LOCOMOTION AND GRASPING IMPAIRMENT IN PRESCHOOLERS WITH AUTISM SPECTRUM DISORDER

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Abstract

Objective: To investigate expressiveness of motor impairment in autism spectrum disorder (ASD) and its correlation with developmental and clinical features of ASD.

Method: Thirty-five male preschoolers with ASD completed the Peabody Developmental Motor Scales-2 (PDMS-2; Folio and Fewell, 2000) and underwent a multidisciplinary assessment including medical examination, standardized assessment of cognitive abilities, administration of Autism_Diagnostic_Observation_Schedule (ADOS) and a parent interview about adaptive skills.

Results: Results revealed a substantial impairment in locomotion and grasping skills. Both fine and gross motor skills were significantly correlated with non verbal IQ and adaptive behaviours ($p<0.01$) but not with chronological age or ADOS scores. Children with weaker motor skills have greater cognitive and adaptive behaviours deficits.

Conclusions: Motor development in ASD can be detected at preschool age and locomotion and grasping skills are substantially the most impaired area. These findings support the need to assess motor skills in preschoolers with ASD in addition to other developmental skill areas. Along with the increasingly acknowledged importance of motor skills for subsequent social, cognitive, and communicative development our findings support the need to consider motor intervention as a key area in therapeutic program to improve outcome in preschoolers with ASD.

Key words: motor impairment, autism spectrum disorder, Peabody Developmental Motor Scales-2, Autism Diagnostic Observation Schedule, preschoolers**.**

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Introduction

Autism Spectrum Disorders (ASD) comprise a complex group of behaviourally defined conditions characterized by social and communication impairment along with presence of repetitive and restrictive behaviours (DSM Fifth Edition, 2013). Although motor impairment is not considered a primary diagnostic criteria for ASD, motor delays or disorders have been observed 1) in infants who later developed ASD (Adrien et al. 1993, Teitelbaum et al. 1998; Baranek 1999, Landa and Garret 2006, Esposito and Venuti 2008, Ozonoff et al. 2008, Esposito et al. 2009), 2) in young children with ASD (Landa and Garret 2006, Esposito and Venuti 2008), 3) in school aged children and adolescents (Green et al. 2002, Green et al. 2009, Staples and Reid 2010).

The greater attention to the characteristics of the motor functioning is based on its practical importance as well as its theoretical significance. Indeed, understanding motor impairment in ASD may improve early diagnosis, the planning of specific treatments and the comprehension of important aspects of social cognition. In particular, very early motor development has emerged as an area of interest due to the increasing evidence of atypical motor features as a sign for early diagnosis (Trevarthen et al. 2013).

Studies focusing on motor impairment in ASD approached the analysis of motor skills through different techniques including cinematic approaches, detailed video-tape analysis and standardized motor test assessments (Fournier et al. 2010). Findings from these studies reported both fine and gross motor disturbances in all age ranges but a single motor symptom as a universal sign or prodrome for ASD was not identified.

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Although different studies suggested that the motor impairment in ASD could vary in type and intensity with age (Minshew et al. 2004, Lloyd et al. 2013), most are focused on preschool age. Generally at this age motor impairment is characterized by an atypical development of both gross and fine motor abilities (Provost et al. 2007b, Vanvuchelen et al. 2007, Jasmin et al. 2009, Zachor et al. 2010). Using a clinical rating scale as the Peabody Developmental Motor Scales (PDMS-2; Folio and Fewell 2000), Provost et al. (2007b) reported some degree of motor delay in at least one area (gross motor skills or fine motor skills) of motor development for each subject stressing a substantial defect in Locomotion, in Visual-Motor Integration and in Manipulation of objects abilities. Subsequently, Jasmin et al. (2009) have also used the PDMS-2 and demonstrated that sixty-three percent of their sample showed a significant gross motor delay, fifty-three percent showed a fine motor delay and fifty-seven percent presented an overall motor delay. This Author pointed out a major impairment in Locomotion, Manipulation of the object and Grasping whereas the Stationary and the Visual-Motor Integration abilities appeared more preserved. Locomotion impairment has been reported as recurring deficit in ASD in pre-school age (Vernazza et al. 2005, Rinehart et al. 2006, Ming et al. 2007, Vilensky et al. 1981) and different Authors reported atypical features from the early stages of acquisition (Esposito et al. 2008, Esposito et al. 2011). Regarding fine motor skills, different studies reported a substantial deficit in grasping skills in pre-schoolers with ASD (Jasmin et al. 2009, David et al. 2012) and difficulties with movements involving simple grasp and reach-to-grasp sequences in children (Mari et al. 2003). Ming et al. (2007) suggested a higher prevalence of fine motor impairment in pre-schoolers than in older children. All this literature focusing on motor impairment in pre-schoolers with ASD suggested a substantial delay in both gross and fine motor skills with particular involvement in locomotion and grasping abilities; nevertheless the hypothesis concerning an age-related pattern of motor disability in ASD has still to be confirmed.

Some researchers have reported that the motor deficits exhibited by children with ASD could be related to the severity of autism (Jasmin et al. 2009, Hilton et al. 2012, MacDonal et al. 2014) whereas others failed to find this relationship (Provost et al. 2007b, Zachor et al. 2010). Furthermore, it has been reported that intellectual disability may contribute to a greater degree of motor impairment in ASD (Green et al. 2009) whereas other Authors suggested that motor impairment may be ASD specific and not related to intellectual disabilities (Staples and Reid 2010). Others suggested that intellectual disabilities may be responsible for defined fine motor patterns within ASD (Mari et al. 2003, David et al. 2012). Unsubstantial and conflicting data could at least partly be related to the use of different methods of investigation, small samples size, lack of homogeneity of samples and diagnosis not confirmed by objective diagnostic measures.

The first purpose of this study is to determine the degree and the type of motor disorder in pre-schoolers with ASD using the standardized motor testing PDMS-2 that has been found useful in quantifying motor development in preschoolers with ASD. A second issue addresses the correlation between motor development and clinical features as chronological age, intellectual abilities, autism severity and adaptive behaviours.

Method

Participants

The sample consisted of 35 male preschoolers with ASD (mean age = 48,5 months, SD =8,8, range 30-60 months), consecutively recruited at ASD unit at the IRCCS Stella Maris Foundation between October 2010 and January 2013. The study was carried out according to the standards for good ethical practice of the IRCCS Stella Maris Foundation and in accordance with the guidelines of the Declaration of Helsinki. Children were selected among patients who received a clinical diagnosis of autistic disorder (AD) and pervasive developmental disorder not otherwise specified (PDD-NOS), based on DSM-IV-TR criteria (APA 2000). Exclusion criteria were: (a) neurological syndromes or focal neurological signs, (b) significant sensory impairment (e.g., blindness, deafness), (c) anamnesis of severe birth asphyxia, head injury or epilepsy, (d) potential secondary causes of ASD revealed by high-resolution karyotyping, DNA analysis of Fragile-X, or screening tests for inborn errors of metabolism.

A multidisciplinary team (a senior child psychiatrist, an experienced clinically trained research child psychologist and a speech-language pathologist) performed the ASD diagnosis during 5-7 days of extensive evaluation. Each participant underwent a multidisciplinary assessment including medical examination, standardized assessment of cognitive abilities, administration of Autism_Diagnostic_Observation_Schedule (ADOS-G, Lord et al. 2000) and a parent interview about adaptive skills via Vineland Adaptive Behaviours Scale (VABS, Sparrow et al. 1984).

Complete cognitive data were available for 34 subjects (mean non-verbal I.Q. = 98,2, SD =22,1, range = 42-144). ADOS-G data were available for 34 subjects (19 subjects were classified as having "Autism disorder" and 14 subjects were classified as having "Autism spectrum disorder", 1 subjects showed ADOS-G scores below of the cut off for "Autism spectrum disorder" on ADOS-G classification but data analyses anyway include this latter subject as the clinical judgment is the gold standard for ASD diagnosis in young preschool children (Klin et al. 2000, Charman and Baird 2000).

VABS data of daily living, socialization and motor skills were available in 69% of the sample.

One physical therapist administered the PDMS-2 to each participating as part of the interdisciplinary developmental evaluation. All enrolled children completed PDMS-2.

Instruments

Motor assessment: The PDMS-2 (Folio and Fewell 2000) consists of six subscales: Reflexes (for children birth through 11 months), Stationary (ability to sustain control of body within its center of gravity), Locomotion (ability to move from one place to another), Object Manipulation (ability to manipulate balls, for children 12 months and older), Grasping (ability to use hands), and Visual-Motor Integration (ability to use visual perceptual skills to perform complex eye–hand coordination tasks). The Reflexes or the Object Manipulation, the Stationary and the Locomotion subscales contribute to the Gross Motor Quotient (QGM) whereas the Grasping and the Visual-Motor Integration subscales contribute to the Fine Motor

Quotient (QFM). The Total Motor Quotient (QTM) is formed by a combination of the results of the gross and fine motor subscales.

For each subscale, the examiner administrated the first PDMS-2 items according to the chronological age of the child (entry point), following the instructions in the manual. Some items required a verbal request whereas others provided for a verbal request associated to the demonstration of the action. Is not allowed to physically help the child to perform the required test, but it allowed to reformulate the verbal instructions or demonstrate the required action making it more understandable (for example through the use of objects). The PDMS-2 final score classifies performance into 7 categories: very superior, superior, above average, average, below average, poor and very poor.

The PDMS -2 has been recognized as a valid measure to identify delays and defects in motor development at preschool age (Tieman et al. 2005) and Wuang et al. (2011) considered the PDMS -2 a valuable investigative tool for the study of motor skills in children with intellectual disabilities. The Italian version of PDMS-2 has been edited by the member of research group. The member of research group translated individually the source text of the English version of the PDMS-2 and sought agreement between individual translations. *Autism diagnostic measures:* The ADOS-G (Lord et al. 2000) was administered to directly assess ASD symptoms. The ADOS-G is a semi structured instrument wherein the examiner attempts to engage the child with a series of social presses and rates the child's social-communicative impairment and restricted and repetitive behaviors. According to published algorithms (Gotham et al. 2009) calibrated severity scores were also calculated. The data of the ADOS-G were available for 34 patients.

Nonverbal cognitive/developmental abilities: A number of standardized tests were used to assess intellectual abilities due to differences in the age, verbal skills and functioning level of children. These included: the Leiter International Performance Scale-Revised (Roid and Miller 1997), the Griffiths Mental Development Scale-ER (GMDS-ER, Luitz et al. 2006), the Italian version of Wechsler Preschool and Primary Scale of Intelligence (WPPSI, Wechsler 1974). When the tool provides a mental age (MA), IQ was estimated dividing MA by the child's chronological age (CA). For this study we consider the non verbal IQ scores.

Daily living skill: The Vineland Adaptive Behaviours Scale (VABS, Sparrow et al. 1984) is a parent interview that assesses the adaptive behaviour of individuals from birth to adulthood. It covers communication (Receptive, Expressive, Written), daily living skills (Personal, Domestic, Community), socialization (Interpersonal Relationships, Play and Leisure Time, Coping Skills), and motor skills (Fine, Gross), the higher the score, the better the skill. Ratio scores for each available subdomain (Personal, Domestic, Community, Interpersonal Relationships, Play and Leisure Time, Coping Skills, Fine motor, Gross motor) were calculated by dividing age equivalent scores by CA.

Data analysis

Correlation analysis was performed between PDMS-2 measures and IQ. Correlation analysis, performed controlling IQ effect were conducted between PDMS-2 measures and 1) chronological age (months) 2) ADOS-G scores 3) VABS scores. A *p value* <.01 was considered. Statistical analysis was performed with IBM-SPSS (version 16).

Results

Results on PDMS-2 confirm motor impairment in ASD

All Quotients mean scores were into Poor category. Locomotion and Grasping mean scores were into Poor category whereas Stationary, Object manipulation and Visual-motor integration mean scores were into below average category .

The Total Motor, the Gross Motor and the Fine Motor quotients scores were into Poor or Very Poor category for 83%, 80% and 80% of the sample respectively. The Locomotion and the Grasping scores were into Poor or Very Poor category for 69% and 77% of the sample respectively. The Stationary, the Object manipulation and the Visual-motor integration scores were into Poor or Very Poor category for 29%, 34% and 46% of the sample respectively. Results are summarized in **table 1**.

Table 1. *Results on PDMS-2*

	PDMS-2		
	Score	Category	Poor or Very Poor Category
	Mean (DS)		% of the sample
Quotients			
Total Motor	71,03(8,99)	Poor	83
Gross Motor	75,11(8,15)	Poor	80
Fine Motor	71,63(11,16)	Poor	80
Subscales			
Static	7,03(2,23)	Below Average	29
Locomotion	5,14(1,11)	Poor	63
Object Manipulation	6,20(1,71)	Below Average	34
Grasping	4,26(2,28)	Poor	77
Visuo-motor integration	6,29(2,03)	Below Average	46

Motor skills are related to clinical features

All PDMS-2 quotients (QTM, QGM and QFM) were significantly directly correlated with non-verbal IQ. Fine-motor subscales (Grasping and Visuo-motor Integration) were significantly directly correlated with non-verbal IQ.

Correlation performed controlling IQ did not reveal significantly correlation between PDMS-2 measure and chronological age. No significantly correlation appeared between PDMS-2 measures and ADOS-G scores or ADOS calibrated severity scores.

The QGM was significantly correlated with domestic daily living skills subscale and the Stationary subscale was significantly correlated with domestic daily living, community and interpersonal relationship subscales. Correlation data are summarized in **table 2.**

Table 2. *Correlation analysis*

	PDMS-2							
	Quotients			Subscales				
	QTM	QGM	QFM	Static	Locomotion	Object Manipulation	Grasping	Visuo-motor Integration
NON VERBAL IQ	.667*	.480*	.627*	.369	.364	.377	.445*	.671*
CHRONOLOGICAL AGE	-.200	-.449	.278	-.556	-.309	-.199	-.023	.491
ADOS-G SCORES								
Social and Communicative	.353	.293	.185	.303	.320	.084	.156	.141
Repetitive behaviours	-.595	- .235	- .659	- .248	- .186	- .109	- .522	- .536
VABS DOMAIN								
Personal	.453	.523	.121	.419	.559	.247	.470	-.309
Domestic	.569	.740*	.009	.754*	.617	.307	.422	-.444
Community	.213	.560	-.378	.727*	.420	.090	-.080	-.547
Interpersonal relationship	.483	.593	.038	.738*	.504	.063	.178	-.131
Play and leisure time	.553	.661	.078	.560	.632	.388	.463	-.374
Coping skills	.473	.059	.014	.666	.550	.111	.249	-.248
Gross Motor	.522	.328	.442	.409	.138	.144	.406	.298
Fine Motor	.632	.382	.580	.203	.381	.290	.633	.283

* $p < 0.01$

Discussion

Based on a structured clinical assessment, the majority of preschoolers with ASD enrolled in this study showed a delayed motor development. According to previous reports performed with similar structured clinical motor tests (Vanvuchelen et al. 2007, Provost et al. 2007b, Jasmin et al. 2009, Zachor et al. 2010), our research suggest an atypical motor development in young children with ASD contrasting previous conclusions that motor skills in young children with ASD are relatively intact (Gillberg et al. 1990, Mayes and Calhoun 2003). However, these conclusions were based on parents' report on the child's age of the postural acquisition such as independent walking rather than on standardized motor assessment (Provost et al. 2007b). Thus, our data sustain the need to assess motor skills of young children with ASD in addition to other developmental areas usually assesses as social, communicative and cognitive areas. The reported atypical motor development in ASD assumes a theoretical relevance as it has been suggested that social cognition rely on the capacity to predict and understand the motor goals and motor intentions of the actions of others, an ability related to the cortical motor system (Gallese et al. 2013). The neural substrate would be represented by the mirror neuron system (Gallese and Goldman 1998, Rizzolatti et al. 2001, Gallese et al. 2004,) that can provide an implicit knowledge of other's intention based on the observation of the motor act (Pierno et al. 2008). The early atypical motor development in ASD could create a deficit in performing the intentional motor acts that may account for their difficulty in understanding the intention of the observed action (Gallese et al. 2013) and thus contribute to the social deficit in ASD.

In this study both Gross Motor (mainly locomotion) and Fine Motor (mainly grasping) skills are impaired in our sample, suggesting that motor difficulties might be due to some specific mechanism underlying motor control (i.e., motor planning). We found that goal-oriented skills such as locomotion and grasping are more impaired than Stationary and Visual-Motor Integration skills. In agreement with previous reports (Provost et al. 2007b, Jasmin et al. 2009,), the locomotion represents the most noticeable gross-motor vulnerability area in

preschoolers with ASD and it may reflect a lack of coordination (Fournier et al. 2010) or a defect of motor planning (Vernazza et al. 2005). On other hand, our findings substantially also confirm previous reports (Jasmin et al. 2009, Provost et al. 2007b) on a relevant impairment in grasping skills. This impairment may reflect a lack of anticipatory control (David et al. 2012) or an asynchronous activation of components of action functionally coordinated (Mari et al. 2003).

Our results suggest that in ASD low levels of cognitive functioning are related to greater motor impairment. The correlation between motor and cognitive abilities appeared particularly relevant for fine motor skills: Grasping and Visuo-motor Integrations subscales appeared significantly correlated with non-verbal I.Q. Our findings support previous report suggesting that intellectual disability in ASD may contribute to a greater degree of motor impairment in fine motor domain (Green et al. 2009, David et al. 2012).

The motor impairment characterized by a relative preservation of static abilities along with a major impairment in locomotion and grasping skills appear to be relative stable at preschool age as correlation analysis between chronological age and PDMS-2 measures did not reveal any significant results. This is in agreement with some studies (Provost et al. 2007a, Jasmin et al. 2009) whereas other reports supposed that motor abilities worse over time (Minshew et al. 2004, Lloyd et al. 2013). Because these latter assumptions are derived from studies with a longitudinal approach or with very wide sample age range, it is possible that the narrow age range considered in our study prevented to highlight modifications of motor impairments over time. So, even if our findings supported the existence of a stable pattern of motor impairment in preschoolers with ASD, further longitudinal studies are needed in order to clarify the issue of motor worsening over time within ASD.

Correlation analysis between severity of autism measured by ADOS scores and PDMS-2 measures did not reveal any significant value. This findings suggest that atypical motor development in ASD is not related to core symptoms of ASD. Although this is consistent with previous report with similar methodological approach (Zachor et al. 2010), other studies reported contrasting data (Jasmin et al. 2009, Sipes et al. 2011, Hilton et al. 2012, Mc Donald et al. 2014). Different reasons could support this contrasting results such as differences in range of age, methods of measurement of motor skills and measures of severity of autism. More recent findings pointed out a positive correlation between motor development in ASD and presence of restricted and repetitive behaviours (RRB) (Radonovich et al. 2013). It has to be considered that RRB were measured with Repetitive behaviors Scale-Revised (RBS-R, Bodfish et al. 2000) and that the assessment of RRB in a short period of time, as ADOS administration, may underrate them. The RBS-R is a detailed parental report which evaluates the full spectrum of RRB in a better way than ADOS-G. Thus the hypothesis that motor abnormalities are relevant in the mechanism underlying RRB (Ravizza et al. 2013) can be supported and further specific studies are needed to clarify this issue.

In our study there is a lack of significant correlation between motor scores assessed by PDMS-2, that is a directed observation and VABS specific motor scores, that is reported by parents. This findings suggest that the assessment of motor disorder in ASD cannot be entrusted to the exclusive parental reports but require a more detailed direct observation. However, consistent with a previous report (MacDonald et al. 2013) motor development has been found correlated with adaptive skills other than motor: static skills assessed by PDMS-2 appeared significantly correlated with some VABS domains. The finding of direct correlation between motor skills and adaptive behaviors supports previous data (Jasmin et al. 2009, Matson et al. 2010) and suggests an interaction between motor and interpersonal skills traditionally weakly associated with motor system.

One of the most important questions in this study if the participant's motor delays are truly functional or an issue of the standardized testing process. The administration of PDMS-2 requires child attend to instructions and/or demonstrations. Several items are generally not performed during daily routines and thus not spontaneously observed. Thus, the motor scores of children with ASD observed in this study could reflect not also the inability to physically perform items but also the inability to attempt the item following the standardized procedures. However, the PDMS -2 has already been used in preschoolers with ASD (Provost et al. 2007b, Provost et al. 2007a, Vanvuchelen et al. 2007, Jasmin et al. 2009, Riou et al. 2009, Zachor et al. 2010) wherein some advantages were reported as the lack of time trials and the less cognitive demand compared to other motor assessment tools (Vancuchelen et al. 2007). The advantage of providing a greater number of clues for the execution of the performance, the relative flexibility of the setting and the repurposing of items are features of the PDMS-2 lead us to judge this instrument enough suitable to estimate the motor level of preschoolers with ASD. In addition, the possible interference of the autistic imitative deficit on the level of motor scores may be considered minimized by the fact that all the PDMS-2 items which provide for the reproduction of gestures are associated with directions and verbal clues. Finally, the clinical features of our sample as a mean value of non verbal IQ of 98,2 have been certainly contributed to report the inability to physically perform items of preschoolers with ASD than their inability to attempt the item following the standardized procedures.

The lack of a control group consisting of preschoolers with non-ASD neurodevelopmental disorder prevent us to consider motor abnormalities found in children with ASD as specific of this disorders. Further research needs to be done to confirm our findings and to determine the specific value of motor impairments in ASD compared to those of children with non-ASD neurodevelopmental disorders (e.g., children with intellectual disability).

Conclusion

Motor development in preschoolers with ASD is characterized by 1) a delayed development as assessed by standardized tests, 2) locomotion and grasping skills are substantially the most impaired area, 3) an impairment that appears to be related to non verbal IQ and adaptive behaviors. All these findings support the need to assess motor skills in preschoolers with ASD in addition to other developmental skill areas. Along with the increasingly acknowledged importance of motor skills for subsequent social, cognitive, and communicative development (Bhat et al. 2012, Cashon et al. 2013, Libertus and Needham 2014), our findings support the need to consider motor intervention as a key area in therapeutic program to improve outcome in preschoolers with ASD.

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