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博 士 論 文

Speech Intonation in Children with Autism Spectrum Disorder

（自閉症スペクトラム障害児における発話イントネーション）

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Title page

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Speech Intonation in Children with Autism Spectrum Disorder

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Abstract:

Objective: The prosody of children with autism spectrum disorder (ASD) has several abnormal features. We assessed the speech tone of children with ASD and of children with typical development (TD) by using a new quantitative acoustic analysis.

Methods: Our study participants consisted of 63 children (26 with ASD and 37 with TD). The participants were divided into 4 groups based on their developmental features and age. We assessed the variety of the fundamental frequency (F0) pattern quantitatively, using pitch coefficient of variation (CV), considering the different F0 mean for each word.

Results: (1) No significant difference was observed between the ASD and TD group at pre-school age. However, the TD group exhibited significantly greater pitch CV than the ASD group at school age. (2) In pitch CV, range and standard deviation of the whole speech of each participant, no significant differences were observed between the type of participants and age. (3) No significant correlation was found between the pitch CV of each word and the Japanese Autism Screening Questionnaire total score, or between the pitch CV of each word and the intelligence quotient levels in the ASD group. A significant correlation was observed between the pitch CV of each word and social reciprocal interaction score.

Conclusions: We assessed the speech tone of children with ASD by using a new quantitative method. Monotonous speech in school-aged children with ASD was detected. The extent of monotonous speech was related to the extent of social reciprocal interaction in children with ASD.

Key words:

autism spectrum disorder; intonation; acoustic analysis; pitch; coefficient of variation; ASQ

Introduction

Speech prosody refers to the qualitative aspects of speech, such as intonation, rhythm, and stress [1]. The prosody of children with autism spectrum disorder (ASD) has several abnormal features. In particular, speech intonation in ASD is monotonous, which has been used as a diagnostic feature of ASD [2, 3]. Speech intonation is expressed as a pitch pattern [1], using acoustic analysis, a pitch pattern can be described as a fundamental frequency (F0) pattern. The F0 is defined as the lowest frequency of a periodic waveform, and it is the physical quantity of a pitch. Therefore the variety of intonation can be interpreted as the variety of the F0 pattern.

In children with typical development (TD), the expressive abilities of falling tones have a different process pattern from those of rising tones. The range of falling tones of 4-year-old children is twice as wide as that of 1-year-old children with 4-year-old children being able to express words with falling tones as adults do [4, 5]. It is more difficult for young children to express words with rising tones [6]. Crystal [7] reported that children start to learn rising tones after acquiring the ability to use falling tones. There is no significant difference in the variety of rising tones between 1-year-old children and 4-year-old children [4], which supports the idea that children begin to learn rising tones after they have reached school age [8].

Monotonous speech in ASD was first described by Kanner, and has since been consistently observed in a large number of studies [9-11]; moreover, it is evident not only in childhood but also throughout adolescence and adulthood [10, 12, 13]. Monotonous speech is considered a core feature of ASD, yet may not be related to the extent of ASD [14]. However, all of the reports relating to monotonous speech in ASD to date were based on subjective observations. The reliability and validity of human auditory abilities are often lower than predicted [15]. Therefore, a method of quantitative assessments has been sought.

In recent years, a few studies have shown that monotonous speech in ASD is strongly linked to the impairment of speech prosody perception [13, 16]. For example, abnormal processing of auditory stimuli in ASD has been speculated to occur at the brainstem level [17], and abnormalities have been revealed by functional magnetic resonance imaging (fMRI) [18]. Furthermore, speech prosody perception in ASD is considered to be relevant to the development of the Theory of Mind [19]. However, monotonous speech in ASD has not been confirmed in any former quantitative studies that used either pitch range (the minimum F0 subtracted from maximum F0) or pitch standard deviation (SD) [20-22].

The objective of this study, therefore, was to clarify the differences in speech intonation between children with ASD and children with TD. We assessed the speech tone of children with ASD by using a new quantitative method. Additionally, we sought to measure F0 using acoustic analysis, and to examine how the variety of the F0 pattern of children with ASD and of children with TD changes between pre-school and school ages. Furthermore, we assessed the relationship between the variety of the F0 pattern and the extent of ASD symptoms.

Methods

1. Participants

ASD participants were recruited from among the 4- to 9-year-old children who visited the Developmental Behavioral Pediatric Clinic of Kobe University Hospital between April and July 2010, after approval from the Medical Ethics Committee of Kobe University Graduate School of Medicine was received. All of the participants had a clinical diagnosis of ASD based on DSM-IV-TR [2] criteria made by an expert child neurologist. Participants were examined routinely by the one-foot standing test, the finger-to-nose test, and the simple drawing test during their first visit. None of the participants revealed obvious neurological symptoms such as involuntary movements. Participants and their parents were provided with a complete written and oral explanation of the aims and protocols of the study prior to providing their informed consent. They were assured that all personal information would be protected, that non-participation in the study would disadvantage them in any way, and that participants or their parents could discontinue the participation in the study at any time. Participation in the study was limited to children who were able to understand simple instructions and who had obtained 30 or more words for analysis to ensure reliability.

The control or TD participants were recruited from among the children studying in the normal class of their preschools or primary schools, which were in the same region as the ASD participants. None had any history of speech, communication, learning problems, or any history of special education.

Our study participants consisted of a total of 63 children (26 with ASD and 37 with TD). We divided the participants into 4 groups based on their developmental features and age. The 4- to 6-year-old (pre-school-aged) ASD group consisted of 6 children, and the 7- to 9-year-old (school-aged) ASD group consisted of 20 children. The pre-school-aged TD group consisted of 16 children, and the school-aged TD group consisted of 21 children (Table 1).

2. Procedures

We conducted experimental tasks based on the word test proposed by the Japan Clinical Articulation Society [23]. In this test, 50 picture cards of animals and objects were displayed, and participants were instructed to verbally name each object or animal. After displaying all 50 picture cards, we took a 3-min break before repeating the exercise. When picture cards were displayed, the examiners asked the participants to name the object shown. Because the examiner did not say the name of the object, the participants were not able to directly imitate their speech. If the participants gave an incorrect response or did not answer, the examiners displayed the next picture card without providing them with the correct term. The participants were told the correct terms after all word tests had been completed. Incorrect answers were excluded from final data analysis.

The recording equipment consisted of a Lavalier Microphone (Sony, ECM-77B/9X), Audio Capture (ROLAND, EDIROL UA-25EX), and a Mobile Note PC (TOSHIBA, Dynabook SS

RX2/T7G). The microphone was fixed to the participant's collar during recording. Recording parameters included a 48 kHz sampling frequency and 24-bit quantization.

Parents of the participants were asked to complete the Japanese Autism Screening Questionnaire (ASQ) [24] while the word test was being conducted with their children. The ASQ is designed for early detection of autism and pervasive developmental disorders based on Autism Diagnostic Interview-Revised (ADI-R) [25]. We examined the correlation between the pitch CV and 3 domains based on ASQ: social reciprocal interaction (S), communication (C), repetitive behavior and stereotyped patterns (R). The Japanese version of the ASQ consists of 39 questions, and a score of ≥13 indicates a high possibility of ASD (sensitivity 0.86, specificity 0.64). The translator of the ASQ granted us permission to use the questionnaire in this study.

3. Acoustic analysis

Audio data for analysis consisted of the spoken words associated with the picture cards. Using sound editing software (free software, spwave version 0.8.2), the sampling frequency of the recorded data was converted to 24 kHz, and audio data was isolated for each word.

We assessed the variety of the F0 pattern quantitatively using pitch coefficient of variation (CV), which is the relative pitch dispersion, considering the different F0 mean for each word. We developed an original acoustic analysis program, based on existing software MATLAB (Version R2008b. Natick, MA, USA), and signal-processing tools (MathWorks. Signal Processing Toolbox) to calculate pitch CV of each word automatically. Fechner's law states that the level of the stimulus perceived by humans is proportional to the logarithm of the magnitude of physical stimuli; this is the basic principle of human psychology with regard to the perception of the senses [26]. In other words, the variety of the perceived F0 pattern is affected by the magnitude of the mean of F0, which is different for each word. Audio data was examined for each word (WAV file format), and the start and end time of the audio data was recorded (CSV file format). We obtained the mean of pitch CV based on the pitch for each

1/100 s of audio data using our new program. When the F0 value was 0, or the speed of change was greater than 50Hz, the value was excluded from our data set. The maximum, minimum, mean, SD, and variance of pitch were also calculated using the program.

Pitch CV was calculated in two different settings: each word and whole speech. The pitch CV of each word was obtained by calculating pitch CV, range and SD of each word first, then calculating the mean of pitch CV, rang and SD for each participant. The pitch CV of whole speech was to calculate the mean of pitch CV, rang and SD for each participant without isolation by each word.

Statistical analysis was performed using a software package (SPSS Statistics 19, Chicago, IL, USA). To verify the reliability of the variety of the F0 pattern, we used a 1-factor analysis of variance between the first and second word test, in each of the 6 indexes for the ASD and TD group. The difference between the boys and girls was assessed by 1-factor analysis of variance. A 2-factor analysis of variance was used to evaluate the changes by age in the ASD and TD group for the 6 indexes. Additionally, we examined the correlation between the pitch CV of each word and the ASQ score in both the ASD and TD group, and between the pitch CV of each word and the intelligence quotient (IQ) in the ASD group, to assess the relationship between the variety of the F0 pattern and the extent of ASD or cognitive ability.

Results

1. Variety of the F0 pattern

No significant difference was observed between the number of words in the ASD and TD group (mean of the ASD group: 88.2, mean of the TD group: 85.2, $p = 0.46$). We performed a 1-factor analysis of variance between the first and second word test for each of the 6 indexes in the ASD and TD group. No significant differences were observed among them (Table 2). No significant differences were observed between the boys and girls in any of the 6 indexes for the ASD and TD group (Table 3).

2. Variety of the F0 pattern and age

A 2-factor analysis of variance was performed between the ASD and TD group (type of participants) at pre-school aged and school-age for each of the 6 indexes (Table 4).

(1) Each word

For pitch CV of each word in each participant, a significant interaction between the type of participants and age was observed (F (1, 59) = 4.13, p < 0.05, Fig. 1). A significant main effect of the type of participants was found (F (1, 59) = 6.13, p < 0.05). From a simple main effect test, no significant difference was observed between the ASD and TD group at pre-school age, but the TD group was found to exhibit significantly greater pitch CV than the ASD group at school age (pre-school: F (1, 59) = 0.10, *n. s.*, school age: F (1, 59) = 10.16, p < 0.01). A significant main effect of age was observed (F (1, 59) = 7.40, p < 0.01). From a simple main effect test, school-aged participants exhibited significantly greater pitch CV than the pre-school-aged participants in the TD group; notably, no significant difference was observed between pre-school-aged participants and school-aged participants in the ASD group (TD group: F (1, 59) = 11.29, p < 0.01, ASD group: F (1, 59) =0 .24, *n. s.*).

In pitch range and SD, no significant interaction between the type of participants and age was observed (pitch range: F (1, 59) =0.10, *n. s.*, pitch SD: F (1, 59) = 0.10, *n. s.*). Moreover, no significant main effect of the type of participants or age was observed (pitch range: F (1, 59) = 2.50, *n. s.*, F (1, 59) = 3.68, *n. s.*, pitch SD: F (1, 59) = 1.01, *n. s.*, F (1, 59) = 2.18, *n. s.*).

(2) Whole speech

In pitch CV, range and SD of the whole speech of each participant, no significant interaction between the type of participants and age was observed (pitch CV: F (1, 59) = 1.17, *n. s.*, pitch range: F (1, 59) = 0.66, *n. s.*, pitch SD: F (1, 59) = 0.06, *n. s.*). Moreover, no significant main effect of the type of participants or their age was observed (pitch CV: F (1, 59) = 3.48, *n. s.*, F (1, 59) = 2.60, *n. s.*, pitch range: F (1, 59) = 0.36, *n. s.*, F (1, 59) = 3.85, *n. s.*, pitch SD: F (1, 59) =

0.23, *n. s.*, *F* (1, 59) = 0.76, *n. s.*).

3. The relationship between the variety of F0 pattern and the other factors

We examined the correlation between the pitch CV of each word and the ASQ scores in both the ASD and TD group (Fig. 2 A). In the TD group, no correlation was observed between the pitch CV of each word and the ASQ scores (S score: r = 0.07, C score: r = -0.14, R score: r = 0.19 total score: r = - 0.01). In the ASD group, a negative correlation was observed between the pitch CV of each word and S score (r= -0.62), however, no significant correlation was observed between those and the other two scores (C score: r = 0.29, R score: r = 0.28, total score: r = 0.12, Fig. 2 B). No significant correlation was found between the pitch CV of each word and the IQ levels in the ASD group (*r* =0 .02).

15 participants of the ASD group showed instability of balance, and 10 of them revealed fine motor clumsiness compared with their age. Furthermore, 6 participants of the ASD group had fine motor training by occupational therapists. However, there were no children who showed major abnormal movements. There was no significant relationship between the pitch CV of each word and the existence of clumsiness.

Discussion

1. Assessment of monotonous speech in ASD using a new quantitative method

It is well known that the intonation of children with ASD has abnormal features. We completed a quantitative assessment using acoustic analysis of pitch CV on each word of children with ASD in this study. Recently, quantitative studies concerning monotonous speech in ASD also employed acoustic analysis. Bonneh et al. [20] reported that children with ASD had significantly greater values for pitch range and SD of whole speech in each participant than did children with TD. They examined the whole sentences of speech spoken by each participant rather than separating out each word. Their results somewhat differed from the findings of many clinical observational studies [9-12]. Their quantitative methods of analyzing whole speech in each participant had some fundamental problems. When the acoustic data is analyzed without separating out each word, the obtained value is influenced by the kind of word. As mentioned previously, according to Fechner's law, the perceived variety of the F0 pattern is affected by the magnitude of the mean of F0 however, the magnitude is different for each word. Therefore, we adopted the "pitch CV" of each word to examine the variety of the F0 pattern in this study. We used the pitch CV of each word as an assessment tool to estimate the variety of the F0 pattern.

2. The difference in speech intonation between children with ASD and TD

A CV refers to a statistical measure of the distribution of data points in a data series around the mean. It represents the ratio of the standard deviation to the mean. A CV is a helpful statistic in comparing the degree of variation from one data series to the other, although the means are considerably different from each other. The pitch CV of each word of children with TD had significantly more value than that of children with ASD at school age, although there were no significant differences at pre-school age. Our results therefore suggest that the children gradually develop expressive abilities, reflected by the variety of the F0 pattern, after they reach school age, but that children with ASD maintain a monotonous speech pattern even after reaching school age.

3. The relationship between monotonous speech and the extent of ASD

In our study, a negative correlation was observed between the pitch CV of each word and the S score of ASQ in the ASD group. Paul et al. [27] reported that the abnormal prosody in ASD was related to the social reciprocal interaction impairment. Other studies have also shown the relationship between the prosody and the empathy in ASD [18, 19]. Our result supported by these previous reports. Tager-Flusberg [14] emphasized the importance of monotonous speech as a core feature of ASD. Our results also suggest that monotonous speech might be an important factor for understanding the development of brain mechanism in ASD children.

4. Limitations and perspectives

There are a number of limitations in our study. It is known that children with ASD often experience deficits in motor skills development, which often manifests as abnormal clumsiness. The Coordination of many muscles in the oral cavity, tongue, pharynx, larynx and so on, are required for speech. There is a possibility that abnormal prosody in ASD is derived from the poor coordination of the muscles related to speech. In this study, we could not find the relationship between monotonous speech and fine motor coordination by using routine soft neurological observational test. In future studies, we should examine more detail using more structured tests such as Movement Assessment Battery for Children - Second Edition [28] and the Developmental Coordination Disorder Questionnaire [29, 30].

As our study inclusion criteria limited the number of enrolled participants, the number of pre-school-aged children with ASD was smaller than that of the other groups. Furthermore, the ratio of male to female participants was different among the 4 groups, although this difference was not significant. We compared the pitch CV changes between pre-school and school aged children not by age. A further study with more participants is required to clarify the developmental changes of speech prosody by age.

While we assessed intonation by using pitch CV in this study, speech prosody consists of not only intonation but also rhythm and stress. We are currently trying to develop a new acoustic analysis system, which is capable of assessing these factors. Such a system would allow a more detailed analysis of prosody.

We assessed the speech tone of children with ASD by using a new quantitative method. Monotonous speech in school-aged children with ASD was detected. However, the extent of monotonous speech was not related to the extent of communication, repetitive behavior and stereotyped patterns, or to the level of cognitive function, it was related to the extent of social reciprocal interaction in children with ASD.

Acknowledgements

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Figure legends

Fig. 1. For pitch CV of each word in each participant, a significant interaction between the type of participants and age was observed (F (1, 59) = 4.13, $p < 0.05$)

Fig. 2. We examined the correlation between the pitch CV of each word and the ASQ scores in both the ASD and TD group (Fig. 2 A). In the ASD group, a negative correlation was observed between the pitch CV of each word and S score (r= -0.62), however, no significant correlation was observed between those and the other two scores (C score: r = 0.29, R score: r = 0.28, total score: r = 0.12, Fig. 2 B).

Fig. 1

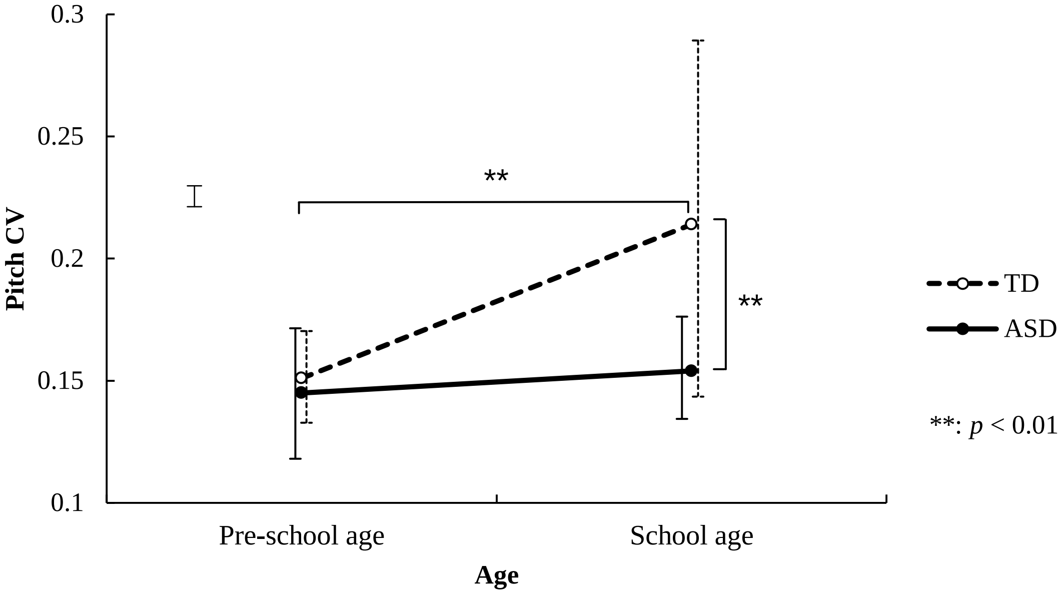
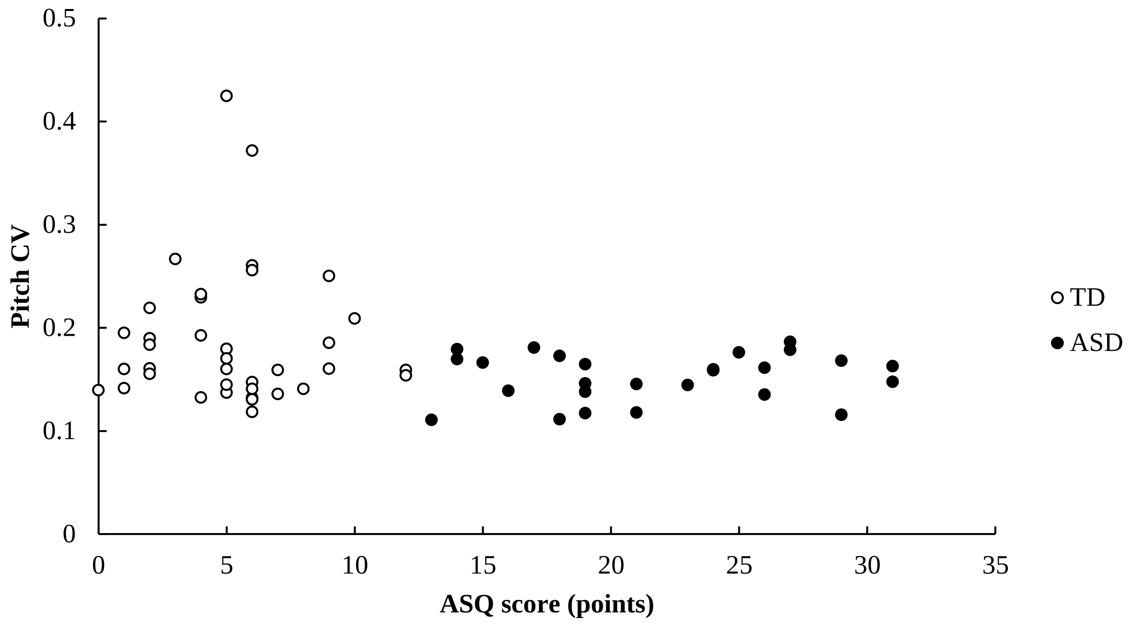
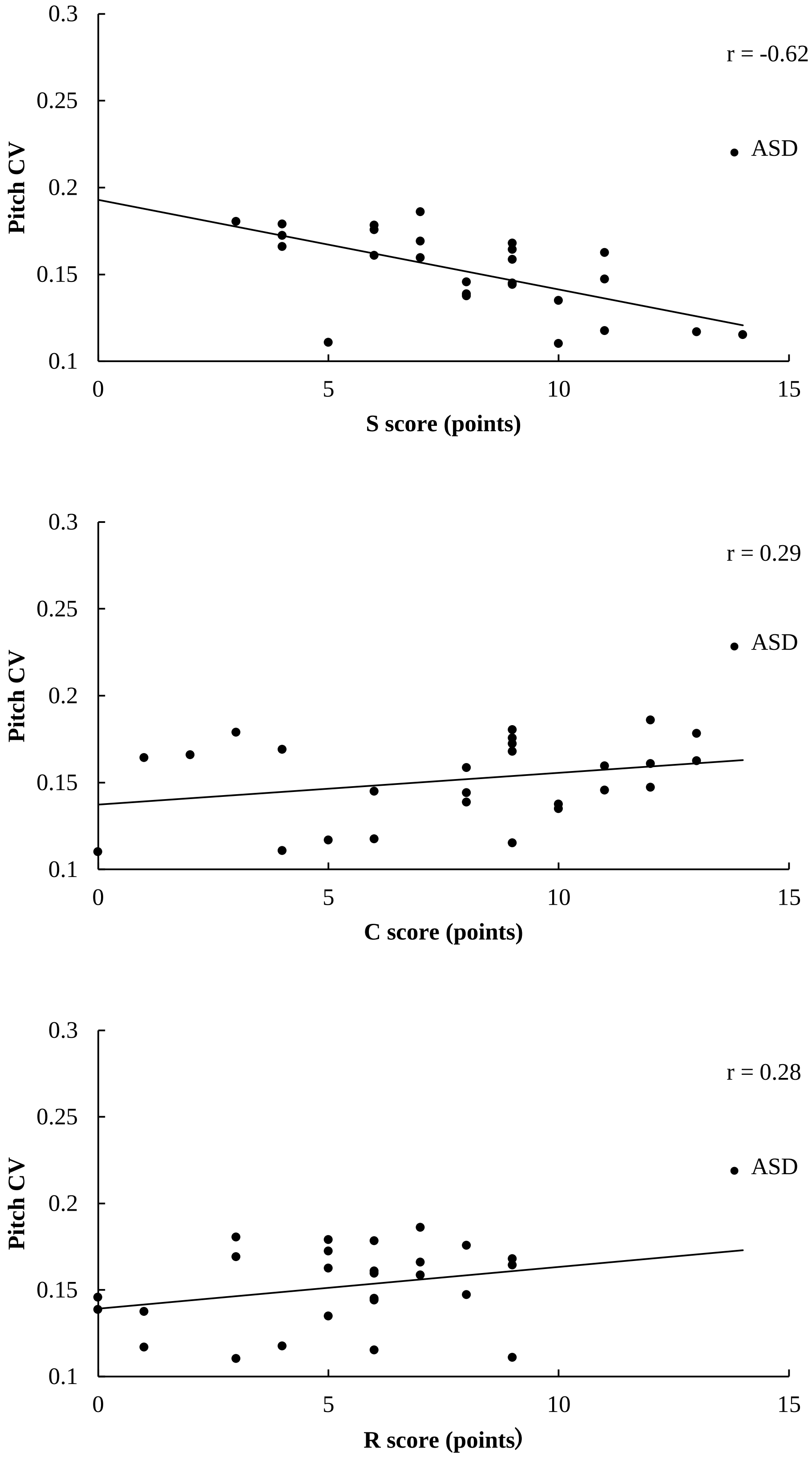

0.3
0.25
0.2
0.15
0.1
Pitch CV
**
**
TD
ASD
**: p < 0.01
Pre-school age
School age
Age

Fig. 2 A



0.5
0.4
0.3
0.2
0.1
0
Pitch CV
0
5
10
15
20
25
30
35
ASQ score (points)
TD
ASD

Fig. 2 B


r = -0.62
ASD
Pitch CV
0.3
0.25
0.2
0.15
0.1
0
5
10
15
S score (points)
r = 0.29
ASD
Pitch CV
0.3
0.25
0.2
0.15
0.1
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5
10
15
C score (points)
r = 0.28
ASD
Pitch CV
0.3
0.25
0.2
0.15
0.1
0
5
10
15
R score (points)

Table 1. Descriptive characteristics of the participants.

		ASD group		TD group	
Age		Pre-school	School	Pre-school	School
N		6	20	16	21
Chronological age	Mean±SD	5:8±0:6	7:9±0:7	5:11±0:8	7:9±0:10
	Range	4:10-6:3	7:1-9:1	4:10-6:5	6:10-9:1
Gender	Male	5	15	10	10
	Female	1	5	6	11
Intelligence quotient	Mean±SD	67.7±17.6	69.8±16.9	-	-
	Range	51-93	52-121	-	-
ASQ	Mean±SD	18.8±6.3	22.6±5.0	5.1±2.8	5.3±3.2
	Range	13-31	14-31	0-12	1-12

Table 2. Variety of the F0 pattern in the first time and the second time.

	ASD group		TD group	
Time	First	Second	First	Second
Each word				
Pitch CV	0.16±.02	0.16±.04	0.18±.06	0.19±.07
Pitch range	200.81±40.20	197.10±39.12	206.49±39.06	212.11±46.56
Pitch SD	50.35±12.07	48.77±15.75	51.14±15.13	53.08±17.78
Whole speech				
Pitch CV	0.20±.03	0.20±.05	0.24±.08	0.24±.09
Pitch range	481.88±30.29	480.87±20.23	471.08±30.51	474.57±31.40
Pitch SD	63.46±17.02	61.00±19.02	63.73±19.74	64.19±22.00

(mean±SD)

Table 3.Variety of the F0 pattern in the boys and the girls.

	ASD group		TD group	
Gender	Boys	Girls	Boys	Girls
Each word				
Pitch CV	0.15±.02	0.16±.02	0.20±.08	0.18±.05
Pitch range	195.32±36.51	205.91±28.45	212.56±52.80	213.64±34.40
Pitch SD	49.02±13.00	49.26±10.70	54.58±20.45	50.38±10.90
Whole speech				
Pitch CV	0.20±.04	0.21±.04	0.26±.10	0.22±.06
Pitch range	486.72±33.36	493.30±14.32	482.65±32.16	495.34±25.96
Pitch SD	63.82±17.55	60.72±13.30	68.10±23.95	61.79±13.03

(mean±SD)

Table 4. Variety of the F0 pattern and age.

	ASD group		TD group	
Age	Pre-school	School	Pre-school	School
Each word		** (School vs TD School)	** (TD Pre-school vs TD School)	
Pitch CV	0.15±.03	0.15±.02	0.15±.02	0.21±.07
Pitch range	183.21±33.90	202.13±34.27	198.18±36.23	224.39±48.13
Pitch SD	45.14±12.20	50.26±12.32	48.19±13.25	56.05±18.48
Whole speech				
Pitch CV	0.20±.04	0.21±.04	0.21±.06	0.27±.10
Pitch range	481.26±33.31	490.33±28.85	479.55±25.01	501.48±23.70
Pitch SD	60.48±18.67	63.89±16.00	61.73±17.09	67.84±21.50

(mean±SD) ** : $p < 0.01$