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ORIGINAL RESEARCH

Windmill-task as a New Quantitative and Objective Assessment for Mirror Movements in Unilateral Cerebral Palsy: A Pilot Study

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Abstract

Objective: To introduce the Windmill-task, a new objective assessment tool to quantify the presence of mirror movements (MMs) in children with unilateral cerebral palsy (UCP), which are typically assessed with the observation-based Woods and Teuber scale (W&T).

Design: Prospective, observational, cohort pilot study.

Setting: Children's hospital.

Participants: Prospective cohort of children (N=23) with UCP (age range, 6−15y, mean age, 10.5±2.7y).

Interventions: Not applicable.

Main Outcome Measures: The concurrent validity of the Windmill-task is assessed, and the sensitivity and specificity for MM detection are compared between both assessments. To assess the concurrent validity, Windmill-task data are compared with W&T data using Spearman rank correlations (ρ) for 2 conditions: affected hand moving vs less affected hand moving. Sensitivity and specificity are compared by measuring the mean percentage of children being assessed inconsistently across both assessments.

Results: Outcomes of both assessments correlated significantly (affected hand moving: ρ=.520; *P*=.005; less affected hand moving: ρ=.488; *P*=.009). However, many children displayed MMs on the Windmill-task, but not on the W&T (sensitivity: affected hand moving: 27.5%; less affected hand moving: 40.6%). Only 2 children displayed MMs on the W&T, but not on the Windmill-task (specificity: affected hand moving: 2.9%; less affected hand moving: 1.4%).

Conclusions: The Windmill-task seems to be a valid tool to assess MMs in children with UCP and has an additional advantage of sensitivity to detect MMs.

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Mirror movements (MMs) are involuntary movements that accompany and mirror voluntary movements of homologous muscles on the opposite side of the body.[1] They commonly occur during typical development and mostly appear during hand movements.[2] In typical development, MMs gradually disappear during the first decade of life.[3] However, in children with unilateral cerebral palsy (UCP), MMs are frequently more pronounced and persistent.[1,4,5] Studies of these "pathological" MMs have predominantly focused on the underlying mechanisms[1,4,6] and their effect on upper limb function.[7-11]

Two general mechanisms of MMs are typically described. First, the motor cortex of the less affected hemisphere also controls the affected hand (AH) by an uncrossed corticospinal tract to

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the ipsilateral side of the spinal cord. This ipsilateral projection might depend on preserved ipsilateral projections to the AH or a branching of crossed corticospinal fibers.[2,12] These "rewiring" profiles are suggested to cause MMs in both, but especially in the AH.[6,13-15] Second, there is widespread and bilateral cortical activation that occurs when actively moving the AH, caused by sensorimotor impairments of this hand and thus increased effort required to move. This lack of "interhemispheric inhibition" is proposed to cause MMs in the less AH.[6,8,13] MMs occurring in only the less AH are therefore thought to be related to sensorimotor impairments of the AH, whereas MMs in the AH have been proposed to indicate 1 motor cortex controlling both hands. Accordingly, MMs detected in the AH may act as a low-risk clinical biomarker to probe corticospinal tract wiring[13] as compared to more invasive and time-consuming neuroimaging methods (eg, transcranial magnetic stimulation, functional magnetic resonance imaging). If accurate, it would have a significant effect on clinical practice, allowing development of individualized therapy programs on the basis of the child's rewiring profile.[16] However, to date, studies[11,17,18] using various assessments for MM detection report conflicting results, challenging its usefulness in probing cortical "rewiring."

With respect to the effect of MMs on upper limb function, the results generally point to an association between pronounced MMs and impaired upper limb function,[7-9] especially in bimanual tasks. However, findings are also not ubiquitous. Some studies[7,9] report correlations between impaired bimanual performance and MMs in both the AH and the less AH, whereas others[8,19] report an association only for MMs appearing in the less AH. Other studies[10,11] report little association between MMs and bimanual performance, whereas some studies[8,19] even indicate that MMs might assist movements of the AH. These contradicting results might be due to the different methods used to assess MMs. To advance our understanding of the mechanisms of MMs and their effect on upper limb function in UCP, it is essential to apply a valid, standardized, objective, and reliable clinical assessment.[8,13]

The universal standard for clinically evaluating MMs is a qualitative observational method based on the Woods and Teuber scale (W&T).[1] It is based on visual evaluation of MMs of one hand during voluntary movements of the other hand.[1] Owing to its easy application and clinical utility, the W&T is widely used in studies[8,9,11] of UCP, offering the potential opportunity for comparison of data. However, its subjective scoring procedure and lack of published guidelines for administration hinder comparison of data. In fact, there is a broad variation in administration and inconsistent use of manual tasks across studies.[1,8,11] The latter is especially problematic, because the severity of MMs is known to be dependent on the type and complexity of movements.[1,5]

List of abbreviations:

AH	**affected hand**
CCC_{max}	**maximum cross-correlation coefficient**
MACS	**Manual Ability Classification System**
MM	**mirror movement**
MVC	**maximal voluntary contraction**
UCP	**unilateral cerebral palsy**
W&T	**Woods and Teuber scale**
$W\&T_{fist}$	**Woods and Teuber fist opening and clenching**
$W\&T_{opposition}$	**Woods and Teuber finger opposition**
$W\&T_{tapping}$	**Woods and Teuber finger tapping**

The observational nature of the W&T may affect the accuracy of detecting MMs and thereby test validity. Likewise, test sensitivity may be suboptimal, thus increasing the likelihood of not detecting MMs that are actually present. This might be due to the extent of mirroring activity (too subtle for visual detection) or the orientation of the hand under observation (eg, persistent wrist flexion of the AH). In addition, test specificity may be compromised, in this case increasing the chance of observing MMs that are not truly mirroring the intended movement of the active hand. Finally, because the close matching of both hand movements in time is not feasible using the W&T, MMs cannot be distinguished from other extraneous movements. These cumulative shortcomings might explain the conflicting results related to the use of MMs to probe cortical rewiring[11,17,18] as well as to the effect of MMs on upper limb function.[7-11,19]

To overcome the shortcomings of the W&T, simultaneous electromyographic recordings of homologous muscles during single hand movements have been applied in earlier studies.[4,6,16] However, it can be argued that these recordings reflect mirror recruitment (muscle activity) rather than actual MMs. Furthermore, the clinical utility remains questionable. To objectively assess actual MMs while being clinically applicable, simultaneous grip force measurements of both hands during single hand movements might offer a solution.[4,6,16]

Here we introduce a new, easy-to-use, objective, standardized, and quantitative assessment for MMs, known as the Windmill-task, using grip force data of both hands during single hand squeeze movements. Quantitative data from the Windmill-task are compared with observation-based data from the W&T on a group level to examine the concurrent validity of the Windmill-data for assessing MMs and on an individual level to estimate differences between both assessment tools in terms of sensitivity and specificity for MM detection. It is hypothesized that the Windmill-task data correlate with the W&T data and that the Windmill-task exhibits higher sensitivity and specificity for MM detection compared to individual data from the W&T.

Methods

Participants

Children with UCP (age range, 6−15y) were recruited from Monash Children's Hospital, Melbourne, Victoria, Australia, from November 1, 2015 through April 30, 2016 as a convenience sample from a cohort of 34 children previously recruited for a larger study (Clinical Trial Registration No.: ACTRN12614000631606). Inclusion criteria for this prospective cohort were diagnosis of UCP with Manual Ability Classification System (MACS)[20] levels I to III. The study was approved by Monash Health Human Research Ethics Committee (HREC: 12167B). Informed consent was obtained before enrollment in the study.

Observation-based assessment of MMs: W&T

Hand movements were videotaped during 3 unimanual tasks: (1) Woods and Teuber fist opening and clenching ($W\&T_{fist}$); (2) Woods and Teuber finger opposition (fingers sequentially touch the thumb; $W\&T_{opposition}$); and (3) Woods and Teuber finger tapping (fingers sequentially tap on the table; $W\&T_{tapping}$).[7,8] Each task was repeated 5 times for each hand, first with the less AH first (MMs in the AH) and then with the AH (MMs in the less AH).

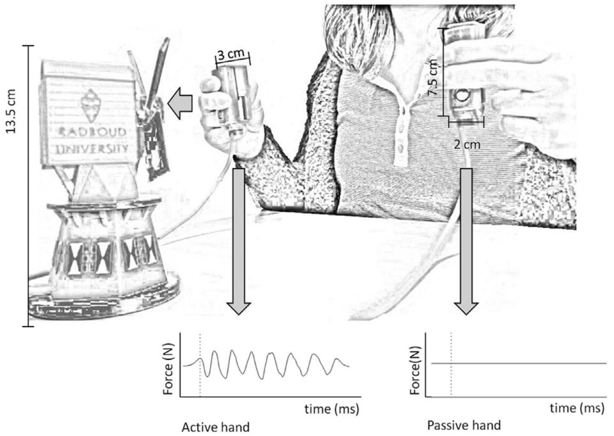

Fig 1 Participant performing the Windmill-task, with the right hand actively squeezing the connected transducer. The participant is holding the grip force transducers in each hand, with the right transducer being connected to the windmill. Both transducers are connected to a computer that digitizes and store the data recorded of both hands' time-locked grip force. This figure presents a squeezing pattern with the passive hand showing no MMs.

The occurrence of MMs was evaluated for each task separately using a 4-point scale ranging from 0 (no clear imitative movement) to 4 (movement equal to that expected for the intended hand),[1] yielding a total score between 0 and 12 (W&T$_{total}$) for each condition separately (AH moving vs less AH moving). Videos were scored by an experienced occupational therapist blinded to the Windmill-task data.

Quantitative assessment of MMs: Windmill-task

The Windmill-task is a custom-made repetitive squeezing task tool developed to quantitatively detect MMs by simultaneously measuring the continuous grip force of both hands via 2 grip force transducers (equipped with micro load cells, 0−20kg; weight, 45g; circumference,10cm) between the thumb and the index plus middle finger (pinch grip). When the child was not able to apply this pinch grip, additional fingers were allowed to stabilize the grip (adapted grip). Grip forces were recorded with a peak of 200N, with an accuracy of 0.2N and a sampling rate of 50Hz. The analog signal was amplified using an INA125p amplifier and converted into a digital signal using the Arduino Nano board.[a] A custom script (PsychoPy v1.83[b]) was used to calibrate the device, set task parameters, run the experiment, and record the data.

Before the MM assessment, the maximal voluntary contraction (MVC) of the pinch grip (or adapted grip) was assessed for each hand separately. One grip force transducer was placed in the child's hand, and the child was asked to press it as hard as possible. This was repeated 3 times, first with the less AH and then with the AH. The average of the 3 squeezes was used as the MVC per hand.

For the MM assessment, children were instructed to hold the transducers in both hands with their forearms or elbows supported on the table. The grip of the less AH was always matched to the grip of the AH (pinch grip vs adapted grip). One transducer was connected to a miniature windmill (fig 1). The motor of the windmill started rotating once the connected transducer was pressed beyond a threshold (20% of the MVC). To speed up rotation, the child's grip needed to return to a lower threshold by loosening the grip (17.5% of the MVC) and again reach the upper threshold within 1000ms so that a repetitive squeezing pattern was induced (≥1Hz frequency). Children were instructed to repetitively squeeze the connected transducer with the active hand ("rotate the windmill as fast as possible") and simply lift and hold the second transducer with the passive hand.

Children performed 10 unimanual squeezing trials with each hand (10s; 5-s rest between trials). A prerecorded voice indicated the start and stop of rotating the windmill. The less AH was tested first. The visual feedback from the rotating windmill guided the children through the task, but instructions contained no information on MMs.

Data processing

The trial started 500ms after the "start" signal and lasted 10 seconds to control the slight delay after the auditory "start" and "stop" signals. To quantify MMs the force pattern of both hands during each squeezing session (10×10s) was cross-correlated.[21] Both grip force signals were correlated by iteratively shifting one signal forward in time against the other signal. A correlation coefficient (Pearson *r*) was calculated for each phase shift (steps of 20ms), resulting in a time series of *r* values. This time series represented a correlation function at each increment of the phase shift between the 2 signals. The maximum cross-correlation coefficient (CCC$_{max}$) of this function was used as the index of similarity between the signals. This value can be directly related to a time lag value showing the match in timing between the 2 signals. A negative time lag indicates the passive hand movement lagging behind the active hand movement. A positive time lag indicates the active hand movement lagging behind the passive hand movement. Furthermore, the mean grip force of the passive hand during each squeezing session was calculated. To ensure a sufficient signal-to-noise ratio, only peaks exceeding the maximum noise values (0.4N) were considered as mirror activity and used.

In a second step, the average of the CCC$_{max}$ for the valid trials (>5 squeezes, with active hand reaching at least 10% of the MVC) was calculated. Hence, CCC$_{max}$ is indicative of the intensity of MMs, with $r=0$ reflecting no mirroring of the passive hand during the active hand movement and $r=1$ reflecting that the passive hand is performing the exact same movement as the active hand. Whenever CCC$_{max}$ was ≥.30, children were classified as having MMs, because correlations <.30 are known to be negligible.[22] CCC$_{max}$ calculations were performed for both conditions separately (AH moving vs less AH moving).

Data analysis

Shapiro-Wilk tests indicated that of the 4 variables—CCC$_{max}$ AH moving, CCC$_{max}$ less AH moving, W&T$_{total}$ AH moving, and W&T$_{total}$ less AH moving—1 variable, W&T$_{total}$ less AH moving, was not normally distributed. In addition, the number of participants was small (N<30); therefore, nonparametric tests were used to compare outcome measures of both assessments.

To estimate the concurrent validity of the Windmill-task, MM scores on the Windmill-task (CCC$_{max}$) were correlated with the total scores on the W&T (W&T$_{total}$) for both conditions separately (AH moving vs less AH moving) by using nonparametric 1-tailed Spearman rank correlations (ρ). Correlation coefficients >.70 were considered as high, .50 to .70 as moderate, .50 to .30 as low, and <.30 as negligible (no MMs).[22]

To estimate the sensitivity of the W&T data as compared with the Windmill-task data, the percentages of children showing MMs

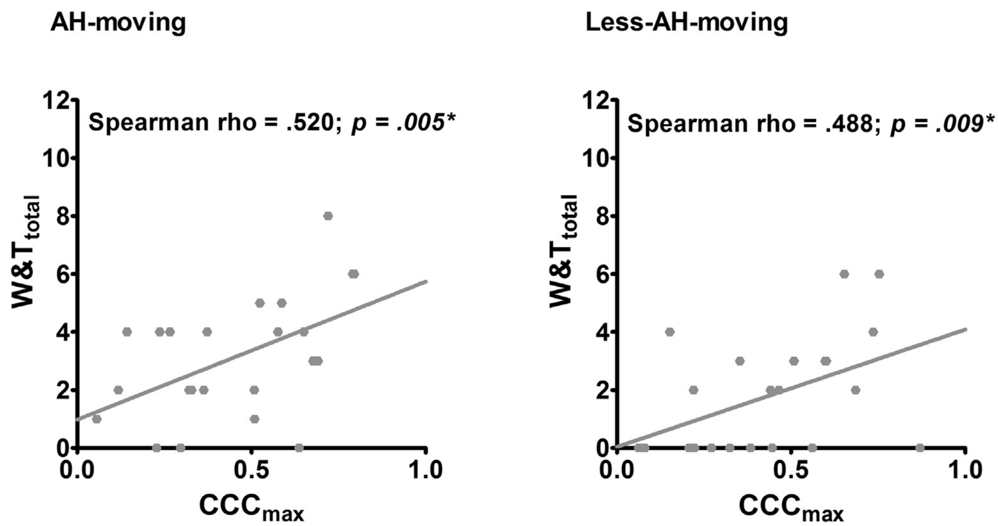

Fig 2 Spearman rank correlations (ρ) between CCC_{max} and $W\&T_{total}$ values for both conditions separately (AH moving vs less AH moving). The asterisk denotes significance ($P<.05$).

on the Windmill-task ($CCC_{max} \geq .30$) but no MMs on the W&T (scores, ≤ 1) were calculated. This was performed for each W&T subscale ($W\&T_{fist}$, $W\&T_{opposition}$, $W\&T_{tapping}$) separately, and in a second step, these percentages were averaged across subscales.

To estimate the specificity, the percentages of children showing MMs on the W&T subscales (scores, ≥ 2)[8] but no MMs on the Windmill-task ($CCC_{max} \leq .30$) were calculated. Again, this was performed for each subscale and averaged afterward.

Results

Participants

Of the 34 children previously recruited for a larger study, 23 children with UCP agreed to participate (13 girls; 16 right AH; mean age, 10.5±2.7y; range, 6.4−14.12y; MACS level I: n=5; MACS level II: n=18). The mean time since the last botulinum toxin A injection was 1.7y (range, 2mo to 6.8y). Two children had previous upper limb surgery (>2y before the assessment).

Mirror movements

The median value of MMs assessed by the W&T ($W\&T_{total}$) is 3 for the AH moving condition and 2 for the less AH moving condition. For the Windmill-task, the median CCC_{max} values are .509 for the AH moving condition and .441 for the less AH moving condition.

Using the Windmill-task, the cross-correlation data of all children who showed MMs ($CCC_{max}>.30$) when moving their less AH had a negative (n=13; median, −10) or zero time lag (n=2), indicating that AH movements are either lagging behind less AH movements or occurring simultaneously. When actively moving the AH, 4 children who showed MMs ($CCC_{max}>.30$) had a positive time lag (median, 16), indicating that in these 4 children the "active" AH movements are actually lagging behind the less AH movements.

Concurrent validity

An evaluation of concurrent validity demonstrated significant correlations between CCC_{max} and $W\&T_{total}$ for both conditions (AH moving vs less AH moving). Correlations were moderate (ρ=.520; P=.005) for the AH moving condition and low for the less AH moving condition (ρ=.488; P=.009) (fig 2).

Sensitivity and specificity

The results for sensitivity demonstrated that for every subscale of the W&T, some children displayed MMs on the Windmill-task ($CCC_{max}>.30$), which were not evident on the W&T subscales (scores, ≤ 1). In the AH moving condition, 27.5% of children demonstrated this pattern ($W\&T_{fist}$: 17.4% [n=4]; $W\&T_{opposition}$: 30.4% [n=7]; $W\&T_{tapping}$: 34.8% [n=8]) and 40.6% of the cases in the less AH moving condition ($W\&T_{fist}$: 21.7% [n=5]; $W\&T_{opposition}$: 52.2% [n=12]; $W\&T_{tapping}$: 47.8% [n=11]).

The results for specificity demonstrated 2 children with clear MMs on at least one of the W&T subscales (score, >1) but no MMs on the Windmill-task ($CCC_{max} \leq .30$). This leads to an average of 2.9% showing this pattern in the AH moving condition ($W\&T_{fist}$: 4.3% [n=1]; $W\&T_{opposition}$: 4.3% [n=1]; $W\&T_{tapping}$: 0% [n=0]) and 1.4% in the less AH moving condition (only $W\&T_{tapping}$: 4.3% [n=1]).

Discussion

We introduced a new method to quantify MMs and compared its data to the commonly used W&T data.[1] Outcomes of this pilot study provide support for the Windmill-task as a valid, standardized, objective, and quantitative assessment of MMs in children with UCP. A comparison to the W&T, which remains the universal standard to detect MMs, demonstrated that both measures seem to detect the same construct, that is, involuntary and simultaneous movements of the mirroring hand, providing support for the concurrent validity of the Windmill-task. Correlations, however, were found to be moderate to low. The comparison of sensitivity and specificity of the W&T with those of the Windmill-task revealed a likely explanation for these findings. With respect to specificity, only 2 children displayed MMs on the W&T but not on the Windmill-task, indicating only a minor advantage of the Windmill-task with regard to test specificity. However, results were different for sensitivity. When actively moving the AH, 27.5% of children displayed MMs in the less AH using the Windmill-task, but none of them on the W&T. For the assessment of MMs in the AH, this percentage increased to 40%. These combined findings indicate that visual observation with the W&T might lead to an underestimation of mirror activity and thus a reduced sensitivity for detecting MMs. These pilot data support the hypothesis that the use of simultaneous grip force recordings using the Windmill-task enhance sensitivity for detecting MMs in children with UCP.

The objective nature, standardized administration procedure, and data for improved sensitivity of the Windmill-task support its use in the future. Previous studies of the effect of MMs on upper limb function or the underlying mechanisms of MMs are not uniform. This is most likely due to different methods used to assess MMs and the subjective nature of the W&T. Understanding the effect of MMs on upper limb function has the potential to improve therapy recommendations on the basis of the individual's MM profile.[23] With respect to the underlying mechanisms, it still needs to be established if MMs appearing in the AH are indicative of 1 motor cortex controlling both hands.[6,13-15] If neuroimaging outcomes (eg, transcranial magnetic stimulation, functional magnetic resonance imaging) are found to be consistent with outcomes from the Windmill-task, this would allow clinicians to quickly and easily assess the rewiring profile of children and allow treatment programs to be individualized on the basis of this profile.[16] Currently, no clinical outcome measure has been found to reliably detect the presence of ipsilateral projections in children with UCP. A further potential advantage of a more sensitive assessment tool lies in the opportunity of detecting subtle changes in MMs after different therapy programs.

The clinical utility of an assessment is an important feature and might favor the use of the W&T because almost no equipment is needed. However, data from this study suggesting a lack of sensitivity show that despite its ease of administration, up to 40% of children with UCP were classified as not having MMs using the W&T, even though these actually seem to be present. If MMs appearing in the AH are indeed indicative of preserved ipsilateral corticospinal projections,[6,13-15] then our pilot data suggest that 40% of children would have been classified with the incorrect rewiring profile by using the W&T. The Windmill-task also presents some potential advantages in its application. The turning of the windmill provides direct feedback so that the task of repetitively moving the hand becomes meaningful and motivating. This design, along with a short, easy, and highly standardized assessment procedure supports the clinical utility of the Windmill-task. Furthermore, the Windmill-task is low cost (∼€100−€150) and it simply needs to be plugged into a computer (via a USB drive) and user-friendly software is available. Data are automatically saved for later analysis when starting the installed program.

In addition to improved sensitivity to detect MMs, the Windmill-task captures the individual strength and timing of these movements. This information can potentially provide evidence for the strategic use of MMs, as previously reported.[19] The information on the timing of the mirroring signal might also help identify different underlying mechanisms of MMs (interhemispheric inhibition vs ipsilateral corticospinal projections). How to use this time lag information for greater understanding of MMs requires further investigation.

An alternative explanation for moderate to low correlation between the data of both assessments may be that they do not measure precisely the same construct. First, compared with the 3 W&T subscales, the Windmill-task assesses only a single movement. One could argue that this one simple squeezing task does not reflect the actual occurrence of MMs during daily routines. However, highly repetitive and simple motor tasks have earlier been suggested to be most appropriate to assess MMs in children with UCP.[13] Second, it is reasonable to propose that on the one hand the W&T subscale should not function as a criterion measure because of its limitations; on the other hand, one could propose that the repetitions and strength required for the completion of the Windmill-task assessment could overestimate the actual occurrence of MMs. It is known that MMs in the less AH are caused by sensorimotor impairments of the AH and increase with effort.[6,8,13] Therefore, the observation that more MMs are detected when using the Windmill-task (27.5%) may also be explained by the increased effort needed for the task. However, even more children did not show any MMs on the W&T when actively moving their less AH (40.6%), which cannot be explained by the increased effort on the Windmill-task. Thus, the argument seems more likely that the higher sensitivity of the Windmill-task is inded responsible for the increased number of MMs detected.

Study limitations

This study is limited by the small number of participants used to draw general conclusions and the lack of inclusion of children at MACS level III. Future studies with a larger sample size should be performed, also enabling to statistically test whether the Windmill-task is indeed more sensitive or specific than the W&T. The reliability of the Windmill-task requires investigation, especially via test-retest and interrater reliability calculation. Also, it should be considered to assess the predictive validity as a potential stronger validation approach. A further limitation is related to the conclusions drawn on the basis of the concurrent validity of the Windmill-task to detect MMs. Because the W&T has many limitations, it is questionable whether it can be used as a criterion measure. However, no alternative assessment is commonly used to assess MMs in children with UCP. For future studies, it could be considered to also compare the outcomes of the Windmill-task to simultaneous electromyographic measurements of the mirror recruitment. Finally and most importantly, reference scores need to be developed by assessing MMs in typically developing children to inform about how strong MMs need to be to be considered as "pathological" and up to which age a certain amount of MMs can be considered as "physiological." This is especially important to validate the claim that a cross-correlation coefficient between the movement patterns of both hands that is $>.30$ does indeed reflect pathological MMs.

Conclusions

This pilot study provides support for the Windmill-task as a valid, standardized, objective, and motivating tool to assess MMs in children with UCP as well as to quantify the timing and intensity of these movements. Outcomes from the Windmill-task suggest enhanced sensitivity and specificity when compared to the observation-based W&T.

Suppliers

a. Arduino Nano board; Farnell, Leeds, UK.
b. PsychoPy v1.83; psychopy.org.

Keywords

Child; Movement; Rehabilitation; Upper extremity

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