



Proposal of a new 3D bimanual protocol for children with unilateral cerebral palsy: Reliability in typically developing children

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ABSTRACT

Introduction: Quantitative evaluation of upper limb (UL) kinematics in children with unilateral cerebral palsy (uCP) remains challenging for researchers and clinicians, especially during bimanual situations. This study proposed a new 3D bimanual protocol dedicated to children with uCP, called “Be an Airplane Pilot” (BE-API protocol) and assessed its reliability for typically developing children (TDC).

Methods: this protocol is composed of four bimanual tasks that allow the exploration of all degrees of freedom of the hemiplegic/non-dominant UL. Twenty TDC (mean age 11.9 ± 3.4) carried out three protocol sessions. Reliability was investigated through three kinematic parameters: angular waveforms (WAVE) using the coefficient of multiple correlation (CMC), range of motion (RoM) and maximum angles (MAX) both using the intra-class correlation coefficient (ICC) and the standard error of measurement (SEM).

Results: A very good reliability was observed for the three kinematic parameters in most cases (WAVE: $CMC \geq 0.90$, RoM & MAX: $ICC \geq 0.81$, $SEM \leq 5.0^\circ$).

Discussion: the very good reliability can be partly explained by the high level of rigor of the protocol. Such promising results open the door to validation tests on children with uCP. The BE-API protocol could pretend to support clinical decisions by objectively assessing the efficiency of therapeutics, e.g. injection of botulinic toxin.

1. Introduction

Quantitative evaluation of upper limb (UL) kinematics in disabled children represents a key question for researchers and clinicians especially in the field of unilateral cerebral palsy (uCP) because of its high prevalence [Himmelmann et al., 2005]. This pathology induces impaired motor control of the UL that hinders the execution of their daily life activities [Franco de Moura et al., 2016]. Although qualitative tools are used in clinical practice, such as the Assisting Hand Assessment (AHA) [Krumlinde-Sundholm et al. 2007], more precise information can be expected for a better understanding of motor limitations and a subsequent adaptation of therapeutics. Fine quantitative evaluation of UL kinematics in children with uCP, through three-dimensional movement analysis (3DMA) technologies, allows such benefits [Brochard et al., 2012; Coluccini et al., 2007; Gaillard et al., 2018; Kreulen et al., 2007; Mackey et al., 2005; Reid et al., 2010] and may pretend to support clinical decisions by objectively assessing the efficiency of therapeutics, e.g. injection of botulinic toxin or surgery. Such analysis depends on the motor tasks performed by children, and therefore the choice of these tasks is a crucial question to solve prior to clinical

evaluation. However, only a few studies have put their clinical protocol into place after investigating the reliability of the tasks. [Jaspers et al. (2011a); Jaspers et al. (2011b)] used a very complete approach, with validations performed on typically developing children (TDC) and children with uCP. Their protocol includes three reach tasks, two reach-to-grasp tasks and three gross motor tasks, for a total of eight standardized unimanual UL tasks. Similarly, Butler et al. [Butler et al., 2010] designed a protocol based on a unique unimanual reach-to-grasp task divided in six sequential subtasks. Both of these studies consist of unimanual tasks for which children are asked to use only their paretic UL. Generally speaking, this methodological choice can be discussed for two reasons. First, it does not correspond to most daily life situations for a child with uCP since he/she would spontaneously use his/her non-paretic UL to execute a unimanual activity [Rudisch et al., 2016]. Second, it does not allow the exploration of the UL movements in bimanual situations, which represent a major issue of execution for this pathological population. The bimanual coordination implies many complex cerebral structures, with possible higher defaults of motor planning, selective motor control and/or mirror movements [Eliasson, 2008; Jaspers et al., 2009].

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<https://doi.org/10.1016/j.jelekin.2019.08.001>

Received 16 April 2019; Received in revised form 6 August 2019; Accepted 9 August 2019

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A few recent studies have explored UL kinematics of children with uCP during bimanual tasks such as decanting cups, moving a box on a desk [Klotz et al., 2014] and opening a box plus pressing a button inside [Rudisch et al., 2016]. Although these tasks are interesting to study due to their connections to activities of daily living and to their association with a playful setting masking the clinical surrounding [Klotz et al., 2014], they suffer from not covering a full spectrum of degrees of freedom (DoF) of the paretic UL. Furthermore, none of these studies has investigated the reliability of the kinematic parameters. Assessing the reliability of a protocol, in TDC and children with uCP, represents a compulsory step before its deployment as a clinical routine, for any follow-up of a therapeutic/treatment. Studying the non-pathological population before the pathological one often represents the first step of validation of a protocol [Jaspers et al., 2011a,b].

The objectives of the present work are (1) to develop a 3DMA bimanual protocol designed for children with uCP and (2) to assess its reliability in TDC. This protocol allows the exploration of all degrees of freedom of the paretic UL, with standardized tasks integrated into a playful game situation of “Be an Airplane Pilot” (BE-API Protocol).

2. Methods

2.1. Participants

Twenty TDC (11 boys, 9 girls) aged from 6 to 18 years (mean age 11.9 ± 3.4 years old) voluntarily participated. Children had no history of UL complaints. Nineteen children were right-handed and one left-handed. Ethical approval was granted by the Ethics Committee of the University Hospital Rennes (France). Parents and children gave their written informed consent for this experiment.

2.2. BE-API protocol

The 3DMA protocol consists of a game scenario “Be an Airplane Pilot” (BE-API protocol) which is divided into four bimanual tasks (Fig. 1). These tasks were designed based on discussions with Physical Medicine and Rehabilitation therapists. Each task is specifically designed to involve a unique and natural bimanual strategy, and to explore one, two or three DoF of UL known as limited in children with uCP (“primary DoF”). For each task, all DoF of the UL can be classified

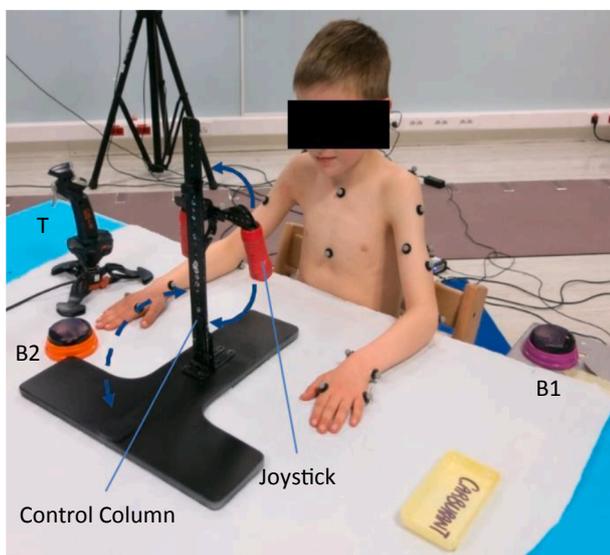


Fig. 1. A child with the set-up of the protocol BE-API. T is the turbo used for task 2, B1 is the buzzer 1 used for task 3 and B2 is the buzzer 2 used for task 4. Blue dash-arrow represents the movement of the control column during task 1, blue solid arrow represents the movement of the joystick during task 2.

as “primary”, “secondary” or “not explored” (Table 1), based on the a priori expected motion used to perform the task. The total set of four tasks allows the exploration of all the DoF of the UL: three DoF in the shoulder joint, two DoF in the elbow joint including the forearm pronosupination, two DoF in the wrist joint.

Prior to the start of the protocol, adjustments of the equipment were done according to the child anthropometry (Fig. 1). First, the seat is adjusted in order to obtain 90° hip and knee flexions. Second, the height of the table corresponds to the height of the elbow when the upper arm of the child is along the trunk. The table is placed in contact with the child’s belly in order to limit trunk motion. According to the child’s anthropometry, the position of all objects (i.e. joystick, turbo, buzzers 1 and 2), the height and the width of the joystick are adjusted.

Task 1, called “mountain passing”, consists of four consecutive forward-backward movements of the control column, ranging from its vertical starting position to its horizontal upward position. This task was carried out in the child’s sagittal plane, holding the joystick with the two hands. The prototype was designed to minimize the resistance or inertia effect from the control column. Primary DoF are elbow flexion-extension (elbFE) and wrist abduction-adduction (wriAA).

Task 2, called “slaloming”, consists of four consecutive rotations of the joystick of 180° with the non-dominant UL, from top to bottom, while the turbo is continuously pressed using the dominant UL. Primary DoF are shoulder elevation (shoEl) and internal external rotations (shoIER), and wrist flexion-extension (wriFE).

Task 3, called “dropping parachutists”, consists of four consecutive reach-and-press movements to buzzer 1 with the non-dominant UL, including a repositioning on the joystick after each pressure, while the joystick is maintained horizontal with the dominant UL. Primary DoF is the plane of elevation of the shoulder (shoPE).

Task 4, called “refueling”, starts with both forearms and hands flat on the table and consists of offering the non-dominant hand to receive a gas coin coming from the top, as soon as the buzzer 2 is pressed by the dominant UL. The coin was given by the operator at the level of the joystick. It is performed once. The primary DoF is forearm pronosupination (elbPS).

Details concerning the movements performed during the four tasks are available in the supplementary material (Supplementary figure).

The playful setting, including the speech of the operator, aims at surrounding the clinical environment [Klotz et al., 2014] and at leading to spontaneous movements of the child. All tasks were performed at self-selected speed. The chronological order of a protocol session was Task 1 – Task 4 – Task 2 – Task 4 – Task 3 – Task 4. Three consecutive sessions were performed by the child (Fig. 2), for a total duration of about 45 min, including a familiarization and briefing of 5 min performed beforehand.

2.3. Motion analysis tracking

An optoelectronic system (Motion Analysis, Rohnert Park, USA, sampling frequency of 100 Hz) with 12 cameras was used to track the 3D position of 15 reflective markers placed on the non-dominant UL and the trunk of the child. The marker placement, the definition of the segment coordinate systems and the choice of Euler sequences followed the recommendations of the international society of biomechanics (ISB) [Wu et al., 2005]. The shoulder joint was defined as the “thoracohumeral joint” and its joint center was estimated using a functional method [Gamage and Lasenby, 2002; Lempereur et al., 2010]. Markers were also placed on the dominant side of the child but were not used in this study.

2.4. Data analysis

Tasks 1, 2, and 3 correspond to a series of 4 consecutive cycles. The second and the third cycles were selected for data processing, to avoid any bias of start/stop strategies [Jaspers et al., 2011a,b]. Task 4

Table 1
Primary (big dashed cross) and secondary (small cross) DoF of the non-dominant UL and the trunk explored through each task.

	DoF	TASK 1	TASK 2	TASK 3	TASK 4
TRUNK	Flex-Ext	X			
	Rot Int-Ext			X	
SHOULDER	Elev	X	X		
	Plane Elev		X	X	X
	Rot Int-Ext		X	X	X
ELBOW	Flex-Ext	X			
	Prono-Sup		X	X	X
WRIST	Flex-Ext	X	X	X	X
	Abd-Add	X			

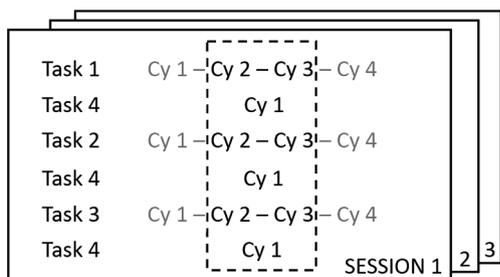


Fig. 2. The second and the third cycles of tasks 1, 2, 3 and all cycles of task 4 were selected for data processing, through the 3 sessions.

corresponds to a unique cycle performed after tasks 1, 2 and 3; these three cycles were retained for data processing. Considering that three consecutive sessions were performed by the child, a total of 6 movement cycles for tasks 1, 2 and 3, and 9 movement cycles for task 4 were retained for data processing (Fig. 2). The analysis was focused on the task execution (e.g. reaching the most anterior position in Task 1) which means we did not consider the “return to neutral position” movement in each cycle.

2.5. Statistical analysis

Reliability of the movement cycles was assessed through three kinematic parameters: the angular waveforms (WAVE), the maximum

Table 2
Within-day reliability of the WAVE parameter through mean CMC values with [95% confidence interval] for primary DoF (bold) and secondary DoF (non-bold).

	DoF	TASK 1	TASK 2	TASK 3	TASK 4
TRUNK	Flex-Ext	0.95 [0.91 - 0.98]			
	Rot Int-Ext			0.94 [0.92 - 0.96]	
SHOULDER	Elev	0.98 [0.96 - 0.99]	0.96 [0.95 - 0.97]		
	Plane Elev		0.97 [0.95 - 0.98]	0.98 [0.98 - 0.99]	0.90 [0.86 - 0.95]
	Rot Int-Ext		0.93 [0.91 - 0.95]	0.95 [0.91 - 0.98]	0.92 [0.87 - 0.96]
ELBOW	Flex-Ext	0.96 [0.94 - 0.98]			
	Prono-Sup		0.95 [0.93 - 0.97]	0.95 [0.91 - 0.97]	0.98 [0.96 - 0.99]
WRIST	Flex-Ext	0.93 [0.89 - 0.96]	0.97 [0.96 - 0.98]	0.75 [0.66 - 0.85]	0.74 [0.45 - 0.90]
	Abd-Add	0.97 [0.95 - 0.99]			

angle value (MAX) and the range of motion of the joint (RoM). It was applied for all DoF (primary and secondary) for all the four tasks. Reliability of WAVE was performed using the coefficient of multiple correlation (CMC) [Kadaba et al., 1990]. The CMC index quantifies the similarity between joint angle waveforms by taking into account differences in shape, offset, correlation, and range of motion. CMC was calculated for each child and the mean value was reported with 95% confidence interval. Four mean CMC thresholds were considered: excellent (≥ 0.90), good (0.80–0.89), moderate (0.60–0.79), and poor (< 0.60) [Jaspers et al., 2011a,b]. Reliability of MAX and RoM was assessed with the intraclass correlation coefficient (ICC(2,k)) and the standard error of measurement (SEM). Mean values were reported with 95% confidence interval. Four ICC thresholds were considered: very high (≥ 0.80), moderately high (0.60–0.79), moderate (0.40–0.59), and low (< 0.40) [Jaspers et al., 2011a,b].

3. Results

3.1. Angular waveforms

Within-day reliability of WAVE was excellent for all primary DoF during all tasks ($0.93 \leq \text{mean CMC} \leq 0.98$) (Table 2). The same observation of excellence is done for all secondary DoF during all tasks ($0.90 \leq \text{mean CMC} \leq 0.98$), with the exception of the wrist flexion–extension during tasks 3 and 4 with moderate values ($0.74 \leq \text{mean CMC} \leq 0.75$).

3.2. RoM and MAX

Within-day reliability of RoM parameter was very high for all primary DoF during all tasks ($0.86 \leq \text{mean ICC} \leq 0.96$), as well as for all secondary DoF during all tasks ($0.81 \leq \text{mean ICC} \leq 0.96$) (Table 3). All mean SEM values were between 1.7° and 4.3° for the primary DoF and between 1.7° and 5.0° for the secondary DoF. Concerning the MAX parameter, ICC values were very high for all primary DoF during all tasks tested ($0.83 \leq \text{mean ICC} \leq 0.98$), as well as for all secondary DoF during all tasks ($0.81 \leq \text{mean ICC} \leq 0.98$) (Table 4). All mean SEM values were between 0.9° and 3.8° for the primary DoF and between 1.3° and 3.5° for the secondary DoF.

The forward shoulder elevation angle is reported with positive

values to ease the understanding, the negative sign involved by the YXY sequence used ([Wu et al., 2005]) has been removed. The shoulder plane of elevation is negative for a posterior position, and positive for an anterior position. A maximal elbow extension is at 0° or slightly negative. The neutral forearm pronosupination is at 0°, a supination position has a negative value and a pronation position has a positive value. “Dev uln” stands for “ulnar deviation”.

4. Discussion

4.1. Development of a new protocol

Any development of a new protocol requires a good definition of the scientific aim [Kontaxis et al., 2009]. The final clinical outcome of this protocol is to be able to study the upper limb movements of TDC and children with uCP during bimanual situations in order to objectively assess the efficiency of clinical uCP-oriented therapeutics. To do so, all tasks were conceived in order to (1) be fully feasible by TDC, induce sufficient motion in DoF known as limited in children with uCP, (3) involve a unique and natural strategy of performance in bimanual condition. All the gaming elements were carefully positioned on the table to guarantee the main action of the asymmetric tasks was performed by the non-dominant limb (or the hemiplegic limb of the child with uCP) and not by the other limb. A special attention was put on the complementarity of tasks regarding the DoF explored. Various conditions of task execution were represented: with symmetrical and asymmetrical tasks, with guided and free trajectories, with and without object grasping. Tasks had also to be homogeneous to fit into a same environment of game (thematic of airplane) and to follow a plausible game scenario. Although it is often disregarded in the literature [Butler et al., 2010; Jaspers et al., 2011a,b; Kreulen et al., 2007], the presence of a playful environment is important to mask the surrounding clinical environment [Klotz et al., 2014] and to lead to spontaneous movements of the child. In the game scenario, the child had to pass four mountains, to perform four slaloms and to drop four parachutists during tasks 1, 2 and 3 respectively. The operator’s speech, the elements used (including noisy buzzers) and the decoration of the table and the room were adapted to the thematic of airplane in our study. The set-up had also to be thought for left and right-handed children, without any restriction of height and weight.

Table 3

Within-day reliability of the RoM parameter expressed with mean ICC with 95% Confidence Interval ([95% CI]), and mean SEM with [95% CI] for primary DoF (bold) and secondary DoF (non-bold). Mean RoM values with standard deviation [SD] are also presented.

	DoF	TASK 1			TASK 2			TASK 3			TASK 4		
		Mean (°) [SD]	mean ICC [95% CI]	mean SEM (°) [95% CI]	Mean (°) [SD]	mean ICC [95% CI]	mean SEM (°) [95% CI]	Mean (°) [SD]	mean ICC [95% CI]	mean SEM (°) [95% CI]	Mean (°) [SD]	mean ICC [95% CI]	mean SEM (°) [95% CI]
TRUNK	Flex-Ext	27.5 [8.1]	0.91 [0.68 - 0.98]	2.6 [1.3 - 4.7]									
	Rot Int-Ext							16.4 [4.0]	0.85 [0.52 - 0.95]	1.7 [1.1 - 2.6]			
SHOULDER	Elev	37.9 [6.1]	0.84 [0.61 - 0.95]	2.7 [1.5 - 4.5]	35.7 [9.0]	0.96 [0.93 - 0.98]	1.8 [1.3 - 2.2]						
	Plane Elev				56.2 [13.9]	0.96 [0.91 - 0.98]	2.7 [1.9 - 3.6]	84.5 [11.6]	0.92 [0.80 - 0.95]	3.5 [2.8 - 4.3]	27.7 [10.1]	0.95 [0.89 - 0.97]	2.3 [2.0 - 2.7]
	Rot Int-Ext				47.8 [21.8]	0.96 [0.92 - 0.98]	4.3 [3.4 - 5.2]	37.7 [12.5]	0.92 [0.82 - 0.95]	3.7 [3.0 - 4.6]	37.2 [13.2]	0.95 [0.89 - 0.97]	2.9 [2.4 - 3.5]
ELBOW	Flex-Ext	42.1 [12.8]	0.94 [0.88 - 0.97]	3.2 [2.3 - 4.3]									
	Prono-Sup				59.5 [16.7]	0.92 [0.80 - 0.96]	5.0 [3.5 - 7.0]	43.9 [11.3]	0.88 [0.71 - 0.94]	4.2 [3.2 - 5.7]	102.0 [10.6]	0.95 [0.90 - 0.97]	2.3 [1.8 - 2.9]
WRIST	Flex-Ext	32.9 [10.5]	0.92 [0.80 - 0.96]	2.9 [2.2 - 3.6]	55.3 [8.6]	0.86 [0.66 - 0.94]	3.5 [2.4 - 4.9]	22.0 [8.1]	0.81 [0.56 - 0.90]	4.1 [3.0 - 5.7]	17.5 [5.6]	0.89 [0.71 - 0.94]	2.0 [1.4 - 2.8]
	Abd-Add	29.9 [5.5]	0.92 [0.82 - 0.96]	1.7 [1.2 - 2.2]									

Table 4

. Within-day reliability of the MAX parameter expressed with mean ICC with 95% Confidence Interval ([95% CI]), and mean SEM with [95% CI] for primary DoF (bold) and secondary DoF (non-bold). Mean MAX values with standard deviation [SD] are also presented.

	Max of DoF	TASK 1			TASK 2			TASK 3			TASK 4		
		MEAN (°) [SD]	mean ICC [95% CI]	mean SEM (°) [95% CI]	MEAN (°) [SD]	mean ICC [95% CI]	mean SEM (°) [95% CI]	MEAN (°) [SD]	mean ICC [95% CI]	mean SEM (°) [95% CI]	MEAN (°) [SD]	mean ICC [95% CI]	mean SEM (°) [95% CI]
TRUNK	Flex (+)	26.7 [8.1]	0.97 [0.94-0.99]	1.3 [0.9-1.7]									
	Rot Ext (+)							14.2 [4.8]	0.89 [0.60-0.96]	1.6 [1.1-2.3]			
SHOULDER	Elev (+)	88.9 [10.4]	0.98 [0.95-0.99]	1.6 [1.1-2.1]	71.8 [10.0]	0.95 [0.91-0.97]	2.3 [1.7-2.8]						
	Plane Elev (posterior) (-)							-19.7 [9.9]	0.89 [0.67-0.94]	3.5 [2.6-4.5]			
	Plane Elev (anterior) (+)				103.8 [11.4]	0.95 [0.91-0.97]	2.5 [2.0-3.0]				68.1 [9.2]	0.93 [0.86-0.97]	2.4 [1.9-3.1]
	Rot Int (+)							-5.1 [13.3]	0.93 [0.88-0.96]	3.5 [2.6-4.4]			
	Rot Ext (-)				-77.6 [19.7]	0.96 [0.92-0.98]	3.8 [3.1-4.5]				-63.3 [14.9]	0.96 [0.93-0.98]	2.8 [2.1-3.6]
ELBOW	Ext	32.5 [6.7]	0.91 [0.81-0.96]	2.1 [1.3-2.9]									
	Prono (+)							42.1 [11.9]	0.97 [0.93-0.98]	2.1 [1.7-2.6]			
	Sup (-)				-41.2 [10.1]	0.90 [0.64-0.96]	3.3 [2.2-5.0]				-63.8 [6.0]	0.98 [0.96-0.99]	0.9 [0.7-1.2]
WRIST	Flex (+)	15.2 [7.7]	0.91 [0.82-0.95]	2.4 [1.9-3.1]				7.8 [6.1]	0.81 [0.55-0.91]	3.0 [2.1-4.0]			
	Ext (-)				-39.5 [7.0]	0.83 [0.63-0.92]	3.2 [2.3-4.3]				-14.9 [5.0]	0.88 [0.63-0.95]	1.9 [1.3-2.7]
	Dev uln (+)	34.1 [3.2]	0.85 [0.71-0.92]	1.4 [0.9-1.8]									

4.2. 3D data tracking and processing

Marker occlusions were minimized by having a capture area above the table and by using thin and small objects. Both upper limbs were equipped with reflective markers, for allowing future complementary investigations and for leaving the child in the dark concerning the purpose of the experiment. Alternative technologies such as inertial sensors may be used, with different advantages and drawbacks [Bouvier et al., 2015]. The attribution for a DoF to be “primary” or “secondary” was established a priori considering the DoF targeted by the task. This attribution was confirmed a posteriori according to the RoM and MAX values actually calculated in the TDC group. Note: the shoulder elevation angle has been remained as “primary” and “secondary” DoF during tasks 2 and 1 respectively, although very convincing results were also obtained for task 1. Indeed, task 2 is a shoulder-oriented task which also involve motions of shoulder plane of elevation and rotations, whereas task 1 is not and may involve passive motion of the shoulder elevation due to high elbow and trunk contributions. These denominations are not strict and may evolve regarding future results on children with uCP.

4.3. Feasibility and adherence

This protocol was easily feasible by all children, aged from 6 to 18. Fifteen minutes for marker equipment and adjustments of the set-up, plus five minutes for briefing and familiarization were needed beforehand. One session lasted about 10 min. It is not recommended to use the protocol for children under the age of 6 due to lower ability of mental focus or a sequential temporal coupling of both hands [Rudisch et al., 2016]. The playful environment on the thematic of “Be an Airplane Pilot” was appreciated by all children. The adherence of children aged from 6 to 12 was very high. Children aged from 13 to 18 were slightly less attracted by the game scenario, the use of a video game or the virtual reality coupled with the set-up would possibly increase their adherence [Kerem et al., 2014].

4.4. Exploration of DoF of the UL

One of the objectives of the BE-API protocol was to explore the DoF of the UL during functional tasks, in range of motion that correspond to activities of daily living (ADL). Generally speaking, the BE-API protocol stimulates all the DoF of the non-dominant UL in ranges of motion and with maximum angle values that correspond to functional ranges of motion during ADL [Gates et al., 2016]. The mean maximal shoulder elevation was measured as 88.9° and 71.8° during tasks 1 and 2 respectively and corresponds to the maximum needed to drink a glass, 62° and 87° as reported by [van Andel et al., 2008] and [Aizawa et al., 2010] respectively. Mean maximal shoulder plane of elevation was reported as -19.7° during the task 3 and is in accordance with most of ADL such as fastening a button at navel level (-13° [Aizawa et al., 2010]), excepted very demanding backward tasks (i.e. “hand to pocket” with -65° [Gates et al., 2016]). Maximal shoulder external rotation, measured during tasks 2 and 4 with values of 77.6° and 63.3° respectively are also compatible with various activities of daily living (32° and 55° as reported by [Petuskey et al., 2007] and [Gates et al., 2016] resp.). The elbow extension explored during task 1 is sub-maximal with a value of 32.5° and corresponds to most of ADL such as pouring from a pitcher (36°) or using a telephone (43°) [Morrey et al. 1981]. Task 1 may involve assistance from one UL to the other since both hands grasped the joystick, as it can occur in certain ADL of the child too. Task 4 succeeds in exploring the forearm supination in depth with mean maximum value of 63.8° higher than those reported by Jaspers et al. (15°) [Jaspers et al., 2011a,b], Klotz et al. during bimanual tasks (28.4°) [16], and higher than the maximal value for five different ADL (53°) [24]. Concerning the wrist joint, maximal and RoM values are not wide due to the limited RoM of this joint and are similar to those reported in the literature [Jaspers et al., 2011a,b]. Lastly, this protocol was not originally conceived to stimulate the trunk motion, the small motion measured on two DoF during tasks 1 and 3 has been reported to leave the door open to a future exploration of compensation movements from children with uCP.

4.5. Reliability on TDC

Results indicated a very high reliability of the three kinematic parameters (WAVE, RoM, MAX), for all DoF, for all tasks, through all statistical indicators (CMC, ICC, SEM). Concerning WAVE parameter, most of CMC values for primary and secondary DoF of shoulder and elbow joints were superior or equal to 0.90 and are higher than the corresponding literature. [Jaspers et al., 2011a,b] and [Vanezis et al., 2015] reported CMC superior to 0.80 for shoulder and elbow joints during reaching tasks, with or without grasping, hand to mouth/head/shoulder tasks, and a throwing task, performed by TDC. Lower CMC values for the wrist flexion-extension during tasks 3 and 4 (0.74–0.75) are also consistent with these two studies, and may be explained by the smaller range of motion of this joint during these two tasks (17.5°–22.0°), in comparison with tasks 1 and 2 (32.9°–55.3°). It must be reminded that CMC index is sensitive to the range of motion, the number of subjects or even the sampling frequency used, therefore any comparison of CMC values from different studies has to be done with precaution [Roislien et al., 2012].

Concerning RoM and MAX parameters, very high ICC values (ICC \geq 0.81) presented in our study were superior to those reported by [Jaspers et al., 2011a,b] for similar UL joint angles measured at a point of task achievement (ICC > 0.60). Similarly, SEM values (SEM \leq 5.0°) were a bit smaller than those presented in the literature, (SEM \leq 7.8° [Vanezis et al., 2015], SEM \leq 8.5° [Jaspers et al., 2011a,b] reported for movements a bit wider than in our study). These small values of SEM are optimistic for investigation on children with uCP, since they would guarantee a minimal detectable change inferior to 14° for any degree of freedom investigated.

These very high values of reliability can be partly explained by the high level of rigor of the protocol, including a strict placement of objects and some trajectories guided by objects (tasks 1 and 2). Ensuring excellent results on reliability for TDC is an important first validation step, since higher variability may be expected for children with uCP. The more reliable the BE-API protocol will be, the more efficient and useful clinical assessment may also be.

Kinematic parameters of this study have been narrowed to UL joint angles to draw a parallel with the methodology adopted for the lower limb during the quantitative gait analysis, already widely spread as a clinical routine. Other parameters such the movement duration, speed [Jaspers et al., 2011a,b], movement efficiency [Butler et al., 2010] represent potential perspectives, by looking at the future results of children with uCP and at the comparison between the two populations. Crossing joint angle information from the two UL of the child may also be of interest to explore the bimanual coordination.

5. Conclusions

The BE-API protocol is a new 3DMA bimanual protocol dedicated to the assessment of the upper limb kinematics of children. It is composed of four complementary bimanual tasks that allow the exploration of all DoF of the non-dominant UL, with a specific focus on DoF known as limited in children with uCP. The UL movements are stimulated in functional ranges of motion consistent with activities of daily living and are carried out during a game scenario of “Be an Airplane Pilot”. A very good reliability of kinematic parameters has been observed on TDC. Such results are very promising and open the door to further tests on children with uCP and to the definition of first normative values based on TDC. Such a protocol would aim at supporting clinical decisions by objectively assessing the efficiency of therapeutics, e.g. injection of botulinic toxin or surgery.

Declaration of Competing Interest

None.

Acknowledgements

The authors would like to thank all children and parents for participating in this study. Special thanks to Bertrand Elie for his help to design the protocol.

Appendix A. Supplementary material

Supplementary data to this article can be found online at <https://doi.org/10.1016/j.jelekin.2019.08.001>.

References

- Aizawa, J., Masuda, T., Koyama, T., Nakamaru, K., Isozaki, K., Okawa, A., Morita, S., 2010. Three-dimensional motion of the upper extremity joints during various activities of daily living. *J. Biomech.* 43 (15), 2915–2922. <https://doi.org/10.1016/j.jbiomech.2010.07.006>.
- Bouvier, B., Duprey, S., Claudon, L., Dumas, R., Savescu, A., 2015. Upper limb kinematics using inertial and magnetic sensors: comparison of sensor-to-segment calibrations. *Sensors* 15, 18813–18833. <https://doi.org/10.3390/s150818813>.
- Brochard, S., Lempereur, M., Mao, L., Rémy-Néris, O., 2012. The role of the scapulothoracic and gleno-humeral joints in upper-limb motion in children with hemiplegic cerebral palsy. *Clin. Biomech.* <https://doi.org/10.1016/j.clinbiomech.2012.04.001>.
- Butler, E.E., Ladd, A.L., LaMont, L.E., Rose, J., 2010. Temporal-spatial parameters of the upper limb during a reach & grasp cycle for children. *Gait Post.* <https://doi.org/10.1016/j.gaitpost.2010.05.013>.
- Coluccini, M., Maini, E.S., Martelloni, C., Sgandurra, G., Cioni, G., 2007. Kinematic characterization of functional reach to grasp in normal and in motor disabled children. *Gait Post.* <https://doi.org/10.1016/j.gaitpost.2006.12.015>.
- Eliasson, A.-C. (Karolinska I., & Burtner, P. A. (University of N. M. (Eds.). Bimanual coordination in children with hemiplegic cerebral palsy. In Improving hand function in children with cerebral palsy: theory evidence and intervention. London: Mac Keith Press, 1st ed., (2008) pp. 160–175.
- Franco de Moura, R.C., Almeida, C.S., Dumont, A.J.L., Lazzari, R.D., Lopes, J.B.P., Duarte, N.A. de C., Oliveira, C.S., 2016. Kinematic upper limb evaluation of children and adolescents with cerebral palsy: a systematic review of the literature. *J. Phys. Therapy Sci.* <https://doi.org/10.1589/jpts.28.695>.
- Gaillard, F., Cretual, A., Cordillet, S., Le Cornec, C., Gonthier, C., Bouvier, B., Rauscent, H., 2018. Kinematic motion abnormalities and bimanual performance in children with unilateral cerebral palsy. *Development. Med. Child Neurol.* <https://doi.org/10.1111/dmcn.13774>.
- Gamage, S.S.H.U., Lasenby, J., 2002. New least squares solutions for estimating the average centre of rotation and the axis of rotation. *J. Biomech.* [https://doi.org/10.1016/S0021-9290\(01\)00160-9](https://doi.org/10.1016/S0021-9290(01)00160-9).
- Gates, D.H., Walters, L.S., Cowley, J., Wilken, J.M., Resnik, L., 2016. Range of motion requirements for upper-limb activities of daily living. *American J. Occupat. Ther.* <https://doi.org/10.5014/ajot.2016.015487>.
- Himmelman, K., Hagberg, G., Beckung, E., Hagberg, B., Uvebrant, P., 2005. The changing panorama of cerebral palsy in Sweden. IX. prevalence and origin in the birth-year period 1995–1998. *Acta Paediatr* 94, 287–294. <https://doi.org/10.1080/08035250410023313>. April 2004.
- Jaspers, E., Desloovere, K., Bruyninckx, H., Molenaers, G., Klingels, K., Feys, H., 2009. Review of quantitative measurements of upper limb movements in hemiplegic cerebral palsy. *Gait Post.* <https://doi.org/10.1016/j.gaitpost.2009.07.110>.
- Jaspers, E., Feys, H., Bruyninckx, H., Cutti, A., Harlaar, J., Molenaers, G., Desloovere, K., 2011a. The reliability of upper limb kinematics in children with hemiplegic cerebral palsy. *Gait Post.* <https://doi.org/10.1016/j.gaitpost.2011.01.011>.
- Jaspers, E., Feys, H., Bruyninckx, H., Harlaar, J., Molenaers, G., Desloovere, K., 2011b. Upper limb kinematics: development and reliability of a clinical protocol for children. *Gait Post.* <https://doi.org/10.1016/j.gaitpost.2010.11.021>.
- Kadaba, M.P., Ramakrishnan, H.K., Wootten, M.E., 1990. Measurement of lower extremity kinematics during level walking. *J. Orthop. Res.* 8 (3), 383–392. <https://doi.org/10.1002/jor.1100080310>.
- Kerem, M., Kaya, O., Ozal, C., Turker, D., 2014. Virtual reality in rehabilitation of children with cerebral palsy. *Cerebral Palsy - Challenges Future.* <https://doi.org/10.5772/57486>.
- Klotz, M.C.M., van Drongelen, S., Rettig, O., Wenger, P., Gantz, S., Dreher, T., Wolf, S.I., 2014. Motion analysis of the upper extremity in children with unilateral cerebral palsy—an assessment of six daily tasks. *Res. Development. Disabilit.* <https://doi.org/10.1016/j.ridd.2014.07.021>.
- Kontaxis, A., Cutti, A.G., Johnson, G.R., Veeger, H.E., 2009. A framework for the definition of standardized protocols for measuring upper-extremity kinematics. *Clin. Biomech.* 24 (3), 246–253. <https://doi.org/10.1016/j.clinbiomech.2008.12.009>.
- Kreulen, M., Smeulders, M.J.C., Veeger, H.E.J., Hage, J.J., 2007. Movement patterns of the upper extremity and trunk associated with impaired forearm rotation in patients with hemiplegic cerebral palsy compared to healthy controls. *Gait Post.* <https://doi.org/10.1016/j.gaitpost.2006.05.015>.
- Krumlinde-Sundholm, L., Holmefur, M., Kottorp, A., Eliasson, A.C., 2007. The Assisting Hand Assessment: Current evidence of validity, reliability, and responsiveness to change. *Development. Med. Child Neurol.* 49 (4), 259–264. <https://doi.org/10.1111/j.1469-8749.2007.00259.x>.
- Lempereur, M., Leboeuf, F., Brochard, S., Rousset, J., Burdin, V., Rémy-Néris, O., 2010. In

- vivo estimation of the glenohumeral joint centre by functional methods: accuracy and repeatability assessment. *J. Biomech.* 43 (2), 370–374. <https://doi.org/10.1016/j.jbiomech.2009.09.029>.
- Mackey, A.H., Walt, S.E., Lobb, G.A., Stott, N.S., 2005. Reliability of upper and lower limb three-dimensional kinematics in children with hemiplegia. *Gait Post.* <https://doi.org/10.1016/j.gaitpost.2004.06.002>.
- Morrey, B.F., Askew, L.J., Chao, E.Y., 1981. A biomechanical study of normal functional elbow motion. *J. Bone Joint Surgery. American* 63 (6), 872–877. <https://doi.org/10.1007/s10439-012-0680-7>.
- Petuskey, K., Bagley, A., Abdala, E., James, M.A., Rab, G., 2007. Upper extremity kinematics during functional activities: Three-dimensional studies in a normal pediatric population. *Gait Post.* <https://doi.org/10.1016/j.gaitpost.2006.06.006>.
- Reid, S., Elliott, C., Alderson, J., Lloyd, D., Elliott, B., 2010. Repeatability of upper limb kinematics for children with and without cerebral palsy. *Gait Post.* <https://doi.org/10.1016/j.gaitpost.2010.02.015>.
- Røislien, J., Skare, O., Opheim, A., Rennie, L., 2012. Evaluating the properties of the coefficient of multiple correlation (CMC) for kinematic gait data. *J. Biomech.* 45 (11), 2014–2018. <https://doi.org/10.1016/j.jbiomech.2012.05.014>.
- Rudisch, J., Butler, J., Izadi, H., Zielinski, I.M., Aarts, P., Birtles, D., Green, D., 2016. Kinematic parameters of hand movement during a disparate bimanual movement task in children with unilateral Cerebral Palsy. *Human Movement Sci.* <https://doi.org/10.1016/j.humov.2016.01.010>.
- van Andel, C.J., Wolterbeek, N., Doorenbosch, C.A., Veeger, D.H., Harlaar, J., 2008. Complete 3D kinematics of upper extremity functional tasks. *Gait Post.* 27 (1), 120–127. <https://doi.org/10.1016/j.gaitpost.2007.03.002>.
- Vanezis, A., Robinson, M.A., Darras, N., 2015. The reliability of the ELEPAP clinical protocol for the 3D kinematic evaluation of upper limb function. *Gait Post.* 41 (2), 431–439. <https://doi.org/10.1016/j.gaitpost.2014.11.007>.
- Wu, G., van der Helm, F.C., Veeger, H.E., Makhssous, M., Van Roy, P., Anglin, C., Buchholz, B., 2005. ISB recommendation on definitions of joint coordinate systems of various joints for the reporting of human joint motion—Part II: shoulder, elbow, wrist and hand. Retrieved from. *J. Biomech.* 38 (5), 981–992. <http://www.ncbi.nlm.nih.gov/pubmed/15844264>.

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