Action performance in children with autism spectrum disorder at preschool age: A pilot study

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Abstract

Motor deficits related to imitation have been observed in autism spectrum disorder (ASD) patients. This pilot investigation focused on motor performances, including daily tool-use actions, performing an action in the absence of the tool, and imitating (copying tool-use action presented visually), in 8 children with ASD and 8 children with typical development (TD), with all of pre-school age (4-6 years). Motor performances were compared between the children with ASD and TD. Differences between an actual tool-use action and performing a tool-use action without the tool according to verbal instruction were also assessed between the groups. Children with ASD showed impairments in imitating, but their actual tool-use actions and tool-use actions without tools following verbal instruction were not different from those of TD children. The spatial error rate in the tasks was higher in children with ASD. The present study indicates that disturbance in imitating actions appears by the age of 4-6 years in children with ASD, possibly as a characteristic symptom affecting motor performance at pre-school age. Generalized apraxia might follow by the age of 8 years or older.

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Key words: ASD; Motor control; Motor Development; Imitating

Introduction

Autism spectrum disorder (ASD) is a neurodevelopment disorder defined by impairment of social and communication skills as well as restricted behaviors and interest (American Psychiatric Association. Deficits in motor control, including gross and fine movements, were also reported in previous studies (DeMyer et al. 1972; Jansiewicz et al. 2006). Another observation in children with ASD is impairment of gross and fine motor skills, observed before the age of 36 months and progressing with age (Lloyd et al. 2013). Motor functions related to motor skills, which include imitating, gesturing/pantomiming, and praxis, were also observed before the age of 48 months (Vivanti et al. 2014; Mastrogiuseppe et al. 2015), and such impairments led to generalized dyspraxia with poor complex movement and imitation on verbal command and during tool-use by school age in children with ASD (Bhat et al. 2018).

Tool-use actions are first observed by the second year of life (Connolly and Dalgleish 1989; Caselli et al. 2012), and the amount of tool-use actions and vocabulary increases in daily life in preschool-aged children. In school-aged children with ASD, studies of imitating and gesturing/pantomiming actions have revealed general dyspraxia in children with ASD between 8 and 12 years of age (Rogers et al. 1996; Mostofsky et al. 2006; Dziuk et al. 2007). Pre-school age between 4 and 6 years old is the most critical and rapid period of motor and cognitive development in human life (UNICEF 2017; Zeng et al. 2017), possibly with increasing opportunities for tool-use and social communication. Therefore, we considered that developmental changes at pre-school age between 4 and 6 years old should be addressed in children with ASD. In view of development of motor performance, 4-year-old children were in a transitional stage regarding the hand posture for tool-use gripping in a previous study (Comalli et

al. 2016), and the age between 4-6 years old might be an important period for the development of tool-use actions. Although previous studies assessed verbal and tool-use skills in children with autism before the age of 2 years old (Libertus et al. 2014; Sparaci et al. 2018), the number of studies focusing on fine motor skills and tool-use actions in pre-school children between 4 and 6 years old is still limited (Rogers et al. 2010; Kana et al. 2011; Paquet et al. 2016). Regarding verbal/social communication, the relationship between motor and language development was previously reviewed (Iverson 2010), and Mody et al. (2017) reported a strong association between fine motor skills and verbal/social communication skills in children with ASD in aged between 2 and 15 years. Pre-school age, in which children learn tool-use actions as well as precise actions, could be an important and critical period to develop fine motor and verbal/social communication skills for children with ASD.

We conducted a pilot study to compare tool-use actions between pre-school children with ASD and TD aged between 4 and 6 years old. We hypothesized that developmental delay of tool-use actions depended on verbal/social communication skill, and that there might be difference of developmental delay of tool-use actions among communication modalities associated with the actions between 4 and 6 years old. The present study focused on tool-use actions following three main different modalities: performing a tool-use action without the tool but according to verbal instruction (Task 1), imitating actions visually presented on a monitor (Task 2), and performing real actions (Task 3), and two sub-modalities: tool naming, tool selection in Task 2, and tool naming in Task 3. Since imitating actions included informing contents of actions to others, we expected to find a significant developmental delay in imitation rather than in actions following verbal instruction or spontaneous action production in children with ASD in aged between 4 and 6 years. By understanding of the characteristic disturbance in

actions in pre-school children with ASD between 4 and 6 years old, if there is any, therapist could conduct specific approaches to the disturbance in tool-use actions before school-age, in which children are exposed with numerous tool-use actions. Preparatory intervention to school life for children with ASD during pre-school age, may help them to adopt daily life in school.

In the present study, a tool-use action was defined as an action to use a tool in a daily activity, such as a spoon, knife, or phone. We asked children to perform a tool-use action in the absence of the tool, with an understanding of what the action was. The performance in the present study indicates a body movement simulating a tool-use action, which corresponds with pantomime/pantomiming in Kendon's continuum (McNeill 1992, Studdert-Kennedy 1993). Imitation was defined as copying an action presented visually regardless of understanding of what the action was.

Methods

Participants

Eight children with ASD (7 boys and 1 girl) and 8 showing TD (3 boys and 5 girls) were enrolled in the present study. The participants were 4 to 6 years of age (mean, ASD: 66 ±8.9 months (SD), TD: 62.5 ±12.4 months). The children with ASD were recruited from domestic daycare centers for children with disabilities. Inclusion criteria for the study specified that a pediatrician and pediatric psychiatrist had to verify the diagnosis using the Diagnostic and Statistical Manual of Mental Disorders - Fifth Edition (DSM-V; American Psychiatric Association 2013) as autism spectrum disorder, Asperger's syndrome, high-functioning autism, or pervasive developmental disorder. Children with diagnoses of attention deficit hyperactivity

disorder, developmental coordination disorder, or any neurologic or psychiatric disorder were excluded. The TD children were recruited from nursery schools near our institute. The Social Responsiveness Scale (SRS) (Constantino and Gruber 2005) was used to exclude participants with probable (SRS score of 60 or higher) and possible (76 or higher) ASD from the TD group. Participants with an IQ lower than 70 on the Wechsler Intelligence Scale for Children, 4th Edition (WISC-4) (Wechsler 2003) were also excluded from the TD group. Eight participants in the ASD group had SRS scores of 60 or higher, and they completed the experiment. Ten participants were initially recruited in the TD group, but one with a high SRS score, 86, was excluded. One TD child did not complete the participation in the experiment and the participant was also excluded. Therefore, eight participants were finally present in both ASD and TD groups.

Profiles of the participants in the two groups are presented in Table 1. Intellectual functioning and basic verbal abilities were assessed using WISC-4. There were significant differences in the full-scale IQ and Verbal Comprehension Index (VCI) between the two groups (Table 1). In addition, the SRS and Developmental Coordination Disorder Questionnaire (DCDQ) (Wilson et al. 2000) showed significant differences between the two groups (Table 1).

All parents of the children were given an explanation of the purpose of the study and consented to the children being videotaped while taking part in it. The protocol of the study was explained orally and in writing to one of the parents, who signed a consent form to participate in the study. We also explained that the video recordings made during the study would not be presented in academic reports including papers. Ethical approval was obtained from the local ethical committees of the National Rehabilitation Centre for Persons with Disabilities and the Faculty of Medicine, Nagoya University.

Experimental procedure

The series of experimental tasks used in the present study was devised based on the Florida Apraxia Screening Test (Power et al. 2010) and Praxis Examination for children (Mostofsky et al. 2006), to assess the ability to perform tool-use actions with or without the tool. The original tasks involved transitive performance of tool-use actions, such as cutting paper, and intransitive performance, such as waving goodbye (Mostofsky et al. 2006). In the present study, the three tasks of transitive performance of tool-use actions were selected based on the Florida Apraxia Screening Test (Power et al. 2010), as follows:

Task 1: perform a tool-use action without a tool, according to verbal instructions (tool-use action following verbal instruction), such as "Show me how you use a comb." The examiner showed the correct action after the performance of the children.

Task 2: imitate a tool-use action without the tool, according to a tool and action displayed on an iPad (9.7-inch iPad, Apple Inc.). Participants then had to verbally give the name of the tool (tool naming) and then select a picture of the tool from 4 pictures (tool selection). Four pictures included a correct tool and three incorrect objects in different categories, semantic, function associated, and motoric categories. Ten sets of four pictures are listed in Table 2, which were modified from the sets used in the previous study (Power et al. 2010). The examiner showed the correct action, name of the tool, and correct selection of the picture after the performance of the children.

Task 3: perform a tool-use action with a tool (using tool). A tool for an action was given by the examiner, and the participant was asked to show how to use it. Then, participants had to

verbally provide the name of the tool (tool naming). The examiner showed the correct action and name of the tool after the performance of the children.

Ten tool-use actions listed in Table 2 were used for each task. Each of the three tasks was conducted on a different day with intervals of at least 7 days, to prevent fatigue of the participant. The order of the tasks was randomly selected for each participant. The participant sat on a chair. An examiner sat beside the participant to give verbal instructions and how them the tablet monitor for Tasks 1 and 2, respectively. Prior to the task, each participant was given an explanation of the procedure.

Scoring and analysis

All performances were video-recorded. Errors in the performances were checked by two independent investigators who were blinded to the participants' diagnoses and objectives of the study. Each participant performed a total of 30 actions, and the total number of errors during the 30 actions was counted. The rates of actions correctly and incorrectly performed are expressed as percentages, the correct and error rate, respectively, for each participant. For incorrect performances, error types were identified using the classification by Power et al. (2010) (Table 3). When an error listed in Table 3 was observed, the action was counted as an incorrect action and the type of error was identified. Incorrect actions and types of errors were recorded in each task, and the answers for tool names were also assessed for correctness. Numbers of incorrect actions counted by two investigators were averaged for each participant, and types of errors and correct answers for the name were evaluated between TD and ASD groups using the t-test followed by multiple comparisons with the false discovery rate (FDR).

Results

Table 4 shows the results of performances of the TD and ASD children. During each of the 3 tasks, the total correct action rate for the 10 tool-use actions (Table 2) in the ASD group was significantly lower than in the TD group ($p = .02$, t-test). In the children with ASD, correct rates were lower for the imitation and picture choice performances in Task 2 ($p < .01$). However, there was no significant difference in Tasks 1 (tool-use action following verbal instruction) and 3 (using tool), and tool naming in Tasks 2 and 3.

Regarding the number of errors and erroneous actions in all 30 actions performed during 3 tasks, the total number of errors during performances was higher in the ASD group than in the TD group ($p = 0.02$), and the rate of the spatial error type in Table 3 was significantly higher among the children with ASD ($p = 0.01$). The error rate for body part for tool (BPT) was lower in the ASD than in TD group ($p = 0.04$).

Discussion

The present pilot study observed the developmental delay of showing a tool-use action without the tool and imitating actions in pre-school ASD compared with TD children between 4 and 6 years of age. Children with ASD showed impairments in imitating tool-use actions, but there was no significant difference in performing an action following verbal instruction and actual tool-use actions, although the number of participants involved was small in the present study. The present results partially support previous studies (Mostofsky et al. 2006; Dzuik et al.

2007; Dowell et al. 2009). In previous studies (Mostofsky et al. 2006; Dzuik et al. 2007; Dowell et al. 2009), children with ASD aged 8 to 14 showed significantly poorer responses for all three modalities tested in the three tasks in the present study. Developmental delay in children with ASD might progress during the pre-school period to generalized dyspraxia by the age of 8 years or older.

In Task 1, we assessed the ability to perform a tool-use action upon verbal command. Zoia et al. (2002) stated that this verbal modality could be used as a strategy to recall and perform a tool-use action by accessing action semantics together with knowledge of the tool and object functions, and that the ability to use such a verbal strategy does not develop before 9 years of age. The usage of the verbal strategy was still considered to be immature in both TD and ASD groups, and so performing a tool-use action following a verbal command was not significantly different between the groups, although the correct answer rate in Task 1 was lower in ASD than TD children.

In Task 2, we examined the ability to imitate actions. The performance was poorer in ASD than TD children. Deficits in imitation were reported in previous studies regarding motor performance from infants to adults with ASD (Rogers et al. 1996, 2003; Hamilton et al. 2007). Similar results were obtained in the present study, suggesting that impairment in imitating actions continues during pre-school in children with ASD. We considered that imitating actions, which contained communicative performance to inform contents of actions to others, might be one of core deficits in motor performance in children with ASD, and that motor deficit in imitating actions relatively more evident than performing a tool-use action without the tool or spontaneous actual performance in children with ASD in aged between 4 and 6 years.

In Task 3, the ability to use a tool correctly was assessed. Children with ASD aged between 4 and 6 suggested a tool-use action with the tool similarly to TD children. Two possibilities for this can be considered: 1) both TD and ASD groups showed immature performance, and 2) both developed similarly until the level of tool-use action typical for pre-school. Performing a tool-use action was reported to develop in the early stage of motor development (Zoia et al. 2002). The correct action rate in Task 3 was high for both TD and ASD groups, although the values were lower, without significance, in children with ASD. We considered that children with ASD showed similar tool-use action performance to that in TD children at least at pre-school age between 4 and 6 years old in the present study. Children with ASD will not reach a level involving difficult action performance when aged over 8 years old, as reported in previous studies (Mostofsky et al. 2006; Dzuik et al. 2007; Dowell et al. 2009). There is a possibility that the difference in tool-use action performance between ASD and TD children becomes significant at the age of 6-8 years old.

Another difference in performance was the poorer tool selection from the four pictures of tools by children with ASD in Task 2 (tool selection). The children with ASD knew the names of tools similarly to TD children, as shown in Task 3 (tool naming). Neither the TD nor ASD group achieved a high correct rate, 33.8 and 55.0%, respectively, but TD children selected the correct pictures more often than children with ASD. Immaturity of the verbal strategy shown in Task 1 in both groups might account for the lack of difference in tool naming. We could not draw conclusions about the pathophysiology of poor tool selection from the results of the present study. In the selection task, participants needed to translate another's movement of an action of tool-use into a picture of a tool in front of them. We consider that children with ASD might have difficulty in translating another's action into an object placed in front of them due to the

impairment of multiple brain systems relating to the understanding of actions (Hamilton et al. 2007). However, there are alternative explanations such as poor inhibition of behavior, poor visual exploration (Heaton and Freeth, 2016), or the use of tasks originally designed for adults.

In order to understand the reasons behind the failures/development of imitating and performing an action without the tool in children with ASD, the types of errors made by the two groups were examined. Spatial errors were more frequent in pre-school children with ASD than in TD children. This finding might be consistent with a previous report, which showed that children with ASD older than 8 years made more spatial errors than in other categories (Mostofsky et al. 2006). The BPT error, in which a part of body was moved as if a part of tool instead tool-using movement of body parts, was the most common error type in the TD group. The number of BPT errors as a percentage of all errors was significantly higher than in children with ASD at the ages considered in the present study. The BPT errors could be observed in patients with motor deficits due to focal brain lesions (Raymer et al. 1997). However, Moftofsky et al. (2006) reported that the BPT errors observed in children with ASD were not due to motor deficit but a part of general dyspraxia. Since BPT errors were seen even in TD children up to 7 years of age (Kaplan 1983), the BPT errors observed in the present study between the ages of 4 and 6 years old were not considered to be pathological and diminished at an older age, i.e., the absolute number of PBT errors was not higher in TD than ASD children, as shown in Table 4. The remaining BPT errors in children with ASD aged between 4 and 6 years old might be a sign of general dyspraxia.

We did not focus on social communication skills in the present study. However, the results provided a hint regarding the association between fine motor skills and social communication skills in children with ASD at pre-school age. The present participants with ASD showed lower

VCI and SRS scores, which indicated poor social communication skills, than TD children. The children with ASD in the present study, with poor social communication skills, showed relatively more impairment in imitation than in performing a tool-use action without the tool and actual performance of tool-use actions. There is a possibility that the difference in tool-use action was a secondary result of poor social communication skills in children with ASD revealed by those batteries. Among tasks in the present study, performing a tool-use action without the tool following verbal instruction and tool-use actions might be difficult and easy, respectively, for both ASD and TD groups at pre-school age, as described above. Although an association between motor and verbal/social communication skills in children with ASD between 2 and 15 years old was observed (Mody et al. 2017), types of motor skills related to social communication skills might be different among children with ASD of different ages. The relationship between motor skills and verbal/social communication skills is still unclear, and should be investigated in further studies.

Limitations

In this pilot study, the tasks were adapted for pre-school-aged children from an adult dyspraxia battery (Power et al. 2010; Mostofsky et al. 2006). A standardized assessment of praxis for children has not yet been developed, and there might be a need to use more pre-school age-appropriate battery. Our results would also be strengthened by increased numbers in each group as well as a broader age range in order to investigate more detailed developmental processes. We did not analyze the correlations among the Full Scale Intelligence Quotient (FSIQ), Verbal Comprehension Index (VCI), and performing an action without a tool. Finally, although individual experiences using specific tools contribute to the ability to perform a tool-

use action, it was difficult to determine the participants' experience of using tools. Nevertheless, we could ascertain that children performed actions without tools differently depending on the input modality. Due to the small number of participants, only the chronological age was matched between children with ASD and TD, which might have affected the results.

Conclusions

Disturbance in imitating actions was first observed in children with ASD at pre-school age between 4-6 years. Such children showed disturbance in imitating tool-use actions but not in performing a tool-use action without the tool and actual performance. Imitating actions, which contained communicative performance to inform contents of actions to others, might be one of core deficits in motor performance continuing throughout in children with ASD. The motor developmental delay or impairment of the motor performance could differ among children during pre-school, school, and adolescence. Intervention and therapeutic approaches specific for each developmental period may be beneficial for children with ASD.

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Conflict of Interest

The authors declare that they have no conflicts of interest.

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Table 1: Profiles of participants

	ASD	TD
Number of participants	8	8
Boys	7	3
Girls	1	5
Age in months; mean (SD)	66 (8.96)	62.5 (12.4)
FSIQ; mean (SD)	82.4 (9.56)	104.8*(10.36)
VCI; mean (SD)	82.1 (13.99)	104.6 *(9.38)
SRS score; (SD)	73.1 (16.49)	45.75*(4.09)
DCDQ score; mean (SD)	39.6 (13.68)	68*(11.47)

ASD: children with ASD
TD: typically developed children
FSIQ: Full Scale Intelligence Quotient.
VCI: Verbal Comprehension Index.
SRS: Social Responsiveness Scale.
DCDQ: Developmental Coordination Disorder Questionnaire.
* $p < .005$

Table 2: Ten sets of tools and actions used in the present study and the pictures used in Task 2 (see text). Each set of four pictures includes a correct tool and three incorrect objects in different categories.

	Tool and action	**Pictures**			
		Correct tool	**Incorrect objects and category of objects**		
			Semantic	**Function associate**	**Motoric**
Practice	**Phone** and calling	Phone	String telephone	Phone book	Hammer
1	**Spoon** and scooping ice cream	Spoon	Chopsticks	Cup & saucer	Scoop
2	**Glass** and drinking water	Glass	Tea cap	Water pitcher	Banana
3	**Knife** and cutting a vegetable	Knife	Scissors	Carrot	Saw
4	**Key** and unlocking a door	Key	Key ring	Lock	Screwdriver
5	**Comb** and combing hair	Comb	Brush	Hair	Hat
6	**Screwdriver** and turning a screw	Screwdriver	Chisel	Screw	Key
7	**Toothbrush** and brushing teeth	Toothbrush	Brush	Cup	Eraser
8	**Hammer** and hitting a nail	Hammer	Spanner	Nail	Drumsticks
9	**Scissors** and cutting a piece of paper	Scissors	Shears	Paper	Pliers
10	**Pencil** and writing	Pencil	Ruler	Notepad	Needle

Table 3: Classification of behavioral error (modified from Rothi et al. 1997)

Error type	Error sub-type	Description
Spatial	Amplitude	Amplification reduction or irregularity of amplitude/position in space
	Internal configuration	Abnormality of finger/hand posture with target tool
	External configuration	Abnormality of finger/hand/arm relationship with the object as the target of the action
	Movement	Any disturbance of the characteristic action required to complete the goal
Content	Concretization	Mimicking the use of a real object not usually used in the task
	Perseverative	Response includes all/part of a previous response
	Related	An accurate mime associated with the target
	Nonrelated	A real and accurate mime not associated with the target
	Hand	Not using a tool, e.g., tearing paper when target is scissors
Temporal	Sequencing	Movement structure recognizable but addition, deletion, or inaccurate order of sequence
	Timing	Alteration of timing/speed (including increase, decrease, or irregular)
	Occurrence	Representative production of single movements or single production of multiple movements
Body part for tool (BPT)		Using fingers, hands, or arms as a part of a tool instead tool-using movement of body parts
Other	No response	Participant shows no response to request
	Unrecognizable response	Shares no spatial or temporal features with target

Table 4: Correct action rate and types of errors during action tasks. Grey rows: significant difference between ASD and typically developed (TD) children ($p<0.05$, t-test with FDR).

	ASD	TD	*p*-value
Correct action rate in 30 actions (%)			
Total	60.4 (33.3 to 80.0)	79.6 (53.3 to 93.3)	.02
Performing an action without the tool following verbal command (Task 1)	43.8 (10 to 80)	67.5 (30 to 90)	.08
Imitating (Task 2)	48.8 (20 to 70)	77.5 (40 to 100)	.007
Using tool (Task 3)	88.8 (60 to 100)	93.8 (90 to 100)	0.56
Tool naming (Task 2)	33.8 (0 to 80)	55.0 (30 to 80)	.08
Tool selection (Task 2)	43.5 (0 to 80)	82.5 (60 to 100)	.005
Tool naming (Tasks 3)	68.8 (20 to 90)	83.3 (80 to 100)	0.46
Number of errors in 30 actions			
Total	95 (6 to 20)	49 (2 to 14)	.02
Spatial	44 (4 to 9)	15 (0 to 3)	.001
Content	9 (0 to 3)	5 (0 to 2)	0.19
Temporal	1 (0 to 1)	1 (0 to 1)	0.5
BPT	26 (0 to 7)	21 (1 to 6)	0.42
Other	15 (0 to 6)	7 (0 to 3)	0.2
Rate of error type (%) in erroneous actions			
Spatial	46.3 (29.4 to 90.0)	30.6 (0 to 50)	0.25
Content	9.5 (0 to 27.3)	10.2 (0 to 25)	0.41
Temporal	1.1 (0 to 14.3)	2.0 (0 to 7.1)	0.93
BPT	27.4 (0 to 53.8)	42.9 (25 to 100)	.04
Other	15.8 (0 to 30)	14.3 (0 to 37.5)	0.54

BPT: Body part for tool