Dystonia and choreoathetosis presence and severity in relation to powered wheelchair mobility performance in children and youth with dyskinetic cerebral palsy

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Abstract

Introduction: Power wheelchairs (PW) with head/foot steering systems are used as an alternative to joysticks in children with severe dyskinetic cerebral palsy (DCP). Mobility training programs are unstandardized to date, and insight on dystonia, choreoathetosis, and mobility performance may lead to greater independent mobility.

Objective: To map the presence and severity of dystonia and choreoathetosis during PW mobility in DCP and their relation with mobility performance.

Methods: Ten participants with DCP performed four PW mobility tasks using a head/foot steering system. Dystonia and choreoathetosis in the neck and arm regions were evaluated using the Dyskinesia Impairment Mobility Scale (DIMS). PW mobility performance was assessed using time-on-task and the number of errors during performance. The Wilcoxon-signed rank test and the Spearman's correlation coefficients were used to explore differences and correlations.

Results: Median levels of dystonia (83.6%) were significantly higher ($p < 0.01$) than median levels of choreoathetosis (34.4%). Positive significant correlations were found between the Arm Proximal DIMS and the PW mobility experience ($r_s = -0.92$, $p < 0.001$), and between the Arm Distal DIMS and the number of errors ($r_s = 0.66$, $p = 0.039$) during mobility performance.

Conclusions: Dystonia is more present and severe during PW mobility than choreoathetosis. The hypertonic hallmark of dystonia may mask the hyperkinetic hallmark of choreoathetosis, resulting in lower median levels. Results may suggest that with an increase in driving experience, children with DCP adopt deliberate strategies to minimize the negative impact of arm overflow movements on mobility performance, however, future research with bigger sample size and additional outcome measures is strongly encouraged.

Keywords: dyskinetic cerebral palsy, dystonia, choreoathetosis, powered wheelchair, mobility, performance

1. Introduction

Dyskinetic cerebral palsy (DCP) is the most limiting type of cerebral palsy (CP) and the second most common type of CP after spastic forms which is characterized by abnormal postures or movements due to impaired muscle tone regulation and impaired movement coordination.¹⁻³

Dystonia and choreoathetosis are the two predominant movement disorders which comprise DCP.³ Both movement disorders are often simultaneously present with typically higher values of presence and severity during activity as opposed to rest.^{4,5} Additionally, dystonia and choreoathetosis can manifest as overflow movements.^{6,7} Overflow is the unintentional contraction of muscles different from the ones used in goal-directed movements, resulting in an unwanted movement associated with an intended movement.⁷

More than 70% of children with DCP are classified within the highest levels of limitations in gross motor functioning and fine manual abilities.^{2,8,9} Consequently, the majority of people with DCP require the use of powered wheelchairs (PW) for independent mobility and/or are fully dependent on care-givers assistance for passive mobility.¹⁰ Access and opportunities to personal mobility with the greatest independence possible is considered a basic human right, and of vital importance toward good well-being and quality of life.¹¹ Indeed, in people with severe motor limitations, improvements towards more independent mobility have been reported to positively affect their cognitive, psychosocial, emotional, and perceptual development, and to increase independent exploration, activity and participation levels, social interaction, play skills, levels of curiosity, confidence, and motivation.^{10,12-16}

People with severe restrictions in arm and hand control and coordination^{16,17}, such as children with DCP face an additional challenge to independent mobility, as it limits the use of widely available PWs with conventional joysticks. Thus, they may benefit from alternative powered wheelchair control interfaces like switches, sip-n-puff or head arrays.^{13,16-18} Some children with DCP who use PW might be able to achieve independent mobility indoors and outdoors after training, despite significant challenges in terms of terrain, infrastructure and other potential hazards.^{12,16} As such, the use of PWs could enhance their activity and participation at school, outdoors, and in the community, however, several burdens and challenges are present.^{16,19,20} In this respect, clinical practice supports the use of an alternating steering method like the use of head/foot steering wheelchairs, whereas basal movements of the head and the feet seem to be better controlled in DCP than the more difficult control movement of the arms.¹ That is, children with DCP could use their head to steer the wheelchair to the right and to the left, and their feet to accelerate and brake.²⁰ To date, however, PW mobility training with head/foot steering wheelchairs in DCP is solely based on clinical expertise where lack of evidence-based knowledge results in unstandardized mobility training and long learning curves.^{13,21,22}

A previous study in CP participants with severe bilateral involvement showed that the level of power mobility independence is related to the length of training time and not to the participants' intellectual and functional abilities.²³ This study had only three participants with DCP and the measures of functional abilities did not include any information on dystonia, choreoathetosis, and their relation with driving competences. Dystonia and choreoathetosis are functionally limiting movement disorders, and from the perspective of body function and structure following the International Classification of Functioning, Disability and Health (ICF) model²⁴,

increasing insights into their presence and severity during various PW mobility tasks may be an added value in standardizing mobility training programs toward increased independent mobility in DCP.PW users need to develop a set of skills necessary to maneuver with the wheelchair safely and competently, thus the assessment of user's driving abilities are equally important in evaluating the efficiency of the mobility training program and the learning progress over time.^{25,26} Based on a recent review, PW mobility performance is best evaluated in terms of speed (often defined by the time-on-task); and in terms of accuracy. Accuracy is typically defined depending on the mobility task, e.g. deviations from a track when asked to follow a track on the floor, number of boundary violations when asked to drive within boundaries, number of non-task related stops when asked to drive without stopping.²⁵

In order to improve our insights into clinical patterns of dystonia and choreoathetosis during PW mobility with head/foot steering systems in children with DCP and its relation with mobility task performance, this study aims (1) to evaluate the presence and severity of dystonia and choreoathetosis during PW mobility (2) to investigate the relation between dystonia, choreoathetosis, PW mobility performance, and participants' characteristics, and (3) to explore dystonia, choreoathetosis, and PW mobility performance during different PW mobility tasks. It is hypothesized that dystonia will be more present and severe than choreoathetosis during PW mobility, and that a positive relation will be found between dystonia, choreoathetosis, and PW mobility performance.

2. Material and Methods

2.1 Participants

A convenience sample of children and youth with DCP, age range 6-21 years old were recruited from two special education schools for children with motor disorders. Inclusion criteria were (1) a DCP diagnosis by a paediatric neurologist, (2) level IV-V in the Gross Motor Function Classification System-Extended and Revised (GMFCS-E&R)^{27,28}, level IV-V in the Manual Abilities Classification System-Extended and Revised (MACS)^{27,29}, and (3) driving a PW with head/foot steering. Exclusion criteria were the inability to understand and follow test instructions, severe visual impairments, and orthopaedic or neurosurgical intervention within the last 12 months. The study was conducted in accordance with the Declaration of Helsinki, and the study protocol received an approval from the Medical Ethics Committee UZ KU Leuven. All participants and/or their parents signed an informed consent.

2.2 Procedure

Data was collected in the participants' respective special education schools. Participants used their own head/foot steering wheelchair to perform four mobility tasks following a standardized order³⁰: independent driving (ID), driving through a created corridor (C), 360 degrees clockwise and counterclockwise turn (CT), and driving through a slalom (S) (see Figure 1). The protocol of the data collection and the set-up are explained extensively elsewhere³¹, and briefly summarized in the caption of Figure 1.

All participants performed the four mobility tasks twice with a two week break in-between the two measurements. Mobility performance was video recorded using Sony Handycam HDR-CX405 (Sony Corporation, Tokyo, Japan) cameras placed on both sides of the task set-up.

2.3 Outcome measures

Presence and severity of dystonia and choreoathetosis during PW mobility performance were evaluated using the Dyskinesia Impairment Mobility Scale (DIMS) protocol.³¹ The DIMS is an extended protocol of the Dyskinesia Impairment Scale (DIS)⁵, developed to measure dystonia and choreoathetosis during PW mobility with head/foot steering systems in DCP with moderately high to good interrater reliability and moderate to excellent test-retest. The DIMS is a reliable video-based scale that assesses presence and severity of dystonia and choreoathetosis in the neck- and arm regions in individuals with DCP who use a head/foot steering wheelchair. The DIMS consists of the dystonia subscale (DIMS-D) and the choreoathetosis subscale (DIMS-CA) which measure duration (i.e. how long dystonia and choreoathetosis were present during the mobility task performance), and amplitude (i.e. the maximum range of motion achieved due to dystonia and choreoathetosis during the mobility task performance). The scoring protocol of the DIMS is presented in Figure 2.

Mobility performance was assessed using (1) the time-on-task (expressed in seconds), and (2) the number of errors during the mobility performance.²⁵ The time-on-task was considered the number of seconds used to perform the respective mobility task. In the few cases where the mobility performance was interrupted mid-performance due to unforeseen circumstances, like malfunction of the wheelchair, the number of seconds between stopping and task continuation were not included in the final score of time-on-task.

Errors during the mobility performance were considered (1) the number of non-task related stops during the performance of all four mobility tasks (excluding the stops due to unforeseen circumstances as mentioned above), and (2) the number of boundary violations during the corridor task (driving over the created corridor), and the slalom task (hitting the cones).

2.4 Data analyses

Data of the first and the second baseline measurement were checked for auto-correlation using the Spearman's rho correlation coefficients (Supplementary material S1). Data for each outcome measurement were averaged across the two repeated measurements, an approach which is commonly done to reduce any measurement error in study designs with repeated measurements.³²⁻³⁵ Thus, averaged values for each participant were used for all statistical analyses (i.e. n=10 participants).

The DIMS scores, the time-on-task, and the number of errors were calculated for the mobility session (all mobility tasks together), as well as for each mobility task separately.

Shapiro-Wilk test indicated that most of the variables were not normally distributed, and consequently, non- parametric statistics were used. Friedman two-way analysis of variance (ANOVA) by ranks was used to determine any significant differences in the median percentage values of the DIMS score between the four mobility tasks.³⁶ In case a significant difference was found, post-hoc test with correction for multiple comparisons ($p < 0.0125$) was conducted.

Wilcoxon signed-rank test was used to determine the statistical comparisons between the percentage scores of the DIMS subscales, between the DIMS scores of the neck- and arm body regions, and between the four different mobility tasks.³⁶ Spearman's rho correlation coefficients (r_s) were used to explore any relationship between the DIMS variables, between the time-on-task and the number of errors, and between the DIMS- and the mobility performance variables for the total session score, and for each mobility task. Whereas the errors outcome during mobility performance is task-specific²⁵, the latter correlations per mobility task were done between the DIMS and the non-task related stops, and between the DIMS and the boundary violations, separately. Spearman's rho correlation coefficients were used to also explore the relationship between the DIMS and the PW mobility performance with the participants' characteristics. A correlation coefficient between 0.80-1.00 was considered very strong, between 0.60-0.79 as strong, between 0.40-0.59 as moderate, between 0.20-0.39 as weak, and <0.20 as very weak/no correlation.³⁶ The level of significance was set at $p < 0.05$. All statistics were calculated using IBM SPSS Statistics 25 (SPSS Inc., Chicago, IL, USA).

3. Results

3.1 Participants

Ten participants with DCP were included in this study. All participants were able to perform the mobility tasks, except for P05 who could not perform a slalom. Participants' characteristics are reported in Table 1.

3.2 Dystonia and choreoathetosis patterns during PW mobility

3.2.1 Dystonia versus choreoathetosis

The total DIMS median percentage level was 59.4% (IQR 51.9–61.8%), ranging between 35.8% and 84.6%. The total median percentage levels were 83.6% (IQR 70.0–89.8%; range 48.0–94.0%) for the DIMS-D subscale, and 34.4% (IQR 29.2–41.7%; range 23.5–82.8%) for the DIMS-CA subscale. Statistical comparisons between the DIMS-D and the DIMS-CA subscales indicated significantly higher levels ($p < 0.01$) of dystonia during PW mobility for the total scores, for the neck-, and for the arm regions. No significant correlations were found between the DIMS-D and the DIMS-CA subscales.

Strong to very strong significant correlations were obtained in the neck and arm regions between the duration and amplitude of both DIMS-D and DIMS-CA, ranging between $0.78 < r_s \leq 0.99$, $p < 0.01$. These correlations were not significant only for the Neck DIMS-D ($r_s = 0.39$, 95% CI -0.32–0.82, $p = 0.270$). Median percentage values and correlations are presented in Table 2.

3.2.2 Neck region versus Arm region

Median percentage values of the DIMS-D total and the DIMS-D amplitude were significantly higher ($p = 0.041$ and $p = 0.005$, respectively) in the arm region compared to the neck region, whereas the difference in the DIMS-D duration was not significant ($p = 0.889$). No significant differences were found between the median percentage values of the DIMS-CA in the neck- and arm regions ($0.262 < p < 0.959$).

3.2.3 Arm Proximal versus Arm Distal

DIMS-D total and DIMS-D duration showed no significant differences between arm regions ($p=0.059$ and 0.107 respectively). Only DIMS-D amplitude was significantly higher in the distal arm compared to the proximal arm, with $p=0.047$. Strong to very strong correlations with a range between $0.69 < r_s \leq 0.85$ with p -values $0.002 < p < 0.029$ were found for the DIMS-D total, duration, and amplitude between the proximal and the distal arm.

Similarly, DIMS-CA total, duration, and amplitude were also significantly higher in the distal arm as opposed to the proximal arm with $p=0.012$, $p=0.009$, and $p=0.017$ respectively. for the three statistical comparisons. DIMS-CA total, duration, and amplitude were not significantly correlated ($0.083 < p < 0.328$) between the proximal and the distal arm with correlation coefficients ranging between $0.35 < r_s < 0.57$.

3.3 Correlations between dystonia, choreoathetosis scores, PW mobility performance, and participants' characteristics

For the total session scores, a moderate correlation ($r_s=0.66$, 95% CI $0.19-0.81$; $p=0.039$) was found between the time-on-task and the number of errors. The only significant correlation for the mobility session scores was between the number of errors and the total DIMS score of the distal arm ($r_s=0.71$, 95% CI $0.10-0.78$, $p=0.021$).

For each mobility task separately, significant correlations were obtained between the time-on-task and the number of errors only during the CT ($r_s=0.72$, 95% CI $0.17-0.93$; $p=0.020$). The correlations between dystonia and choreoathetosis with the time-on-task and the number of errors per mobility task are reported in Table 3.

Participants' age and their GMFCS levels showed no significant correlations with the DIMS-D, DIMS-CA, and the PW mobility performance. Experience with driving a head/foot steering system showed strong negative significant correlations with both DIMS-D ($r_s=-0.76$, 95% CI $-0.94-0.25$; $p=0.010$) and DIMS-CA ($r_s=-0.69$, 95% CI $-0.92-0.11$; $p=0.028$). Strong to very strong positive correlations were obtained between participants' MACS levels and both time-on-task ($r_s=0.64$, 95% CI $0.02-0.90$; $p=0.046$) and number of errors ($r_s=0.86$, 95% CI $0.50-0.97$; $p=0.002$). Results are reported in Table 4.

3.4 Dystonia versus choreoathetosis per mobility task

The median percentage levels of dystonia and choreoathetosis during ID, C, CT, and S, and the statistical comparisons between the four mobility tasks are reported in Table 5.

For the total DIMS score, Friedman ANOVA by ranks between the four mobility tasks was $p=0.006$. The statistically significant differences were between C and CT ($p=0.007$), and between CT and S ($p=0.008$). For the neck region, Friedman ANOVA by ranks showed no significant differences over the four mobility tasks. For the arm proximal region, Friedman ANOVA by ranks was significant only for the DIMS-CA ($p=0.001$) whereas for the arm distal region, both for the DIMS-D ($p=0.001$) and for the DIMS-CA ($p=0.014$).

4. Discussion

The first aim of this study was to map the presence and severity of dystonia and choreoathetosis during PW mobility in individuals with DCP using a head/foot steering system. Total dystonia levels during PW mobility were significantly higher than total choreoathetosis levels, both in duration and amplitude, supporting the first hypothesis posed in this study. This result agrees with, and compliments the previously published presence and severity levels of dystonia and choreoathetosis during rest postures, voluntary-asked activities, and overflow movements in DCP.^{4,6} In the interpretation of this result, it is worth considering that, as previously reported, the hypertonic hallmark of dystonia may mask the hyperkinetic hallmark of choreoathetosis, likely resulting in lower choreoathetosis median scores.³⁷ In addition, the reported levels of dystonia and choreoathetosis in the neck region during PW mobility are in line with the reported values during neck voluntary-asked activities.⁴ Similarly, the median percentages of dystonia and choreoathetosis overflow movements during PW mobility for the proximal and the distal arm align with previous findings.⁶

The current study found no significant correlations between dystonia and choreoathetosis of the neck and the arm overflow movements. Previous research⁴ did not report on the correlations between dystonia and choreoathetosis during the neck voluntary-asked activities, therefore comparisons cannot be made. As for the overflow movements, the result is somewhat in line with previous findings where only poor to fair correlations were obtained.⁶ This discrepancy could be attributed to the difference in the number of participants between the two studies, where an increase in data points increases the likelihood for significant correlations to be obtained. The non-significant correlations between the dystonia- and choreoathetosis subscale indicate their independent occurrence, supporting the added value to map both movement disorders during powered mobility with head/foot steering in DCP.⁴ Dystonia overflow movements of the proximal and distal arm showed strong to very strong correlations, which was not the case for choreoathetosis as all correlation coefficients were not significant. This might indicate that the sustained muscle contractions which characterize dystonia are simultaneously present in both the proximal and distal arm during powered mobility. On the other hand, choreoathetosis seems to occur independently with significantly higher levels in the distal arms compared to the proximal arms. It is worth emphasizing that the proximal arms have a higher support due to the wheelchair seating which might limit to an extent the manifestation of the hyperkinetic choreoathetotic movements in the proximal- as opposed to the distal arms region.

The second aim of the study was to investigate the relationships between the movement disorders, PW mobility performance-based outcome, and participants' characteristics. The time-on-task and the number of errors were positively correlated for the mobility session score, but also separately for the (counter)clockwise turns. These results are not surprising as it would be expected that with more errors during performance, the time to finish the task is prolonged. Similarly, the (counter)clockwise turn requires from the participant to maintain an uninterrupted contact of the head with the left or right array, therefore, any non-task related stop would significantly prolong the time to finish the 360 degrees turn.

Only distal arm overflow movements were moderately correlated with the summed score of the number of errors. This correlation was obtained for the total DIMS score of the distal arm,

indicating the negative contribution of both dystonia and choreoathetosis overflow movements to a lower PW mobility performance. The finding aligns with previous research stating the negative impact of the overflow movements on the execution of voluntary activities and overall activities of daily living in DCP.⁶ The results in the current study also suggest that children with MACS level V needed more time to perform the mobility tasks and made more errors during the mobility performance compared to those with MACS level IV. The MACS measures the ability of the participants to handle objects²⁹ thus it is plausible that children with higher levels of dystonia and/or choreoathetosis in the distal arms classify in the higher MACS levels. Even though with an almost even distribution of participants in MACS level IV (n=4) and V (n=6), the sample size is small and future research with higher representation in both MACS levels is necessary to confirm these results.

In addition, video observations of the mobility performance seem to indicate that some participants bring their arms to a mid-position with extended elbows during head/foot driving. The significant negative relationship between participants' PW mobility experience and arm proximal dystonia and choreoathetosis may further support this hypothesis. Based on reports by clinicians involved in the current study, attempts to minimize arm overflow movements start from the initial stage of mobility training with head/foot steering systems. That is, in the initial learning phase, arms are mainly strapped whereas in later learning phases, children reportedly adopt this strategy to deliberately 'lock' their arms in mid-position. This may possibly be an efficient way to minimize any negative impact that arm overflow movements may have on their head control and consequently on their driving performance. The chronological age of the participants showed no significant correlations with PW mobility performance. This might emphasize the importance to consider many other factors like cognitive and intellectual abilities³⁸, readiness to drive the wheelchair and motivation levels²², training methods and opportunities³⁹ when assessing PW driving competences. The wide range in PW mobility experience and age, the small number of participants, and the lack of other abovementioned variables limit to an extent the interpretation of these results. Future research with a bigger sample and additional outcome measures is therefore highly recommended to explore in-depth the mechanisms of any head/foot driving strategies in DCP..

In the current study, higher levels of dystonia and choreoathetosis seem to be mainly associated with boundary violations (which mimic environmental barriers) than with non-task related stops (which depend mainly on the motor control of the user). As this has safety implications, mobility training may need to incorporate similar mobility tasks with boundary violations first in a controlled and safe set-up before the user drives the wheelchair in surroundings of daily living. It is important to note that children with DCP may show presence of primitive reflexes, including asymmetrical tonic neck reflex (ATNR), symmetrical tonic neck reflex (STNR), and Moro (startle) reflex.⁴⁰ A case study on a participant with severe CP who used head-steering wheelchair reported a negative impact of the startle-like reflex which appeared during sudden visual stimuli.²² This startle-like reflex caused an extension of the trunk, head, and arms, thus the participant was unable to stop and avoid collisions due to the persistent contact of the head with the control array.²² The presence of reflexes has also been reported as a possible cause to accidental activation of the joystick in PW users.⁴¹ Primitive reflexes were not explored in the current study, thus claims cannot be made whether they had any impact on mobility

performance. Future studies assessing the primitive reflexes and their relationship with mobility performance in DCP may be interesting.

The results from the third aim suggest that the total DIMS score lowered significantly from performing the corridor to performing the turns, and then increased again significantly when participants started performing the slalom. This is mainly seen in the arm overflow dystonia and choreoathetosis compared to the neck region where no significant differences were obtained after correcting for multiple comparisons. A possible explanation could be that participants perceived the turns as less demanding compared to the other tasks, given that acquiring the skill to turn is one of the first skills taught in maneuvering with a head steering system.^{13,22} However, the study lacks a measurement of perceived task demand during mobility tasks (e.g. by using the NASA-TLX scale⁴²) and of additional subjective variables like stress or fatigue, thereby warranting a cautious interpretation of possible reasons why dystonia and choreoathetosis significantly changed between tasks.

. This is the first study to map the presence and severity of dystonia and choreoathetosis during PW mobility in individuals with DCP who use a head/foot steering system. Relationships were explored between the movement disorders, PW mobility performance, and participants' characteristics. The reported findings may be a step closer toward a more standardized PW mobility training in DCP. Nevertheless, this study warrants some reflections to consider. First, the sample size is relatively small with a large range in age and PW mobility experience, considering the challenging inclusion criteria like the DCP diagnosis which is rare, and the use of an own head/foot steering wheelchair, which is not widely available. However, the experimental set-up allowed the data of the two baseline measurements to be averaged, hence reducing any measurement error and increasing confidence in the reported results. Second, the current study design enabled mapping clinical patterns and relations between the movement disorders, the PW mobility performance, and participants' characteristics. These insights may be promising and a valuable addition to the very limited existing research in head/foot steering mobility in DCP. Evidence-based knowledge on structured guidelines toward more efficient PW mobility training methods are not lacking only in head/foot steering, but also in general for individuals with CP who use PWs with any control interface.³⁹ There is therefore a clear need for future intervention studies to find the best strategies in customizing a structured PW mobility training programs individually for each child with DCP. Of importance may be to explore and compare different mobility training programs and their efficacy in PW users with DCP, the frequency of training per week, the number of task repetitions per session, the best training environment for the child, and the best ways to optimize the interaction between the therapist and the user.^{39,43} Third, performing fast and accurately is perceived as a central goal of motor performance, thus speed-accuracy trade-off is a widely used concept to measure task execution in clinical populations.^{44,45} A review on PW performance-based outcome measures reported time-on-task as a measure of speed, and the number of errors as a measure of accuracy during PW mobility.²⁵ As the current study investigated the between-participant differences, inferences cannot be made with regards to a speed-accuracy trade-off during head/foot steering, which might be a topic of interest to consider for future studies. Finally, number of studies

suggest that subjective variables like cognitive and emotional development, mood, attention, fatigue, and stress are related to performance.^{16,38,46-48} The above-mentioned factors were not explored in the current study, thus future research on the impact of any subjective variables on PW mobility in DCP is strongly encouraged.

5. Conclusion

The present study aimed to map the presence and severity of dystonia and choreoathetosis during PW mobility in DCP, and to explore their relation with PW mobility performance and participants' characteristics. Dystonia and choreoathetosis showed occurrence in all participants during PW mobility, with dominating dystonia presence and severity. To interpret this result, it is worth considering that the hypertonic hallmark of dystonia may mask to an extent the hyperkinetic hallmark of choreoathetosis, resulting in lower choreoathetosis median levels. Results suggest that distal arm overflow movements have a negative impact on PW mobility performance due to their significant relationship with the number of errors. Furthermore, the negative relationship between proximal arm overflow movements and PW mobility driving experience may imply that over time, children with DCP adopt deliberate strategies to bring their arms in mid-position with extended elbows. This might possibly be an efficient way to increase the control of the head during driving by minimizing as much as possible the negative impact of arm overflow movements. The small sample size limits to an extent the interpretation of the results, thus future studies with a bigger sample size and additional outcome measures are strongly recommended.

Acknowledgements

The authors express immense gratitude to the participants who were included in this study and their supportive parents. A special thank you goes to the therapists of the participants for their invaluable help from the screening process to data collection. The authors immensely thank biostatistician, Dr. Steffen Fieuws for his guidance and support in the statistical approach.

Funding

This research was funded by a C3 grant from the Research Council of KU Leuven, grant number C32/17/056.

Declaration of interest

The authors declare no conflict of interest. The funders had no role in the design of the study; in the collection, analyses, or interpretation of data; in the writing of the manuscript, or in the decision to publish the results.

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Figure 1. Data collection set-up with four standardized power wheelchair mobility tasks using a head/foot steering wheelchair: (A) **ID**, Independent Driving; (B) **C**, driving through a created Corridor; (C) **CT**, 360° Clockwise and counterclockwise Turn, (D) **S**, driving through a Slalom. The set-up of ID, C, and S is 10 meters in length (20 meters of driving performance from- and back to the initial starting point). The width of the created corridor in C, and the distance between the cones in S is calculated for every participant as follows: total length of their head/foot steering wheelchair + 50% of that length, both expressed in centimeters.

Figure 2. The scoring protocol of the Dyskinesia Impairment Mobility Scale (DIMS) to measure presence and severity of dystonia and choreoathetosis during powered wheelchair mobility in dyskinetic cerebral palsy

Table 1: Participants' characteristics

	Gender (M/F)	Age (years)	GMFCS	MACS	Experience with powered mobility (years)	DIMS-D (%)	DIMS-CA (%)	Time on task (mean)	Number of errors (mean)
P01	M	19	IV	IV	10	53,5	36,0	154,5 s	11,0
P02	M	16	IV	IV	6	48,0	23,5	150,0 s	2,5
P03	M	8	V	V	1,5	86,5	82,8	427,0 s	36,0
P04	F	6	IV	IV	1,5	90,3	50,5	250,5 s	11,0
P05	M	15	V	V	6,5	80,6	37,2	308,0 s	64,5
P06	M	17	V	V	7	66,5	32,8	471,5 s	35,5
P07	M	15	V	V	6	90,3	29,0	279,5 s	20,0
P08	F	11	V	V	1,5	88,5	29,8	273,5 s	11,5
P09	F	21	V	IV	4,5	94,0	28,5	387,5 s	10,5
P10	F	8	IV	V	0,5	80,8	43,3	390,0 s	17,5

P, participant; M, male; F, female; GMFCS, Gross Motor Function Classification System to measure gross motor functioning; MACS, Manual Abilities Classification System to measure fine manual abilities; DIMS-D, the Dyskinesia Impairment Mobility Scale, dystonia subscale; DIMS-CA, the Dyskinesia Impairment Mobility Scale, choreoathetosis subscale; s, seconds

Table 2: Median and interquartile range of the outcome measures on the Dyskinesia Impairment Mobility Scale, statistical comparisons, and correlation coefficients between dystonia and choreoathetosis during powered wheelchair mobility

	DIMS-D(%) Median (IQR)	DIMS-CA (%) Median (IQR)	Wilcoxon	r_s (95% CI); p -value
DIMS total	83.6 (70.0–89.8)	34.4 (29.2–41.7)	0.005	0.04 (-0.60–0.65); 0.907
DIMS neck	71.6 (69.1–80.0)	31.3 (26.6–53.7)	0.009	0.41 (-0.30–0.82); 0.244
DIMS arms	86.1 (77.7–91.9)	33.3 (29.2–38.0)	0.005	0.12 (-0.55–0.62); 0.738
DIMS arm proximal	77.4 (67.7–89.5)	27.2 (22.7–34.5)	0.005	0.51 (-0.18–0.86); 0.132
DIMS arm distal	93.1 (89.4–94.9)	38.1 (35.0–52.7)	0.007	-0.31 (-0.79–0.40); 0.382
	Duration	Amplitude		
DIMS-D total	84.8 (74.8–96.9)	79.3 (65.0–83.4)		0.87 (0.53–0.97); 0.001
DIMS-D neck	97.5 (78.1–99.4)	50.6 (41.3–66.3)		0.39 (-0.32–0.82); 0.270
DIMS-D arms	85.0 (81.4–96.1)	83.4 (73.3–92.5)		0.86 (0.50–0.97); 0.002
DIMS-D arm proximal	80.6 (68.8–95.6)	73.6 (66.6–89.4)		0.88 (0.56–0.97); 0.001
DIMS-D arm distal	94.4 (88.4–97.0)	93.1 (87.2–94.7)		0.78 (0.30–0.95); 0.008
	Duration	Amplitude		
DIMS-CA total	36.0 (29.6–40.1)	33.8 (29.5–43.4)		0.92 (0.69–0.98); 0.000
DIMS-CA neck	32.5 (26.3–60.5)	30.0 (24.4–46.9)		0.99 (0.96–0.99); 0.000
DIMS-CA arms	32.2 (28.3–39.6)	34.5 (29.7–40.0)		0.83 (0.42–0.96); 0.003
DIMS-CA arm proximal	26.9 (22.5–28.6)	27.1 (24.1–40.9)		0.81 (0.37–0.95); 0.005
DIMS-CA arm distal	37.5 (34.4–53.5)	38.8 (34.4–49.9)		0.95 (0.80–0.99); 0.000
DIMS, Dyskinesia Impairment Mobility Scale; DIMS-D, dystonia subscale of the DIMS; DIMS-CA, choreoathetosis subscale of the DIMS; IQR, interquartile range; CI, confidence interval; r_s , Spearman's ρ correlation coefficient (to test strength of the relationship between groups); Wilcoxon, Wilcoxon signed-rank test to test significant differences between groups.				

Table 3: Correlations between dystonia and choreoathetosis with the mobility performance per mobility task

	ID r _s (95% CI); p-value		C r _s (95% CI); p-value			CT r _s (95% CI); p-value		S r _s (95% CI); p-value		
	Time-on-task	Stops	Time-on-task	Stops	Boundary violations	Time-on-task	Stops	Time-on-task	Stops	Boundary violations
Neck										
DIMS-D total	0.17 (-0.51–0.72); 0.636	-0.17 (-0.72–0.51); 0.643	0.29 (-0.42–0.78); 0.419	-0.11 (-0.69–0.56); 0.760	0.53 (-0.15–0.87); 0.117	-0.25 (-0.76–0.45); 0.485	0.32 (-0.39–0.79); 0.376	0.01 (-0.66–0.67); 0.982	0.05 (-0.64–0.69); 0.892	0.46 (-0.29–0.86); 0.214
DIMS-D duration	-0.20 (-0.74–0.49); 0.577	-0.22 (-0.75–0.48); 0.546	-0.23 (-0.75–0.47); 0.524	0.12 (-0.55–0.70); 0.733	0.00 (-0.63–0.63); 0.993	-0.46 (-0.84–0.24); 0.182	0.04 (-0.60–0.65); 0.917	-0.05 (-0.69–0.64); 0.893	0.23 (-0.51–0.78); 0.559	0.26 (-0.49–0.79); 0.497
DIMS-D amplitude	0.14 (-0.54–0.70); 0.708	-0.27 (-0.77–0.43); 0.457	0.34 (-0.37–0.80); 0.336	-0.18 (-0.73–0.51); 0.615	0.64 (0.18–0.90); 0.044*	-0.29 (-0.78–0.42); 0.421	0.21 (-0.48–0.74); 0.555	0.33 (-0.43–0.82); 0.381	0.08 (-0.62–0.71); 0.843	0.44 (-0.32–0.85); 0.231
DIMS-CA total	-0.18 (-0.73–0.51); 0.621	-0.29 (-0.78–0.42); 0.419	-0.03 (-0.65–0.61); 0.946	-0.11 (-0.69–0.56); 0.759	0.33 (-0.38–0.79); 0.358	-0.04 (-0.65–0.60); 0.919	0.29 (-0.42–0.78); 0.418	0.30 (-0.45–0.80); 0.427	-0.04 (-0.69–0.64); 0.922	0.10 (-0.60–0.72); 0.794
DIMS-CA duration	-0.28 (-0.77–0.42); 0.432	-0.32 (-0.79–0.39); 0.368	-0.14 (-0.71–0.54); 0.704	-0.07 (-0.67–0.59); 0.839	0.23 (-0.47–0.75); 0.523	-0.10 (-0.69–0.57); 0.783	0.23 (-0.47–0.75); 0.527	0.42 (-0.34–0.85); 0.262	0.11 (-0.60–0.72); 0.774	0.13 (-0.58–0.73); 0.749
DIMS-CA amplitude	-0.08 (-0.68–0.58); 0.837	-0.18 (-0.73–0.51); 0.624	0.12 (-0.55–0.70); 0.736	-0.23 (-0.75–0.47); 0.527	0.51 (-0.18–0.86); 0.130	-0.06 (-0.66–0.59); 0.878	0.25 (-0.45–0.76); 0.480	0.30 (-0.45–0.80); 0.427	-0.05 (-0.69–0.64); 0.896	0.12 (-0.59–0.73); 0.768
Arm Proximal										
DIMS-D total	-0.22 (-0.75–0.48); 0.542	-0.29 (-0.78–0.42); 0.425	-0.25 (-0.76–0.45); 0.490	-0.48 (-0.85–0.21); 0.157	0.36 (-0.35–0.81); 0.315	-0.44 (-0.84–0.26); 0.204	-0.36 (-0.81–0.35); 0.307	0.49 (-0.26–0.87); 0.183	-0.23 (-0.78–0.51); 0.554	0.28 (-0.47–0.80); 0.466
DIMS-D duration	-0.12 (-0.70–0.55); 0.740	-0.18 (-0.73–0.51); 0.618	-0.15 (-0.71–0.53); 0.677	-0.28 (-0.77–0.42); 0.436	0.36 (-0.35–0.81); 0.313	-0.47 (-0.85–0.23); 0.166	-0.28 (-0.77–0.42); 0.429	0.45 (-0.31–0.86); 0.227	-0.03 (-0.68–0.65); 0.946	0.37 (-0.39–0.83); 0.324
DIMS-D amplitude	-0.20 (-0.74–0.49); 0.586	-0.37 (-0.81–0.34); 0.294	-0.31 (-0.79–0.40); 0.388	-0.62 (-0.90–0.02); 0.057	0.33 (-0.38–0.79); 0.348	-0.37 (-0.81–0.34); 0.300	-0.32 (-0.79–0.39); 0.364	0.48 (-0.27–0.87); 0.197	-0.44 (-0.85–0.32); 0.231	0.35 (-0.41–0.82); 0.355
DIMS-CA total	-0.08 (-0.68–0.58); 0.828	-0.30 (-0.78–0.41); 0.406	-0.01 (-0.64–0.62); 0.980	-0.33 (-0.79–0.38); 0.359	0.68 (0.09–0.92); 0.030*	-0.26 (-0.76–0.44); 0.476	-0.35 (-0.80–0.36); 0.326	0.04 (-0.64–0.69); 0.929	-0.51 (-0.88–0.23); 0.162	0.42 (-0.34–0.85); 0.264
DIMS-CA duration	0.05 (-0.60–0.66); 0.890	-0.15 (-0.71–0.53); 0.686	0.12 (-0.55–0.70); 0.736	-0.21 (-0.74–0.48); 0.568	0.67 (0.07–0.91); 0.035*	-0.44 (-0.84–0.26); 0.199	-0.43 (-0.83–0.27); 0.215	0.02 (-0.65–0.68); 0.964	-0.47 (-0.86–0.28); 0.205	0.38 (-0.38–0.83); 0.320
DIMS-CA amplitude	-0.10 (-0.69–0.57); 0.775	-0.28 (-0.77–0.42); 0.429	-0.03 (-0.65–0.61); 0.940	-0.30 (-0.78–0.41); 0.406	0.71 (0.15–0.93); 0.023*	-0.23 (-0.75–0.47); 0.532	-0.34 (-0.80–0.37); 0.335	0.04 (-0.64–0.69); 0.929	-0.51 (-0.88–0.23); 0.162	0.42 (-0.34–0.85); 0.264
Arm Distal										
DIMS-D total	-0.12 (-0.70–0.55); 0.738	-0.20 (-0.74–0.49); 0.572	0.55 (-0.12–0.88); 0.098	0.25 (-0.45–0.76); 0.486	0.51 (-0.18–0.86); 0.131	-0.09 (-0.68–0.57); 0.802	0.26 (-0.44–0.76); 0.467	0.76 (0.19–0.95); 0.017*	0.38 (-0.38–0.83); 0.319	0.29 (-0.46–0.80); 0.448
DIMS-D duration	-0.02 (-0.64–0.62); 0.947	-0.20 (-0.74–0.49); 0.583	0.70 (0.13–0.92); 0.025*	0.18 (-0.51–0.73); 0.627	0.48 (-0.21–0.85); 0.160	-0.26 (-0.76–0.44); 0.461	0.03 (-0.61–0.65); 0.946	0.61 (-0.09–0.91); 0.083	0.57 (-0.15–0.90); 0.113	0.14 (-0.58–0.74); 0.715
DIMS-D amplitude	-0.17 (-0.72–0.51); 0.649	-0.35 (-0.80–0.36); 0.317	0.55 (-0.12–0.88); 0.098	0.25 (-0.45–0.76); 0.486	0.51 (-0.18–0.86); 0.131	0.08 (-0.58–0.68); 0.828	0.37 (-0.34–0.81); 0.291	0.71 (0.09–0.93); 0.033*	-0.03 (-0.68–0.65); 0.948	0.55 (-0.18–0.89); 0.122
DIMS-CA total	0.04 (-0.60–0.65); 0.920	-0.05 (-0.66–0.60); 0.892	0.63 (0.00–0.90); 0.051	0.10 (-0.57–0.69); 0.793	0.19 (-0.50–0.73); 0.606	-0.15 (-0.71–0.53); 0.676	0.04 (-0.60–0.65); 0.907	0.56 (-0.17–0.89); 0.116	0.04 (-0.64–0.69); 0.923	0.34 (-0.42–0.82); 0.374
DIMS-CA duration	0.07 (-0.59–0.67); 0.854	-0.06 (-0.66–0.59); 0.865	0.54 (-0.14–0.87); 0.104	0.20 (-0.49–0.74); 0.586	0.13 (-0.54–0.70); 0.717	-0.15 (-0.71–0.53); 0.673	0.00 (-0.63–0.63); 0.993	0.56 (-0.17–0.89); 0.116	0.10 (-0.60–0.72); 0.804	0.28 (-0.47–0.80); 0.468
DIMS-CA amplitude	-0.14 (-0.71–0.54); 0.699	-0.14 (-0.71–0.54); 0.707	0.67 (0.07–0.91); 0.033*	0.08 (-0.58–0.68); 0.832	0.24 (-0.46–0.76); 0.511	-0.17 (-0.72–0.51); 0.638	0.04 (-0.60–0.65); 0.906	0.64 (-0.04–0.92); 0.063	0.06 (-0.63–0.70); 0.870	0.25 (-0.50–0.78); 0.515

DIMS, Dyskinesia Impairment Mobility Scale; DIMS-D, dystonia subscale of the DIMS; DIMS-CA, choreoathetosis subscale of the DIMS; ID, Independent driving; C, Corridor; CT, (counter)clockwise turn; S, Slalom; r_s, Spearman's rho

correlation coefficient (to test strength of the relationship between variables); *, significant correlations p<0.05

Table 4. Correlations between dystonia, choreoathetosis, and PW mobility performance with participants' characteristics

		Age	Experience with PW	GMFCS	MACS	
		rs (95% CI); p-value	rs (95% CI); p-value	rs (95% CI); p-value	rs (95% CI); p-value	
DIMS	Neck	DIMS	-0.37 (-0.81–0.34); p=0.297	-0.07 (-0.67–0.59); p=0.852	-0.04 (-0.65–0.60); p=0.922	0.43 (-0.27–0.83); p=0.218
		DIMS-D	-0.18 (-0.73–0.51); p=0.613	-0.22 (-0.75–0.48); p=0.538	-0.14 (-0.71–0.54); p=0.695	0.36 (-0.35–0.81); p=0.314
		DIMS-CA	-0.40 (-0.82–0.31); p=0.257	-0.07 (-0.67–0.59); p=0.853	-0.21 (-0.74–0.48); p=0.554	0.21 (-0.48–0.74); p=0.554
	Arm Proximal	DIMS	-0.57 (-0.88–0.09); p=0.083	-0.92 (-0.98–0.69), p<0.001**	0.28 (-0.42–0.77); p=0.426	0.07 (-0.59–0.67); p=0.845
		DIMS-D	-0.28 (-0.77–0.42); p=0.432	-0.76 (-0.94–0.25); p=0.010*	0.36 (-0.35–0.81); p=0.314	-0.14 (-0.71–0.54); p=0.695
		DIMS-CA	-0.60 (-0.89–0.05); p=0.072	-0.69 (-0.92–0.11); p=0.028*	-0.14 (-0.71–0.54); p=0.694	-0.07 (-0.67–0.59); p=0.845
	Arm Distal	DIMS	-0.60 (-0.89–0.05); p=0.069	-0.34 (-0.80–0.37); p=0.337	0.39 (-0.32–0.82); p=0.262	0.50 (-0.19–0.86); p=0.142
		DIMS-D	-0.06 (-0.66–0.59); p=0.867	-0.41 (-0.83–0.30); p=0.236	0.57 (-0.09–0.88); p=0.086	0.21 (-0.48–0.74); p=0.554
		DIMS-CA	-0.56 (-0.88–0.11); p=0.093	-0.09 (-0.68–0.57); p=0.805	0.00 (-0.63–0.63); p=1.000	0.32 (-0.39–0.79); p=0.365
PW mobility performance	Time-on-task	-0.07 (-0.67–0.59); p=0.854	-0.07 (-0.67–0.59); p=0.853	0.57 (-0.00–0.88); p=0.086	0.64 (0.02–0.90); p=0.046*	
	Number of errors	-0.31 (-0.79–0.40); p=0.380	0.12 (-0.55–0.70); p=0.747	0.57 (-0.09–0.88); p=0.085	0.86 (0.50–0.97); p=0.002**	

DIMS, Dyskinesia Impairment Mobility Scale; DIMS-D, dystonia subscale of the DIMS; DIMS-CA, choreoathetosis subscale of the DIMS; PW, powered wheelchair; GMFCS, Gross Motor Functional Classification System; MACS, Manual Ability Classification System; *rs*, Spearman’s *rho* correlation coefficient (to test strength of the relationship between variables); *, significant correlations p<0.05; **, significant correlations p<0.01

Table 5: Median and interquartile range of the outcome measures on the Dyskinesia Impairment Mobility Sale (DIMS), and statistical comparisons for the neck and arms region for the different mobility tasks during powered mobility

	ID Med (IQR)	C Med (IQR)	CT Med (IQR)	S Med (IQR)	Wilcoxon (ID-C)	Wilcoxon (C-CT)	Wilcoxon (CT-S)	Wilcoxon (ID-S)	Friedman (ID, C, CT, S)
Total DIMS	62.2 (60.9–70.9)	63.1 (61.4–69.5)	53.2 (42.0–60.9)	66.3 (55.0–73.2)	0.326	0.007**	0.008**	0.767	0.006**
Neck									
DIMS	60.9 (47.7–64.8)	57.8 (51.6–71.9)	50.8 (44.9–57.4)	53.1 (50.0–59.4)	0.391	0.025*	0.236	0.858	0.237
DIMS-D	71.9 (62.5–79.7)	71.9 (64.1–81.3)	71.9 (66.4–81.3)	68.8 (68.8–81.3)	0.783	0.496	1.000	0.726	0.922
DIMS-CA	46.9 (25.0–54.7)	46.9 (31.3–60.9)	20.3 (18.8–48.4)	37.5 (18.8–62.5)	0.201	0.017*	0.030*	0.779	0.047*
Arms									
DIMS	71.1 (62.3–72.3)	68.4 (63.7–73.0)	53.5 (37.6–60.8)	64.1 (57.0–78.1)	0.414	0.007**	0.008**	0.813	0.008*
DIMS-D	93.0 (73.4–95.3)	93.0 (82.4–98.0)	88.7 (63.1–91.2)	89.1 (82.8–95.3)	0.101	0.173	0.123	0.833	0.071
DIMS-CA	57.8 (52.3–65.6)	48.4 (37.1–55.9)	18.0 (14.5–29.1)	39.1 (29.7–62.5)	0.059	0.005**	0.008**	0.138	0.001**
Arm Proximal									
DIMS	62.5 (46.1–66.8)	63.3 (52.7–68.4)	53.1 (44.1–61.1)	50.0 (46.9–56.3)	0.188	0.092	0.552	0.212	0.012
DIMS-D	87.5 (52.3–93.8)	92.2 (65.6–96.1)	80.5 (59.8–89.1)	87.5 (81.3–96.9)	0.188	0.260	0.068	0.361	0.100
DIMS-CA	34.4 (28.9–46.1)	34.4 (28.9–46.1)	28.1 (25.0–34.0)	12.5 (12.5–31.3)	1.000	0.235	0.008**	0.017*	0.005**
Arm Distal									
DIMS	78.9 (70.3–80.9)	71.9 (67.2–81.3)	59.4 (47.5–69.7)	65.6 (60.9–85.9)	0.358	0.012*	0.038*	0.091	0.005**
DIMS-D	95.3 (91.4–99.2)	100 (96.9–100)	91.4 (77.3–93.8)	93.8 (84.4–93.8)	0.083	0.008**	0.206	0.027*	0.001**
DIMS-CA	60.9 (54.7–62.5)	45.3 (36.7–63.3)	23.4 (16.4–48.4)	37.5 (34.4–75.0)	0.212	0.016*	0.028*	0.159	0.014*

DIMS, Dyskinesia Impairment Mobility Scale; DIMS-D, dystonia subscale of the DIMS; DIMS-CA, choreoathetosis subscale of the DIMS; ID, Independent driving; C, Corridor; CT, (counter)clockwise turn; S, Slalom; Friedman, Friedman two-way analysis of variance by ranks (to asses the significant differences between the four mobility tasks); Wilcoxon, Wilcoxon signed-rank test (to test significant differences pairwise between the mobility tasks); *p<0.05; **p<0.0125, p-value adjusted for multiple comparisons

Supplementary material S1

(Auto)correlations between the first- and the second baseline measurement

Neck						
	DIMS-D duration	DIMS-D amplitude	DIMS-D	DIMS-CA duration	DIMS-CA amplitude	DIMS-CA total
r _s (95% CI); p-value (W1-W2)	0.94 (0.76–0.99), p<0.001	0.67 (0.07–0.91), p=0.033	0.89 (0.59-0.97), p=0.001	0.45 (-0.25–0.84), p=0.184	0.53 (-0.15–0.87), p=0.112	0.49 (-0.20–0.86), p=0.151
Arm proximal						
	DIMS-D duration	DIMS-D amplitude	DIMS-D total	DIMS-CA duration	DIMS-CA amplitude	DIMS-CA total
r _s (95% CI); p-value (W1-W2)	0.76 (0.25–0.94), p=0.011	0.58 (-0.08–0.89), p=0.077	0.68 (0.09-0.92), p=0.030	0.66 (0.05–0.91), p=0.039	0.61 (-0.03–0.90), p=0.060	0.63 (0.00–0.90), p=0.049
Arm distal						
	DIMS-D duration	DIMS-D amplitude	DIMS-D total	DIMS-CA duration	DIMS-CA amplitude	DIMS-CA total
r _s (95% CI); p-value (W1-W2)	0.91 (0.66–0.98), p<0.001	0.93 (0.73–0.98), p<0.001	0.94 (0.76–0.99), p<0.001	0.75 (0.23–0.94), p=0.013	0.62 (-0.02–0.90), p=0.057	0.71 (0.15–0.93), p=0.021
PW mobility performance						
	Time-on-task	Non-task related stops	Boundary violations			
r _s (95% CI); p-value (W1-W2)	0.64 (0.02–0.90), p=0.046	0.61 (-0.03–0.90), p=0.063	0.88 (0.56–0.97), p=0.001			

DIMS, Dyskinesia Impairment Mobility Scale; DIMS-D, dystonia subscale; DIMS-CA, choreoathetosis subscale; r_s, Spearman’s rho correlation coefficients to assess the correlations between the two baseline measurements; W1, week 1 (first baseline measurement); W2, week 2 (second baseline measurement); PW, powered wheelchair