RESEARCH PAPER

THE MOTOR MELODY IN ACTION PLANNING: THE CASE OF AUTISTIC CHILDREN AND THEIR NON-AUTISTIC SIBLINGS

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Abstract

Objective: it is well known that during an intentional behavior, the final goal of the action shapes the entire sequence of motor acts. This chained organization has been previously demonstrated to be altered in school-age autistic children, who modulate only the final motor act according to the action goal. Here, we investigate the temporal modulation during the intentional action in three groups of preschoolers: neurotypical, autistic, and non-autistic siblings of autistic children.

Method: the participants engaged in a simple task of reaching and grasping an object and placing it into two containers of different sizes.

Results: neurotypical children adjusted both reaching and placing times according to the width of the containers, indicating an action-chained organization. In contrast, both autistic children and non-autistic siblings adapted only the placing - but not the reaching- time according to the container size, exhibiting an unchained organization of intentional actions.

Conclusions: despite not being included among the diagnostic criteria, motor alterations are present in a large number of autistic individuals, detectable from an early age. Being motor signs also expressed by non-autistic siblings, our findings suggest a potential link between motor abnormalities and the pathogenesis of autism. Thereby, tasks similar to the one employed here could be valuable for screening children with an increased likelihood of atypical neurodevelopment.

Key words: action planning, motor abnormalities in autism, endophenotype, atypical motor developmental trajectories, preschoolers

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Introduction

The existing body of literature increasingly indicates that autistic individuals experience early and persistent motor atypicality (80% of the cases; see Bhat, 2020; Lai et al., 2014), spanning from basic altered motor development to higher order alterations (Fabbri-Destro et al., 2013; Lloyd et al., 2013; Mosconi & Sweeney, 2015). In this vein, Leary and Hill (1996), in exploring the relationship between movement disturbances and autism symptoms, emphasized how motor abnormalities can impact life experiences and potentially precede the emergence of social-communication deficits in autism.

While it is conceivable that social/communicative and motor deficits are independent of each other, there is substantial evidence that - through the mirror mechanism - the motor system plays a pivotal role in cognitive and social functions like action and intention understanding (see for a review Rizzolatti et al., 2014). This evidence suggests that dysfunction in the organization of the motor system may impact both domains. Contrarily, a well-developed motor system brings cascading benefits on multiple domains, encompassing the capacity to interact with other individuals in different environments, interpret their emotions and actions, and engage in joint attention (Leonard & Hill, 2014; Libertus & Needham, 2010;

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Michel et al., 2016). Accordingly, early examination of children's motor development offers a unique opportunity to identify potentially atypical developmental trajectories by detecting early motor signs, the appearance of which may precede the markers typically used as clinical indicators of neurodevelopmental disorders.

This scenario appears to be particularly fitting in the case of autism. However, even if early motor signs are successfully identified, it remains uncertain whether these signs are intrinsic to autism spectrum conditions or if they represent adaptive behaviors in response to the primary symptoms. An attempt to answer this question is to evaluate children with an increased likelihood of developing autism, e.g., siblings of autistic children (Lauritsen et al., 2005; Ozonoff et al., 2014; Persico & Sacco, 2014) who are known to exhibit certain subtle traits or characteristics related to autism (Georgiades et al., 2013; Gizzonio et al., 2014; Pisula & Ziegart-Sadowska, 2015) that are more stable and closer to the underlying genetic influences.

Given these premises, we sought atypical motor signs during action sequence implementation in autistic preschoolers and tested whether this altered motor pattern is shared with non-autistic siblings of autistic children. This investigation evaluated behavioral modulations in reaching and grasping an object and placing it into containers of different sizes. The capacity to adjust movement performance according to the action goal has been previously demonstrated to be a function inherent to the motor system organization in neurotypical individuals (De Marco et al., 2020), partially disrupted in school-aged autistic children (Fabbri-Destro et al., 2009).

Methods

Participants

Eighy-eight Italian children (53f and 35m, age 2-5, M = 3.48 years) were enrolled in the study. The sample was composed of autistic children (AU), non-autistic siblings of autistic children (SIB), and typically developing children (TD). Twenty-three participants were subsequently excluded from the original cohort due to technical issues encountered during the video recording of the task (9AU, 3 SIB, and 11 TD). Thus, the final sample included 65 children: 18 AU (M = 3.84 years; SD = 0.64; 1f and 17m), 21 SIB (M = 3.98 years; SD = 0.86; 12f and 9m), and 26 TD (M = 3.38 years; SD = 0.83; 13f and 13m). The inclusion criteria and the neuropsychological evaluation were described in the Supplementary materials.

The local ethical committee approved the study (prot. Number 1069/2018/SPER/AUSLMO), which was conducted according to the Declaration of Helsinki principles. The parents of all participants provided written informed consent.

Procedure

Participants were seated in front of a table with their right hand facing downwards on the starting position. They were asked to reach, grasp, and place a small cylinder into a big or small box (Size: Big Box and Small Box, **figure 1A**), positioned within the arm's length of the participant to minimize trunk adjustments during movement execution, thus making the execution time comparable. The experiment was administered in a block design (15 repetitions *per* Box), whose order was randomized across participants, with a brief training session performed initially to ensure task comprehension.

The subjects' performance was recorded with a digital camera (25 frames per second 'fps'), and two variables - Reaching and Placing Time - were extracted through video processing software (VirtualDub, http://www.virtualdub.org). Reaching Time was calculated as the duration between the hand's first movement from the starting position and the contact with the object; Placing Time was intended as the duration between lifting and dropping the object into one of the two boxes (**figure 1A**). The number of frames was then converted into seconds according to the fps. Trials whose grasping or placing failed were excluded from the analysis (rate of discarded trials: 16% TD, 10% SIB, 33% AU).

Statistical analysis

A one-way ANOVA and Chi-squared test were conducted to assess the balance of age and gender distribution among groups. The distribution of discarded trials was tested with repeated measures ANOVA with Group (TD, SIB, AU) as a between-subjects factor and Size (Big, Small) as a within-subjects factor.

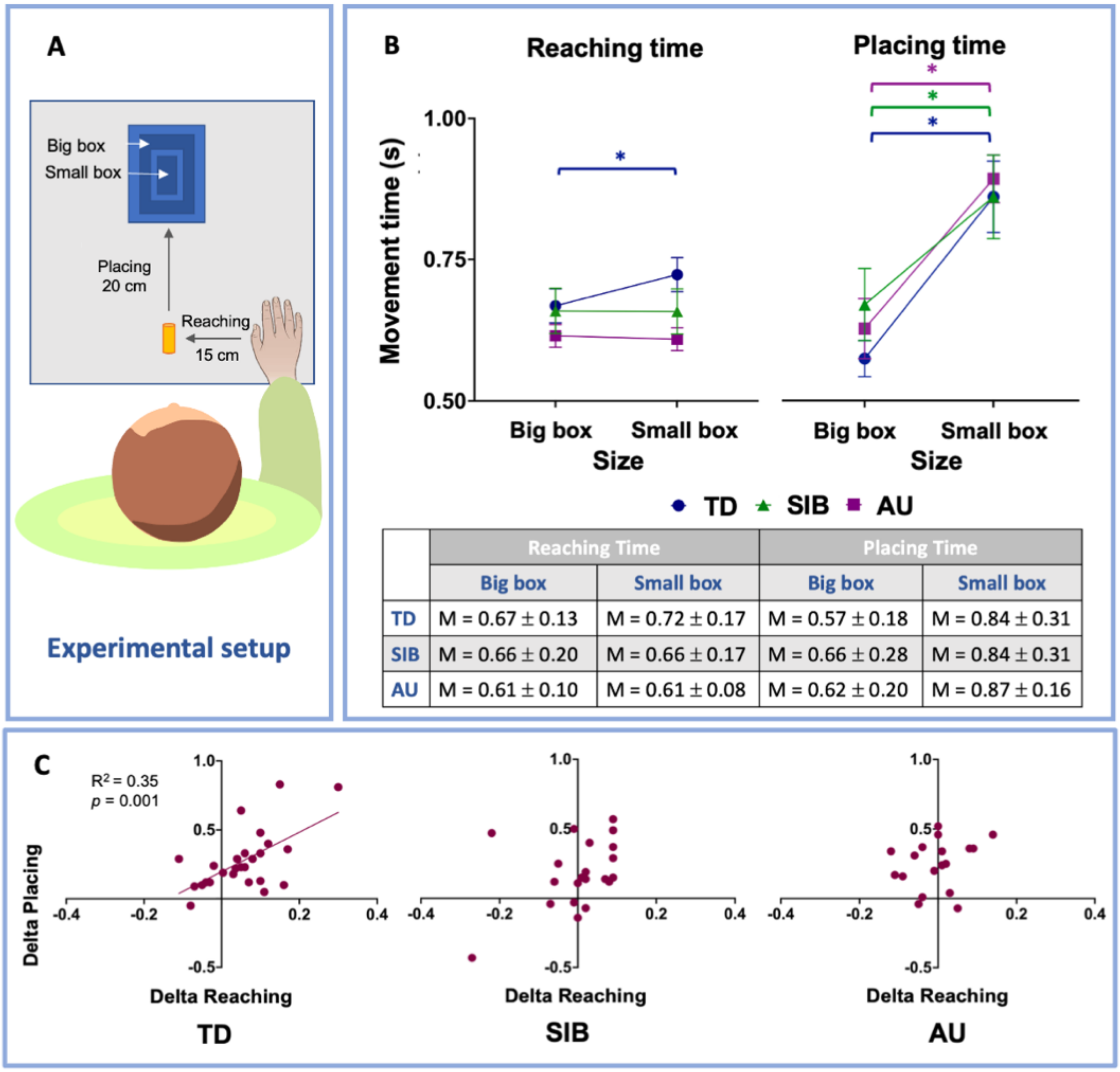
The difference in movement duration induced by the box size was investigated by two repeated measures ANOVAs on Placing Time and Reaching Time, with Size as a within-subjects factor and Group as a between-subjects factor. The effect size was measured using partial η^2, and post-hoc analysis was performed with Bonferroni correction. In case of significant interactions, we applied a General Linear Model with age as a covariate to verify that the effect was not driven by age imbalance. Finally, a linear regression analysis was conducted for each group to evaluate the dependency between Reaching and Placing time.

Results

The three groups differed in terms of age (Group: F(2,62 = 3.77; p = 0.03), with TD (M = 3.38 ± 0.83) younger than SIB (M = 3.98 ± 0.86; p = 0.03) and AU (M = 3.84 ± 0.64; p = 0.18) and prevalence of male, significantly higher in AU compared to TD and SIB (all $p < 0.01$). The performance accuracy differed across groups [$F(2,62) = 6.72$; $p < 0.002$], with AU being systematically more inaccurate than others (all $p < 0.02$). However, no significant main effects of the Size or Size*Group interactions were found, indicating an even distribution of discarded trials between the two sizes.

The results of the ANOVA indicated that the three groups equally modulated the duration of placing according to the box size [$F(2,62) = 74.66$; $p < 0.001$; *Partial* $\eta^2 = 0.55$] (**figure 1B**). Differently, considering the Reaching time, a significant Size*Group interaction [$F(2,62) = 3.38$; $p = 0.04$; *Partial* $\eta^2 = 0.1$] was found, in which only TD exhibiting a timing modulation between the two conditions (p = 0.03, **figure 1B**). This finding was confirmed even when adding age as a covariate ($F(2,61) = 4.82$; $p = 0.01$; *Partial* $\eta^2 = 0.14$]). The linear regression analyses revealed that the modulations driven by the box size on Reaching and Placing Time were interrelated only for the TD group ($R^2 = 0.35$; p = 0.001) but not for AU ($R^2 = 0.06$, $p = 0.33$) and SIB (R^2 = 0.17; p = 0.06) (**figure 1C**).

Figure 1. *Experimental setting and results about the temporal organization of the action. Panel A: Schematic representation of the experimental setting; each child was instructed to place the palm of the right hand facing downwards on the table surface at the starting position and to reach, grasp, and place a cylinder into a big (Big box, sized 16 × 11.2 × 4 cm) or small (Small box, sized 7.3 × 6.5 × 4 cm) box (left inset). Panel B: The graphs report the movement duration of the three groups' reaching and placing phases; error bars indicate the standard error. Significant modulations between the Big and Small boxes ($p < 0.05$) are indicated on top in the corresponding color code. The table below reports for each group (TD, SIB, and AU) the means and standard deviation of movement duration (Reaching and Placing) for the Big and Small boxes. Panel C shows the scatterplots of the temporal difference (Delta) between Big and Small box durations in the reaching (x-axis) and placing (y-axis) phases for the TD, SIB, and AU groups separately. The red line indicates the significant linear trend followed by data distribution*



	Reaching Time		Placing Time	
	Big box	Small box	Big box	Small box
TD	M = 0.67 ± 0.13	M = 0.72 ± 0.17	M = 0.57 ± 0.18	M = 0.84 ± 0.31
SIB	M = 0.66 ± 0.20	M = 0.66 ± 0.17	M = 0.66 ± 0.28	M = 0.84 ± 0.31
AU	M = 0.61 ± 0.10	M = 0.61 ± 0.08	M = 0.62 ± 0.20	M = 0.87 ± 0.16

Discussion

Our study demonstrated that at preschool age, typically developing children exhibit a chained organization during the execution of intentional actions, adjusting both the reaching and placing time according to the action goal. On the contrary, autistic children and their non-autistic siblings modulated only the placing phase, exhibiting an unchained organization of intentional actions.

A vast literature has tackled the neurotypical motor development of intentional actions. The first movement adjustments due to intentional drives become noticeable during late infancy and toddlerhood across various tasks (Örnkloo & Von Hofsten, 2007; Rosenbaum et al., 2012). As children progress through preschool years, they tend to deal with higher task complexity by increasing the time to plan, formulate, and select a correct motor program (Krajenbrink et al., 2020). Finally, the late preschool/early school years witness a further motor progression in the performance of goal-directed sequential tasks, with actions temporally and kinematically organized according to the overarching goal (Domellöf et al., 2013; Jongbloed-Pereboom et al., 2013; Jung et al., 2018; Krajenbrink et al., 2020; Wilmut et al., 2013). Our findings position the TD sample within this framework, as they exhibited the capacity to modulate not only the placing time according to the box size but also the reaching time.

Concerning the implementation of intentional actions in autistic children, different studies have reported atypical features like excessive adjustments of prehension when grasping objects of different sizes, poor interdependency across the different phases composing sequential actions (Cattaneo et al., 2007), and less adjustment to end-state-comfort (ESC) than controls (Scharoun & Bryden, 2016). Our autistic sample confirmed these observations, resulting in a reaching phase implemented regardless of the action goal, contrary to the placing phase, whose duration was modulated according to the specific box size. These results confirm that autistic children can adjust their motor intentional behavior, but only for isolated motor acts, thus lacking an overarching modulation impacting the whole chain of motor acts forming the action. One might argue that our findings reflect a delayed motor maturation for autistic children. Nevertheless, this conclusion seems unlikely, given that previous studies using a similar task showed that such motor modulations are also absent in older autistic children [age 7-12 (Fabbri-Destro et al., 2009)] and that deficits in motor planning become more pronounced with age (Rodgers et al., 2019).

The most striking result of our study was that siblings showed a motor pattern overlapping with the autistic population. Siblings of autistic children display, on average, 25-fold higher autism risk rates than the general population (Persico & Sacco, 2014). Although siblings do not have a diagnosis of autism and do not exhibit any behavioral or social impairments, their motor organization resembles that proper of their neurodiverse siblings, thus indicating that the lack of chained motor organization could be regarded as a motor endophenotype associated with autism. In other words, some genetic predisposition shared between autistic children and their siblings might underlie the expression of this altered motor organization, positioning this motor abnormality nearer to the primary features of autism.

Along this line of research, exploring similarities and differences in motor abnormalities and social cognition deficits across psychiatric (see, for example, Romeo et al., 2014 in schizophrenia) and neurological disorders could help uncover the mediating mechanisms relevant to autism. This cross-disorder approach is particularly valuable in identifying both shared and unique pathways that contribute to the complex symptomatology seen in autism.

Identifying motor tasks that are extremely easy to administrate (see also Brisson et al., 2012) and adaptable to different ages and contexts could be a way to anticipate screening of motor organization in infants and toddlers in the widest population possible. From our findings, we can conclude that the impairment of intentional motor organization is early detectable and seems proper for children with an increased likelihood of developing autism. To date, autistic children are typically diagnosed at 4-5 years (Ozonoff et al., 2018), but modern trends point to autism diagnosis already in toddlers because a timely identification would enable early interventions, significantly impacting the later developmental trajectories (McPheeters et al., 2016; Rojas-Torres et al., 2021; Siu et al., 2016), alleviating parental distress, and mitigating the secondary consequences of autism spectrum conditions (Anderson et al., 2014; Bradshaw & Enticott, 2014; Brasher & Stapel-Wax, 2020; Schreibman et al., 2015). Although the task implemented in our study is not intended as a diagnostic tool, it could serve as an early "magnifying lens" for raising warnings about motor development in children. These observations, possibly complemented by behavioral measures (stopwatches, wearable sensors, or video-tracking), could aid the identification of children diverging from the neurotypical development of motor organization. Such a warning would not imply a neurodiversity diagnosis but could let them enter deeper, more focused screening programs.

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