized strategies). Oliveira et al.[14] showed the positive influence of interest in children's bimanual performance. Lidman et al.[9] explored the experiences of children and adolescents with USCP in learning bimanual activities. The participants described that the process of learning bimanual activities involves the awareness of their own abilities as well as the boundaries of their disability, the possibility to develop strategies and practice activities, considering the extra time needed, and the support from people around them.[8] Brandão et al.[15] analyzed the characteristics of the home and clinical environments that permeate the use of the affected extremity by children with USCP. The lack of organization of the family routine, added to the insufficient time and negative attitudes of the child (e.g., low sense of self-efficacy, lack of interest) to perform daily activities, led to the need for parents to provide excessive assistance to their children.[15]

Despite the fact that previous qualitative studies highlight the contribution of personal and contextual factors in the bimanual performance of children with USCP, most of the literature used objective measures to identify the contributors for hand function of children with USCP in outcomes such as dexterity and efficacy of the affected hand use in bimanual activities.[11,16] Subjective parameters such as perception of efficacy, perception of time spent in performing activities compared to other children, and feeling bothered during performance, which are important outcomes to the understanding of children's bimanual performance, have not been properly explored.[16,17] These outcomes may be influenced by the child's age, manual ability, and the provision of caregiver assistance. The aim of this study was to investigate the influence of age, sex, side of hemiparesis, classification of hand function, and provision of assistance during bimanual activities on the efficacy of hand use, time spent, and feeling bothered with the performance by children and adolescents with USCP.

Methods

Study design

This was an observational cross-sectional study.

Participants

Families of 102 children and adolescents with USCP being assisted in pediatric rehabilitation centers, clinics, and institutes in Brazil, such as Associação Mineira de Reabilitação (AMR), Associação de Deficientes Físicos de Poços de Caldas (ADEFIP) and Instituto Nossa Casa, were a sample of convenience.

The participants were caregivers and their children or adolescents with USCP. Children had a medical diagnosis of USCP; ages between 6 and 18 years; hand function classified as levels I, II, or III of the Manual Ability Classification System (MACS).[18] According to Eliasson et al.,[18] children/adolescents classified at MACS level I can perform manual activities independently. Those classified at level II are able to handle different objects, but with reduced movement quality or speed. Children classified at level III have difficulties to manipulate objects of daily routine and need preparation or support.[18] Children who had undergone upper extremity surgery in the previous 12 months were excluded.

Procedures and instrumentation

This study was approved by the Research Ethics Committee from Universidade Federal de Minas Gerais (CAAE: 62,319,616.8.0000.5149). Prior to the beginning of data collection, the caregivers of the children and adolescents invited to participate provided informed consent. Data were collected from August 2019 to December 2020. At the rehabilitation centers, therapists selected potential families

who had children with the diagnosis of USCP. Moreover, social media advertisements were posted by Instituto Nossa Casa . Families who agreed to participate, contacted the researchers in person at the rehabilitation centers. Those who chose the online data collection, were contacted to certify the inclusion and exclusion criteria in the online meeting.

We interviewed caregivers of children/adolescents with USCP, in person or online, according to their preference. The following information was collected: sex of the child/adolescent; age of the child/adolescent; side of hemiparesis; classification of the manual ability of the child/adolescent, according to the Manual Ability Classification System (MACS).[18] In addition, caregivers answered the Children's Hand-Use Experience Questionnaire- CHEQ 2.0.[19]

The CHEQ 2.0 is an online questionnaire (available at www.CHEQ.se) translated and adapted to Portuguese − Brazil.[19] This test is validated for individuals with USCP (6 to 18 years)[20,21] to assess their experience using the affected extremity during the performance of bimanual activities, according to children (>8 years) or caregivers.[15] The CHEQ is composed of 27 daily living activities that involve the collaborative use of both hands (e.g., tying shoelaces, opening a bag).[19] For each task, the caregiver indicates whether or not the child/adolescent receives assistance to perform the activity.[19] If the task is performed independently, the respondent also indicates whether the activity is performed with one or two hands.[19] In addition, the caregiver answers three sub-questions for each activity, assessing the use of the affected hand regarding three outcomes: (a) efficacy of the use of the affected hand (1: bad, 4: good); (b) time spent by the child/adolescent to perform the activity compared to peers (1: considerably longer; 4: equally long); and (c) feeling bothered with their performance (1: bothers them a lot, 4: it does not bother them at all). For each outcome, raw scores are transformed into continuous scores (i. e., logits) in a 0−100 scale.

Data analysis

We collected information on the children/adolescents including sex, age, manual ability classification (MACS), and side of hemiparesis. To understand the assistance profile provided by caregivers in the CHEQ 2.0 activities, we conducted cluster analysis, with the complete linkage method.[22] Cluster analysis classifies individuals into groups, based on information from a set of variables and maximizes similar intragroup information and intergroup differences.[22] In the present study, the caregiver's answers to how the child performed each activity (with one hand, with two hands, receives assistance) was dichotomized into: child receives help, child performs the activity independently.

Subsequently, the Shapiro-Wilk test was used to test data normality.[22] The Kruskal-Wallis test[22] was used to identify differences in efficacy scores, time, and feeling uncomfortable with performance in relation to sex, age, side of hemiparesis, MACS level, and care clusters.[22] Multiple linear regression analysis using the Stepwise method[22] examined the association among the variables sex, age, side of hemiparesis, MACS level, and clusters of assistance with each of the CHEQ 2.0 scales: efficacy, time, and feeling bothered.

Table 1 Descriptive characteristics of children and adolescents with unilateral spastic cerebral palsy (USCP) (n = 102) regarding sex, side of hemiparesis, age, manual ability, as well as CHEQ outcomes (efficacy, time, feeling bothered).

Children's/ adolescents' characteristics	
Sex[1]	
Boys	40 (39.2%)
Girls	62 (60.8%)
Age (years)	
Range	6- 17.7
Mean (SD)	10.8 (3.40)
MACS[1]	
I	16 (15.6%)
II	61 (59.8%)
III	25 (24.6%)
Side of hemiparesis[1]	
Right	42 (41.2%)
Left	60 (58.8%)
CHEQ 2.0: Efficacy (logits)	
Mean (SD)	44.4 (20.2)
CHEQ 2.0: Time (logits)	
Mean (SD)	45.8 (18.1)
CHEQ 2.0: Feeling bothered (logits)	
Mean (SD)	61.1 (23.7)

SD: standard deviation; MACS: Manual Ability Classification System; CHEQ: Children's Hand Use Experience Questionnaire.
[1] numbers indicate frequency (%).

The Spearman's correlation coefficient estimated the correlation between the three CHEQ 2.0 scales (efficacy, time, and feeling bothered).[22] All statistical analyzes were performed using Stata software, version 14.2, with a significance level of α=0.05.

Results

The descriptive characteristics of caregivers and children/ adolescents with USCP are presented in Table 1.

Clusters of assistance

Cluster analysis identified three groups of children and adolescents, considering the use of one or two hands during bimanual activities versus the need for assistance (Table 2). Cluster 1 included children and adolescents who performed most bimanual activities independently. Cluster 2 was composed of children and adolescents who performed most bimanual activities independently, but received help in more complex activities (e.g., buttoning pants, opening popsicle wrappers, cutting food on the board, peeling oranges, opening packages, removing protective paper, cutting meat, opening milk carton, tying shoelaces). Finally, cluster 3 consisted of children and adolescents who needed assistance to perform most bimanual activities. There was a significant difference among clusters in relation to age (p = 0.0008) and manual ability (MACS) (p<0.0001). (Table 2)

Table 2 Age and MACS level of children/adolescents in the clusters of assistance.

Cluster	N	Age Range	Mean Age (SD)	MACS I	MACS II	MACS III
I	54	6- 17.6	12 (3.6)	15	35	4
II	37	6- 16.5	9.9 (2.8)	1	23	13
III	11	6.7- 12	8.3 (1.6)	0	3	8
Difference among clusters			p = 0.0008	p = 0.0001		

MACS, Manual Ability Classification System.

Efficacy, time, and feeling bothered to perform bimanual activities

The Shapiro-Wilk test showed that the variables efficacy (p = 0.204), time (p = 0.087), and feeling bothered (p = 0.091) followed normal distribution. The assumptions of normality of residual homoscedasticity were valid for these outcomes, and no multicollinearity was observed between the covariates included in the model (mean VIF=1.60).

The results of the linear regression analysis are presented in Table 3. The model showed a significant relationship among the efficacy of hand use and the MACS and the clusters of assistance. These two combined variables (i.e., MACS and clusters of assistance) explained 31.18% of the efficacy variability. For the time spent outcome, significant correlations were observed for MACS and clusters of assistance. These two variables together explained 37.54% of the time variability. Regarding the analysis of feeling bothered, child's manual ability (MACS) explained 22.76% of the outcome variability. The variables sex, age, and side of hemiparesis did not contribute to additional explanation of any investigated outcome.

Discussion

This study investigated factors that contributed to explain the efficacy of using the affected upper extremity, the time spent, and feeling bothered to perform bimanual activities of children and adolescents with USCP. The results showed that the child's manual ability classification (e.g., MACS) and clusters of assistance were significant predictors of the investigated outcomes. The variables sex, age, and side of hemiparesis did not contribute to an additional explanation of any of the models.

To our knowledge, this is the first study which analyzed the influence of manual classification on outcomes of efficacy, time, and feeling bothered through the CHEQ 2.0. Previous studies have analyzed the relationship between the classification of hand function (i.e., MACS) and outcomes related to manual dexterity[23] and assisting hand performance in bimanual activities.[20,21] In these studies, children classified as MACS level III had lower scores than those classified as MACS I and II regarding the performance of the assisting hand in bimanual activities, with the use of the Assisting Hand Assessment (AHA),[24,25] corroborating our results. Although both instruments, AHA and CHEQ 2.0, provide

Table 3 Linear regression analysis for CHEQ outcomes (efficacy, time, and feeling bothered scales).

| Outcomes	Variables	VIF	Coefficient	p-value	CI (95%)
Efficacy (CHEQ) (0−100); R^2=0.31	Sex	1.08	2.23	0.530	−4.8, 9.2
	Age	1.2	−0.04	0.938	−1.0, 1.1
	Side of hemiparesis	1.06	−56	0.871	−7.4, 6.3
	[1]MACS II	2.15	−16.65	0.001*	−26.5, −6.8
	[1]MACS III	2.74	−19.72	0.003*	−32.4, −7.0
	Cluster II	1.43	−13.88	0.001*	−22.0, −5.7
	Cluster III	1.56	−20.5	0.004*	−34.4, −6.6
Time (CHEQ) (0−100) R^2=0.37	Sex		−3.07	0.312	−9.1, 2.9
	Age		−0.40	0.383	−1.3, 0.5
	Side of hemiparesis		−3.2	0.283	−9.1, 2.9
	[1]MACS II		−15.93	0.0001*	−24.4, −7.5
	[1]MACS III		−15.53	0.005*	−26.4, −4.7
	Cluster II		−12.28	0.001*	−19.3, −5.3
	Cluster III		−26.41	0.0001*	−38.2, −14.6
Feeling bothered (CHEQ) (0−100) R^2=0.22	Sex		0.19	0.965	−8.5, 8.9
	Age		0.40	0.546	−0.9, 1.7
	Side of hemiparesis		−2.03	0.638	−10.6, 6.5
	[1]MACS II		−14.27	0.023*	−26.5, −2.0
	[1]MACS III		−15.43	0.054	−31.2, 0.3
	Cluster II		−5.29	0.302	−15.4, 4.8
	Cluster III		−32.49	0.0001*	−49.7, −15.3

MACS: Manual Ability Classification System; CHEQ: Children's Hand Use Experience Questionnaire, CI: Confidence Interval.
* statistical significance.

A
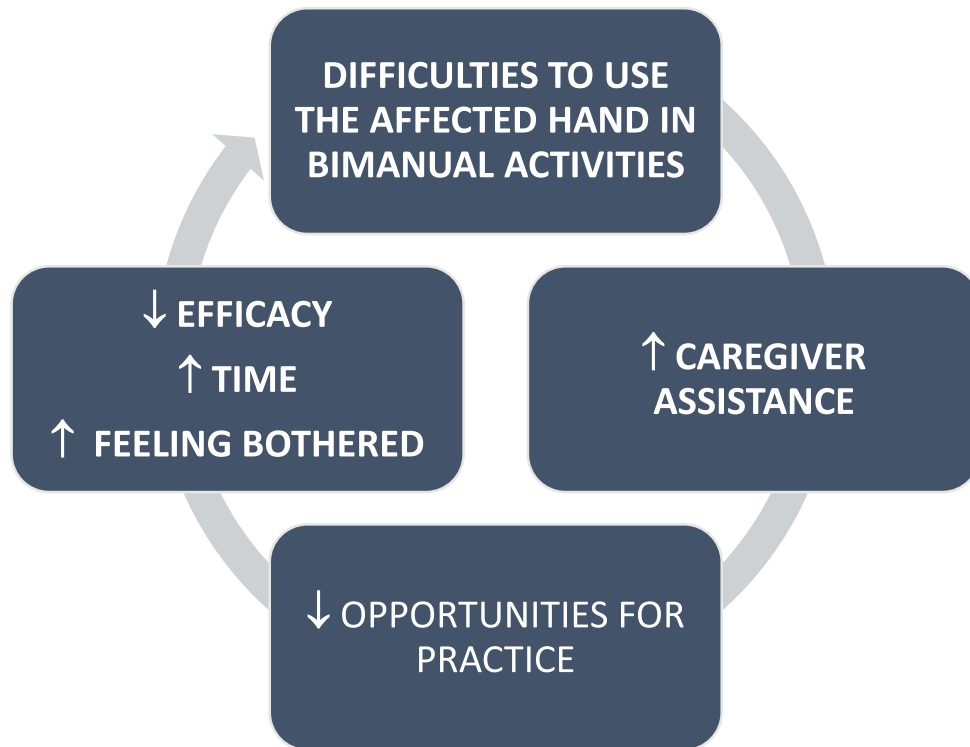
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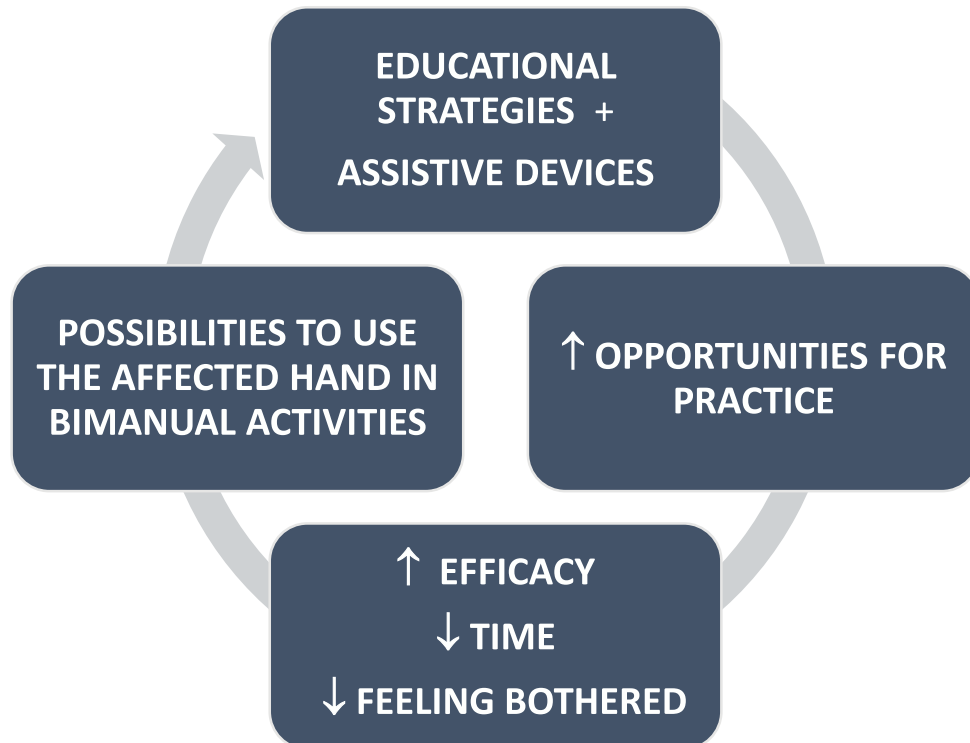
Fig. 1 Proposed cycles of the relationship between assistance provision and bimanual performance of children/adolescents with unilateral spastic cerebral palsy (USCP) (A), as well as of intervention strategies to promote bimanual performance (B).

information about assisting hand use in bimanual tasks, the AHA test is administered directly to the child, and it requires formal training. The CHEQ 2.0, in turn, is a questionnaire that assesses caregivers'/adolescents' perception of efficacy, time, and feeling bothered with their performance in bimanual activities in their daily living contexts.[26] As an online questionnaire available without costs, the CHEQ may be more feasible to use in clinical practice.[16] Thus, the combined use of these instruments may add valuable information about the child's performance in bimanual activities.

The provision of assistance explained the lower efficacy, longer time, and greater feeling bothered of the child/adolescent in the performance of bimanual activities. There is a clinical assumption guiding practice that the provision of assistance is a consequence of the child's limitations in performing activities. However, previous studies have questioned the linear relationship between the abilities' repertoire of children with CP and caregivers' provision of support.[14] In fact, provision of support seems to be context-dependent, and children may receive various types and amount of support based on others' attitudes.[4,14] According to Eliasson et al.,[18] children classified as MACS level III often need preparation or supervision in the performance of activities due to their limited repertoire. While the provision of assistance may be necessary, excessive help can reinforce the child's low sense of efficacy.

When parents offer excessive assistance during the performance of bimanual activities, there is a decrease in the opportunities for the child to experience these activities, which may limit their development of alternative solutions for more effective performance. Fig. 1 illustrates the cyclical relationship between the provision of assistance and the bimanual performance of children with USCP, as well as ways to overcome the negative aspects of this cycle. The assistance provided to children/adolescents with USCP during bimanual activities reduces their opportunity to practice. The lack of sufficient practice impacts on children's confidence to use their hands. Educational strategies targeting caregivers, associated with the use of assistive technology resources, may help the child/adolescent practice bimanual activities. Specifically, informing parents on alternative ways to foster the child to use their affected extremity, even with the use of compensatory strategies (e.g., support of objects using parts of the body, help with surfaces), and encouraging the child to discover active problem-solving solutions (e.g., verbal, visual cues) may lead to greater child involvement in bimanual activities.[7]

In our study, age was not a variable that added additional explanation to the efficacy of use, perception of time, or feeling bothered with performance. Previous studies have documented significant differences on longitudinal bimanual performance of children classified at different levels of manual function (i.e., MACS).[24-29] Holmefur et al.[25] and Klevberg et al.[28] observed that children with USCP showed greater gains in bimanual performance over the first three years of life. According to Klevberg et al.[28] and Eliasson et al.,[27] the developmental trajectories of bimanual performance among children classified as MACS level I and II showed gains up to 5 years of age. Children classified as MACS level III showed stability of skills at 7 years of age.[29] It is possible that child's age shows a direct influence on bimanual performance in the first years of life. During school age and adolescence (i.e., age range of our sample), the effect of age may occur indirectly, and environmental factors may be more relevant to explain the use of the upper extremity in bimanual activities.

This study has some limitations. It is important to consider that the analysis of efficacy, time, and feeling bothered were reported in this study by caregivers, who completed the CHEQ 2.0. In the present study, we chose to interview caregivers, to include parents of children as young as age 6 years. It is possible that older children and adolescents may have different perspectives of their performance in bimanual activities. Future studies may investigate the perception of children and adolescents, to capture the individual needs of these individuals.

Conclusion

This study investigated the factors that interfere in the efficacy of use, time, and feeling bothered in performing bimanual activities by children and adolescents with USCP. While the provision of assistance from caregivers and the child's/adolescent's manual ability significantly contributed to these outcomes, age, sex, and side of hemiparesis did not. Intervention strategies aimed at fostering bimanual performance in activities of daily routine of children with USCP should consider the efficacy, time spent in use of the affected upper extremity, as well as the child's feeling bothered during such activities. It is possible that educational strategies aimed to children and caregivers and the use of adaptations could contribute to improve children's performance in daily living activities.

Conflicts of interest

The authors declare no conflicts of interest.

Acknowledgements

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