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Abstract: In this multiple single-cases study, we used dance to train Sensorimotor Synchronization (SMS), motor and cognitive functions in children with Developmental Cerebellar Anomalies (DCA). DCA are rare dysfunctions of the cerebellum that affect motor and cognitive skills. The cerebellum plays an important role in temporal cognition including SMS which is, critical for motor and cognitive development. Dancing engages the SMS neuronal circuitry, composed of the cerebellum, the basal ganglia and the motor cortices. Thus, we hypothesized that dance has a beneficial effect on SMS skills and associated motor and cognitive functions in children with DCA. Seven children (aged 7-11) with DCA participated in a 2-month dance training protocol (3h/week). A test-retest design protocol with multiple baselines was used to assess children's SMS skills as well as motor, cognitive and social abilities. SMS skills were impaired in DCA before the training. The training led to improvements in SMS (reduced variability in paced tapping), balance and executive functioning (cognitive flexibility), as well as in social skills (social cognition). The beneficial effects of the dance training were visible in all participants. Notably, gains were maintained two months after the intervention. These effects are likely to be sustained by enhanced activity in SMS brain networks due to the dance training protocol.

Keywords: Cerebellar anomalies, sensorimotor synchronization, rhythm, rehabilitation, dance

1. Introduction

Developmental Cerebellar Anomalies (DCA) are rare diseases that affect cerebellar functions in less than one in 2000 (Musselman et al., 2014). The cerebellum is a pivotal brain region that contributes to motor and cognitive control as well as social and affective functions (Bareš et al., 2018; Hoche, Guell, Sherman, Vangel, & Schmahmann, 2016; Koziol et al., 2014; Manto et al., 2012; Schmahmann & Sherman, 1998; Therrien & Bastian, 2019). It achieves maturity a few years after birth, making it particularly vulnerable to developmental disorders (ten Donkelaar, Lammens, Wesseling, Thijssen, & Renier, 2003; Wang & Zoghbi, 2001). Unfortunately, very few specific remediation strategies to compensate for DCA currently exist (Brenna, 2014), possibly because of the rarity of this disease. The current research focuses on an innovative training protocol using dance to improve motor, cognitive and social skills in children with DCA.

DCA can have various causes (e.g., Joubert and Dandy-Walker syndromes, intrauterine trauma or intoxication). Cerebellar syndromes found in DCA are often explained by structural and volumetric anomalies of the cerebellum (Boltshauser, 2004; Poretti, Boltshauser, & Doherty, 2014). They can also be diagnosed on the basis of clinical observations in the absence of neuroimaging evidence (Ryan & Engle, 2003). Predominantly, DCA is associated with congenital forms of cerebellar ataxia (Fogel, 2012; Marsden, 2018). Ataxia refers to abnormal organization of movement and impaired coordination of balance, gait, extremity and eye movements, and dysarthria. DCA is also consistently associated with cognitive deficits (e.g., in executive functions, visual-spatial skills, linguistic abilities; Bodranghien et al., 2016; De Smet, Paquier, Verhoeven, & Mariën, 2013; Schmahmann, 2004; Schmahmann, Guell, Stoodley, & Halko, 2019; Schmahmann & Sherman, 1998) and emotional, social and communication deficits (Bolduc & Limperopoulos, 2009; Guell, Hoche, & Schmahmann, 2015; Hoche et al., 2016; Koziol et al., 2014; Schmahmann, 2004; Schmahmann & Sherman, 1998).

The cerebellum plays a crucial role in motor control (Manto et al., 2012); for example, it is involved in the control of oculomotor movements (Thier & Markanday, 2019), speech (Ackermann, 2008), grasping (Nowak, Topka, Timmann, Boecker, & Hermsdörfer, 2007), and voluntary limb movements (Ebner, Hewitt, & Popa, 2011). A specific contribution of the cerebellum to these tasks is the control of precise, online adaptive and predictive timing (Bastian, 2006; Manto et al., 2012; Therrien & Bastian, 2019). The cerebellum is involved in coordinating an action with temporally predictable, rhythmic events (Bareš et al., 2018; Braitenberg, Heck, & Sultan, 1997; Damm, Varoqui,

Cochen De Cock, Dalla Bella, & Bardy, 2019; Manto et al., 2012; Paquette, Fujii, Li, & Schlaug, 2017), especially in auditory–motor interactions (Doyon, Penhune, & Ungerleider, 2003; Provasi et al., 2014; Schwartze, Keller, & Kotz, 2016; Zatorre, Chen, & Penhune, 2007). This is referred to as Sensorimotor Synchronization (SMS, Repp, 2005; Repp & Su, 2013). SMS is usually studied via finger tapping to predictable stimuli such as a metronome or music sequences (Dalla Bella et al., 2017). The cerebellum is involved in the perception of sequences (Molinari et al., 2008), a capacity that is required for SMS. In addition, cerebellar circuitry is necessary to produce and adapt the next motor response using error information (i.e., discrepancy between the tap and the sound) of the previous response (Shadmehr, Smith, & Krakauer, 2010).

More generally, the cerebellum is part of a distributed neural system, including the basal ganglia, motor and premotor cortices, that enables temporal processing (Breska & Ivry, 2016; Coull & Nobre, 2008; Coull, Cheng, & Meck, 2011; Grahn, 2009; Grahn & Brett, 2007; Kotz & Schwartze, 2011). The cerebellum is crucial for timing and rhythmic tasks as well as for other motor and cognitive capacities in which timing and SMS are involved, such as gait (Damm, Varoqui, Cochen De Cock, Dalla Bella & Bardy, 2019; Bengtsson, Ehrsson, Forssberg, & Ullén, 2005; Dalla Bella, Benoit, Farrugia, Schwartze, & Kotz, 2015; Lang et al., 2017), language (Murdoch, 2010; Schwartze & Kotz, 2013) and social interactions (Van Overwalle, Manto, Leggio, & Delgado-García, 2019). Therefore, it is not surprising that patients with DCA are usually impaired in a variety of the aforementioned motor, cognitive and social capacities associated with SMS (Bodranghien et al., 2016; Koziol et al., 2014; Schmahmann, 2004; Schmahmann & Sherman, 1998).

To date, there is no direct evidence that SMS skills are affected in DCA, as no research studied SMS skills such as tapping to a metronome or music in these patients. Nevertheless, studies have reported other timing impairments with other cerebellar acquired or degenerative pathologies such as spino cerebellar ataxia or infarction. They are visible in tasks requiring precise timing, such as eye movements, spatio-temporal prediction, predictive motor timing and finger movements in overarm throwing (Broersen et al., 2016; Ivry & Keele, 1989; Ivry, Spencer, Zelaznik, & Diedrichsen, 2006; Timmann, Watts, & Hore, 1999). Interestingly, timing disorders are not confined to motor tasks, as cerebellar anomalies are also associated with poorer performance in perceptual tasks such as duration discrimination (Ivry & Keele, 1989; Ivry, Spencer, Zelaznik, & Diedrichsen, 2002). Provasi et al. (2014) studied SMS and rhythm perception in children with acquired (not developmental) cerebellar tumors (medulloblastoma) and showed that patients displayed

more tapping variability in SMS task, despite normal rhythm perception. Therefore, it is likely that patients with DCA also have difficulties in SMS.

Because of the role played by the cerebellum in motor and sensorimotor skills, dance is of particular interest for persons with cerebellar dysfunctions. Dance is an engaging activity involving an elaborate form of full body synchronization to a musical beat in a nonverbal communicative, aesthetic context (Joufflineau, Vincent, & Bachrach, 2018; Laland, Wilkins, & Clayton, 2016). It engages neuronal networks including the premotor and parietal cortices ("action observation network"), the supplementary motor area (SMA), the motor cortex, the basal ganglia and the cerebellum (Bachrach, Jola, & Pallier, 2016; Brown, Martinez, & Parsons, 2006; Cross, Hamilton, & Grafton, 2006; Cross, Kraemer, Hamilton, Kelley, & Grafton, 2009; Giacosa, Karpati, Foster, Penhune, & Hyde, 2016; Karpati, Giacosa, Foster, Penhune, & Hyde, 2015). The anterior cerebellar vermis and cerebellar lobules V and VI appear as key structures for the entrainment of movement to external auditory timing cues in dance (Brown et al., 2006) and expert dancers reveal brain changes, structural and functional, in these regions. Expertise in dance is also associated with improved sensorimotor functions (Bläsing et al., 2012; Jin et al., 2019; Karpati, Giacosa, Foster, Penhune, & Hyde, 2016), and proprioceptive body representation (Jola, Davis, & Haggard, 2011), thus supporting the tight link between brain networks affected by dance training and the sensorimotor and proprioceptive functions.

During the last decade, promising results showed that dance training can be used successfully for improving motor and cognitive functions in a variety of conditions such as aging (Chuang, Hung, Huang, Chang, & Hung, 2015; Coubard, Duretz, Lefebvre, Lapalus, & Ferrufino, 2011), neurodegenerative diseases (de Natale et al., 2017), stroke (Sampaio, Subramaniam, Arena, & Bhatt, 2016) and neurodevelopmental diseases (Grönlund, Renck, & Weibull, 2005). For example, dance training showed beneficial effects on motor and cognitive skills in Parkinson's disease (dos Santos Delabary, Komeroski, Monteiro, Costa, & Haas, 2018; Hackney et al., 2015; Hackney, Kantorovich, Levin, & Earhart, 2007; McKee & Hackney, 2013; Shanahan, Morris, Bhriain, Saunders, & Clifford, 2015), a condition associated with poorer rhythm and timing skills (Benoit et al., 2014; Grahn & Brett, 2009; Jones & Jahanshahi, 2014). Beneficial effects of a dance training protocol on motor functions were also observed in children with ADHD (Grönlund et al., 2005), a condition also affecting the cortico-cerebello-striatal pathways and rhythmic capacities (Bledsoe, Semrud-Clikeman, & Pliszka, 2011; Puyjarinet, Bégel, Lopez, Dellacherie, & Dalla Bella, 2017), and in children with cerebral palsy (López-Ortiz, Egan, & Gaebler-Spira, 2016; López-Ortiz et al., 2012). Moreover, consistent with the notion that

dance is a social and highly engaging activity (Chauvigné, Walton, Richardson, & Brown, 2019; Hackney & Gammon, 2011; Himberg, Laroche, Bigé, Buchkowski, & Bachrach, 2018; Joufflineau et al., 2018), positive effects of this activity on psychosocial behavior and emotional responses are reported, especially in children with developmental disorders (Grönlund et al., 2005; Jeong et al., 2005; Teixeira-Machado, Azevedo-Santos, & De Santana, 2017). Yet, it is not clear whether these motor and socio-affective improvements are mediated by SMS skills, because most studies did not include SMS testing. More generally, the effect of dance on cerebellar deficits has not been evaluated so far, except in one recent case study reporting the effects of an eight-week dance training protocol (based on an adapted form of tango) in an adult with a non-congenital cerebellar ataxia (Song, Ryu, Im, Lee, & Park, 2018). After the dance training, this patient displayed improved standing balance, gait characteristics, and functional mobility as well as self-declared improvements in emotion regulation and quality of life. This result is promising for the use of dance in cerebellar dysfunctions. However, so far, other physical therapy programs for patients with cerebellar dysfunctions, especially DCA, are scarce (Brenna, 2014), and, to the best of our knowledge, none included SMS or rhythmic components (Marsden & Harris, 2011; Martin, Tan, Bragge, & Bialocerkowski, 2009; Sartor-glittenberg & Brickner, 2014). We aimed to respond to this lack of rehabilitation methods in DCA by designing a sensorimotor training protocol based on dance.

The goal of this study was to test the effect of dance on a range of functions in children with DCA. This small-scale study included seven children with DCA who participated in a customized seven-week dance training program. We implemented a multiple baseline design adapted to small sample studies. With this method, the number of baselines (i.e., evaluation sessions before the protocol begins) was set randomly for each participant (between 4 and 7 baselines). This allows an accurate and rigorous assessment of the patient's initial performance. Therefore, the effects of the intervention can be highlighted more reliably than with a single baseline or with the same number of baselines for all participants (Himle, Miltenberger, Flessner, & Gatheridge, 2004; Morgan & Morgan, 2008). In addition, analyses methods adapted to single-case studies were used (Crawford & Garthwaite, 2007). Finally, a control group was tested in order to compare patients' skills with those of age-matched typically developing children and to control for the test-retest effect and the normal developmental evolution of the main tested domains.

Children SMS's skills when tapping to an external stimulus were evaluated. A battery of fine motor, cognitive tasks and social questionnaires was also used. We expected improved SMS skills after the training, with possible associated effects on motor, cognitive and social skills.

2. Methods

2.1.1. Participants

Patients (CEREB)

Case histories: Seven children (2 females, all right-handed) with developmental cerebellar anomalies, aged between 7.5 and 10.3 years at the inclusion stage participated in the study (Table 1). Patients were recruited at the Centre de Référence Maladies Rares: Malformations et Maladies Congénitales du Cervelet (CRMR, University Hospital of Lille) where they each received a multidisciplinary assessment including a neurological standard examination and a neuropsychological evaluation. No specific scale to evaluate congenital ataxia currently exists. Cerebellar functionality was evaluated with the items of the Scale for the assessment and rating of ataxia (SARA) (Schmitz-Hübsch et al., 2006), although the quantitative items of this scale were not specifically developed for this type of congenital ataxia. The results at the SARA revealed the presence of cerebellar signs in all patients (see Table 1). All patients were diagnosed with cerebellar ataxia of unknown etiology (Patients 1 to 6) or, in one case (Patient 7), with cerebellar ataxia in the context of a Joubert's syndrome linked to mutations of the gene CC2D2A (see Table 2). Cerebellar ataxia was associated with malformations observable on structural brain imagery (MRI) in Patients 1 (cisterna magna and slightly enlarged fourth ventricle), 4 (vermian dysplasia), 5 (abnormal fissuring of the superior vermis) and 7 (molar tooth sign, cerebellar dysplasia of superior vermis, and malformation of the cerebellar peduncles) whereas Patients 2, 3 and 6 did not present any morphological malformation visible on a structural MRI. The exclusion criteria were as follows : (1) radiological picture or medical history consistent with a diagnosis of acquired lesions due to pre-natal or perinatal issues; (2) progressive cerebellar pathology; (3) metabolic diseases associated with congenital malformation of the cerebellum; (4) malformation affecting other cerebral structures besides the cerebellum; (5) epileptic seizures or febrile convulsions or even focal or generalized paroxysmal EEG abnormalities (spikes or spike-wave complexes), (6) Intellectual disability (IQ<70) in the patients clinical history. Table 2 presents IQ indexes for each participant. All indexes are within the norms, although some patients' performances were in the low norms in verbal (VCI) and non-

verbal (PRI) and visual-spatial (VSI) indexes. Most patients displayed borderline performances in processing speed (PSI).

Control group (CONTROL):

A group of 20 children (CONTROL) aged between 7.00 and 10.58 years (M = 8.72, SD = 1.20, 7 females) were tested twice with a 8-week interval on a subset of tasks in order to control the test-retest effect and the effect of normal development on the childrens' performances. No children who displayed impairments in verbal comprehension and/or perceptual reasoning, with perceptual or motor impairments, neurological, psychiatric or developmental disorders were included in the control group.

The study was approved by the National Ethics Committee (CPP Sud Est III, France, EudraCT: 2017-A03300-53). All participants and their parents gave their informed consent before testing and intervention in accordance with the Declaration of Helsinki.

2.2. Procedure

Figure 1 shows the experimental design for the CEREB group. Children in the CEREB group took part in a seven-week dance training protocol at the Eurasport center (University of Lille, France). Before and after the training, a complete individual testing session was administered to each participant. Testing included the evaluation of movement, sensorimotor and cognitive skills. Questionnaires assessing social cognition, executive functioning, and anxiety were also administered. Two months after the training, a two-hour evaluation session was administered in order to evaluate the long-term effects of the training.

For a subset of tasks, a multiple baseline design was used. We administered randomized baseline periods (i.e., each participant received a different number of short pre-intervention evaluations). Children were randomly assigned from 4 to 7 multiple baselines. These short evaluation sessions lasted 15 minutes. Repeated measures with the same subtest of tasks were also administered on a weekly basis during treatment. Since the intervention starts for all patients at the same time after a different number of baselines between participants, if an improvement is observed after the start of the intervention regardless of the number of baselines, this improvement can be attributed with more certainty to the

intervention. In other words, this multiple baseline methodological design reinforces the inference of causal links between the intervention and the observed improvement.

2.3. Training

CEREB children participated in a seven-week dance training protocol with a total of fourteen dance classes of one and a half hours each given regularly by a professional dance teacher. Dance training consisted in exercises implying SMS (drumming and synchronization to music and partners, walking in rhythm), body and space exploration (progressive movements, jumps, balance), learning and creating choreographies, as well as improvisation and singing.

2.4. Experimental measures

2.4.1. Rhythm assessment

Perceptual rhythm assessment: the Beat Alignment Test (BAT, Iversen & Patel, 2008) was taken from The Battery for the Assessment of Auditory and Sensorimotor Timing Abilities (BAASTA, Dalla Bella et al., 2017). The task was to judge whether a metronome was aligned or not with the beat of music. Two 20-beat musical excerpts from Bach's « Badinerie » and two from Rossini's « William Tell Overture » were played at a tempo of 600ms of Inter-Beat Interval (IBI). After 7 beats, a metronome (isochronous tones with a triangle timbre) was superimposed onto the music. When unaligned, tones occurred earlier or later than the beat by 33% of the quarter note duration (phase shift, 12 stimuli), or the interval between the tones was increased or decreased by 10% of the quarter note duration (period shift, 12 stimuli). Stimuli were presented in pseudo-randomized order with PsychoPy 2 (http://www.psychopy.org/) installed on a Dell computer (XPS 13 9360 Model). Auditory stimuli were delivered via headphones (Sennheiser HD280) at a comfortable sound level (60 dB). Responses were provided verbally by the children, and entered by the Experimenter by pressing one of the two response keys ("yes" or "no"). "Yes" indicated the situation when the child considered that the metronome was aligned with the musical beat. The sensitivity index (*d'*) was used as an unbiased measure of detection performance. It is obtained by calculating the standardized difference between the Hits (i.e., when a misaligned metronome was correctly detected) and False alarms (i.e., when a misalignment was erroneously reported): $z(H) - z(FA)$.

Motor and sensorimotor rhythmic tasks: motor and sensorimotor rhythmic skills were assessed using finger tapping tasks from the BAASTA (Dalla Bella et al., 2017). A self-paced finger tapping (referred to as Unpaced tapping) was used to assess participants' spontaneous motor tempo and variability. Sensorimotor rhythmic skills were assessed

using finger tapping to the beat of a metronome or music (referred to as Paced tapping). For tapping tasks, a tablet version of BAASTA was used (Acer Iconia Tab 10 model) (Bégel, Verga, Benoit, Kotz, & Dalla Bella, 2018; Puyjarinet et al., 2017). Note that the timing precision of the presentation of the stimuli on the tablet was 1% of the inter onset interval. Auditory stimuli were delivered via headphones (Sennheiser HD280). Stimuli were presented at a comfortable sound level (60 dB). Children had to tap with their index finger on the tablet. In Unpaced Tapping, participants had to produce regular finger taps with their dominant hand at a comfortable rate for 60 seconds. In Paced Tapping to metronome and music, children's capacity to track the beat was tested with finger tapping synchronization tasks. The metronome sequences included 60 piano tones (tone frequency = 1319 Hz) of 600 ms IBI. Two additional conditions with slow (750 ms) and fast (450 ms) IBIs were used at pre and post testing (but not in weekly evaluations). 64-beats musical excerpts taken from Bach's « Badinerie » and Rossini's « William Tell Overture » (Beat = quarter note) were used in Paced Tapping with music. The IBI was 600 ms, which is within the range for optimal rhythm perception (Drake & Botte, 1993; London, 2012; Repp, 2006). All stimuli were repeated twice. Artifacts and outliers were discarded as follows. Artifacts were taps leading to Inter-Taps Intervals (ITIs) smaller than 100 ms. In addition, taps were considered as outliers when the ITI between the actual tap and the preceding tap was smaller than Q1 – 3*Interquartile range (IQR) or greater than Q3 + 3*IQR, where Q1 is the first quartile and Q3 is the third quartile (Dalla Bella et al., 2017). The first ten taps of each trial were discarded. In Unpaced tapping, the mean ITI was computed. The coefficient of variation (CV) of the ITI (the ratio of the SD of the ITIs over the mean ITI) was calculated as a measure of motor variability. In Paced tapping, the CV of the ITI was used as a measure of motor variability during a synchronization task. In Paced tapping with a metronome, the mean performance at the two trials was calculated for each tempo. The four music trials were averaged for tapping with music.

2.4.2. Balance and fine motor control

Subtests from the Movement Assessment Battery for Children (M-ABC-II, Henderson, Sugden, & Barnett, 2007) (Static and dynamic balance, Manual dexterity) and the NEPSY-II (Brooks, Sherman, & Strauss, 2010) (Hand imitation) were used to assess balance and motor skills. Three subtests from the M-ABC-II (Placing pegs in a peg board, Threading a lace and Drawing a line into a trail) were used to evaluate Manual dexterity. The percentile score at manual dexterity on the French version of the M-ABC-II (Soppelsa & Albaret, 2005) was calculated for each patient. In Static and dynamic balance (M-ABC-II), three subtests from the M-ABC-II (One-leg balance, Walking on a line

and Hopping on mats) were used. The final scores were the average time the child kept on the balance board for the One-leg balance task (Maximum = 30 seconds), the total number of steps for the Walking on a line task (Maximum = 15) and the number of hops averaged across the two legs for the Hopping on mats task (Maximum = 5). The percentile score at static and dynamic balance on the French version of the M-ABC-II (Soppelsa & Albaret, 2005) was also calculated for each patient. In Hand imitation (NEPSY-II), the experimenter presented up to 12 different positions with his hand. Children were asked to reproduce the position. The task was done first with the dominant hand and was repeated with the non-dominant hand. A total score (Maximum = 24) was calculated.

2.4.3. Neuropsychological assessment

A battery of neuropsychological tests was submitted. Subtests were taken from the Test of Everyday Attention for Children (TEA-ch, Manly et al., 2001) (Motor inhibition, Cognitive flexibility) and the NEPSY-II (Brooks et al., 2010) (Verbal inhibition and switching, Verbal and Non-verbal fluencies). The Motor inhibition task (TEA-ch) is a go-no go task consisting of pointing at squares while listening to short sequences of tones and stopping to point when a different timbre occurs. The final score is the number of good responses. In Cognitive flexibility (TEA-ch), Children had to count the number of "creatures" (i.e., green little monsters) visible all along their burrow. Arrows are interspersed among the creatures and pointed either upwards or downwards. They were instructed to begin counting the creatures one by one and to change the direction of counting when the sense of the next arrow was downwards, until the last creature was presented. The final score was calculated based on the number of good responses (Quality index) and the time spent (Speed index) to complete the task. A composite score of speed processing was also computed. Verbal inhibition and switching (NESPY-II) consisted in a series of black and white shapes (squares and circles or upwards or downwards arrows) that children had to denominate or tell the direction (denomination condition). In two other conditions, they were instructed to give the name of the other shape or of the other direction (inhibition), and to name the shape or give the direction when it was black and to give the name of the other shape or direction when it was white (change condition). The total number of errors and the time of execution were calculated for each condition. The total of errors across conditions was also calculated. In Verbal fluencies (NESPY-II), participants were asked to generate as many words as they could within a specific semantic category (Animals and foods/drinks) or initial letter (S and F) category for 60 seconds. A score of semantic fluency and a score of initial letter category were calculated by cumulating the total number of correct words generated in each category. In Non-verbal fluencies (NESPY-II),

children had to connect structured or random dots presented in squares taking care not to form the same pattern as in previous squares. The total number of correct squares filled was considered as the final score for each condition, and a composite score for non-verbal fluencies was calculated by adding the two conditions.

2.4.4. Questionnaires

Questionnaires filled by the children and by the parents were used to assess executive functions in everyday life, social cognition, and anxiety. The Behavior Rating Inventory of Executive Function parent version (BRIEF, Gioia, Isquith, Guy, & Kenworthy, 2000), a validated questionnaire, was used to measure the child's executive function competence in the real-world setting. Six sub-scores (the higher the score the better) were provided: Inhibition, Cognitive flexibility, Emotional control, Initiation, Working Memory, Planification/Organisation, Organisation of materials and self-control. A global composite score was also computed. A Social cognition & theory of mind questionnaire created by our research team (Dellacherie et al, in preparation) was administered. It consisted in a series of 15 five-level Lickert scale questions evaluating children capacities in social cognition and theory of mind. The final score is the sum of the scores obtained for each question. Higher scores indicate higher estimated capacities. Finally, anxiety reported by the children was evaluated using the Revised Children's Manifest Anxiety Scale (R-CMAS, Reynolds & Richmond, 1979). A global score was calculated. Low scores indicated high anxiety.

2.5. Statistical analysis

Group statistical analyses: for all tasks, we compared CEREB children's performances before and after the training with Wilcoxon signed-rank test to highlight overall group differences. When a difference was observed, we also compared the difference between the pre-test and the follow-up test and between the post-test and the follow-up test. For tasks that have been included in the multiple baselines, the mean of the last four multiple baselines was used as the pre-training measure. In addition, we calculated the difference between the post and the pre-training (Delta pre/post), the follow-up and pre-training (Delta pre/follow-up) in the CEREB group, and between the retest and the test (Delta test/retest) in CONTROL. For tasks in which a significant difference between the pre- and the post- training evaluations was observed, we used Wilcoxon-Mann-Whitney U test to compare the Delta of the CEREB and the CONTROL groups.

Single-cases statistical analyses: CEREB performance at the pre and post-training were compared to controls' using statistics adapted for the analysis of single cases (DissocBayes program, Crawford & Garthwaite, 2007). For each

patient, a subgroup of six to eight age-matched (i.e., they were less than six months older or one year younger than the patient) control participants was constituted.

3. Results

3.1. Group analyses

Table 3 shows tasks for which a significant difference between the pre and the post training results were found. Note that almost all patients displayed a significant impairment before the training at these tasks (see below, "Single-case analysis" section).

Perceptual, motor and sensorimotor rhythmic tasks: When patients synchronized with a metronome and with music, tapping variability (CV) was significantly reduced after the training. The change was significantly greater in CEREB than in CONTROL. The differences with baseline were maintained at follow-up (Metronome: follow-up, $M = .18$, $SD = .17$, $Z = 2.20$, $p < .05$, $r = 0.83$; Music: follow-up, $M = .30$, $SD = .16$, $Z = 2.36$, $p < .01$, $r = 0.89$). No overall significant change was observed in perceptual rhythmic skills (BAT) and unpaced tapping ($p > .05$) in CEREB.

Balance and fine motor control: Hopping on mats was improved after the training in CEREB, and the difference was maintained at follow-up (follow-up, $M = 3.79$, $SD = 1.87$; $Z = 2.38$, $p < .05$, $r = 0.90$). The change in hopping on mats was significantly larger in CEREB than in CONTROL. No differences were observed between the pre and the post tests in the other variables ($p > .05$).

Neuropsychological assessment: in neuropsychological tests, cognitive flexibility was improved after the training, both in terms of number of correct answers and time; the difference was maintained at follow-up for good answers (follow-up, $M = 5.66$, $SD = 1.03$; $Z = 2.13$, $p < .01$, $r = 0.87$). The change was significantly greater in CEREB than in CONTROL for both correct answers and time. A difference was also found in the number of errors of the change condition in verbal inhibition and switching (before, $M = 8.62$, $SD = 5.64$; after, $M = 5.71$, $SD = 5.68$; $Z = 2.37$, $p < .01$, $r = 0.89$) and in the total number of errors (before, $M = 16.36$, $SD = 13.29$; after, $M = 10.86$, $SD = 9.62$; $Z = 1.95$, $p < .05$, $r = 0.74$). However, these differences were not confirmed at follow-up (number of errors of the change condition, follow-up, $M = 8$, $SD = 12.61$; $Z = 1.10$, $p < .05$, $z = 0.42$; total number of errors, follow-up, $M = 9.33$, $SD = 7.03$; $Z = 1.16$, $p < .05$, $r = 0.47$). In addition, the change was similar in CONTROL and in CEREB for both the

number of errors in the change condition (CEREB, *M* = -2.9, *SD* = 2.92; CONTROL, *M* = -1.5, *SD* = 2.29; 7 = 0.72, *p* > .05, r = 0.14) and the total number of errors (CEREB, *M* = -5.5, *SD* = -5.96; CONTROL, *M* = -2.05, *SD* = 5.45; Z = 1.27, *p* > .05, r = 0.24). No improvements were observed in other neuropsychological tests (*p* > .05).

Questionnaires: no effect of the training was observed on the overall score of the executive function questionnaire, but there was an improvement in the cognitive flexibility sub-score (Cognitive flexibility at home). Scores reported on the social cognition questionnaire were also better after the training. In these two questionnaires, the differences were not maintained at follow-up (Cognitive flexibility at home, follow-up, *M* = 13.6, *SD* = 3.78; Z = 0.27, *p* > .05, r = 0.10; Social cognition questionnaire, follow-up, *M* = 59.33, *SD* = 10.38; Z = 1.26, *p* > .05, r = 0.52). The changes were significantly different in CEREB (Delta pre/post) as compared with CONTROL for both Cognitive flexibility at home and Social cognition. No effects were found at the R-CMAS (*p* > .05).

3.2. Single-case analysis

Motor rhythmic skills: statistics calculated with the DissocsBayes program (Crawford & Garthwaite, 2007) are presented in Table 4. In tapping with a metronome (CV), Patients 2, 3, 4, 6 and 7 were impaired at pre-training, but only patient 3 was still impaired at post training. Only patients 1 and 5 did not have a significant dissociation between pre and post-testing. In tapping with music (CV), patients 2, 3, 5, 6 and 7 were significantly impaired at pre-training. Patients 5 and 6 did not show measurable impairment after the training.

Neuropsychological assessment: single-case statistics are presented in table 5. In Balance (Hopping on mats), all patients displayed an impairment before the training. Patients 1, 4 and 6 had comparable performances after the training. Note that most controls participants obtained the maximum score (5) at this task. In Cognitive flexibility (Speed), Patients 5, 6 and 7 had a performance significantly poorer than controls at pre-training. This impairment was still found at post-training. At the Quality index of Cognitive flexibility, Patients 2, 4, 6 and 7 were impaired before the training, and patient 1 and 5's performances were borderline. Patient 4 and 5 displayed a normal performance at post-training in comparison with the control group. Note that Patient 3 did not pass the Cognitive flexibility task. At the Cognitive flexibility at home index, Patients 1, 2, 3 and 7 were significantly impaired before the training. Among them, Patient 3 and 7 displayed a normal performance after the training. Finally, in Social cognition, Patients 2, 3 and 5 had significantly lower scores than controls at pre-training.

4. Discussion

This interventional study explored the effect of a dance training protocol in children with DCA. Seven patients participated in fourteen dance classes over a period of seven weeks. Before and after the training, we tested children with DCA's movement, sensorimotor and cognitive skills, as well as social and affective functions. The results showed a positive change in rhythmic sensorimotor (Finger tapping with a metronome and music), motor (balance), cognitive (Cognitive flexibility) and social (Social cognition) skills in most patients. We compared these changes with the evolution of these skills in a group of age-matched control participants over the same period of time. This confirmed that the improvement was greater in the CEREB patients than in typically developing children, and not merely due to a test retest effect. Given the rarity of DCA and the complexity of the training protocol, only a small number of participants could be included in this study. We used single case analyses with a multiple baseline experimental design to compare the performance of each patient before and after the training.

Children with DCA's difficulties in SMS were manifest: six patients out of seven displayed a more variable performance than controls when tapping to the beat of a metronome or music. To the best of our knowledge, this is the first time that rhythmic difficulties are reported in developmental anomalies affecting the cerebellum. This result highlights the important role played by the cerebellum in synchronization already suggested by studies showing rhythmic deficits in acquired cerebellar dysfunction (Provasi et al., 2014). Recent neuroimaging evidence also support the idea that the cerebellum is important for rhythm perception and neural tracking of beat and rhythm (Nozaradan, Schwartze, Obermeier, & Kotz, 2017; Paquette et al., 2017; Schwartze et al., 2016). This result contrasts with those obtained in other studies in patients with acquired cerebellar lesions who had difficulties in timing of single intervals (duration-based timing) but not in rhythmic processing (beat-based timing) (Breska & Ivry, 2016, 2018; Grube, Cooper, Chinnery, & Griffiths, 2010). However, the fact that rhythmic processing is preserved in patients with acquired lesions of the cerebellum does not rule out the implication of the cerebellum in the development of rhythmic abilities.

After the dance training protocol, most patients had SMS performances comparable to those of controls. Their motor variability while synchronizing with a metronome or music was reduced (i.e., the performance was improved). This study is the first to show that SMS skills can be improved in children with DCA. This confirms the hypothesis that

dance, which enhances sensorimotor functions in other populations (Jin et al., 2019; Karpati et al., 2016), may boost these functions in cerebellar disorders. This result could indicate that the cerebral network sustaining SMS can be trained to match an average functioning with specific remediation techniques. The cerebellum is belonging to this cerebral network that also involves the basal ganglia, the motor and premotor cortices (Breska & Ivry, 2016; Coull & Nobre, 2008; Coull, Cheng, & Meck, 2011; Grahn, 2009; Grahn & Brett, 2007; Kotz & Schwartze, 2011). Dance is a complex activity that recruits a vast array of brain regions, including the cerebellar-cortical and basal ganglia-cortical networks (i.e., the supplementary motor area, the motor cortex, the basal ganglia and the cerebellum; Bachrach et al., 2016; Brown et al., 2006; Cross et al., 2006, 2009; Giacosa et al., 2016; Karpati et al., 2015). It is reasonable to hypothesize that the recruitment of some of these dance associated regions would underlie the observed positive effects.

One question that remains unanswered is whether the training had a direct effect on the cerebellum. The observed improvement can also be underpinned by compensatory mechanisms in the brain involving the other structures such as the basal ganglia. Compensatory brain mechanisms have been proposed to explain the effect of rhythmic training protocols in Parkinson's Disease (Benoit et al., 2014a; Cochen De Cock et al., 2018; Dalla Bella, 2020; Dalla Bella et al., 2015; Nombela, Hughes, Owen, & Grahn, 2013). Parkinson's Disease serves as a model of basal ganglia dysfunction as the cerebellum function remains relatively unaffected (Breska & Ivry, 2018; Wu & Hallett, 2013). In this disease, hyperactivation of the cerebellar-cortical network is believed to act as a compensatory mechanism for the basal ganglia partial loss of function (Benoit et al., 2014; Cochen De Cock et al., 2018; Dalla Bella, 2020; Dalla Bella et al., 2015; Nombela et al., 2013; Wu & Hallett, 2013). The opposite pattern, namely the recruitment of the basal ganglia-cortical network to compensate for impaired cerebellar-cortical pathways, may be involved in the positive effect of our dance protocol on sensorimotor training. Further studies involving neuroimaging could be used to confirm this hypothesis.

The cerebellar-cortical and basal ganglia-cortical networks that sustain SMS are also involved in various motor (Bengtsson, Ehrsson, Forssberg, & Ullén, 2005; Dalla Bella, Benoit, Farrugia, Schwartze, & Kotz, 2015; Lang et al., 2017), cognitive (Murdoch, 2010; Kotz & Schwartze, 2010; Schwartze & Kotz, 2013) and social (Van Overwalle et al., 2019) skills. Therefore, it is not surprising that a dance protocol would affect other seemingly unrelated skills, confirming the idea that the sensorimotor training may exhibit a broader spectrum of remediation effects. For example,

balance involves some common structures with the SMS brain network such as the cerebellum and basal ganglia (Surgent, Dadalko, Pickett, & Travers, 2019). The effect of dance on balance associated brain network might be directly linked to the improvement of the SMS skills. Nevertheless, even though SMS is involved in dance training, the changes are not necessarily limited to SMS brain networks. Dance involves balance training as well and hence stimulates several other balance associated cerebral structures distinct from the SMS ones, such as the frontal, temporal and parietal cortices and the thalamus (Brown et al., 2006; Karpati et al., 2015; Surgent et al., 2019).

Cognitive skills (Cognitive flexibility) were also improved by the dance training. This might seem surprising at first sight, as dance is a physical activity that mainly involves motor and SMS skills. Cognitive flexibility is sustained by a frontoparietal network (Dajani & Uddin, 2015; Mogadam et al., 2018) of which involvement has not been shown as critical to movement control and SMS. It is important to note, however, that cognitive flexibility is a complex ability that involves different subdomains such as inhibition, working memory and switching (Dajani & Uddin, 2015). Recent studies showed that these capacities correlate with SMS skills in children (Puyjarinet et al., 2017; Tierney & Kraus, 2013; Bégel et al, under review). Therefore, we cannot rule out the possibility that the gain in cognitive flexibility is linked to the SMS training, even though some other factors such as the exercise of creativity through body improvisation might have acted in synergy. Indeed, cognitive flexibility is linked to creativity (De Dreu et al., 2011). Moreover, in a review on interventions to aid executive functions in children, Diamond & Lee (2011) concluded that addressing emotional, social and physical development was more efficient than focusing narrowly on executive functions only (Diamond & Lee, 2011). Therefore, with its social, physical, and creative components, dance appears as an ideal candidate to boost executive functions, and more specifically cognitive flexibility. Note that a previous study reported the effect of dance on cognitive flexibility in aging people (Coubard et al., 2011). This suggests that the effect of dance on cognitive flexibility is not limited to the current study's protocol and patients.

The improvement produced by the training extended to social skills. More specifically, parents reported enhancement of their children's social cognition and theory of mind, such as the capacities to express their own emotions, to adapt their behavior to others in a social context, and to infer on other's mental states. The collective training protocol proposed in this study may have contributed to develop children social skills by stimulating their capacity to anticipate others' movement. Motor synchronization between individuals leads to increased pro-social behaviour (Hove & Risen, 2009; Cirelli, Einarson & Trainor, 2014; Wiltermuth & Heath, 2009), In addition, interpersonal synchronization

involved in dance increases the sense of connectedness with partners (Himberg, Laroche, Bigé, Buchkowski, & Bachrach, 2018). Dance also fosters the allocation of attention to others' cognitive and mental states (Joufflineau et al., 2018), and dance training is linked to enhanced empathic abilities (Gujing et al., 2019). Notably, cognitive flexibility is important for social behavior, especially in the ability to shape behavior to the social context (Turner, Oakes, Haslam, & McGarty, 1994). Cognitive flexibility is also correlated with measures of empathy (Grattan, & Eslinger, 1989; Milders, Ietswaart, Crawford, & Currie, 2008). In contrast, other cognitive skills, such as memory, processing speed, or attention, play a limited role in social behaviour (Milders, Ietswaart, Crawford, & Currie, 2008). Therefore, the concurrent improvement in synchronization, social skills and cognitive flexibility is not surprising. However, the causal relations between these competences is still to be explored.

The functions that were enhanced after the dance training are crucial for everyday life activities. This transfer effect may in turn significantly improve patients' quality of life. Notably, the effect found on a behavioral test of cognitive flexibility is corroborated by the parents' evaluation, meaning that the effect in everyday life was noticed by the parents and reflects a functional improvement. Cognitive flexibility is of great importance for the executive adaptation of cognitive and behavioral responses to the environment. It is of particular importance for different skills such as mathematics (Verschaffel, Torbeyns, De Smedt, Luwel, & Van Dooren, 2007), language (Deák, 2004) and planification (Snow, 1992). Enhancing cognitive flexibility abilities would improve patients' quality of life and boost their academic performance. In general, executive functions are a good predictor of school achievement (Becker, Miao, Duncan, & McClelland, 2014). In addition, parents' responses at the social skills questionnaire point towards a positive effect of the dance training protocol on children's social skills. Social skills are also a good predictor of school achievement (Agostin & Bain, 1997; Bramlett, Scott, & Rowell, 2000). The fact that movement and balance skills were also improved further indicates that patients' everyday life can be improved by the dance training protocol on several dimensions.

Because of their difficulties, children with DCA cannot take part in most extracurricular physical activities with children of their age. This in turn reduces their chances to progress. Thus, in addition to the lack of specific treatment or training available for DCA (Brenna, 2014), the range of activities for these children to participate in is limited. Some types of dance can be too challenging for them, but the results of this study confirm that dance can be proposed to DCA patients. The dance training protocol we proposed is promising because it is adapted to each child's skills.

Children with various disorders and symptoms can participate in the same dance class. Besides, there is no competition and no judgment on the aesthetic production of the children, which helps to set trustful environment. Dance is also an activity that can be tailored to participants' special needs. Training protocols can therefore be adapted to different populations, such as adolescents or adults with cerebellar anomalies, using various types of dance.

In summary, very encouraging sensorimotor, motor, cognitive and social improvements were exhibited by children with DCA as a consequence of a dance training protocol. The cerebellum is a key structure of the central nervous system and its role in various motor (Bastian, 2006; Manto et al., 2012; Therrien & Bastian, 2019), cognitive (Murdoch, 2010; Schmahmann, 2004; Schmahmann, Guell, Stoodley, & Halko, 2019) as well as social (Van Overwalle et al., 2019) and affective functions is well known. Consequently, some or all of these domains are affected in patients with DCA. Finding ways to enhance patients with DCA's motor, cognitive and social skills is a challenge that we addressed using dance in this study. Dance is an activity that stimulates different aspects of behavior, especially sensorimotor functions. The beneficial effect of dance is likely to be partly due to increased activity in the sensorimotor cortico-cerebello-striatal pathways. This research paves the way for larger scale studies on dance as a tool for reeducation in cerebellar dysfunctions.

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Conflict of interest

The authors have no relevant financial or non-financial interests to disclose

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Tables

Table 1. Demographics and Cerebellar signs at the latest neurological evaluation

Patient	Age	Genre	A	HT	CD	DM	DA	T	S	F	N	OA	OM	OBF
1	10.3	M	+	-	+	-	-	-	+	+	-	-	+	-
2	7.8	F	+	-	+	-	+	-	-	-	-	-	+	+
3	7.5	F	+	+	+	-	+	-	-	-	-	-	-	+
4	8.2	M	+	+	+	-	-	-	-	+	-	-	-	-
5	9.0	M	+	+	+	+	+	+	+	+	-	-	-	+
6	7.11	M	+	+	+	-	+	-	+	-	-	-	+	-
7	8.3	M	+	+	+	+	+	-	+	+	+	+	+	+

A = Ataxia, HT = Hypotonia, CD = Coordination disorder/Dysdiadochokinesis, Dysmetria, Dysarthria/Speech disorder (articulation), T = Intentional tremors, S = Slowness, F = Fatigability, N = Nystagmus, OA = Oculomotor apraxia, OM = Other Oculomotor disorders, OBF = Oro-bucco-Facial apraxia

Table 2. IQ scores of children with DCA. IQ indexes were calculated with the WISC-IV for patients 1, 4,5 and 6, and with the WPPSI-IV for patients 2, 3 and 7 in accordance with the age of the patient at last intellectual evaluation.

Patient	VCI	PRI	VSI	FRI	WMI	PSI
1	98	94	NT	NT	97	112
2	84	NT	106	118	106	74
3	95	NT	77	95	97	77
4	138	114	NT	NT	100	96
5	82	79	NT	NT	NT	76
6	89	NT	102	91	88	77
7	100	NT	85	95	NT	71

VCI = Verbal Comprehension Index, **PRI** = Perceptual Reasoning Index, **VSI** = Visual Spatial Index, **FRI =** Fluid Reasoning Index, **WMI** = Working Memory Index, **PSI** = Processing Speed Index; note that in the WISC IV, the main indices are VCI, PRI, WMI and PSI. In the WPPSI IV, the main indices are VCI, VSI, FRI, WMI and PSI. NT = not tested (the index does not exist in the battery used).

Table 3. **Differences between the pre and the post training and between CEREB and CONTROLS group. Tasks for which a significant difference were observed are presented**

	CEREB Pre-Post Comparison					Comparison CEREB and CONTROLS				
Variable	Mean Pre *(SD)*	Mean Post *(SD)*	Z	*p*	Effect Size (r)	Delta Patients *(SD)*	Delta Controls *(SD)*	Z	*p*	Effect Size (r)
Tapping with a metronome (CV)	0.35 *(0.05)*	0.19 *(0.11)*	2.37	< 0.01**	0.89	-0.16 (0.08)	0.03 (0.14)	3.37	< 0.001***	0.65
Tapping with music (CV)	0.41 *(0.12)*	0.22 *(0.09)*	2.37	< 0.01**	0.89	-0.19 *(0.11)*	-0.01 *(0.14)*	2.77	< 0.005***	0.53
Hopping on mats	1.86 *(1.38)*	3.57 *(2.09)*	2.30	< 0.05*	0.87	1.71 *(1.19)*	-0.05 *(0.28)*	3.96	< 0.005***	0.76
Cognitive flexibility Speed	7.24 *(2.87)*	12.8 *(10.3)*	2.20	< 0.05*	0.90	-5.19 *(8.47)*	-1.04 *(1.19)*	1.95	< 0.05*	0.37
Cognitive flexibility Quality	4.67 *(1.86)*	3 *(1.91)*	2.27	< 0.05*	0.93	1.33 *(0.52)*	0.3 *(0.73)*	2.78	< 0.01**	0.53
Cognitive flexibility at home	16.57 *(3.6)*	13.86 *(3.39)*	1.87	< 0.05*	0.71	2.71 *(3.30)*	0.18 (*1.13)*	2.11	< 0.05*	0.41
Social cognition	59.43 *(8.28)*	55.14 *(7.78)*	1.86	< 0.05*	0.70	4.29 *(4.89)*	-0.44 *(3.03)*	2.76	< 0.01**	0.53

Table 4. Individual scores in Perceptual and motor rhythmic skills for each DCA patient in comparison with an age-matched control group. Statistics are calculated with Bayesian Single-Case Methods (Crawford & Garthwaite, 2007). Columns Pre show the comparison between the control group and patients' performance before the testing and columns Post show the comparison between the control group and patients' performance after the testing.

Patient 1			
	Pre		Post

Variable	Patient's score	Control Group Mean (*SD*)	*p*	Patient's score	Control Group Mean (*SD*)	*p*
Tapping with a metronome (CV)	0.29	0.12 (*0.14*)	= 0.15	0.10	0.12 (*0.13*)	= 0.38
Tapping with music (CV)	0.42	0.15 (*0.15*)	= 0.07	0.13	0.14 (*0.12*)	= 0.47
Patient 2						
	Pre			Post		
Variable	Patient's score	Control Group Mean (*SD*)	*p*	Patient's score	Control Group Mean (*SD*)	*p*
Tapping with a metronome (CV)	0.33	0.01 (*0.01*)	< 0.005***	0.25	0.11 (*0.07*)	= 0.06
Tapping with music (CV)	0.43	0.12 (*0.05*)	< 0.005***	0.26	0.12 (*0.04*)	< 0.05*
Patient 3						
	Pre			Post		
Variable	Patient's score	Control Group Mean (*SD*)	*p*	Patient's score	Control Group Mean (*SD*)	*p*
Tapping with a metronome (CV)	0.42	0.07 (*0.01*)	< 0.005***	0.39	0.11 (*0.07*)	< 0.01**
Tapping with music (CV)	0.55	0.12 (*0.05*)	< 0.005***	0.34	0.12 (*0.04*)	< 0.005***
Patient 4						
	Pre			Post		
Variable	Patient's score	Control Group Mean (*SD*)	*p*	Patient's score	Control Group Mean (*SD*)	*p*
Tapping with a metronome (CV)	0.31	0.07 (*0.2*)	< 0.001***	0.09	0.11 (*0.14*)	= 0.42
Tapping with music (CV)	0.15	0.14 (*0.07*)	< 0.001***	0.13	0.13 (*0.07*)	0.500
Patient 5						
	Pre			Post		

Variable	Patient's score	Control Group Mean (*SD*)	*p*	Patient's score	Control Group Mean (*SD*)	*p*
Tapping with a metronome (CV)	0.31	0.07 (*0.03*)	< 0.001***	0.11	0.06 (*0.01*)	< 0.005***
Tapping with music (CV)	0.46	0.14 (*0.09*)	< 0.005***	0.13	0.09 (*0.04*)	0.19
Patient 6						
	Pre			Post		
Variable	Patient's score	Control Group Mean (*SD*)	*p*	Patient's score	Control Group Mean (*SD*)	*p*
Tapping with a metronome (CV)	0.39	0.07 (*0.02*)	< 0.001***	0.26	0.11 (*0.14*)	= 0.19
Tapping with music (CV)	0.39	0.14 (*0.08*)	< 0.05*	0.25	0.13 (*0.07*)	= 0.1
Patient 7						
	Pre			Post		
Variable	Patient's score	Control Group Mean (*SD*)	*p*	Patient's score	Control Group Mean (*SD*)	*p*
Tapping with a metronome (CV)	0.39	0.07 (*0.02*)	< 0.001***	0.13	0.12 (*0.14*)	0.47
Tapping with music (CV)	0.42	0.14 (*0.08*)	< 0.01**	0.31	0.13 (*0.07*)	0.026*

Table 5. Individual scores in Motor and cognitive skills for each DCA patient in comparison with an age-matched control group. Statistics are calculated with Bayesian Single-Case Methods with the DissocsBayes program (Crawford & Garthwaite, 2007). Columns Pre show the comparison between the control group and patients' performance before the testing and columns Post show the comparison between the control group and patients' performance after the testing.

Patient 1

Variable	Pre			Post		
	Patient's score	Control Group	*p*	Patient's score	Control Group	*p*

		Mean (*SD*)			Mean (*SD*)	
Hopping on mats	4.5	5.00 (*0.00*)	< 0.001***	5	5.00 (*0.00*)	0.5
Cognitive flexibility Speed	5.19	5.31 (*0.76*)	= 0.44	4.5	3.96 (*0.46*)	0.15
Cognitive flexibility Quality	4	6.00 (*1.00*)	= 0.06	4	6.43 (*0.79*)	0.07
Cognitive flexibility at home	14	19.33 (*2.25*)	< 0.05*	13	20 (*2.83*)	< 0.05*
Social cognition	61	68.14 (*4.77*)	= 0.11	69	65.67 (*3.27*)	= 0.19

Patient 2

Variable	Pre			Post		
	Patient's score	Control Group Mean (*SD*)	*p*	Patient's score	Control Group Mean (*SD*)	*p*
Hopping on mats	2	4.92 (*0.20*)	< 0.000***	3.5	4.67 (*0.52*)	< 0.05*
Cognitive flexibility Speed	9.43	6.24 (*1.91*)	= 0.09	6.78	5.31 (*1.24*)	= 0.16
Cognitive flexibility Quality	4	6.33 (*0.42*)	< 0.005***	5	6.83 (*0.41*)	< 0.005***
Cognitive flexibility at home	10	20.83 (*2.4*)	< 0.005***	13	20.67 (*2.73*)	< 0.05*
Social cognition	39	67.17 (*8.93*)	< 0.05*	48	67.50 (*5.86*)	< 0.05*

Patient 3

Variable	Pre			Post		
	Patient's score	Control Group Mean (*SD*)	*p*	Patient's score	Control Group Mean (*SD*)	*p*
Hopping on mats	0	4.92 (*0.20*)	< 0.005***	0.5	4.67 (*0.62*)	< 0.005***
Cognitive flexibility Speed	NA	NA	NA	NA		
Cognitive flexibility Quality	NA	NA	NA	NA		
Cognitive flexibility at home	12	20.83 (*2.40*)	< 0.005***	21	20.67 (*2.73*)	= 0.46

Social cognition	61	67.17 *(8.93)*	< 0.05*	62	67.50 *(5.86)*	= 0.212

Patient 4

Variable	Pre			Post		
	Patient's score	Control Group Mean *(SD)*	*p*	Patient's score	Control Group Mean *(SD)*	*p*
Hopping on mats	2	4.86 *(0.25)*	< 0.001***	5	4.71 *(0.48)*	= 0.30
Cognitive flexibility Speed	6.05	5.92 *(1.97)*	= 0.48	5.35	4.38 *(1.02)*	= 0.20
Cognitive flexibility Quality	5	6.29 *(0.49)*	< 0.05*	7	6.71 *(0.49)*	= 0.30
Cognitive flexibility at home	14	19.29 *(3.55)*	= 0.11	15	19.6 *(4.16)*	= 0.18
Social cognition	58	65.29 *(7.63)*	= 0.20	68	64.67 *(4.18)*	= 0.24

Patient 5

Variable	Pre			Post		
	Patient's score	Control Group Mean *(SD)*	*p*	Patient's score	Control Group Mean *(SD)*	*p*
Hopping on mats	2	4.94 *(0.18)*	< 0.001***	4	5 *(0.00)*	< 0.001***
Cognitive flexibility Speed	8.60	4.76 *(0.80)*	< 0.001***	6.43	4.02 *(0.69)*	< 0.01**
Cognitive flexibility Quality	5	6.37 *(0.74)*	= 0.06	6	6.37 *(0.52)*	= 0.26
Cognitive flexibility at home	20	18.87 *(3.52)*	= 0.38	20	19.28 *(0.59)*	= 0.43
Social cognition	59	66.37 *(2.67)*	< 0.05*	60	66.12 *(2.70)*	< 0.05*

Patient 6

Variable	Pre			Post		
	Patient's score	Control Group Mean *(SD)*	*p*	Patient's score	Control Group Mean *(SD)*	*p*

Hopping on mats	2	4.86 *(0.25)*	< 0.001***	5	4.71 *(0.49)*	= 0.30
Cognitive flexibility Speed	10.33	5.93 *(1.97)*	< 0.05*	7.77	4.38 *(1.02)*	< 0.05*
Cognitive flexibility Quality	1	6.28 *(1.07)*	< 0.001***	3	6.71 *(0.49)*	< 0.001***
Cognitive flexibility at home	16	19.29 *(3.55)*	= 0.22	20	19.6 *(4.16)*	= 0.45
Social cognition	52	65.29 *(7.63)*	= 0.08	49	64.67 *(4.18)*	< 0.01**

Patient 7

Variable	Pre			Post		
	Patient's score	Control Group Mean *(SD)*	*p*	Patient's score	Control Group Mean *(SD)*	*p*
Hopping on mats	1	4.86 *(0.24)*	< 0.001***	1	4.71 *(0.49)*	< 0.001***
Cognitive flexibility Speed	35	5.93 *(1.97)*	< 0.001***	12.62	4.38 *(1.02)*	< 0.001***
Cognitive flexibility Quality	1	6.29 *(0.)*	0.000***	2	6.71 *(0.49)*	0.000***
Cognitive flexibility at home	11	19.29 *(3.55)*	< 0.05*	14	19.6 *(4.16)*	= 0.15
Social cognition	56	65.29 *(7.63)*	= 0.16	60	64.67 *(4.18)*	= 0.17

Figures

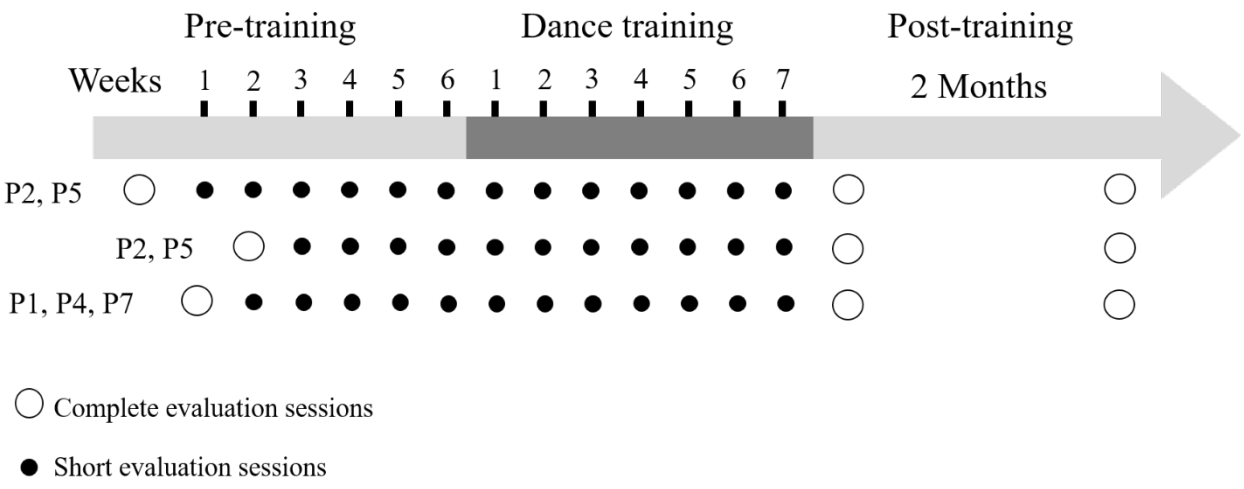

Figure 1. Experimental design for the CEREB children.