Longitudinal Case-Control Study of Imitation in Younger Siblings of Children with Autism Spectrum Disorder

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Background: *Siblings of children with autism spectrum disorders (ASD) have higher prevalence of ASD with a recurrence of 19%. Children with ASD demonstrate significant impairment in all types of imitative skills. Imitation is markedly developed in the first few years of life; therefore, a study of imitation in younger siblings in this period may reveal early deviation.*
Objective: *To study the development of imitation skills from 9- to 18-months, specifying types of imitation, in siblings of children with ASD compared with typically developing children.*
Method and Material: *A longitudinal case-control study was conducted on eight siblings of children with ASDs and nineteen typically developing children who were age- and gender- matched. Data collection consisted of parental recording of emerging imitative abilities and structured direct observation of imitative skills at 9, 12 and 18 months. Three types of imitative skills were targeted including vocal, object and gesture imitation.*
Results: *The development of vocal imitation in siblings of children with ASD between 12 to 18 months was delayed in comparison with typically developing children with significant statistical difference at 18 months. Object and gesture imitations were not significantly different between the two groups.*
Conclusion: *Siblings of children with ASDs had some delays in vocal imitation skills at the age of 12 to 18 months, compared with typically developing children.*

Keywords: *Imitation, Sibling, Autism, Autism spectrum disorder*

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Autism spectrum disorder (ASD) is a neurodevelopmental disorder characterized by impairments in communication and social interaction with a restricted pattern of behaviors and interests. Although prevalence of ASD in the general population is 1.1%[1], younger siblings of children with ASD are at increased risk of developing ASD (19%)[2]. In addition, up to 30% of the siblings exhibit broader autism phenotype features, which include social difficulties, rigidities, and language delays[3,4]. The exact cause of ASD is still unknown, but it is now well established that ASD is a complex heritable disorder. Actual candidate genes and genomic regions are being evaluated for their potential role in autism[5].

Currently, many researchers are focusing on the identification of early markers in infants and young toddlers because research suggested that early intervention leads to better outcomes for children with ASD[6-8]. Typically developing children demonstrate the ability to imitate other persons' behaviors from birth and progress during the first two years of life; imitation is considered an important component of both social and cognitive development[9,10]. From a social perspective, imitation is one of the earliest forms of reciprocal interaction between infant and caregivers, and from a cognitive perspective, imitation is a precursor to learning about using objects as tools and the development of both play and language. Given the theoretical importance of early imitation to social and cognitive development, imitation deficit was proposed as one of the fundamental ASD symptoms[11].

A systematic review of imitation in ASD showed that children with autism demonstrated significant impairment in all types of imitative skills[12]. Problems with imitation were seen as early as 2 years[13,14] and continued into adulthood[15]. Studies

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of imitation both in typically developing children and in children with ASD classify forms of imitation in different ways. Principal distinctions were object vs. gesture; meaningful vs. non-meaningful; and immediate vs. deferred[12]. Although every type of imitation deficit was reported in children with ASD, some types of imitation were more susceptible to impairment. For example, problems in gestural imitation (imitation of action without object) were more severe than in procedural imitation (imitation of object manipulation)[16]; imitation of non-meaningful gestures appears more difficult than meaningful gestures[17]; and imitation of non-meaningful action on objects was more challenging than meaningful action on objects[14]. Another crucial imitative skill is vocal imitation because imitation of sound is an important process in the early-speech period of infants. A previous study also mentioned poor imitation of voice and sound in 24-month-old children with ASD[17].

Many siblings of children with ASD exhibited autism phenotype features in the first few years, both in a group that later were diagnosed with ASD and a group that belong to broader autism phenotype[2]. Features of autism, which were seen in previous research, included: lower verbal skills, poor eye contact, limited functional play, repetitive behaviors and impaired imitation[18]. However, it is not clear when and how imitative impairment in siblings of children with ASD emerges, either as a result of a loss of skills or a failure to acquire new skills[19,20], nor which type of imitative skill is weakened in siblings of children with ASD.

Our research studiedthe developmental trajectory, specifically three types of imitation, in siblings of children with ASDs who were a risk-group compared with typically developing children in the period of rapidly developing of imitative skills.

Material and Method

Subjects

Children in the sibling group were enrolled from Developmental and Behavioral Pediatrics Units, Ramathibodi Hospital, Bangkok, from May to December, 2008. The sibling group was recruited at ages of 9 or 12 months (±2 weeks). Their older biological siblings were diagnosed based on DSM-IV criteria by developmental and behavioral pediatricians or child psychiatrists. The control group was age- and gender-matched typically developing children enrolled from the Well Child Clinic and daycare center. Children in the control group had no history of first degree relative suspected or diagnosed with ASD. Children who were born prematurely (gestational age less than 37 weeks) and who had significant developmental delays (developmental quotient <70) were excluded. Children who had risk of hearing impairment due to JCIH criteria[21] were sent for auditory evaluation and would be excluded if the result was abnormal.

Methods

The project has been reviewed and approved by the committee on human rights related to researches involving human subjects, Faculty of Medicine, Ramathibodi Hospital. After informed consents were obtained, participants' and parents' demographic data were collected by questionnaire. The Capute Scales[22,23] were used to evaluate the developmental status at the first visit (age 9- or 12-month) and at 18-month-old visit. Studying imitation was comprised of two structures: the parental recording data and the direct observation at the clinic. In regard to the parental recording data, parents received the form at the first visit and were informed on how to record the child's age in months when they saw their child begin to perform the imitative skills listed in the recording form. Parents were phoned for data collection on a monthly basis. The direct observation was performed three times at age 9-, 12- and 18- month-old at the Developmental and Behavioral Pediatrics Unit, Ramathibodi Hospital by fellows in Developmental and Behavioral Pediatrics. Inter-observer reliability of each item in direct observation was tested and weighted kappa (w) was calculated. The kappas ranged from 0.86 to 1.0 which revealed excellent reliability. At 18-months of age, the Modified Checklist for Autism in Toddlers (M-CHAT) was screened for ASD[24].

Materials and procedure

The capute scales[22,23]

The Capute Scales is a 100-item developmental assessment tool to quantitatively measure language and non-verbal problem-solving skills in infants from birth to 3 years of age. The Capute Scales are composed of the Capute Linguistic and Auditory Milestone Scales (CLAMS) and the Capute Adaptive Test (CAT). To generate the developmental quotients (DQ), the total CLAMS and CAT scores are divided by the chronological age. A DQ of 100 indicates that the child is performing at what is expected at his/her age. A standard deviation of DQ is 15, so a DQ less than 70 (2 standard deviations) represents significant impairments in development.

Modified checklist for autism in toddlers (M-CHAT)[24]

The M-CHAT was designed as a simple, self-administered, parental questionnaire. The M-CHAT is validated for screening toddlers between 16 and 30 months of age, to assess risk for autism spectrum disorders (ASD). M-CHAT is consisting of 23 yes/no items. The sensitivity and specificity for criterion 1 were 0.95-0.97 and 0.95-0.99, respectively.

Parental recording form

The parental recording form of imitation is designed as a longitudinal recording form for parents to continually record their child's emerging imitative ability at home (or elsewhere) from 9 to 18 months. The form consisted of items of three types of imitative skills: (1) gesture imitation (tongue protrusion, facial expression, hand waving, hand movement with opening and closing), (2) object imitation (hair brushing, telephoning, spoon-feeding, block banging), and (3) vocal imitation (non-word sounds, and word sounds). Parents were informed to demonstrate the actions to their child and encourage their child to imitate them frequently. If the child could imitate the action at least twice (to ensure that this action did not accidentally occur), the parents would then record the age at which their child could imitate the action.

Direct observation

Each session of the direct observation was approximately 45-60 minutes at the Developmental and Behavioral Clinic. One parent accompanied the child in each session. Three types of imitative skills; vocal imitation, object imitation and gesture imitation were observational targets. For vocal imitation, the researcher demonstrated how to make a "popping" sound by rapidly opening her mouth and then encouraged the child to imitate makingthe sound. Five trials were demonstrated, the child was scored as a "pass" if he/she could correctly imitate it twice.

Concerning object imitation, a squeezing car toy was used for two target actions: (1) tapping on the top of the car to make squeezing sound which would be classified as "non-functional object imitation", and (2) holding and pushing the car forward and backward which was classified as"functional object imitation". After five trials, if the child did not imitate at all, the researcher would physically prompt him/her to do the action and subsequently gave him/her another five trials to imitate the action.

Regarding gesture imitation, a social gesture i.e. hand waving ("bye-bye") was demonstrated. The researcher waved her own hand without saying anything and let the child imitate. If the child did not imitate after five trials, the researcher would then say "bye-bye" together with waving and encouraged the child to imitate her. If he/she still had no response, the researcher would attempt to prompt the child by manipulating the child's hand to wave bye-bye. The child was again given five trials for each setting. The passing criteria for object and gesture imitation were the same as that for vocal imitation.

Statistical analysis

Chi-square and Fisher exact tests of significance were used for univariate analysis of discrete variables. T-tests were used to compare case and control differences on continuous variables. Group differences regarding age in parental record were checked with the Mann-Whitney U-test.

Results

A total of eight (8) siblings of children with ASD, and nineteen age- and gender-matched typically developing children participated in this study. Two children in the siblings group and fourin the control group were enrolled in the project at 12-months. Table 1 demonstrated the demographic data of both groups; no significant differences in sex, maternal education, family's income and DQ scores between two groups were noted. The mean ages of the first direct observation in the sibling and control groups were 9.7 months (SD = 0.9) and 9.9 months (SD = 0.7), respectively: again, no significant difference between groups was seen. The mean ages of the second and third direct observation in both groups were similar and also demonstrated no statistical differences.

Parental recording data

Fig. 1 shows the box-plot comparing the age in months when parents of each group indicated their children's ability to perform the specified imitative skills. The median ages of children in both groups as reported by the parents do not present significant differences in all types of imitation. The range of age of the items "tongue protrusion" and "facial expression" in gesture imitation was quite extensive. The ages were reported as early as 2 months to 17 months. The other two gesture imitations, including "hand waving" and" alternating hand opening and closing", were mostly reported by parents at around ages of 8 to12 months. Regarding vocal imitation, most children (25^{th}-75^{th}

Table 1. Sample characteristics for siblings of children with ASD and control group

Demographic data	Sibling		Control		*p*-value
	n	Percent	n	Percent	
Sex					
Male	4	50	10	52.6	1
Female	4	50	9	47.4	
Maternal education					
≤12 years	3	37.5	3	15.8	0.32
≥12 years	5	62.5	16	84.2	
Family income					
<30,000	6	75	7	36.8	0.1
≥30,000	2	25	12	63.2	
	Mean	SD	Mean	SD	*p*-value
DQ score					
9-12 months	101.1	14.7	100.5	8.6	0.91
18 months	101.6	9	106.1	8.5	0.23

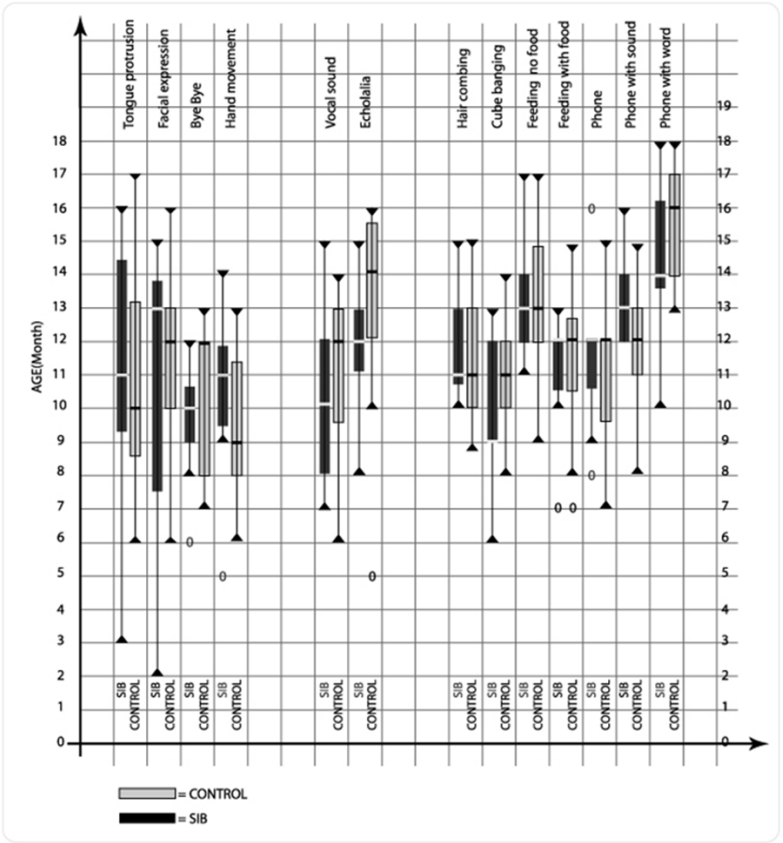

Fig. 1 Box-plot comparing age in months that parents of each group indicated their children's ability to perform the imitative skills in parental recording data.

percentile) in both groups began to imitate "vocal sounds" at the ages of 8-13 months and "word sounds (echolalia)" at around 11-15 months. On the topic of object imitation, the range of ages at which most children (25^{th}-75^{th} percentile) from both groups could imitate combing, block banging, feeding a doll with real food and feeding a doll without real food were 10-13, 9-12, 10.5-12.5 and 12-15 months old, respectively. The set of items concerning telephoning reveals that children could imitate "telephone talking without sound" earlier than "telephone talking with sounds" and "telephone talking with words".

Direct observation of imitation

Regarding vocal imitation, at age 9 months, no child in either group imitated the "pop sound" demonstrated by the researcher. In the control group, from the age 12 to 18 months, the typically developing

Table 2. Number and percentage of subjects in each group passing the vocal imitation test in the direct observation session

Age at direct observation	Sibling		Control		*p*-value
	n	Percent	n	Percent	
9 months	0/6	0.0	0/15	0.0	
12 months	1/8	12.5	3/19	15.8	1.00
18 months	0/8	0.0	9/19	47.4	0.03*

Table 3. Number and percentage of subjects in each group passing the non-functional and functional object imitation test in the direct observation sessions

Type of object imitation skill	Number/ percent passing	Sibling			Control		
		9 months	12 months	18 months	9 months	12 months	18 months
Non-functional object imitation							
No prompt	n	3/6	7/8	6/8	6/15	12/19	15/19
	Percent	50	87.5	75	40	63.2	78.9
Prompt	n	4/6	7/8	7/8	10/15	14/19	19/19
	Percent	66.7	87.5	87.5	66.7	73.7	100
Functional object imitation							
No prompt	n	1/6	5/8	5/8	1/15	10/19	17/19
	Percent	16.7	62.5	62.5	6.7	52.6	89.5
Prompt	n	1/6	6/8	6/8	3/15	11/19	17/19
	Percent	16.7	75.0	75.0	20.0	57.9	89.5

Table 4. Number and percentage of subjects in each group passing the gesture imitation in the direct observation sessions

Type of gesture imitation	Number/ percent passing	Sibling			Control		
		9 months	12 months	18 months	9 months	12 months	18 months
Without sound cue	n	1/6	1/8	1/8	1/15	7/19	10/19
	Percent	16.7	12.5	12.5	6.7	36.8	52.6
With sound cue	n	1/6	2/8	5/8	8/15	12/19	17/19
	Percent	16.7	25.0	62.5	53.5	63.2	89.5
With sound cue and prompt	n	2/6	2/8	5/8	9/15	13/19	17/19
	Percent	33.3	25.0	62.5	60.0	68.4	89.5

children had demonstrated the vocal imitation skill increasingly (15.8% to 47.4%), while, almost all of the siblings of the children with ASD did not imitate vocal sound. As a result, at 18-months-old, significant difference in vocal imitation was seen between 2 groups (Table 2).

Concerning object imitation, no group difference was seen in performing non-functional and functional object imitation, before and after physical prompting (Table 3). For non-functional object imitation, nearly half of the children in both groups could do this task at 9 months and about 75% could pass this task at 18 months without prompting. Functional object imitation was acquired later than non-functional. At 9-month-old, only one subject from each group was passed without prompting. From 9 to 18 months, percentage of children in both groups who passed this test increased over time.

In gesture imitation (Table 4), we divided the direct observation into 3 steps; gesture without sound cue, with sound cue (bye-bye), and with physical prompt. Concerning gesture imitation without sound cue, at 9-months old, only one subject in each group could imitate hand-waving gesture demonstrated without any sound cue. Between 12- to 18-months, many children in the control group acquired this skill, whereas, the number of children in the sibling group who passed the item remained the same (one child). Later, at 18 months, percentages of children in control and sibling groups who passed the test without the sound cue were 52.6 and 12.5 percent, respectively, at this age the group difference almost reached the significant value (p-value = 0.09). Once the sound "bye-bye" was added to gesture demonstration, many children in the control group could take advantage of this cue at the age 9- and 12- months. The percentage passing increased more in the control group (42.8%, 27.4%) than in the sibling group (0%, 12.5%). Interestingly, at 18-months old, the sibling group seemed to get a bigger advantage from the sound cue than in younger ages. The percentage passing after adding the sound cue increased from 12.5 to 62.5%. Regarding physical prompt, when adding physical prompt to sound cue, no significant effect was seen in either group at any time of direct observation.

At 18-months old, one girl in the sibling group failed the screening for ASD with the Modified Checklist for Autism in Toddlers (M-CHAT). She had participated in our research since she was 12-months old. She rarely demonstrated use of any imitation between 12- and 18-month-old in the direct observation. Her behavior contrasted with other children in the sibling group, who imitated more frequently any various points in our direct observation, though not as much as the control group.

Discussion

In this study, we aimed to investigate the development trajectory of imitation in young siblings of ASDs in comparison with typically developing children at early ages. The authors found that the deviant imitative skills in siblings of children with ASDs may be seen around 12 months, when behavioral symptoms usually emerge in siblings who are later diagnosed as ASD or a broader phonotype of ASD[25]. We specified three types of imitation because evidence suggests that some types of imitation are more deficits in children with ASD[12,16]; therefore, detailed examination of imitation by type may help in gaining more understanding of imitation weakness.

The study revealed no group difference of imitation from the parental reports, while direct observation showed that the sibling group had less vocal imitation. Notably, we found dissimilarity between the parental report and the direct observation. For instance, by the age 12 months, almost all of the parents reported that their children could imitate the hand waving (bye-bye) gesture at home, but in the directed observation setting, only a few children did. Also, in vocal imitation, all of the parents from both groups reported that their children could imitate vocal sound before 18 months, but only half of the children in the control and none in the sibling group displayed the skill in direct observation. The parental report and the direct observation had some distinctions: the directed observation setting is a novel situation for children, including an observer, and an unfamiliar place, and/or sounds. These factors would all seem to impact imitation of the children in both groups.

Our finding on the subject of vocal imitation revealed significant group differences at 18 months. Unlike the sibling group, vocal imitation of the typically developing children progressed rapidly between 9 and 18 months. At 9 months, no children from either group imitated the vocal sound of the experimenter. Later, the number of the children in the control group who could imitate vocal sound expanded rapidly from 12 to 18 months, while almost all of the children in sibling group still couldn't perform the vocal imitation. In fact, one child in the sibling group, who could imitate vocal sound at 12 months, displayed frustration and did not imitate vocal sound at 18 months. A recent study also found

that the impairment of the vocal imitation correctly at the age of 12 months reported by parent was recognized more in the high-risk than in the typically developing children[26]. The hypothesis about the weaker performance of vocal imitation in sibling group was that the sibling group might have some difficulty in auditory processing. From a neurobiological standpoint, children with ASDs are proposed to have a deficit in auditory processing which underpins language impairment[27]. Sharing with some genetic components, siblings of children with ASDs may also have weakness in auditory processing that makes them face difficulty in vocal imitation with unfamiliar sounds particularly in novel situations.

Regarding gesture imitation and object imitation, although many studies in ASDs revealed weaker performance in this type of imitation[12,15,16], group difference was not found between the sibling and control group in our study. However, when looking in detail at gesture imitation (hand waving), we found an interesting point: the control group appeared to benefit more from the sound cue "bye-bye" than the sibling group. A study of young typically developing children demonstrated that understanding of goal of action impacted on imitation in children[28,29]. When a gesture was demonstrated alone, children might not clearly understand the goal of the action, so only a few of them succeed in imitating it. Once the sound cue "bye-bye" was added, the goal of the action was clearer and enhanced the imitation in the control group. Unlike the sibling group, they could not get more meaning from the verbal cue. Many research studies revealed that siblings of children with ASD have less social skill proficiency[4]; they might not be interested in saying "bye-bye" in a social situation. As a result, the sibling group might gain benefit from the sound cue "bye-bye" to help them understand the goal, thus, enhancing the gesture imitation.

The study of Young et al found that different types of imitation varied in difficulty for children in sibling groups and also typically developing children: object imitation is easier than gesture imitation and vocal imitation, respectively[30]. We also found the same trend from the direct observation, children from both groups could perform object imitation earlier than gesture and vocal imitation. Furthermore, we also found that children in both groups gradually gained ability to imitate during the consecutive observation sessions. Easy items, such as object imitation, could be performed by most children from both groups at 9 months old. For more challenging items, such as gesture imitation, the sibling group appeared to get the ability later than the control group, but an improving trend was seen from 12 to 18 months. Consistent with the previous study, although the sibling who was later diagnosed as ASD or broader phenotype of ASD had weaker imitative skills when compared with typically developing children at the same age, the development trajectory of imitation in both groups were similar[30]. It is possible that the pattern of development of vocal imitation in the sibling group might not differ from the typically developing children, but it is only delayed to gain the ability. Thus, further follow-up study probably reveals the answer.

We recognize some limitations in this study. First, the sample size of both groups was small. Second, we didn't use an ADI-R and ADOS to evaluate the probands because in Thailand (up to our research study period), there was no validated ADI-R and ADOS to use in our culture. However, diagnoses of the probands were performed by experienced developmental and behavioral pediatricians. Lastly, definite diagnoses of ASD could not clearly made at 18 months, so follow-up is needed until the time when an exact diagnosis could be given. Nevertheless, this is a preliminary finding about imitation in siblings of children with ASD and might have value for further research.

What is already known on this topic?

Siblings of children with Autism Spectrum Disorder (ASDs) have limitations in many skills including imitation, which can be observed as early as age 1-2 years.

What this study adds?

Among various types of imitation, siblings of children with ASDs had some delays in vocal imitation skills at the age of 12 to 18 months.

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Potential conflicts of interest

None.

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การศึกษาติดตามระยะยาวทักษะการเลียนแบบในน้องของเด็กกลุ่มอาการออทิซึมเปรียบเทียบกับเด็กปกติ

อิสราภา ชื่นสุวรรณ, นิชรา เรืองดารากานนท์, ทัศนวัต สมบูรณ์ธรรม, ปิยวรรณ วัฒนสุนทรสกุล, ปราณี อมรชัยชาญ

ภูมิหลัง: *น้องของเด็กกลุ่มอาการออทิสซึมมีความเสี่ยงในการเกิดโรคมากกว่าเด็กปกติ เด็กกลุ่มอาการออทิซึมมีความผิดปกติของการเลียนแบบในทุกด้าน โดยพบความผิดปกติได้ตั้งแต่ในช่วงวัยเตาะแตะ ดังนั้นการศึกษาการเลียนแบบของน้องเด็กกลุ่มอาการออทิซึม ในช่วงวัยนี้อาจจะแสดงให้เห็นความผิดปกติได้*

วัตถุประสงค์: *เพื่อศึกษาพัฒนาการในการเลียนแบบในแต่ละด้านของน้องของเด็กกลุ่มอาการออทิซึม ตั้งแต่ช่วงอายุ 9 ถึง 18 เดือน เปรียบเทียบกับเด็กปกติ*

วัสดุและวิธีการ: *เป็นการศึกษาในระยะยาวเปรียบเทียบทักษะการเลียนแบบของน้องของเด็กกลุ่มอาการออทิซึมจำนวน 8 ราย กับเด็กปกติ 19 ราย โดยเก็บข้อมูลใน 2 ส่วน ได้แก่ แบบบันทึกพัฒนาการการเลียนแบบโดยผู้ปกครอง และการสังเกตพฤติกรรมการเลียนแบบโดยผู้วิจัยที่คลินิกเมื่ออายุ 9, 12 และ 18 เดือน โดยการเลียนแบบที่ศึกษามี 3 ด้าน ได้แก่ การเลียนแบบเสียง การเลียนแบบท่าทาง และการเลียนแบบการกระทำกับวัตถุ*

ผลการศึกษา: *พัฒนาการด้านการเลียนแบบเสียงของน้องเด็กกลุ่มอาการออทิซึมมีความล่าช้าในช่วงอายุ 12 ถึง 18 เดือนเมื่อเปรียบเทียบกับเด็กปกติ และพบความแตกต่างของพัฒนาการด้านดังกล่าวอย่างมีนัยสำคัญเมื่ออายุ 18 เดือน ส่วนการเลียนแบบท่าทางและการกระทำกับวัตถุไม่พบความแตกต่างกัน*

สรุป: *น้องของเด็กกลุ่มอาการออทิซึมมีความล่าช้าของพัฒนาการการเลียนแบบเสียงในช่วงวัยเตาะแตะ*