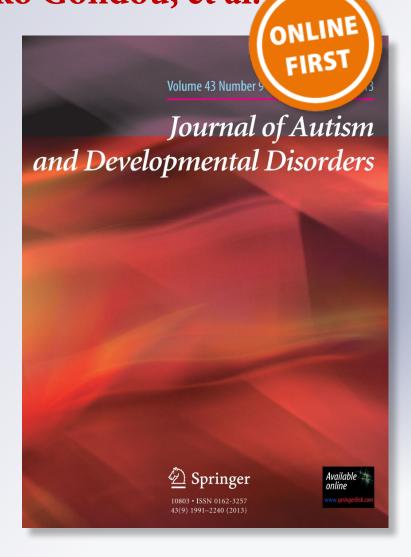
## Quantitative Aspects of Communicative Impairment Ascertained in a Large National Survey of Japanese Children

Manabu Oi, Hiroshi Fujino, Naotake Tsukidate, Yoko Kamio, Yuko Yoshimura, Mitsuru Kikuchi, Chiaki Hasegawa, Keiko Gondou, et al.

Journal of Autism and Developmental Disorders

ISSN 0162-3257

J Autism Dev Disord DOI 10.1007/s10803-017-3226-x





Your article is protected by copyright and all rights are held exclusively by Springer Science+Business Media, LLC. This e-offprint is for personal use only and shall not be selfarchived in electronic repositories. If you wish to self-archive your article, please use the accepted manuscript version for posting on your own website. You may further deposit the accepted manuscript version in any repository, provided it is only made publicly available 12 months after official publication or later and provided acknowledgement is given to the original source of publication and a link is inserted to the published article on Springer's website. The link must be accompanied by the following text: "The final publication is available at link.springer.com".



#### ORIGINAL PAPER



# **Quantitative Aspects of Communicative Impairment Ascertained** in a Large National Survey of Japanese Children

Manabu Oi¹ · Hiroshi Fujino² · Naotake Tsukidate³ · Yoko Kamio⁴ · Yuko Yoshimura⁵ · Mitsuru Kikuchi⁵ · Chiaki Hasegawa⁵ · Keiko Gondou⁶ · Tomoko Matsui⁵

© Springer Science+Business Media, LLC 2017

Abstract The Japanese version of the Children's Communication Checklist-2 (CCC-2) was rated by caregivers in a large national population sample of 22,871 children aged 3–15 years. The General Communication Composite (GCC) of the CCC-2 exhibited a distribution with a single-factor structure. The GCC distribution between autism spectrum disorders (ASD) and language impairment (LI) groups in the general population fit inside a bell curve with significant overlap with the general population, and a continuum was evident between groups. No evidence of a natural cutoff that would differentiate categorically affected from unaffected children was seen. The Social Interaction Deviance Composite (SIDC) supported the notion that

ASD and LI are on the opposite endpoints of a SIDC continuum of communication impairment.

**Keywords** Autism · Language impairment · Communication · Checklist

### Introduction

Whether aspects of communicative impairment are continuously distributed between typically developing (TD) children and those with developmental disorders remains unclear. However, except for Bishop and Norbury (2002), few studies have directly addressed the question of whether communicative impairments in otherwise healthy children are better conceptualized as a dimensional or a categorical phenomenon. Weismer (2007) showed a language endowment spectrum between children with specific language impairment (SLI), late talkers, and typical talkers. Henrichs et al. (2011) investigated the continuity and discontinuity of vocabulary skills in a population-based cohort focusing on the relationships between late talkers who developed normal vocabulary skills and those with persistent delay. Dollaghan (2011) showed that children with SLI are at the lower end of a continuous distribution of language skills rather than being part of a qualitatively distinct group. In addition, Kalnak et al. (2012) reported the existence of a broad phenotype of SLI in families who have children with SLI. These findings suggest that in studies examining structural aspects of language such as lexicon, syntax, and speech (Bishop 2014), whether aspects of communicative impairment are continuously distributed also needs to be investigated. According to Crystal (1987), linguistic communication has two aspects, language structure and language use, which are linked by pragmatics. Regarding

- Manabu Oi oimanabu@ed.kanazawa-u.ac.jp
- United Graduate School of Child Development, Kanazawa University, Bldg D, 13-1 Takaramachi, Kanazawa, Ishikawa 920-8640, Japan
- Faculty of Education, Tokyo Gakugei University, 4-1-1 Nukuikitamachi, Koganei, Tokyo 184-0015, Japan
- <sup>3</sup> Department of Humanities, Yamanshi Eiwa College, 888 Yokonemachi, Kofu, Yamanashi 400-8555, Japan
- Department of Child and Adolescent Mental Health, National Institute of Mental Health, National Center of Neurology and Psychiatry, 4-1-1 Ogawahigashimachi, Kodaira, Tokyo 187-8551, Japan
- Research Center for Child Mental Development, 13-1 Takaramachi, Kanazawa, Ishikawa 920-8640, Japan
- Department of Child Studies, Faculty of Home Economics, Kyoritsu Women's University, 2-2-1 Hitotsubashi, Chiyoda, Tokyo 101-8437, Japan
- Center for Research in International Education, Tokyo Gakugei University, 4-1-1 Nukuikitamachi, Koganei, Tokyo 184-0015, Japan

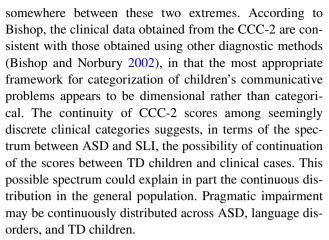
Published online: 10 July 2017



pragmatic aspects of language such as speech acts, conversational maxims, and implicature, in recent research on autism spectrum disorder (ASD), continuity between normal and clinical samples has been suggested. Pragmatic communication impairment is a core symptom of ASD, and has been seen as a dimensional rather than a categorical entity since epidemiological research by Wing and Gould (1979). Kim et al. (2011) reported a high prevalence of categorically defined ASD in a total population sample, finding a continuous distribution of symptoms throughout the population. In addition, quantitative autistic traits have been ascertained in a national survey of Japanese schoolchildren (Kamio et al. 2013), where the Japanese version of the Social Responsive Scale was used (SRS; Constantino and Gruber 2005). The SRS contains subscales to test for nonverbal communication problems and social impairments. Pragmatic impairment may represent a quantitative autistic trait that is continuously distributed between TD children and children with ASD. This possibility is consistent with the results from pragmatic impairment research by Perkins (2007), in which he insists that pragmatic impairment in general would be continuous rather than discontinuous, as it is considered an emergent property resulting from interactions between linguistic, cognitive, and social factors.

On the other hand, Perkins (2007) also insists that pragmatic impairment is caused by the communicator's compensatory adaptation to their brain disorder in cases other than those involving ASD, such as SLI and aphasia. In such cases, structural aspects of language are considered primarily impaired, while pragmatic language impairment (PLI) is regarded as secondary or collateral impairment. Bishop (2003) revised the original Children's Communication Checklist (CCC) into the CCC-2 and found that the parent ratings tended to be substantially lower for SLI groups than for TD groups on the pragmatic composite. It is probable that both structural and pragmatic aspects of language are closely intertwined in children with communicative impairment.

In addition to the suggested continuity between TD children and clinical cases in terms of communicative impairment, continuity is also suggested in clinical cases such as SLI, PLI, and ASD. Regarding structural aspects of language such as lexicon, syntax, and speech, overlapping impairment between ASD and SLI has been suggested (Bishop 2010; Boucher 2012; Leyfer et al. 2008; Loucas et al. 2008; McGregor et al. 2012; Tomblin 2011). In establishing the CCC-2, Bishop (2003) indicated that research at that time supported a more dimensional view toward PLI in SLI, PLI, and ASD cases. She claimed that, through this view, one might observe an entire spectrum of impairments with typical SLI at one end and core autism at the other, with many children having patterns of impairment



Therefore, to propose an epidemiologic framework for interpreting the diversity of communicative impairments seen in children, the present study aimed to determine whether aspects of communicative impairment are continuously distributed in a population-based sample.

#### Methods

## **Participants**

The participants comprised a normative sample (N=22,871) of children ranging in age from 3 to 15 years, children with autism spectrum disorder (n=48), children with language impairment (LI; n=30), and TD children (n=64) (Tables 1, 2). The TD sample was needed for principal component analysis (PCA) and analysis in terms of the relationship between

**Table 1** Frequency distribution of respondents by age and sex on the Children's Communication Checklist-2 (CCC-2)

Sex					
Male Age (years)	Female				
	$\overline{n}$	%	n	%	
3	60	0.5	62	0.5	
4	123	1.1	137	1.2	
5	149	1.3	150	1.3	
6	270	2.3	261	2.3	
7	1590	13.8	1456	12.8	
8	1470	12.7	1407	12.4	
9	1366	11.8	1432	12.6	
10	1358	11.8	1338	11.8	
11	1360	11.8	1254	11.1	
12	1168	10.1	1211	10.7	
13	1023	8.9	1009	8.9	
14	916	7.9	999	8.8	
15	677	5.9	625	5.5	
Total	11,530	100.0	11,341	100.0	



Table 2 Demographics of the clinical sample and the TD group

	ASD group	LI group	TD group
N	48	30	64
Boys	37	24	44
Girls	11	6	20
Mean age in years (SD)	6.10 (1.60)	9.06 (1.53)	6.37 (1.60)
Mean K-ABC mental processing scale score (SD)	95.27 (20.92)	- (-)	104.23 (10.27)
Mean WISC-III FIQ (SD)	- (-)	89.73 (9.22)	- (-)
SCQ (SD)	- (-)	5.10 (3.17)	2.00 (2.00)

ASD autism spectrum disorder, LI language impairment, TD typical development, K-ABC Kaufman Assessment Battery for Children, SCQ Social Communication Quotient

CCC-2 score and cognitive development. Although some preliminary studies have been conducted on Japanese SLI (Fukuda and Fukuda 2001; Ito et al. 2009), it is not yet established as a diagnostic category in Japan, as no standardized test for grammatical development in Japanese is available. Thus, in the present study, we use the term LI instead of SLI. All assessments were made using the Japanese version of the CCC-2 (Bishop 2003; Tsukidate et al. 2015). Regarding the normative sample, questionnaires were distributed by mail to the caregivers of all children attending nursery schools, kindergartens, primary schools, and secondary schools that sought to be included in the survey in the 13 geographical areas of Japan in 2010 (n=91,196). Nursery schools and kindergartens were all local institutions attended by more than 92% of the children living in the community, according to the Japan Cabinet Secretary (2010), and all schools were community schools attended by more than 93% of the children living in the community, according to the annual report of the Ministry of Education, Culture, Sports, Science and Technology (2010). Questionnaires were returned for 26,586 children from 26 nursery schools, 17 kindergartens, 187 primary schools, and 71 secondary schools (response rate, 29.15%). Questionnaires with missing answers were excluded, leaving a total of 24,263 participants (12,330 boys, 11,933 girls), with CCC-2 data provided by mothers (n=22,072), fathers (n=1780), both parents (n=173), other caregivers (n=144), or unspecified (n=94). In addition, to ensure that all analyses were based on a complete data set, questionnaires involving children with hearing impairment or unknown age were excluded, as were those that failed to clear a consistency check (Bishop 2003), leaving a final normative sample of 22,871 participants (11,530 boys, 11,341 girls). Each of the 13 age levels comprised a minimum of 60 participants of each sex; each sex was proportionally represented (Table 1). The number of participants under 6 years of age was much smaller than the number of those over 7 years of age; this was due to the fact that kindergartens and nursery schools are much smaller in size than primary and secondary schools. The clinical sample consisted of 48 children diagnosed with ASD (ASD group) and 30 children diagnosed with LI (LI group), as shown in Table 2. Children in the ASD group (37 boys, 11 girls) ranged in age from 3.33 to 9.25 years. The diagnosis of ASD was made by a psychiatrist and a clinical speech therapist using American Psychiatric Association's Diagnostic and Statistical Manual of Mental Disorders IV-TR (DSM-IV-TR) criteria. The speech therapist, who has more than 5 years of experience in ASD treatment and is well trained and certified in assessment using the Autism Diagnostic Observation Schedule (ADOS), employed the Autism Diagnostic Observation Schedule-Generic (ADOS-G; Lord et al. 1999). The psychiatrist and the speech therapist were both blinded to the study purpose. The definitive diagnosis of ASD was made by the psychiatrist, who has more than 10 years of experience in ASD, using the Diagnostic Interview for Social and Communication Disorders (DISCO; Wing et al. 2002) at the time of data acquisition using the Kaufman Assessment Battery for Children (K-ABC). Twenty-four children satisfied the diagnoses of autism and another 24 satisfied the diagnosis of autism spectrum in accordance with the ADOS-G. K-ABC mental processing scale scores in the ASD group ranged from 58 to 144. Children in the LI group were diagnosed by their school system as having difficulties in speaking and listening, but no intellectual disabilities. Individual full-scale Intelligence Quotient (IQ) was obtained for children with LI using Wechsler's Intelligence Scale for Children III (WISC-III). Full-scale IQ ranged from 74 to 108. LI group children all attended a language unit in their school system. They were evaluated as having less than 15 points on the Japanese version of the Social Communication Questionnaire (SCQ; Rutter et al. 2003). Their SCQ scores ranged from 0 to 11. They were rated as problematic or disabled according to the Learning Disabilities Inventory-Revised (LDI-R; Ueno et al. 2008) based on responses to items related to speaking and listening. The children (24 boys, 6 girls) ranged in age from 6.58 to 12.00 years. In addition, 64 TD children participated. These children (44 boys, 20 girls) ranged in age from 3.17 to 10.17 years. Their K-ABC mental processing scale scores ranged from 86 to 130. Their SCQ scores ranged from 0 to 10.

#### Measures

## The Children's Communication Checklist-2 (CCC-2)

The CCC-2 (Bishop 2003) is a 70-item questionnaire regarding communicative impairment. It is intended for use with 4–16-year-olds, and can be completed in 15 min by an adult who has observed the child over time in



natural social settings. The CCC-2 has ten subscales, with each subscale comprising seven items. Based on CCC-2 scaled scores, two parameters were devised by Bishop to identify communicative impairments and to indicate the need for more precise assessment in regard to ASD. One of these parameters is the General Communication Composite (GCC), which is the sum of scaled scores for "speech," "syntax," "semantics," "coherence," "inappropriate initiation," "stereotyped speech," "use of context," and "nonverbal communication". The other is the Social Interaction Deviance Composite (SIDC), which is the difference of the sum of "speech," "syntax," "semantics," and "coherence" scaled scores from the sum of "inappropriate initiation," "nonverbal communication," "social relations," and "interests" scaled scores. The CCC-2 was developed to provide a general measure for communicative impairments and to identify pragmatic/social interaction deficits, and has been validated for use in clinical child populations in UK samples (Norbury et al. 2004). Norbury et al. demonstrated that the CCC-2 provides a useful screening measure for communication impairment and can be helpful in identifying children who should be referred for more detailed assessment of possible ASD. However, their data highlighted substantial overlap between groups with "distinct" diagnoses such as SLI, PLI, and ASD. The CCC-2 can be used as a tool to detect broader autism phenotypes (Bishop et al. 2006).

We have composed a Japanese version of the CCC-2 with some modifications on items in which linguistic or cultural differences between the UK and Japan should be taken into account (Oi et al. 2016). Back-translation and verification procedures were conducted for the Japanese version. This modified version has demonstrated internal consistency for Japanese children (Cronbach's  $\alpha = 0.533$ to 0.761) (Tsukidate et al. 2015). Cronbach's  $\alpha$  is lower than the original UK version ( $\alpha$ =0.661 to 0.804) when the CCC-2 is translated into languages other than Norwegian (Helland et al. 2009), including Dutch (Geurts and Embrechts 2008), Serbian (Glumbić and Brojčin 2012), and Québec French (Vézina et al. 2013). The Japanese version was used in this study for children aged 3-15 years. Higher scaled scores on the CCC-2 indicate a lower degree of communicative impairment. The 70 CCC-2 items were categorized into the following ten subscales: speech, syntax, semantics, coherence, inappropriate initiation, stereotyped language, use of context, nonverbal communication, social interaction, and interests. Regarding the standardization of the CCC-2, the raw score was converted to the standard score with a mean of 10 and a standard deviation (SD) of 3 for each subscale. This procedure is based on that outlined in the original CCC-2 manual (Bishop 2003).



## **Data Analysis**

Based on CCC-2 data, and referring to the algorithm proposed by Bishop (2003), we examined continuity in the GCC and SIDC through a comparison between the ASD, LI, and TD groups.

The first step of the analysis was factor analysis, which was performed on children in the ASD, LI, and TD groups using PCA with data from the GCC subscales. In the second step, the most parsimonious model was examined by confirmatory factor analysis (CFA) in the normative sample using data from the GCC subscales. Spearman's correlation coefficients were computed to examine associations between K-ABC mental processing scale scores and GCC in the ASD and TD groups, respectively. Spearman's correlation coefficient was also computed to examine the association between WISC-III FIQ and GCC in the LI group. In addition, a receiver operating characteristic (ROC) analysis was conducted for GCC to determine the cutoff point, where the sum of sensitivity and specificity was the largest. ROC analysis was also conducted for the SIDC to determine the cutoff point that discriminates ASD from LI between clinical groups, excluding the TD group. Analysis was performed using SPSS Statistics 22.

## **Ethical Approval**

The current study was approved by the medical research ethics committee at Kanazawa University and performed in accordance with the ethical standards laid down in the 1964 Declaration of Helsinki and its later amendments. The study protocol was also approved by the Ethics Committee of the National Center of Neurology and Psychiatry, Japan. Informed consent was obtained from the parents of the participant children before the study began.

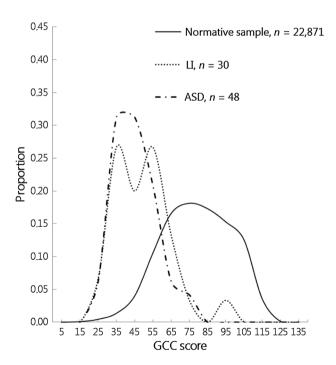
## **Results**

## **Population Distribution**

Sex differences in terms of raw scores on the ten subscales of the CCC-2 were found (Tsukidate et al. 2015). Accordingly, the Japanese version of the CCC-2 was standardized for the boy and girl subsamples across the entire age range (Oi et al. 2016). The GCC distribution among 3–15-year-old children in the Japanese general population is shown in Fig. 1.

#### **Factor Structure**

We confirmed that the GCC subscales were compiled into a single factor from the viewpoint of a dimensionality reduction. PCA suggested a single-factor solution for 142 children comprising the clinical and TD groups (Table 2). The first factor explained 64.736% of the variance (Table 3). These results suggested the single-factor model, which was then subjected to CFA using data from the normative sample. In fact, the single-factor model suggested by dimensionality reduction with PCA was replicated by a model with CFA: a single construct in GCC affected the observed subscale scores. As a result, factor loadings ranged from 0.67 to 0.83, and all estimated values were significant at the 5% level (Fig. 2). The goodness-of-fit index (GFI), the adjusted goodnessof-fit index (AGFI), the root mean square error of approximation (RMSEA), the comparative fit index (CFI), and the standardized root mean square residual (SRMR) were 0.971, 0.948, 0.077, 0.973, and 0.026, respectively, for the 56 items constituting the GCC. Furthermore, the  $\chi^2$ associated with the model was significant:  $\chi^2$  (22,871, df = 20) = 2707.187, p < .001. This suggests that  $\chi^2$  values are inflated with a very large sample size, not that the model is inconsistent with the observed data. Although the RMSEA indicated a mediocre fit (p < .08; MacCallum et al. 1996), the GCC single construct indicated an acceptable fit. The finding lends support to the notion of a unitary factor influencing multiple aspects of communicative impairment in children in the general population.



**Fig. 1** Distribution of Childrens Checklist-2 (CCC-2) General Communication Composite (GCC) standard scores in a normative sample

**Table 3** Principal component analysis of CCC-2 data

Component	Eigenvalue	% of variance	Cumulative %
1	5.179	64.736	64.736
2	0.742	9.275	74.012
3	0.461	5.764	79.775
4	0.443	5.538	85.314
5	0.389	4.864	90.177

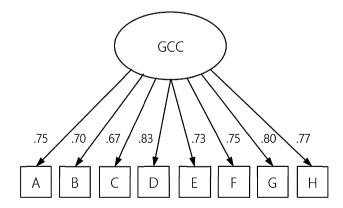
The clinical sample consisted of participants with ASD (n=48) and LI (n=30)

ASD autism spectrum disorder, TD typical development

### **Other Psychometric Properties**

The GCC scores of the ASD (mean = 49.65; SD = 11.683) and LI (mean = 53.23; SD = 15.106) groups were lower than that of the TD group (mean = 76.53; SD = 17.183). One-way analysis of variance (ANOVA) indicated differences between the groups (F=50.766, df=2, p<.001). Multiple comparison by Scheffe's test showed that GCC scores were lower in the ASD (p < .001) and LI groups (p < .001) than in the TD group. As shown in Fig. 1, the GCC scores of both the ASD and LI groups were distributed widely and significantly overlapped the general population distribution. Table 4 shows the GCC cutoffs by sex for the 1st, 5th, and 10th percentile values for the normative sample and the proportion of children diagnosed with ASD or LI who fell within the respective cutoffs. About 50% of the children in the LI group and about 70% of the children in the ASD group did not reach the 10th percentile.

One-way ANOVA revealed a significant difference (F=15.137, df=2, p<.001) between the mean SIDC scores for the ASD, (mean = -4.40; SD=7.709), LI (mean = -7.36; SD=10.18), and TD groups (mean = 1.75;



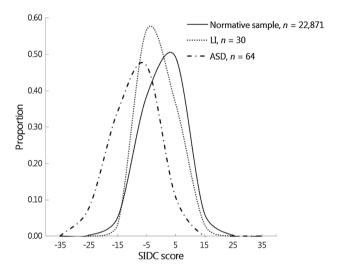
**Fig. 2** General Communication Composite (GCC) subscales for the single-factor model. Latent construct is shown in ellipses and observed variables are shown in rectangles. A speech; B syntax; C semantics D coherence; E inappropriate initiation; F stereotyped speech; G use of context and H nonverbal communication



**Table 4** Proportion of children with autism spectrum disorder or language impairment (ASD/LI) corresponding to the 1st, 5th, and 10th percentile values for the GCC

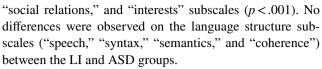
Normative sample ( $N = 22,871$ )		ASD/LI (n = 78		
Percentile value	GCC cutoff	N (%)		
		$\overline{\text{ASD} (n=48)}$	LI (n = 30)	
≤1	34	4 (8.3%)	4 (13.3%)	
≤5	48	24 (50.0%)	12 (40.0%)	
≤10	54	33 (68.8%)	16 (53.3%)	
>10		48 (100%)	30 (100%)	

GCC General Communication Composite, ASD autism spectrum disorder, LI language impairment



**Fig. 3** Distribution of Childrens Checklist-2 (CCC-2) Social Interaction Deviance Composite (SIDC) scores in a normative sample

SD=6.688). Multiple comparison with Scheffe's test revealed that SIDC scores were lower in the ASD than in the LI (p < .001) and TD groups (p < .001). Figure 3 shows the distribution of SIDC scores in the ASD and LI groups in the normative sample. The SIDC scores of both groups were distributed widely and significantly overlapped with the general population, while that of the ASD group tended to be at the lower extreme of the SIDC, and that of LI group tended to be in the middle of the SIDC distribution. Posthoc ANOVA revealed that the LI group also performed significantly worse than the TD group on the "inappropriate initiation," "nonverbal communication," "social relations," and "interests" subscales (F=62.146, df=2, p<.001), as well as in the language structure subscales ("speech," "syntax," "semantics," and "coherence") (F=29.151, df=2, p < .001). The same post-hoc ANOVA revealed that the LI group performed significantly better than the ASD group on the "inappropriate initiation," "nonverbal communication,"



GCC scores did not correlate with K-ABC mental processing scale scores in either the ASD group ( $r_s = 0.028$ ) or the TD group ( $r_s = 0.234$ , p = .062). In addition, GCC scores in the LI group did not correlate with their WISC-III full-scale IQs ( $r_s = -0.031$ ).

From ROC analysis, we obtained a cutoff point of 61.50 for GCC scores on the CCC-2 (sensitivity=0.797; specificity=0.808). The area under the curve (AUC) was 0.882, indicating moderate accuracy of the GCC for predicting the existence of ASD or LI. We also obtained a cutoff point of -0.50 for SIDC scores on the CCC-2 (sensitivity=0.688; specificity=0.700) in ROC analysis. The AUC was 0.765, indicating moderate accuracy of the SIDC for discriminating between ASD and LI.

## **Discussion**

To the best of our knowledge, this is the first study to investigate the distribution of aspects of communicative impairment in a nationwide representative sample of children in the general population. The findings suggest that aspects of communicative impairment measured by the Japanese version of the CCC-2 are continuously distributed, and that ASD and LI fit inside the bell curve of the GCC. These results involving quantitative aspects of communicative impairment add substantial evidence in support of the continuous nature of the impairments in the general population. However, this does not mean that individual ASD or LI cases cannot be discretely or categorically determined. It is well known that categorical, relatively rare causes of ASD or SLI exist. For example, ASD has been diagnosed secondary to fragile X syndrome, Rett syndrome, and tuberous sclerosis; in these cases, ASD is caused by singlegene abnormalities. Ullman and Gopnik (1999) and Kabani et al. (1997) reported the existence of familial SLI that shows abnormalities in inflectional morphology. However, based on the findings from the present study, the notion of a GCC continuum remains consistent with the existence of such discrete entities.

In the present study, no evidence was seen of a natural cutoff that differentiated children categorically affected from those unaffected by ASD or LI. The parent-report Japanese CCC-2 cutoff score from our ROC analysis was 61.5 in terms of the GCC; this analysis comprised 19% of our normative sample, suggesting the existence of subthreshold conditions in children that might warrant clinical attention. This percentage was larger than that found by Kamio et al. (2013), who only investigated ASD distribution. Based on



the highest sensitivity for their study, 10.9% of their normative sample would be cut off. Regarding SLI, our number was smaller than the language screening failure rate (26.2%) reported by Tomblin et al. (1997). Concerning the prevalence of SLI, their study showed 7.4% for whole sample, with 8% for boys and 6% for girls. When adding 7.4% for SLI and 10.9% for ASD, the percentage of affected sample reached 18.3% of normative sample. This value was very close to the value of 19% in the present study. Taken together, these findings suggest that the Japanese version of the CCC-2 estimates a similar number of children with suspicion of ASD or SLI compared with findings from previous studies. Our ROC analysis showed no clear-cut border between those with and without ASD or LI, as the specificity, sensitivity, and accuracy were relatively low.

The results of exploratory factor analysis for the clinical sample are consistent with those from previous studies (Constantino and Todd 2003; Kamio et al. 2013), even though those studies only investigated ASD. In addition, the results of CFA for a very large general population suggest the presence of a primary underlying factor that influences the CCC-2 subscales. Factor structure has important implications for understanding the core neuropsychological mechanisms underlying communicative impairment. Unitary factor structure was not expected because the GCC is composed of eight subscales that greatly differentiate from one another. These subscales are based on phonology, morphology, syntax, semantics, and pragmatics of language. Despite these differences, GCC scores were shown to be a single primary factor that significantly influences the eight subscales of the CCC-2. Despite the linguistic and cultural differences between the UK and Japan, the validity of the GCC in clinical usage was assured in the present study. Cross-cultural consistency in terms of the validity of the GCC should be tested with languages other than English and Japanese, because factor analysis showed that the Serbian CCC-2 had three factors ("General Communication Ability", "Pragmatics", and "Structural Language Aspects"), which accounted for only 29.39% of the total variance (Glumbić and Brojčin 2012).

Regarding the relationship between cognitive development in children and GCC scores, no significant correlation was found between K-ABC mental processing scale and GCC scores in the ASD group or TD group. In addition, full-scale IQ did not correlate with GCC scores in the LI group. The lack of correlation in the ASD group suggests that their GCC scores were independent of their cognitive development. This deficit was also identified in a study by Fujino and Oi (unpublished data). In that study, no correlation was found between GCC scores and full-scale IQ (WISC-IV) in schoolchildren with ASD.

SIDC and GCC scores were continuously distributed, while SIDC scores were higher in LI than in ASD cases.

This supports the notion by Bishop (2003) that an entire spectrum of impairments, with typical SLI at one end and core autism at the other, can be observed in CCC-2 scores, with most children having patterns of impairment between these two extremes. SIDC scores can therefore be useful in further studies on the relationship between ASD and SLI. Data from a large population sample tell us that ASD and SLI are not entirely separate categories, but rather points on a continuum, as the SIDC scores showed a normal distribution (the so-called "bell curve").

The present study had five major limitations. First, although the response rate was consistent with what can be expected from a population-based survey, it was still relatively low (29.15%). Second, other than relationship to the child, no additional information on caregiver characteristics, such as educational level or socioeconomic status, was collected. Third, the size of clinical sample was relatively small, particularly in the LI group. A larger clinical sample would be expected to make the contrast between the TD, ASD, and LI groups more clear-cut in terms of differences in SIDC scores. Fourth, the low scores among the 22,871 Japanese children were not confirmed using any type of diagnostic instrument. Studies designed to assess Japanese language impairment are limited because SLI has not been established as a diagnostic category in Japan. The establishment of SLI in Japan is expected to be attained soon because both GCC and SIDC scores on the CCC-2 appear to be extremely useful for understanding the continuum of communicative impairments across cultures. Fifth, using a measurement scale such as CCC-2 would result in a continuous distribution, which indicates that, as suggested by Pickles and Angold (2003), "the same pathology can have some properties that are most easily understood using a dimensional conceptualization while at the same time having other properties that are best understood categorically".

In conclusion, despite these limitations, the present study is the first to provide strong evidence of the continuous nature of aspects of communication impairment in the general population. The findings underscore the notion that paradigms for categorical case assignment are superimposed on the continuous distribution seen in the general population in regard to the GCC. The findings also support the notion that ASD and LI are not fully discrete entities that exclude each other; rather, they are located at opposite ends of an assumed SIDC continuum of communication impairment, with a considerable amount of cases falling in between. Both GCC and SIDC scores obtained from the CCC-2 are therefore considered to offer promising prospects in understanding the diversity seen in developmental disorders, including ASD and LI, from wider perspectives such as neurology or genetics.



**Acknowledgments** We would like to thank Professor Dorothy Bishop for her comments on our manuscript.

**Funding** This research was supported by a grant (Grant-in-Aid for Scientific Research No. 23330276) from the Japan Society for the Promotion of Science to Dr. Oi; and by research grants from the Ministry of Health, Labour, and Welfare of Japan to Dr. Kamio (H19-KOKORO-006 and H20-KOKORO-004).

**Authors Contributions** MO has written the manuscript. HF has conducted primary component analysis, NT has conducted confirmatory factor analysis. YK has provided framework for the manuscript. YY, MK and CH have provided data for the ASD group including ADOS and K-ABC scores. KG and TM have helped MO writing the manuscript.

#### **Compliance with Ethical Standards**

Ethical Approval The current study was approved by the medical research ethics committee at Kanazawa University and performed in accordance with the ethical standards laid down in the 1964 Declaration of Helsinki and its later amendments. The study protocol was also approved by the Ethics Committee of the National Center of Neurology and Psychiatry, Japan.

**Informed Consent** Informed consent was obtained from the parents of the participant children before the study began.

#### References

- Bishop, D. V. (2003). *The Children's Communication Checklist* (2nd edn.). London: Harcourt Assessment.
- Bishop, D. V. (2010). Overlaps between autism and language impairment: phenomimicry or shared etiology? *Behavior Genetics*, 40(5), 618–629. doi:10.1007/s10519-010-9381-x.
- Bishop, D. V. (2014). Ten questions about terminology for children with unexplained language problems. *International Journal of Language & Communication Disorders*, 49(4), 381–415.
- Bishop, D. V., Maybery, M., Wong, D., Maley, A., & Hallmayer, J. (2006). Characteristics of the broader phenotype in autism: A study of siblings using the Children's Communication Checklist-2. American Journal of Medical Genetics Part B: Neuropsychiatric Genetics, 141B(2), 117–122. doi:10.1002/ajmg.b.30267.
- Bishop, D. V., & Norbury, C. F. (2002). Exploring the border-lands of autistic disorder and specific language impairment: A study using standardised diagnostic instruments. *Journal of Child Psychology and Psychiatry*, 43(7), 917–929. doi:10.1111/1469-7610.00114.
- Boucher, J. (2012). Research review: Structural language in autistic spectrum disorder—characteristics and causes. *Journal of Child Psychology and Psychiatry*, 53(3), 219–233. doi:10.1111/j.1469-7610.2011.02508.x.
- Constantino, J. N., & Gruber, C. P. (2005). Social responsiveness scale (SRS). http://eric.ed.gov/.
- Constantino, J. N., & Todd, R. D. (2003). Autistic traits in the general population: A twin study. Archives of General Psychiatry, 60, 524–530. doi:10.1001/archpsyc.60.5.524.
- Crystal, D. (1987) The Cambridge encyclopedia of language. Cambridge: Cambridge University Press.
- Dollaghan, C. A. (2011). Taxometric analyses of specific language impairment in 6-year-old children. *Journal of Speech, Language,* and Hearing Research, 54, 1361–1371. doi:10.1044/1092-4388.

- Fukuda, S., & Fukuda, S. (2001). The acquisition of complex predicates in Japanese specifically language-impaired and normally developing children. *Brain and Language*, 77(3), 305–320. doi:10.1006/brln.2000.2404.
- Geurts, H., & Embrechts, M. (2008). Language profiles in ASD, SLI, and ADHD. *Journal of Autism and Developmental Disor*ders, 38, 1931–1943. doi:10.1007/s10803-008-0587-1.
- Glumbić, N., & Brojčin, B. (2012). Factor structure of the Serbian version of the Children's Communication Checklist-2. *Research in Developmental Disabilities*, *33*, 1352–1359. doi:10.1016/j.ridd.2012.03.010.
- Helland, W. A., Biringer, E., Helland, T., & Heimann, M. (2009).
  The usability of a Norwegian adaptation of the Children's Communication Checklist Second Edition (CCC-2) in differentiating between language impaired and non-language impaired 6- to 12-year-olds. Scandinavian Journal of Psychology, 50, 287–292. doi:10.1111/j.1467-9450.2009.00718.x.
- Henrichs, J., Rescorla, L., Schenk, J. J., Schmidt, H. G., Jaddoe, V. W., Hofman, A., ... Tiemeier, H. (2011). Examining continuity of early expressive vocabulary development: The generation R study. *Journal of Speech, Language, and Hearing Research*, 54(3), 854–869. doi:10.1044/1092-4388.
- Ito, T., Fukuda, S., & Fukuda, S. (2009). Differences between grammatical and lexical development in Japanese specific language impairment: A case study. *Poznań Studies in Contemporary Linguistics*, 45(2), 211–221. doi:10.2478/v10010-009-0005-7.
- Kabani, N. J., Macdonald, D., Evans, A., & Gopnik, M. (1997). Neuroanatomical correlates of familial language impairment: A preliminary report. *Journal of Neurolinguistics*, 10(2–3), 203–214. doi:10.1016/S0911-6044(97)00009-2.
- Kalnak, N., Peyrard-Janvid, M., Sahlén, B., & Forssberg, H. (2012). Family history interview of a broad phenotype in specific language impairment and matched controls. *Genes, Brain and Behavior*, 11(8), 921–927. doi:10.1111/j.1601-183X.2012.00841.x.
- Kamio, Y., Inada, N., Moriwaki, A., Kuroda, M., Koyama, T., Tsujii, H., ... Constantino, J. N. (2013). Quantitative autistic traits ascertained in a national survey of 22 529 Japanese schoolchildren. *Acta Psychiatrica Scandinavica*, 128, 45–53. doi:10.1111/acps.12034.
- Kim, Y. S., Leventhal, B. L., Koh, Y. J., Fombonne, E., Laska, E., Lim, E. C., ... Grinker, R. R. (2011). Prevalence of autism spectrum disorder in a total population sample. *American Journal of Psychiatry*, 168(9), 904–912. doi:10.1176/appi. ajp.2011.10101532.
- Leyfer, O. T., Tager-Flusberg, H., Dowd, M., Tomblin, J. B., & Folstein, S. E. (2008). Overlap between autism and specific language impairment: comparison of autism diagnostic interview and autism diagnostic observation schedule scores. *Autism Research*, 1, 284–296. doi:10.1002/aur.43.
- Lord, C., Rutter, M., DiLavore, P., & Risi, S. (1999). Autism diagnostic observation schedule. http://eric.ed.gov/.
- Loucas, T., Charman, T., Pickles, A., Simonof, E., Chandler, S., Meldrum, D., & Baird, G. (2008). Autistic symptomatology and language ability in autism spectrum disorder and specific language impairment. *Journal of Child Psychology and Psychi*atry, 49, 1184–1192. doi:10.1111/j.1469-7610.2008.01951.x.
- MacCallum, R. C., Browne, M. W., & Sugawara, H. M. (1996).Power analysis and determination of sample size for covariance structure modeling. *Psychological Methods*, 1, 130–149.
- McGregor, K. K., Berns, A. J., Owen, A. J., Michels, S. A., Duff, D., Bahnsen, A. J., & Lloyd, M. (2012). Associations between syntax and the lexicon among children with or without ASD, and language impairment. *Journal of Autism and Developmental Disorders*, 42(1), 35–47. doi:10.1007/s10803-011-1210-4.



- Ministry of Education, Culture, Sports, Science and Technology. (2010). Annual report on school basic survey. Tokyo: Nikkei.
- Norbury, C. F., Nash, M., Baird, G., & Bishop, D. V. (2004). Using a parental checklist to identify diagnostic groups in children with communication impairment: a validation of the Children's Communication Checklist-2. *International Journal of Communication Disorders*, 39(3), 345–364. http://onlinelibrary.wiley.com/journal/10.1111/(ISSN)1460-6984.
- Oi, M., Fujino, H., Tsukidate, N., Kamio, Y., Gondou, K., & Matsui, T. (2016) *Japanese version of Children's Communication Checklist-2*. Tokyo: Nihon Bunka Kagakusha.
- Perkins, M. (2007). Pragmatic impairment. Cambridge: Cambridge University Press.
- Pickles, A., & Angold, A. (2003). Natural categories or fundamental dimensions: On carving nature at the joints and the rearticulation of psychopathology. *Development and Psychopathology*, *15*(3), 529–551. doi:10.1017/S0954579403000282.
- Rutter, M., Bailey, A., & Lord, C. (2003). The social communication questionnaire. http://eric.ed.gov/.
- Japan Cabinet Secretary (2010). Retrieved May 29, 2015 from http:// www.cas.go.jp/jp/seisaku/youji/dai2/sankou1.pdf.
- Tomblin, B. (2011). Co-morbidity of autism and SLI: Kinds, kin and complexity. *International Journal of Language & Communication Disorders*, 46(2), 127–137. doi:10.1111/j.1460-6984.2011.00017.x.
- Tomblin, J. B., Records, N. L., Buckwalter, P., Zhang, X., Smith, E., & O'Brien, M. (1997). Prevalence of specific language impairment in kindergarten children. *Journal of Speech, Language, and Hearing Research*, 40, 1245–1260. doi:10.1044/jslhr.4006.1245.
- Tsukidate, N., Oi, M., Gondo, K., Matsui, T., & Kamio, Y. (2015).

  Development of the Japanese version of the Children's

- Communication Checklist-2 (CCC-2): Consideration of standardized score. *Japanese Journal of Communication Disorders*, 32(2), 99–108.
- Ueno, K., Takamura, M., & Kaizu, A. (2008). *Learning disabilities Inventory–Revised*. Tokyo: Nihon-Bunka Kagakusha.
- Ullman, M. T., & Gopnik, M. (1999). Inflectional morphology in a family with inherited specific language impairment. Applied Psycholinguistics, 20(1), 51–117. doi:10.1017/S0142716499001034.
- Vézina, M., Sylvestre, A., & Fossard, M. (2013). Développement de la version québécoise francophone du Children's Communication Checklist-2 (CCC-2). Normalisation et équivalence métrique [Development of a Quebec French Version of the Children's Communication Checklist-2 (CCC-2). Normalisation and metric equivalence]. Revue Canadienne d'Orthophonie et d'Audiologie, 37, 156–168. http://cjslpa.ca/.
- Weismer, E. S. (2007). Typical talkers, late talkers, and children with specific language impairment: a language endowment spectrum? In R. Paul (Ed.), The influence of developmental perspectives on research and practice in communication disorders: A Festschrift for Robin S. Chapman (pp. 83–102). Mahwah: Erlbaum.
- Wing, L., & Gould, J. (1979). Severe impairments of social interaction and associated abnormalities in children: Epidemiology and classification. *Journal of Autism and Developmental Disorders*, 9, 11–29. doi:10.1007/BF01531288.
- Wing, L., Leekam, S. R., Libby, S. J., Gould, J., & Larcombe, M. (2002). The diagnostic interview for social and communication disorders: Background, inter-rater reliability and clinical use. *Journal of Child Psychology & Psychiatry*, 43(3), 307–325. doi:10.1111/1469-7610.00023.

