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Functional reaching discloses perceptive impairment in diplegic children with cerebral palsy

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ABSTRACT

The currently accepted definition classifies Cerebral Palsy (CP) as a mere posture and movement disorder. Conversely, some authors have recently associated the presence of several motor dysfunctions exhibited by diplegic children with CP to an impairment in the perceptive system. The aim of the present study was to investigate the influence of the Perceptive Impairment (PI) on motor control and to appraise if the PI can be revealed by a reaching task.

A functional reach and touch experiment was accomplished from sitting posture considering different directions and distances. Typically developing and diplegic children with CP were enrolled and, the latter, a priori divided in two subgroups considering a positive or negative diagnosis of PI. The reaching trials were quantified by means of centre of pressure analysis in terms of the overall quality of the task, and accuracy and effectiveness of postural adjustments and Anticipatory Postural Adjustments (APAs).

The three groups showed statistically significant differences in terms of percentage of touched target, and of time spent and maximum distance covered to reach the target. In particular, PI caused a major difficulty in accomplishing the reaching tasks, thus a lower autonomy level in action. Overall, the PI strongly affected the anticipatory control system. Children with PI, rarely recruited APAs, each of which was characterized by small amplitude and inaccuracy in direction. The lack of effective APAs indicated how PI strongly influenced the motor control strategy.

The present study demonstrates that the PI is a primary syndrome responsible for the long-term prognosis beside the motor and the postural disorders in CP.

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1. Introduction

The currently accepted definition classifies Cerebral Palsy (CP) as a mere posture and movement disorder [1], disregarding the influence of perceptive as well as cognitive, communicative and emotional problems [2,3]. Conversely, some authors stress the role played by the perception disorders while assessing the quality (nature) and the quantity of the functional impairment exhibited by CP children [2,3]. As asserted by some authors [2,4–7], Perceptive Impairment (PI) in CP is the failure of a complex neurological process that enables the individual to collect, integrate, interpret and use the spatial-temporal aspects of sensory information from one's body and environment, in order to plan, control, guide and produce organized motor behavior. In other words, PI is a cognitive–perceptual–motor dysfunction that entails with the inability to adapt and to integrate environmental experiences [7]. Some studies highlighted the presence of perceptive dysfunctions in CP children [7,8], however in literature the following issues remain unclear: (i) which disabilities arise from disorders in the locomotor apparatus, and which ones from disorders in the perceptive system, (ii) if PI affects uniformly the hemiplegic, diplegic and tetraplegic cluster or whether pathological differences are present among and within each cluster, and (iii) which is the mainly responsible deficit for the long-term prognosis between the motor impairment and the PI. Only recently some authors have begun to face these issues. Ferrari and Cioni [2] assessed that the PI is very frequent in tetraplegia, somewhat

Abbreviations: AE, angle error; APA, anticipatory postural adjustment; M, APA Magnitude; *dysp*, dyplegic CP children affected by PI; non*dysp*, dyplegic CP children unaffected by PI; MD, maximum distance; %APA, percentage of APA onset; %Touched, percentage of touched target; PI, perceptive impairment; RD, reaching direction; RT, reaching time; TD, typically developing.

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present in diplegia [9], and almost absent in hemiplegia. In particular, these authors proposed a categorization of the different forms of diplegia based on kinematic criteria [9], in which children were classified as "*dysperceptive*" and "*nondysperceptive*" based on a positive or negative diagnosis of PI [2]. The PI diagnosis is carried out through the detection of seven clinical signs each assessed on the basis of an operative definition [2] in the course of specific environmental stimuli. The signs are: (i) exaggerated and low threshold startle reaction, (ii) upper limb in startle position, (iii) averted eye gaze, (iv) frequent eye blinking and closing, (v) facial grimaces, (vi) posture freezing, and (vii) verbal expressions.

There is a strong evidence from the literature that movement and perception are like two sides of the same coin [5]. The theory of sensory integration [6] states that perception is not merely the acquisition of sensory information, but an active process aimed at guiding the execution of a correct movement, i.e. coherent with the motor programme planned before the action. This process is carried out through the integration of sensory information properly representing the state of one's body and of the environment. Visual, somatosensory and vestibular systems provide the primary redundant sources of sensory information, of which integration within the Central Nervous System (CNS) must result consistent. In cases of disagreement, the CNS may lead to execute incorrect movements with abrupt adaptations [4] thus also preventing the construction of *perceptive representations* of the actions [5], consequently, a "secondary" incapacity to effectively guide the subsequent movement on the basis of accurate perceptive representations. On a prognostic level, therefore, PI may significantly affect the patient's development of independent life skills [4,10]. At a kinematic level, furthermore, the PI may cause an overall fear of moving and the onset of strongly unnatural walking schemes, with the upper limbs raised in a lateral position swinging in the frontal plane [9].

Besides walking, the reaching task is one of the most studied tests to assess the motor and perceptive disorders in CP children [11–13]. In particular, reaching from a sitting position, is used to investigate the postural control ability and it strongly deals with the development of independent life skills [14]. Postural control during reaching has been often assessed by looking at both (i) overall movements by means of the analysis of the Centre Of Pressure (COP) [8,15], and (ii) specialized control strategy by means of electromyography (EMG) [15–17].

Motor control during voluntary movements, such as reaching, is carried out through the performance of Postural Adjustments (PAs). Complex PAs are exhibited by Typically Developing (TD) children from the age of 4 months [11]. For TD, experience and self-learning activities play a key role in fine-tuning modulation of muscle contraction during postural adjustments [11] resulting in a variable recruitment of the direction-specific muscles [13]. Conversely to TD, diplegic CP children (i) use proximal to distal muscle recruitment strategy [13,16,18], (ii) show a reduced capacity in fine-tuning their PAs to task specific conditions [19,20], (iii) usually perform jerky movements [21], and (iv) present stereotyped and direction-inappropriate muscular activities [19,22] more pronounced in diplegic rather than hemiplegic children [14].

The trajectory of the end-effector during reaching exhibited by TD children is straight and characterized by smooth, bell-shaped velocity profiles [23], suggesting that reaching is mostly feedforward controlled [24]. Anticipatory PA (APA), that is the feedforward parallel command aimed at minimizing the equilibrium disturbance occurring in correspondence of a voluntary movement, is one of the main clues of the adoption of this feedforward control strategy [25]. It appears as a movement opposite to the target direction, and it is mainly tuned by the perceptive representations of the PA that is about to happen [4,5]. Riach and Hayes [26] and Johnston et al. [27] detected APAs in TD children during arm movement in standing. Conversely, other authors found that the development of APAs during a reaching task is absent throughout pre-adolescent age [28]. Overall, the postural control of CP children seems less guided by feedforward processes based on prior experience and more by feedback mechanisms [13].

To the authors' knowledge, no study has ever investigated the topic of motor control in CP children during reaching *a priori* considering the presence of perceptive as well as postural-movement disorders, thus interpreting the performances considering independently these two complementary disability sources. Furthermore, no relation between the presence/absence of APAs in CP children and the diagnosis of a PI has been investigated so far.

Two groups of diplegic children, one with and one without a diagnosis of PI as assessed through the criteria provided by Ferrari and Cioni [2], and a control group of TD children were enrolled for a functional reaching task experiment from sitting position. The aim of the study was to assess if the *dysperceptive* children present poorer perceptive organization abilities and lower autonomy in actions than aged-matched TD and *nondysperceptive* children. In particular, by means of COP analysis we studied if the PI causes differences in the motor control strategies and in the quality of the motor performance. The reaching trials were quantified in terms of the overall quality of the task and accuracy and effectiveness of PAs and APAs.

2. Materials and methods

2.1. Participants

Twenty-four diplegic children with purely spastic forms of CP and nine asymptomatic typically developing (TD) children (four males, age range 7–10 years) participated in the study. Eligibility criteria included: a documented CT or MRI diagnosis of CP, ability to sit independently on a stool, acquired walking with or without devices, no major sensory, cognitive or visual field deficits, ability to sustain visual fixation for at least 3 s and no functional surgery or botulinum toxin injection during the previous year.

During routine clinical evaluation sessions, the diagnosis of PI was assessed by the same operator according to the standardized criteria described in [2]. Hence, out of the 24 diplegic CP children, 12 were positive to PI (*dysperceptive* group—*dysp*; seven males, age range 6–14 years), and 12 were negative (*nondysperceptive* group—non*dysp*; 10 males, age range 5–11 years). Each *dysp* child exhibited all the seven clinical signs associated to the diagnosis of the PI (see Section 1), conversely non*dysp* children never showed more than three signs. TD, non*dysp* and *dysp* groups resulted both age matched, as tested by one-way ANOVA ($p > 0.05$), and anthropometrically matched, as tested by one-way ANOVA ($p > 0.05$) on the dominant arm length (distances from the acromion up to the tip of the middle finger).

None of the participants had prior knowledge of the purpose of the study. An informed consent was obtained from parents before participation.

2.2. Experimental protocol

The experimental setting (Fig. 1) consisted of a functional reach and touch task from a sitting position. The participants were seated on a height adjustable stool without any back, foot, and arm support, while a force platform (Bertec 4060A, sampling rate 100 Hz), placed under the stool, registered the COP trajectories during the trials. The reaching target was an appealing small ball (8 cm diameter) containing an accelerometer synchronized with the force platform. The ball was hung from an adjustable arm (Fig. 1), the fulcrum of which was positioned above the greater trochanter of the subject's dominant side (spatial correspondence set with a laser-pointer). A climbing-rope was tied to a non-hampering body-harness to protect the subjects from falling.

The subjects were asked to sit still, gazing at an eye-level fixed point, and then reinstate the same initial position of head, trunk and hands during the trials. A whistle blew 3 s after the beginning of the recording, triggering the subject to reach and touch the ball with the dominant hand at a natural speed. The acquisition ended when the subject reinstated the starting position once reached the object (detected with the accelerometer) or once performed the maximum reachable displacement. The reaching trials were obtained from the combination of two directions (*dir*) and two distances (*dist*). Specifically, *dir* was set to *frontal* or *lateral* with respect to the dominant hand (movements occurring in the sagittal and frontal plane, respectively); *lateral* being more difficult because of the reduced support base. The target *dist* was set at 120% or 140% of the arm length. The shorter distance was chosen as an easy reaching, the farther to evaluate the perceived functional limit of

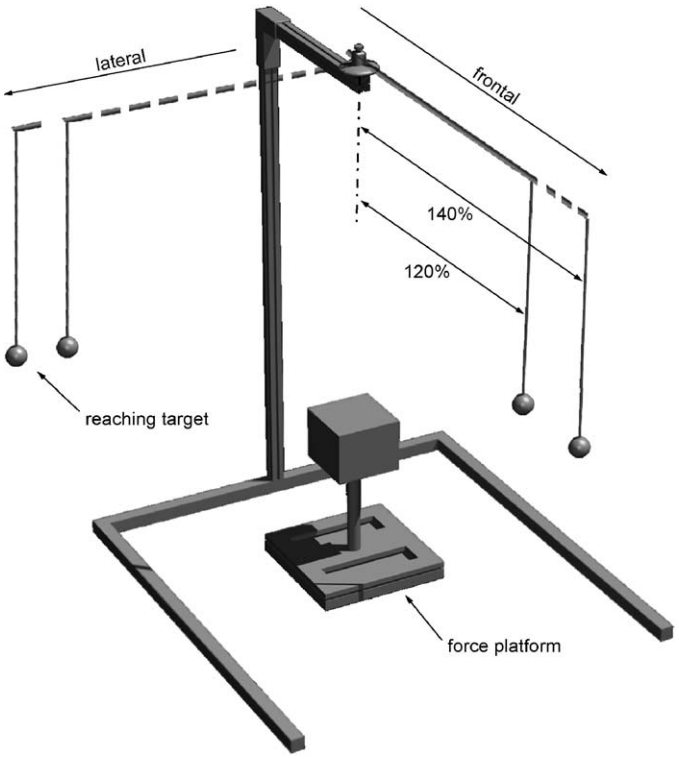

Fig. 1. Set-up of the functional reaching experiment. The reaching target (ball) and the arm adjustable in direction and length are shown in the four configurations resulting from the combination of the two *dir* (frontal and lateral) and the two *dist* (120% and 140% the arm length).

stability. Six repetitions were executed for each trial condition. The stool height *h* was set at 100 cm for each child.

2.3. Data reduction

The raw COP trajectory was low-pass filtered at 25 Hz with a zero-phase, second-order Butterworth digital filter. Each trial, and its relevant COP trajectory, was divided (offline) in three different intervals: (i) the *waiting interval* during the first 3 s of still sitting (W_{int}), (ii) the *APA interval* (APA_{int}) if present, and (iii) the *reaching interval* (R_{int}), from the end of the APA to the end of the task in case of APA detection, or from the whistle to the end of the task in case of no APA detection. The COP trajectory was then re-centred to its midpoint during W_{int}, while its direction of maximum variance during R_{int} was interpreted as the Reaching Direction (RD) performed by the subject.

The reaching performances were quantified by means of six different parameters. The overall functional abilities were assessed through (1) the percentage of times the target was touched (%Touched), (2) the Reaching Time (RT), (3) the Maximum Displacement (MD) of the COP during R_{int} (calculated only for the trials with *dist* = 140% being the trials with *dist* = 120% not representative of the functional limits of stability), and (4) the percentage of APA onsets (%APA). This latter was automatically identified as the COP shift opposite to the RD during the initial phase of the gesture (2 standard deviations higher than the baseline value during W_{int}). In case APAs were detected, the following parameters were calculated to quantitatively describe their features: (5) the Angle Error (AE), being the angle between the RD and the APA direction (computed as the maximum variance direction during APA_{int}), and 6) the APA Magnitude (M), being the maximum COP displacement reached during APA_{int} from the midpoint (see Fig. 2).

2.4. Statistical analyses

In order to filter out the effects of lack of attention, habituation and fatigue, only the most representative trial among the six repetitions was considered for the statistical analysis. This was detected in a space of four dimensions created from the parameters RT, MD, M and AE each normalized with respect its own standard deviation (calculated considering the total number of trials). In this space, the closest point of the six repetitions to the midpoint was considered as the most representative trial [29]. If RT, M and AE were "not-a-number" due to the failure of the task or to the absence of APA, the trial corresponding to the median value of MD among the six was considered as the most representative.

A logarithmic transformation of RT, MD, M and AE was performed in order to normally distribute these parameters. The normality and the homoschedasticity were then ascertained respectively with Kolmogorov–Smirnov and Levene tests. The effects of direction, distance and diagnosis were tested with 3-way ANOVAs on the logarithmically transformed parameters. The post hoc Bonferroni correction was then used for multiple comparison. Non-parametric Kruskal–Wallis tests were carried out to detect differences among the TD, *dysp* and non*dysp* groups of the non-normally distributed parameters (%Touched, %APA), and the Mann–Whitney *U* test was performed to ascertain the between-groups significant differences. All statistical analyses were performed with NCSS (NCSS, Kaysville, USA).

3. Results

Fig. 2a reports a typical pattern of the COP trajectory in the platform reference frame. Fig. 2b–d reports three typical COP trajectories along the RD relative to the performances with *dir* = frontal, *dist* = 140% as collected from a TD, a non*dysp* and a *dysp* child, respectively.

Fig. 3 reports the %Touched resulted among the three groups considering all the trials performed. *Dysp* children resulted functionally more compromised: the Kruskal–Wallis test highlighted that %Touched was lower in all the trials with respect to both non*dysp* and TD. Moreover, the trial factors (*dir, dist*) influenced %Touched: if no statistically significant difference among the groups was found in the easiest trial (*dist* = 120%, *dir* = frontal), the difference became significant by increasing task difficulty. In the worst case (*dist* = 140%, *dir* = lateral) the %Touched revealed significant differences between non*dysp* and TD and

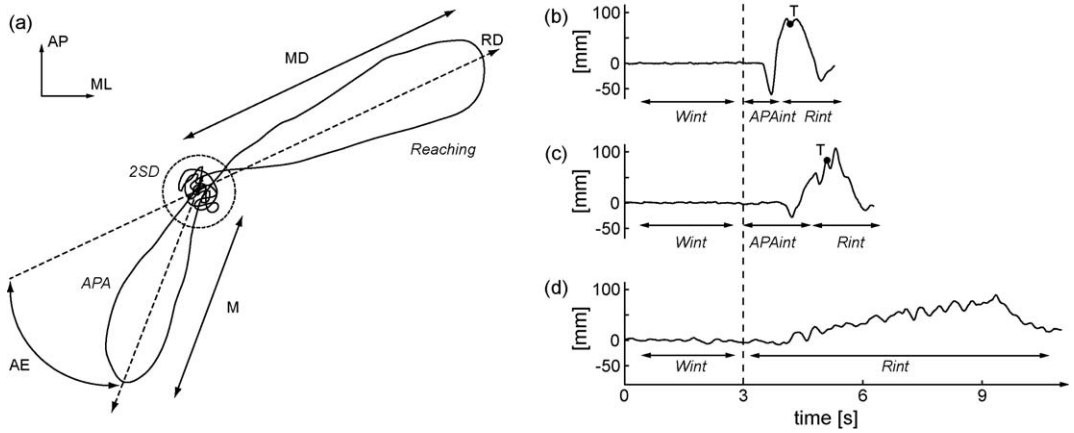

Fig. 2. (a) A typical pattern of the COP trajectory with respect to the medio-lateral and antero-posterior directions in the force platform reference frame. Three typical COP trajectories after a projection along the RD relative to the performances with *dir* = 90, *dist* = 140%, as collected from a TD (b), a non*dysp* (c) and a *dysp* (d) child respectively, are also reported. The trials are divided in W_{int}, APA_{int} when present, and R_{int}. 'T' indicates the instant of touching of the target.

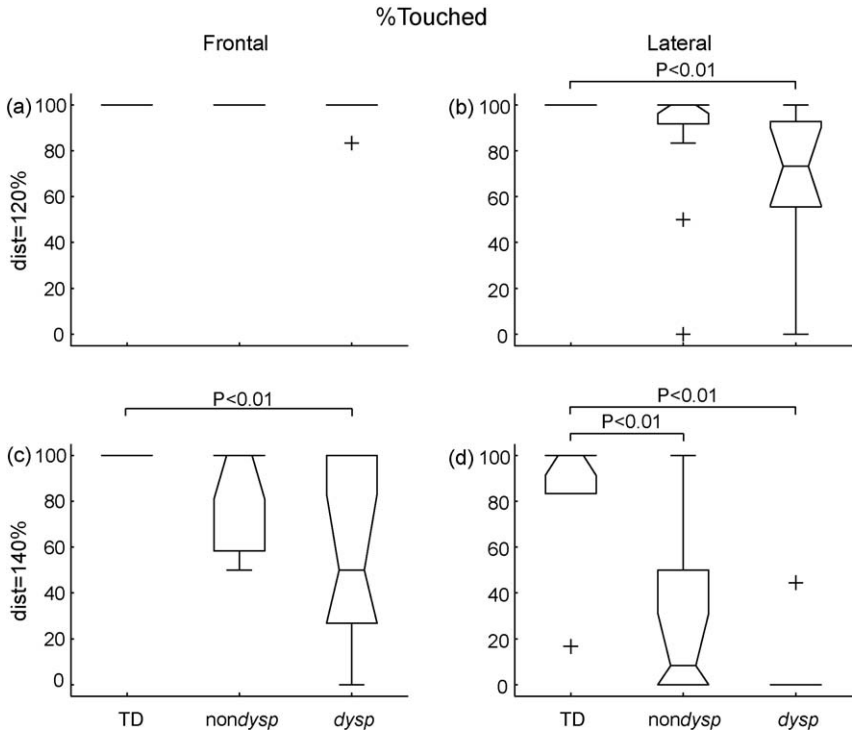

Fig. 3. Distributions of the %Touched of the most representative trials for each of the TD, non*dysp* and *dysp* groups. All the four configurations resulting from the combination of the two *dir* and *dist* are shown.

between *dysp* and TD. Moreover, when *dysp* reached the target, they were slower than the other groups: no differences were revealed for RT for the cases with *dist* = 120% (mean RT = 2.39 s), while significant differences were obtained with *dist* = 140% ($p < 0.01$, mean RT equal to 2.53 s, 3.02 s, 4.68 s for TD, non*dysp*, *dysp*, respectively).

Functional limits of stability quantified by MD during the tasks at *dist* = 140% showed an increasing discrepancy between the groups shifting from the frontal to the lateral direction (see Fig. 4). In this last case, significant differences were obtained between *dysp* and TD and also between non*dysp* and *dysp*.

Fig. 5 reports %APA, AE and M distributions. Considering both *dir*, for *dysp* the APA was detected in fewer cases (41%) as compared to non*dysp* (81%) and TD (87%). These differences were statistically significant as assessed through the Kruskal–Wallis test (Fig. 5a). Statistically significant differences were observed for the parameters quantified on the detected APA. Fig. 5b and c reports the results for AE and M with *dist* = 120%, since *dist* had no significant effect. With respect to AE and M, the diagnosis had an effect statistically significant ($p < 0.05$ and $p < 0.01$ respectively). With respect to AE, a statistically significant effect of *dir* was found just for *dysp* ($p < 0.01$, Fig. 5b).

Considering both *dir*, a statistically significant difference was obtained between TD and *dysp* on AE ($p < 0.01$), and between *dysp* and non*dysp* on M ($p < 0.01$). Considering *dir* = lateral, a statistically significant difference between *dysp* and TD was observed on AE (Fig. 5b). Finally, with *dir* = frontal, a statistically significant difference was obtained between *dysp* and non*dysp* on M (Fig. 5c).

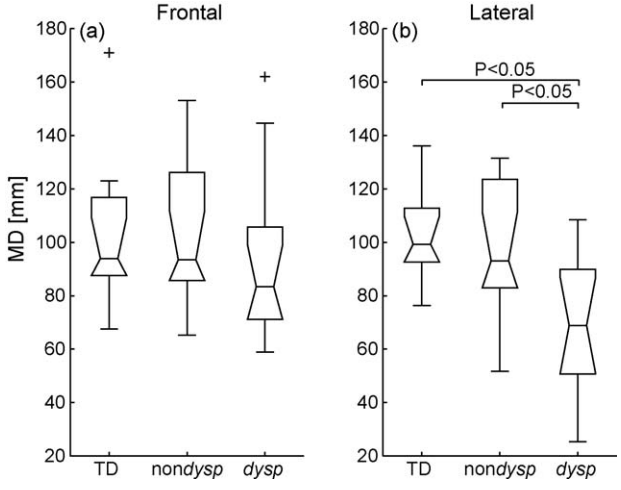

Fig. 4. Distributions of the MD of the most representative trials for each of the TD, non*dysp* and *dysp* groups when *dist* = 140% and divided with respect to *dir*.

4. Discussion

Functional disabilities exhibited by some forms of CP children cannot be thoroughly explained on the basis of a pure motor dysfunction but should be ascribed to an impaired perceptive system [7,12,13]. The PI is considered as a syndrome that entails the failure of a complex multisensory process involving the integration and use of the spatial-temporal aspects of the sensory information to plan and guide organized motor behavior [7].

A functional reaching experiment aimed at appraising the influence of PI on motor control was accomplished. TD and diplegic CP children were enrolled and, the latter, a priori divided into the subgroups *dysperceptive* and *nondysperceptive* on the basis of the presence of the PI as clinically diagnosable through the operative definition provided by Ferrari and Cioni [2]. To investigate global postural control strategy, COP parameters were preferred to EMG allowing better test–retest reliability [15].

By looking at the functional abilities, the three groups showed significant differences in terms of %Touch, RT and MD, with a trend that increased with task difficulty. In particular, during the increase of the difficulty, the *dysp* revealed a significantly slower

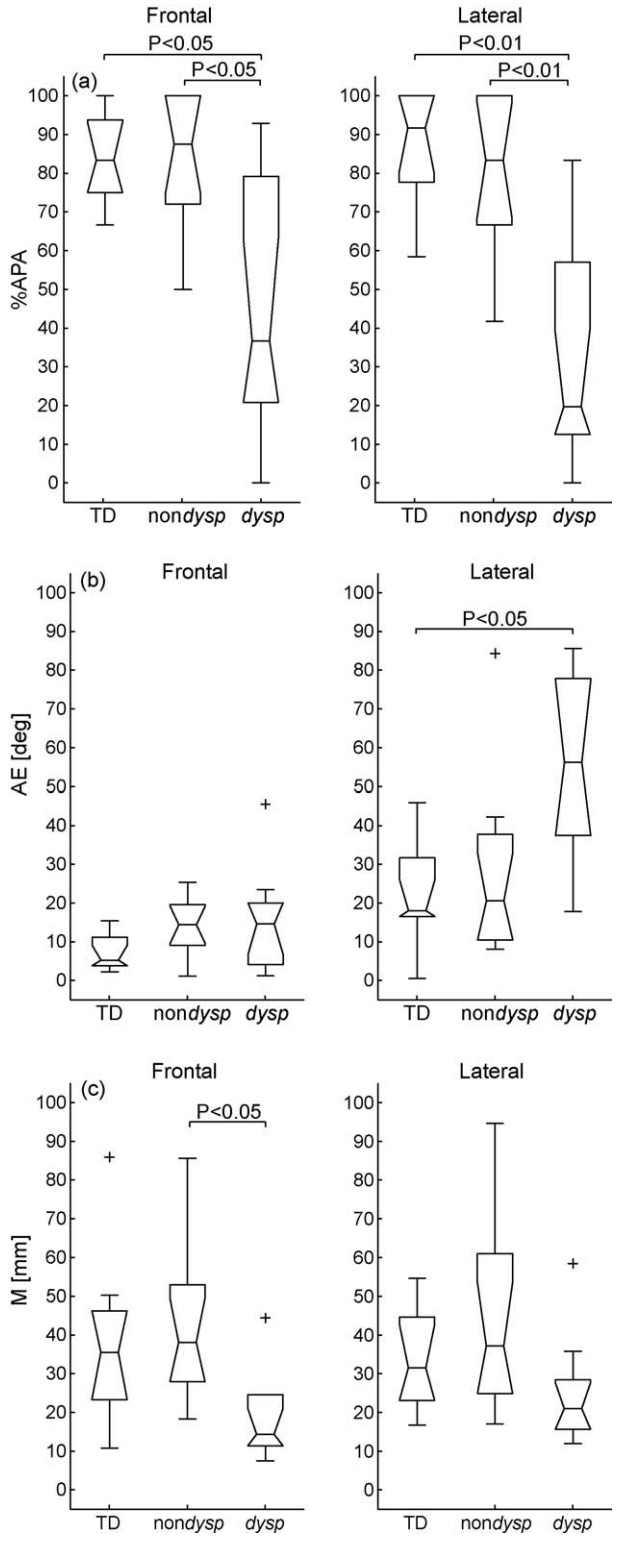

Fig. 5. Distributions of the %APA divided with respect to *dir* of the most representative trials for each of the TD, non*dysp* and *dysp* groups (a). On the detected APA, the distributions of the AE (b) and M (c) with *dist* = 120% are also reported.

and less effective reaching performance also with respect to the non*dysp* group, demonstrating a poorer functional autonomy. With lateral *dir*, they also showed a significant MD drop exhibiting a freezing posture in correspondence of a perception of depth.

The TD children showed APAs basically in every trial (Fig. 5). These results sustain the findings of [26,27] but are in contrast with [28]. The PI negatively influenced the anticipatory control system: the anticipatory strategies of the *dysp* resulted frequently ineffective. As compared to non*dysp* and TD, they rarely recruited APAs, each of which was characterized by low magnitude and direction inaccuracy. The lack of effective APAs testifies that *dysp* less often rely on feedforward programming of reaching than non*dysp* and TD, thus indicating how PI strongly influenced the motor control strategy. Hence, PI caused a major difficulty in accomplishing reaching tasks, that is related to a lower action autonomy level.

In the authors' opinion these findings sustain the hypothesis that PI might have played a "confounding effect" role in previous studies that explored the reaching performances in diplegic children without assuming perceptive skill differences within the cluster.

In conclusion, the results of the present study demonstrate that the PI might be considered as a primary factor responsible for the long-term prognosis and that its diagnosis is relevant both in clinic and in research.

PI in diplegic CP children seems related to a lack of 'perceptual approval' to motion directly related to a major difficulty in solving the sensory-motor integration problem. Further research aimed at quantitatively characterizing the influence of the PI on the routine functions should be addressed.

Conflict of interest

None.

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