TABLE 1. SUBJECTS

Grade	Corresponding Age (mo)	Male	Female	Total
–1	62-77	56	57	113
1	76-88	27	33	60
2	85-100	26	25	51
3	98-112	22	35	57
4	112-126	19	21	40
5	125-140	23	37	60
6	136-147	13	20	33
Total		186	228	414

Grade –1 — preschool toddlers (ie, 1 year younger than school entrance age).

language development can be difficult to assess, because the concept of language includes a wide range of aspects. Hence, to address these difficulties, test batteries have been created that comprise many language tests, covering major language domains and providing a structured review for evaluating both conversation and academic achievement.

In Japan, however, very few language tests are available, especially for school-age children, although some tests have been directly translated from English into Japanese, and others have been developed under the influence of English-language tests. However, because the structure of Japanese syntax is very different from that of English, use of English tests with Japanese speakers presents problems. In addition, each language test was created separately. Therefore, the relationships among the tests and their correlations with other developmental factors, including communication and learning ability, have not been established. To address these shortcomings, we organized previously established language tests representing different language domains to form a set of tests called the Assessment Package for Language Development in Japanese Hearing-Impaired Children (ALADJIN). In this report, Japanese children in a standard sample were evaluated with the tests in this package, and the correlations between the test results, interpersonal communication skills, and academic achievement were examined.

SUBJECTS AND METHODS

Subjects. The study subjects included 113 preschool children and 316 school-age students without hearing impairment. Fifteen children were excluded from the sample because of low scores on Raven's Coloured Progressive Matrices (RCPM) or the Screening Test of Reading and Writing for Japanese Primary School Children (STRAW; less than –2 SD). In total, 414 children were included in this study (Table 1). The study was approved by the Institutional Review Board of Okayama University.

Methods. Verbal communication function was measured with the Test of Question-Answer Interaction Development (TQAID),[4] and academic achievement was measured with the Japanese Language by Criterion Referenced Test–II (CRT-II).[5] The results of these two tests were used as objective variables. For vocabulary, the Picture Vocabulary Test–Revised (PVT-R),[6] the Standardized Comprehension Test of Abstract Words (SCTAW),[7] and the Word Fluency Test (WFT)[8] were performed. For syntax, the Syntactic Processing Test of Aphasia (STA)[9] was also performed. The results of the vocabulary and syntax tests were used as explanatory variables.

Because mental retardation and reading and writing difficulties (dyslexia or dysgraphia) can be confounding factors in language development, children were screened with the RCPM[10] and the STRAW.[11] All test results are summarized in Table 2. All tests except for the CRT-II were administered in a face-to-face setting by trained doctors or speech therapists. The CRT-II was administered in a group setting.

Statistical Analysis. All test results are presented as means and standard deviations. Elemental factors that can affect the results of the TQAID and CRT-II were evaluated by multiple linear regression analy-

TABLE 2. ALADJIN LANGUAGE TESTS ADMINISTERED AT DIFFERENT GRADE LEVELS

Grade	TQAID	CRT-II	PVT-R	SCTAW	WFT	STA	STRAW	RCPM
–1	+	–	+	–	+	+	+*	–
1	+	–	+	+	+	+	+†	+
2	+	+	+	+	+	+	+	+
3	+	+	+	+	+	+	+	+
4	+	+	+	+	+	+	+	+
5	+	+	+	+	+	+	+	+
6	+	+	+	+	+	+	+	+

See text for abbreviations.

*STRAW was partially applied in grade –1: reading and writing for one letter of *hiragana*.

†STRAW was partially applied in grade 1: reading and writing for one letter of *hiragana* and *katakana*.

TABLE 3. MEANS AND STANDARD DEVIATIONS ON ALADJIN

	Grade –1	*Grade 1*	*Grade 2*	*Grade 3*	*Grade 4*	*Grade 5*	*Grade 6*
CRT-II			85.4 ± 11.9	84.3 ± 14.3	82.8 ± 10.1	81.3 ± 11.9	81.7 ± 9.4
TQAID	228.0 ± 30.7	252.1 ± 28.3	261.3 ± 22.5	270.1 ± 19.4	278.4 ± 14.4	282.4 ± 13.4	286.4 ± 12.6
PVT-R	27.0 ± 8.7	35.6 ± 8.8	43.5 ± 8.4	52.1 ± 9.5	60.3 ± 8.1	66.6 ± 7.5	70.6 ± 6.5
SCTAW		11.5 ± 4.3	16.1 ± 3.5	19.9 ± 3.3	23.1 ± 4.3	24.9 ± 3.3	28.0 ± 2.3
WFT	19.4 ± 7.6	33.8 ± 10.9	38.1 ± 9.2	39.2 ± 13.1	51.0 ± 14.4	53.4 ± 14.2	58.2 ± 13.4
STA(C)	28.1 ± 6.4	31.7 ± 5.0	33.1 ± 3.8	36.1 ± 3.6	37.8 ± 2.3	37.4 ± 2.7	37.9 ± 2.6
STA(P)	35.2 ± 10.4	45.1 ± 7.5	48.5 ± 6.9	51.7 ± 5.5	53.6 ± 3.6	54.5 ± 3.8	54.5 ± 4.4
STRAW(R)	17.4 ± 4.8	39.3 ± 2.5	97.5 ± 2.9	98.9 ± 2.8	98.2 ± 0.6	98.5 ± 1.2	99.7 ± 0.6
STRAW(W)	10.8 ± 6.5	37.7 ± 3.2	88.7 ± 11.9	95.2 ± 6.4	95.8 ± 6.6	94.2 ± 4.8	96.6 ± 3.6
RCPM		27.0 ± 4.4	29.4 ± 4.2	30.9 ± 3.5	33.1 ± 2.0	30.9 ± 3.0	33.0 ± 2.1

STA(C) — STA comprehension; STA(P) — STA production; STRAW(R) — STRAW reading; STRAW(W) — STRAW writing.

sis. All statistical values were calculated with JMP 9 software (SAS Institute Inc, Cary, North Carolina).

RESULTS

Test Results. The test results according to grade level are outlined in Table 3. The mean (±SD) score on the TQAID in preschool children (5 to 6 years of age) was 228 ± 30.7. More than 70% of the children in the entire sample had nearly perfect scores on this test. In the children in grade 3 (8 to 9 years of age), the mean score was 270.1 ± 19.4. A ceiling effect was observed at this age. All three vocabulary tests (PVT-R, SCTAW, and WFT) demonstrated incremental increases in vocabulary by grade, whereas the scores for the comprehension test for syntactic structure (STA) almost reached ceiling values (33.1 ± 3.8 and 36.1 ± 3.6 in grades 2 and 3, respectively). The STA production test showed marked improvement from preschool age to grade 1 (35.2 ± 10.4 and 45.1 ± 7.5, respectively). Again, a ceiling effect was observed at grade 3 (51.7 ± 5.5).

Wide interpersonal variation was observed in the results on the STRAW for the 5- to 6-year-old group (17.4 ± 4.8 in reading and 10.8 ± 6.5 in writing). Because the test for *kanji* characters (Japanese ideograms) was included in STRAW testing of the children in grade 2 (7 to 8 years of age) and older, the difference between reading and writing scores did not become apparent until that grade level.

Multiple Regression Analysis. The TQAID scores were analyzed for children who were in grades lower than grade 4 (9 to 10 years of age), because an apparent ceiling effect was observed among children of this age and older. The results of the PVT-R, WFT, STA (comprehension), and STA (production) were used as explanatory variables. The results (Table 4) indicated that the WFT, PVT-R, and STA (production) had predictive value for the TQAID ($R = 0.59$; $R^2 = 0.58$; $p < 0.0001$). The standard regression variables (standardized partial regression coefficients) were significant: 6.97 for STA (production; $p < 0.0001$), 5.47 for the WFT ($p < 0.0001$), and 4.37 for the PVT-R ($p < 0.0001$). A CRT-II analysis was performed separately for each grade because of differences in content according to grade level. The results of the SCTAW, WFT, STA (comprehension), and STA (production) were used as explanatory variables. Among these variables, those of the SCTAW and STA (comprehension) had high predictive value for the scores on the CRT-II (Table 5).

TABLE 4. RESULTS OF MULTIPLE REGRESSION ANALYSIS OF TQAID FOR GRADES –1 TO 3 (N = 281)

	SPRC	*95% CI*	*p*
PVT-R	4.37	0.31 to 0.82	<0.0001
WFT	5.47	0.57 to 1.23	<0.0001
STA(C)	0.75	–0.32 to 0.715	0.45
STA(P)	6.97	0.84 to 1.51	<0.0001
CD	0.59		<0.0001

SPRC — standardized partial regression coefficient; 95% CI — 95% confidence interval; CD — coefficient of determination.

DISCUSSION

In this study, the ALADJIN was used to evaluate the effect of language development on interpersonal communication skills and academic achievement in a normative sample of Japanese-speaking children. The impact of vocabulary, syntax, and reading and writing abilities in these two areas was examined.

The results of the TQAID demonstrated that children 5 to 6 years of age have fairly well-developed interpersonal communication skills. A ceiling effect was observed on this test at grade 3 (8 to 9 years of age). Thus, in standard samples, the TQAID may not be appropriate for children more than 9 years of age. Alternatively, this test could be used for screening of children more than 9 years of age to detect crucial delays in interpersonal communication skills.

The scores on the vocabulary tests in this study (PVT-R, SCTAW, and WFT) increased incrementally by grade level. The results also indicated that

TABLE 5. RESULTS OF MULTIPLE REGRESSION ANALYSIS OF CRT-II

	Grade 2 (n = 51)		Grade 3 (n = 57)		Grade 4 (n = 40)		Grade 5 (n = 60)		Grade 6 (n = 33)	
	SPRC	95% CI	SPRC	95% CI	SPRC	95% CI	SPRC	95% CI	SPRC	95% CI
SCTAW	2.67*	0.30 to 2.10	3.67†	0.75 to 2.58	1.58	–0.17 to 1.38	2.55*	0.25 to 2.09	2.93†	0.57 to 3.22
WFT	–0.66	–0.40 to 0.20	1.74	0.75 to 2.58	1.11	–0.10 to 0.35	0.63	–0.13 to 0.26	1.12	–0.09 to 0.31
STA(C)	3.16†	0.54 to 2.46	2.57*	0.25 to 2.01	0.24	–1.25 to 1.60	2.18†	0.10 to 2.36	1.33	–0.44 to 2.05
STA(P)	–0.61	–0.64 to 0.35	0.80	–0.37 to 0.85	0.97	–0.47 to 1.35	1.53	–0.18 to 1.31	1.48	–0.18 to 1.10
CD	0.43‡		0.47‡		0.16		0.40‡		0.56‡	

*p < 0.05.
†p < 0.01.
‡p < 0.0001.

vocabulary building may play an important role in development of more complicated language skills in older children.[12] A ceiling effect was also observed on the test for syntax (STA) in grade 3. School-age children acquire the basic structures of syntax relatively early.

The results of the multiple regression analysis suggested that different language domains may play different roles in interpersonal communication skills, as indicated by the results on the TQAID, and in academic achievement, as indicated by the results on the CRT-II. The results on the WFT and STA (production) tests were strongly correlated with those on the TQAID, and the results on the CRT-II were highly correlated with those on the SCTAW and STA (comprehension). Interpersonal communication skills as measured by the TQAID may include the ability to select or use appropriate words or sentences in response to given situations or questions. In other words, the results on the TQAID may reflect the development of productive language (both vocabulary and syntax). By contrast, the results on the CRT-II were well correlated with those of the SCTAW and STA (comprehension), suggesting the importance of both vocabulary and syntax in comprehension.

This study was conducted with a standard sample of preschool and school-age children attending a mainstream school with no specialist support. However, some children in each grade demonstrated very poor language development (more than –2 SD). Further inquiries demonstrated that the teachers and caregivers were already aware of these children's difficulties, but were unaware of the causes of these difficulties or how to offer appropriate support. The present data demonstrate the possibility that domain-based language development assessment can predict the results of academic achievement. The ALADJIN may therefore play an important role in identifying children who need language support and in indicating in what areas the support is most needed.

CONCLUSIONS

Using the ALADJIN, we evaluated language ability in different language domains in Japanese-speaking children. Development in the productive language domains (productive vocabulary and syntax) had an effect on interpersonal communication skills, and development in the receptive language domains (receptive vocabulary and syntax) had an effect on academic achievement, together with reading and writing ability. The aim of the ALADJIN is to provide assistance or guidance for diagnosis and intervention. The ALADJIN allows important insights into language delay in children and provides a guide to intervention by indicating weak points that must be overcome.

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REFERENCES

1. Sasamori H, Gokami T, Kuboyama S, et al. Current situation and problems for early detection and early support for children with developmental disability. National Institute of Special Needs Education, 2010;38:3-15.

2. Hargrove ML. Learning disabilities. Early identification signals for parents and teachers. Education 1982;102:366-8.

3. Koed T, Tomita Y, Takeshita K. Prognosis of three-year-old children with developmental speech delay: reading and writing abilities at eight years of age [in Japanese]. No To Hattatsu 1990;22:235-40.

4. Satake T, Higachie H, Chinen H. Manual for Test of Query-Answering Relationship [in Japanese]. Tokyo, Japan: Escor Co, 1996.

5. Tatsuno C, Kitao M. The manual for Criterion Referenced Test (CRT-II) [in Japanese]. Tokyo, Japan: Toshobunka, 2007.

6. Ueno K, Nagoshi S, Konuki S. Picture Vocabulary Test–Revised. Hiroshima, Japan: Saccess/Bell Co Ltd, 2008.

7. Haruhara N, Kaneko M, Uno A. The Standardized Comprehension Test of Abstract Words. Tokyo, Japan: Interuna Publishers, 2002.

8. Koren R, Kofman O, Berger A. Analysis of word cluster-

ing in verbal fluency of school-aged children. Arch Clin Neuropsychol 2005;20:1087-104.

9. Fujita I, Miyake T. The manual for the syntactic processing test of aphasia [in Japanese]. Tochigi, Japan: The Japanese Workshop of Verbal Communication, 2009.

10. Raven's Coloured Progressive Matrices in Japanese Children as a screening intelligence test for children with learning disorder and acquired childhood aphasia. Jpn J Logop Phoniatr 2005;46:185-9.

11. Uno A, Haruhara N, Kaneko M. Reading/writing screening for school aged children: an approach to detect developmental dyslexia [in Japanese]. Tokyo, Japan: Interuna Publishers, 2006.

12. Noboru T. Vocabulary development in elementary school children. Communication Association of Japan, 2006;23:118-25.

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Language development in Japanese children who receive cochlear implant and/or hearing aid

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ABSTRACT

Objectives: This study aimed to investigate a wide variety of factors that influence auditory, speech, and language development following pediatric cochlear implantation (CI).
Study design: Prospective collection of language tested data in profound hearing-impaired children.
Hypothesis: Pediatric CI can potentially be effective to development of practical communication skills and early implantation is more effective.
Methods: We proposed a set of language tests (assessment package of the language development for Japanese hearing-impaired children; ALADJIN) consisting of communication skills testing (test for question–answer interaction development; TQAID), comprehensive (Peabody Picture Vocabulary Test-Revised; PVT-R and Standardized Comprehension Test for Abstract Words; SCTAW) and productive vocabulary (Word Fluency Test; WFT), and comprehensive and productive syntax (Syntactic processing Test for Aphasia; STA). Of 638 hearing-impaired children recruited for this study, 282 (44.2%) with >70 dB hearing impairment had undergone CI. After excluding children with low birth weight (<1800 g), those with >11 points on the Pervasive Developmental Disorder ASJ Rating Scale for the test of autistic tendency, and those <2 SD on Raven's Colored Progressive Matrices for the test of non-verbal intelligence, 190 children were subjected to this set of language tests.
Results: Sixty children (31.6%) were unilateral CI-only users, 128 (67.4%) were CI–hearing aid (HA) users, and 2 (1.1%) were bilateral CI users. Hearing loss level of CI users was significantly ($p < 0.01$) worse than that of HA-only users. However, the threshold level, maximum speech discrimination score, and speech intelligibility rating in CI users were significantly ($p < 0.01$) better than those in HA-only users. The scores for PVT-R ($p < 0.01$), SCTAW, and WFT in CI users were better than those in HA-only users. STA and TQAID scores in CI–HA users were significantly ($p < 0.05$) better than those in unilateral CI-only users. The high correlation ($r = 0.52$) has been found between the age of CI and maximum speech discrimination score. The scores of speech and language tests in the implanted children before 24 months of age have been better than those in the implanted children after 24 months of age.
Conclusions: We could indicate that CI was effective for language development in Japanese hearing-impaired children and early CI was more effective for productive vocabulary and syntax.

1. Introduction

Management of CI in infants and children is one of the most striking advances for congenital severe to profound hearing loss. Several studies have shown that early implantation can be beneficial not only for speech perception, but also for the development of speech and language [1–3]. Moreover, early intervention for children with hearing loss facilitates successful educational integration at the earliest possible age [4].

More than 20 years have passed since the first pediatric CI surgery was performed in Japan. Many hearing-impaired children are now benefiting from this device. However, the long-term benefits for Japanese CI users have rarely been reported. In particular, language development after CI among Japanese children has not often been investigated. Language development outcomes among children with prelingual hearing impairment have been

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studied in Indo-European languages, but language differences may have an effect on language development in children with CI. In addition, differences in national and local education systems may make a difference to language development. To determine the effect of CI, we examined language development in different language and/or social systems.

Language differences may add other difficulties; for example, interpretation of Japanese language test results may not be comparable with that of English or other European language tests. To reduce these difficulties, we have established the assessment package of the language development for Japanese hearing-impaired children (ALADJIN) as a language performance evaluation tool for hearing-impaired children. ALADJIN includes several Japanese language tests that are directly comparable with previously reported English tests, including the Peabody Picture Vocabulary Test-Revised (PVT-R) and Test for Reception of Grammar (TROG)-like syntax tests (e.g., the Syntactic processing Test for Aphasia; STA). These tests all have their own distinctive emphasis and evaluate different aspects or domains of language.

In 2010, we assessed the current status of hearing-impaired children in Japan through a project called Research on Sensory and Communicative Disorders (RSCD). ALADJIN was used in this nationwide research project. The RSCD was originally intended to assess the effectiveness of interventional methods for hearing-impaired children. As part of the RSCD survey, we evaluated the domain-specific language status of Japanese hearing-impaired children with CI, not only in selected institutes and schools that potentially yield biases, but in a wide variety of institutes in Japan.

Thus, the objective of this study was to evaluate the development of interpersonal communication skills (IPCS) in hearing-impaired children with CI using the ALADJIN data set from the RSCD nationwide research project.

2. Materials and methods

All ALADJIN tests were conducted by trained audiologists, speech pathologists, or deaf school teachers in a noise-minimized compartment. Audiometry for evaluation of hearing level, pure-tone threshold, speech discrimination test, and speech intelligibility rating [5] were measured in a sound-attenuated room of the relevant hospital. The study design was approved by the ethics review board of the Association of Technical Aids.

2.1. Subjects

In 2009, 124 institutes were participated in the RSCD project and 638 hearing-impaired children were registered; written informed consent was obtained from their parents. Open recruitment was conducted not only in institutes for hearing-impaired children, i.e., deaf schools and hard of hearing schools, but also in mainstream schools, day-care nurseries, and hospital/clinic training programs.

Most children included in this project were within the age range from 4 years (2 years before elementary school entrance; −2 grade) to 12 years (6th grade of elementary school; +6 grade) and confirmed to have congenital hearing impairment (average hearing level >70 dB at 4 years of age). Children who were discernibly unable to complete the ALADJIN tests due to additional handicaps were excluded. 282 (44.2%) participating children were CI users, and about 45% of the hearing-impaired children of each age group were CI users (Fig. 1). Subjects were classified into four groups as follows: (1) "unil CI-only" group with unilateral CI users, (2) "CI–HA" group with CI plus conventional HA users (also called the bimodal stimulation group), (3) "bil-CI" group with bilateral CI users, and (4) "HA-only" group with HA users. The number of CI children in each age group is given in Fig. 2. 84 children (35 males and 49 females, 29.8%) in the unil CI-only group were diagnosed as

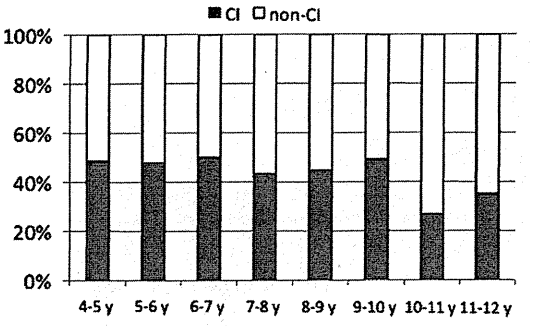

Fig. 1. The rate of CI-only users among the participating hearing-impaired children of each age group. About 45% of hearing-impaired children (>70 dB hearing level) in this study were CI-only users. CI: cochlear implant.

hearing-impaired at 12.5 months on average. In the CI–HA group, 196 children (99 males and 97 females, 69.5%) were diagnosed as hearing-impaired at 10.9 months on average. Two children (1 male and 1 female, 0.7%) were bilateral CI users (bil-CI group). In the HA-only group, 356 children were diagnosed as hearing-impaired at 13.3 months and fitted at first hearing aids at 17.2 months (0–74 months) on average. Age at first fitting hearing aids in the children with CI was 15 months (2–47 months).

In order to reduce the influence of developmental disabilities in our evaluation of the language tests (ALADJIN), participating children with birth weights <1800 g, PARS scores >11 points, and RCPM scores <2 SD of the average were excluded. The numbers of subjects in each group were evaluated in the language tests as follows: 60 unil CI-only users, 128 CI–HA users, and 203 HA-only users. No significant differences in the scores of PARS and RCPM among the unil CI-only, CI–HA, and HA-only users were found (Fig. 3).

2.2. Test battery

We used the test for question–answer interaction development (TQAID) as a tool to measure IPCS function objectively. To let children understand a content of task, their favorite mode of communication (aural, sign language, total communication) were used to perform the language tests. 80% of subjects used aural communication as major mode in the domestic life. The following tests were also used to evaluate IPCS the day after administration of the TQAID.

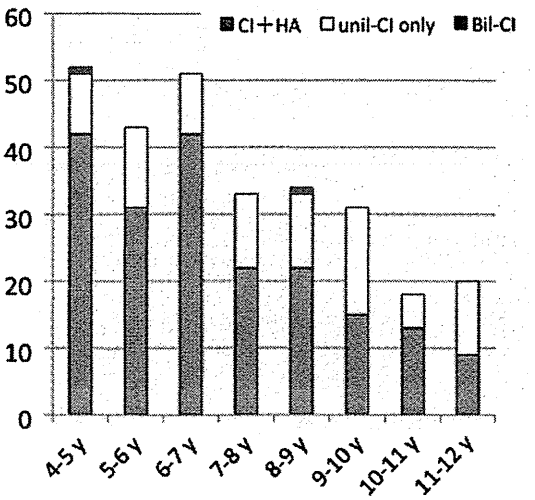

Fig. 2. Number of children in the CI–HA, unil CI-only, and bil-CI groups in each age range. CI plus HA users (bimodal stimulation) make up the majority of CI users. CI: cochlear implant; unil CI: unil CI-only users; bil-CI: bilateral CI users; CI–HA: HA and CI users.

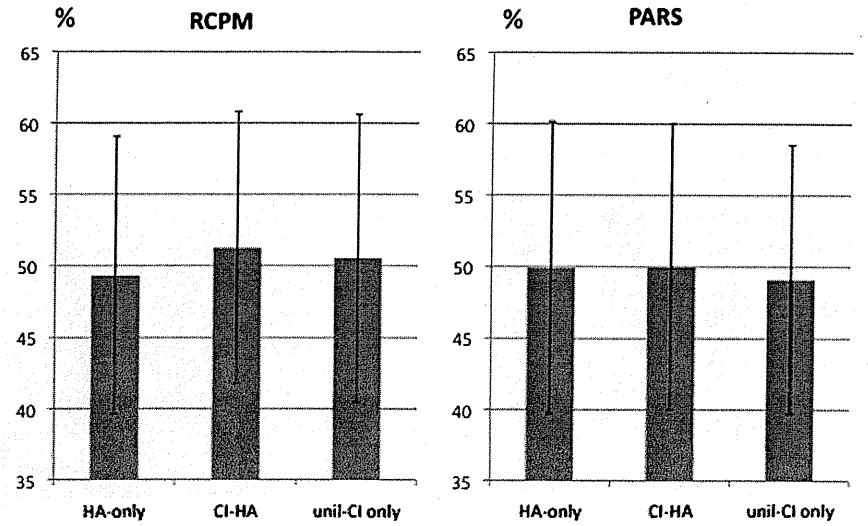

Fig. 3. The scores of PARS and RCPM tests in the HA-only, CI–HA, and unil CI-only groups. There were no significant differences in average scores among the groups. PARS: Pervasive Developmental Disorder ASJ Rating Scales for evaluating autistic tendency; RCPM: Raven's Colored Progressive Matrices test for evaluating non-verbal intelligence; CI: cochlear implant; HA: hearing aid.

The Word Fluency Test (WFT) was conducted as a measure of productive vocabulary [6,7]. Children were asked to produce as many words as possible from a certain category in 60 s. The words, represented either orally or manually, were carefully counted, excluding onomastic words. The Japanese version of the PVT-R [8] and the Standardized Comprehension Test for Abstract Words (SCTAW) [9] were also conducted to evaluate comprehensive vocabulary. An adjusted score was used in this study. The SCTAW consists of 32 or 45 abstract words selected from Japanese school textbooks. The details of how this method has been adapted for hearing-impaired children have been reported in previous studies [9,10]. Only school-aged children were subjected to this test.

The STA evaluates comprehension and production of syntactic structures. The children were asked to choose one of the four pictures appropriate to the tester's presentation (comprehension test) or to express a sentence according to a picture that the tester indicated (production test) [11]. The tests evaluated understanding and expression of irreversible sentences, reversible sentences, Japanese suffixes (Jyo-Shi), and other syntactic structures, including relative pronouns.

To evaluate additional handicaps other than hearing impairment, the Pervasive Developmental Disorder ASJ Rating Scale (PARS) test for autistic tendency [12] and Raven's Colored Progressive Matrices (RCPM) test of non-verbal intelligence [13] were used only in school-aged children.

2.3. Statistical analyses

All statistical values were calculated using IBM SPSS Statistics 18 software (IBM Corp., Armonk, NY, USA). Correlations and standard deviations within each group were examined. The scores of the language tests (PARS, RCPM, PVT-R, SCTAW, WFT, STA, and TQAID) were translated as *Z*-scores from the results of each test in each age group.

3. Results

There were significant ($p < 0.01$) differences in the scores of average hearing loss level, average threshold level with hearing devices, maximum speech discrimination score, and speech intelligibility rating between CI users (unil CI-only or CI–HA users) and HA-only users (Fig. 4). Hearing loss level of CI users was significantly lower than that of HA-only users. However, the threshold level, maximum speech discrimination scores, and speech intelligibility rating of CI users were significantly better than those of HA-only users. The scores of the PVT-R, SCTAW, and WFT tests, which evaluate vocabulary, were higher in CI users than in HA-only users (Fig. 5). There was a significant difference ($p < 0.01$) in the results of the PVT-R test. The scores of the STA (Fig. 6) and TQAID (Fig. 7) in CI–HA users were significantly higher ($p < 0.05$) than those in the unil CI-only group.

The high correlation ($r = 0.52$) has been found between the age of CI and maximum speech discrimination score (Fig. 8). The average scores of speech and language tests in the implanted children before 24 months of age have been better than those in the implanted children after 24 months of age (Table 1). The average scores of WFT (evaluation of productive vocabulary) and comprehension and production tests of STA (evaluation of syntactic structure) were significantly better in the implanted children before age of 24 months compared with the implanted children after age of 24 months.

4. Discussion

To evaluate the language development in the typical hearing-impaired children, we have made exclusionary criteria to standardize the subjects in this study. We excluded the hearing-impaired children with birth weights <1800 g who scored >11 points on the PARS test and <2 SD on the RCPM. Very low birth weight children are at a high risk of neurosensory disability, including developmental delay, behavioral problems, and learning disabilities [14]. Long-term follow-up studies have also emphasized the prevalence of significant neuropsychological and behavioral deficits at school age in children of very low birth weight [15]. Therefore, we excluded children with birth weights <1800 g to reduce the influence of developmental disabilities in our evaluation of communication skills. The PARS and RCPM tests determine the presence of pervasive developmental disorders and non-verbal intelligence, respectively. The scores in these tests were not significantly different among unil CI-only, CI–HA, and HA-only users. Consequently, children with developmental disabilities were excluded from the present study. However, children with ANSD (auditory neuropathy spectrum disorder) could not been excluded, because we did not get the data of ABR and OAE in this study.

Speech development for prelingual deaf children depends on optimal amplification with a CI or HA. Language acquisition is a

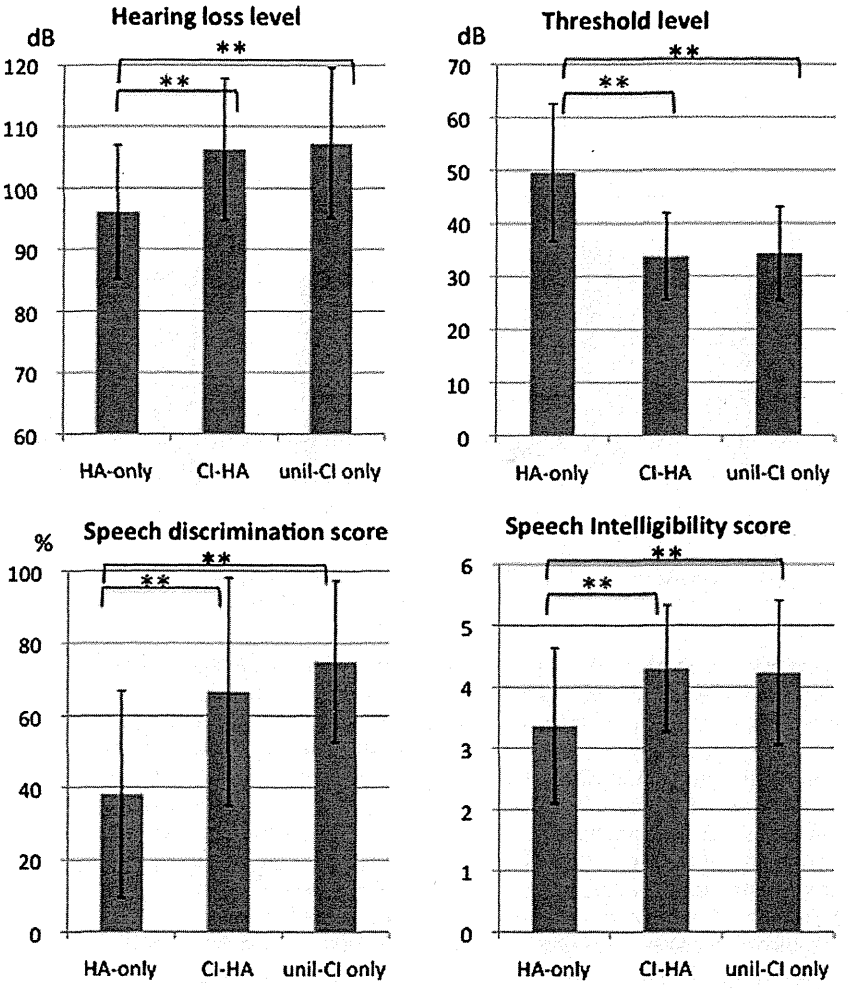

Fig. 4. Hearing loss levels, threshold levels, maximum speech discrimination scores, and speech intelligibility scores in the HA-only, CI–HA, and unil CI-only groups. There are significant differences ($p < 0.01$) in hearing levels, threshold levels, speech discrimination scores, and intelligibility scores between the CI group (CI–HA or unil CI-only groups) and HA-only group. Children with CI achieve better threshold levels, speech discrimination, and intelligibility compared with HA-only users. $^{**}p < 0.01$, CI: cochlear implant; HA: hearing aid.

high priority among deaf children who receive CI. During the 1990s, the following factors were considered to be associated with good speech development: age at implantation, duration of deafness, amount of daily use, mode of communication, and absence of other handicaps. Dettman et al. [16] reported that infants with implantation during the first year of life had significantly faster rates of receptive and expressive language development than those with implantation in the second year of life. On the other hand, another study found no significant differences in the performance in terms of spoken word recognition and expressive language development between children with implantation in the first and second years of life [17]. In our study,

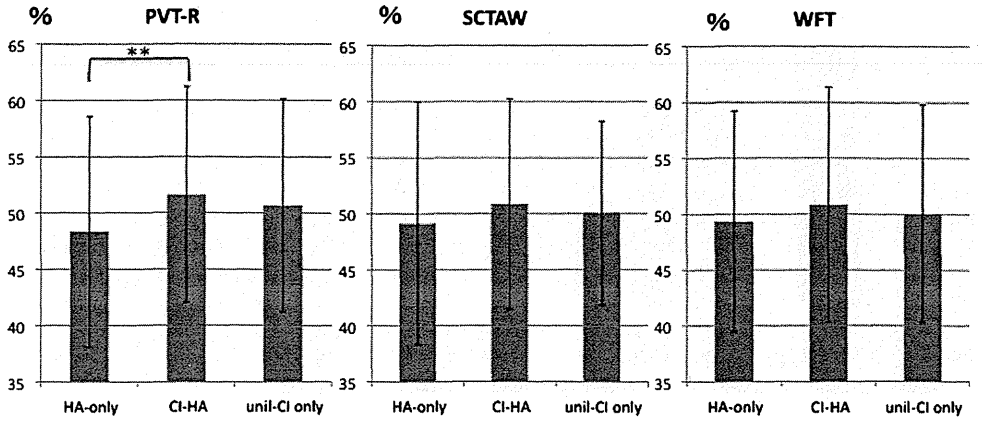

Fig. 5. Scores of the PVT-R, SCTAW, and WFT tests in the HA-only, CI–HA, and unil CI-only groups. Scores of the PVT-R, SCTAW, and WFT tests in the CI–HA and unil CI-only groups were better than those in the HA-only group. A significant difference ($p < 0.01$) was found in the scores of the PVT-R test. $^{**}p < 0.01$, PVT-R: Peabody Picture Vocabulary Test-Revised; SCTAW: Screening Test for Abstract Words; WFT: Word Fluency Test. Values in the longitudinal line indicate Z-score.

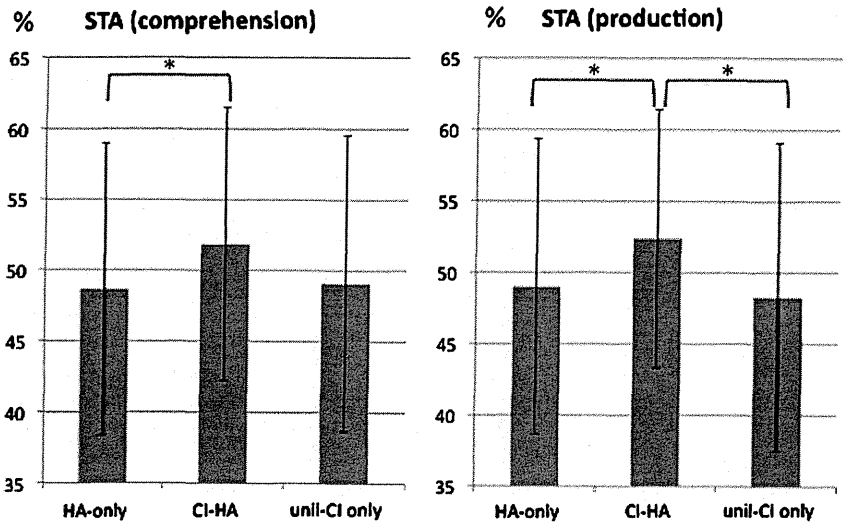

Fig. 6. Scores of the STA test (comprehension and production) in the HA-only, CI–HA, and unil CI-only groups. STA test scores (comprehension) in the CI–HA group were significantly higher ($p < 0.05$) than in the HA-only group. STA test scores (production) in the CI–HA group were significantly higher ($p < 0.05$) than those in the unil CI-only and HA-only groups. *$p < 0.05$. STA: Syntactic processing Test for Aphasia test. Values in the longitudinal line indicate Z-score.

early CI was more effective for better speech discrimination and children with CI before the second year of life had significantly better scores of productive vocabulary and comprehensive and productive syntax.

Early intervention has a strong influence on language outcomes in most, but not all, hearing-impaired children. The degree of hearing loss is an important factor in the modeling of speech production and spoken language outcomes. Several studies have demonstrated a clear relationship between the degree of hearing loss and language outcome [18]. In our study, the average age at diagnosis of hearing loss in children with CI was 11.4 months. Age at diagnosis in CI–HA users (10.9 months) was earlier than in unil CI-only users (12.5 months) and HA-only users (13.3 months).

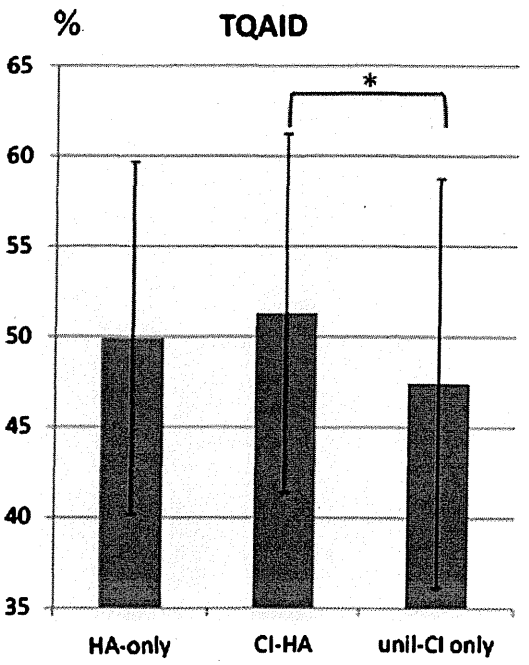

Fig. 7. The scores of TQAID test in the HA-only, CI–HA, and unil CI-only groups. The score of TQAID test in the CI–HA group is significantly ($p < 0.05$) better than that in the unil CI-only group. *$p < 0.05$, TQAID: test for question-answer interaction development is for evaluating the IPCS (interpersonal communication skills) function. Values in the longitudinal line indicate Z-score.

Better speech and language development was found in CI–HA users compared with unil CI-only users.

The degree of hearing loss in CI users was higher than in HA-only users. Speech discrimination score and intelligibility rating were higher in CI users than in HA-only users. The degree of hearing loss was significantly negatively correlated with speech discrimination and intelligibility. However, no clear relationship between the degree of threshold with the amplification devices and speech discrimination and intelligibility was found. The degree of threshold with amplification is thus a predictive factor of speech discrimination and intelligibility. It is beneficial for the CI to establish the better threshold level because fitting method is completely different. This study confirmed that CI has a positive influence on speech discrimination and intelligibility in severely hearing-impaired children. However, 124 institutions were participated in this study as nationwide research project, so there might be a confounding variable for selection of amplification devices (CI/HA vs CI/CI vs unil CI).

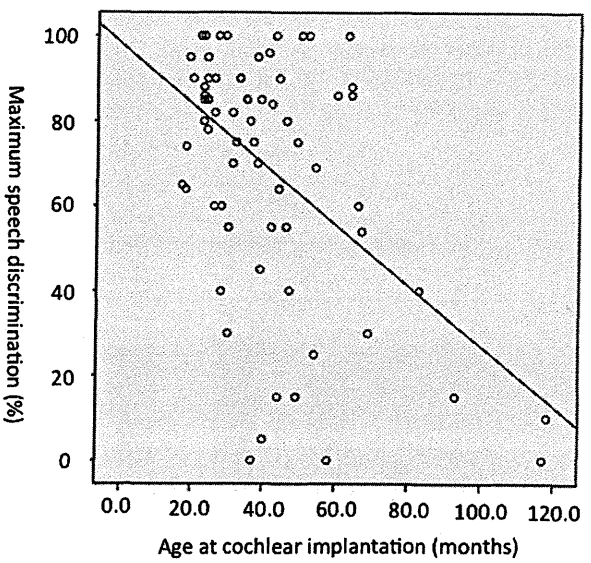

Fig. 8. The correlation between the age of cochlear implantation and maximum speech discrimination score. The high correlation ($r = 0.52$) has been found between the age of CI and maximum speech discrimination score.

Table 1
Average scores of language tests (ALADJIN) in children with CI before and after age of 24 months.

	PVT-R	WFT	SCTAW	STA (Com)	STA (Pro)	TQAID	RCPM	PARS
CI after 24 mo (N=29)	29.5	12.9	13.7	23.1	34.1	209.4	27.5	5.3
CI before 24 mo (N=161)	32.2	15.6	13.8	26.2	39.0	229.7	28.1	4.5
t-Value	0.19	0.02	0.99	0.04	0.04	0.06	0.77	0.30

PVT-R: Peabody Picture Vocabulary Test-Revised; WFT: Word Fluency Test; SCTAW: Standardized Comprehension Test for Abstract Words; STA (Com): Syntactic processing Test for Aphasia (Comprehension); STA (Pro): Syntactic processing Test for Aphasia (Production). TQAID: test for question–answer interaction development; RCPM: Raven's Colored Progressive Matrices; PARS: Pervasive Developmental Disorder ASJ Rating Scale; CI: cochlear implantation; mo: months; N: number.

In evaluating auditory performance, formal speech perception tests, such as open-set and closed-set tests, are often used in children with CI. Communication skills, including auditory, speech, and language development for congenital and prelingual deaf children with CI, are influenced by a wide variety of factors. Several studies have reported that factors such as gender, nonverbal intelligence, estimated family income, communication mode, performance IQ, working memory capacity, articulation rate, and verbal rehearsal speed may predispose a child to better or poorer outcomes with a CI [3,17].

We developed ALADJIN as a set of language tests to evaluate IPCS ability. Results of this assessment showed that CI was more effective for the development of comprehensive and productive vocabulary compared with HA, and bimodal hearing with CI and HA positively influence the development of vocabulary (comprehensive and productive), syntax (comprehensive and productive), and IPCS compared with unilateral hearing with CI. Consequently, we can conclude that early CI, especially in combination with HA, is useful in the development of communication skills.

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References

[1] K.I. Kirk, R.T. Miyamoto, C.L. Lento, E. Ying, T. O'Neill, Effects of age at implantation in young children, Ann. Otol. Rhinol. Laryngol. Suppl. 189 (2002) 69–73.
[2] S. Zimmerman-Phillips, A.M. Robbins, M.J. Osberger, Assessing cochlear implant benefit in very young children, Ann. Otol. Rhinol. Laryngol. Suppl. 185 (2000) 42–43.
[3] I.J. Moon, E.Y. Kim, H. Chu, W.H. Chung, Y.S. Cho, S.H. Hong, A new measurement tool for speech development based on Ling's stages of speech acquisition in pediatric cochlear implant recipients, Int. J. Pediatr. Otorhinolaryngol. 75 (2011) 495–499.
[4] A.G. Geers, J.G. Nicholas, J.S. Moog, Estimating the influence of cochlear implantation on language development in children, Audiol Med 5 (2007) 262–273.
[5] C. Allen, T.P. Nikolopoulos, D. Dyar, G.M. O'Donoghue, Reliability of a rating scale for measuring speech intelligibility after pediatric cochlear implantation, Otol. Neurotol. 22 (2001) 631–633.
[6] D. Riva, F. Nichelli, M. Devoti, Developmental aspects of verbal fluency and confrontation naming in children, Brain Lang. 71 (2000) 267–284.
[7] S. Arffa, The relationship of intelligence to executive function and non-executive function measures in a sample of average, above average, and gifted youth, Arch. Clin. Neuropsychol. 22 (2007) 969–978.
[8] K. Ueno, S. Nagoshi, S. Konuki, Picture Vocabulary Test – Revised, SACCESS Bell Co., Ltd., Hiroshima, Japan, 2008 (in Japanese).
[9] N. Haruhara, M. Kaneko, A. Uno, The Standardized comprehension test of abstract words, Interuna Publishers, Inc., Tokyo, 2002 (in Japanese).
[10] K. Kunisue, K. Fukushima, A. Kawasaki, Y. Maeda, R. Nagayasu, Y. Kataoka, et al., Comprehension of abstract words among hearing impaired children, Int. J. Pediatr. Otorhinolaryngol. 71 (2007) 1671–1679.
[11] R. Nakajima, T. Horai, T. Sugata, H. Tatsumi, T. Hamanaka, Syntactic and communication abilities of aphasics as assessed by CADL, STA and CADL family questionnaire, Jpn. J. Logop. Phoniatr. 38 (1997) 161–168.
[12] A. Yamada, M. Suzuki, M. Kato, M. Suzuki, S. Tanaka, T. Shindo, et al., Emotional distress and its correlates among parents of children with pervasive developmental disorders, Psychiatry Clin. Neurosci. 61 (2007) 651–657.
[13] V. Weichbold, H. Herka, Performance of hearing impaired children on Raven's Coloured Progressive Matrices Test, Int. J. Pediatr. Otorhinolaryngol. 67 (2003) 1213–1217.
[14] S. Tamaru, A. Kikuchi, K. Takagi, M. Wakamatsu, K. Ono, T. Horikoshi, et al., Neurodevelopmental outcomes of very low birth weight and extremely low birth weight infants at 18 months of corrected age associated with prenatal risk factors, Early Hum. Dev. 87 (2011) 55–59.
[15] N. Marlow, D. Wolke, M.A. Bracewell, M. Samara, Neurologic and developmental disability at six years of age after extremely preterm birth, N. Engl. J. Med. 352 (2005) 9–19.
[16] S.J. Dettman, D. Pinder, R.J. Briggs, R.C. Dowell, J.R. Leigh, Communication development in children who receive the cochlear implant younger than 12 months: risks versus benefits, Ear Hear. 28 (2) (2007) 11S–18S.
[17] R.F. Holt, M.A. Svirsky, An exploratory look at pediatric cochlear implantation: is earliest always best? Ear Hear. 29 (2008) 492–511.
[18] Y.S. Sininger, A. Grimes, E. Christensen, Auditory development in early amplified children: factors influencing auditory-based communication outcomes in children with hearing loss, Ear Hear. 31 (2010) 166–185.

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人工内耳装用時期と言語発達の検討

—全国多施設調査研究結果—

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要旨：感覚器障害戦略研究「聴覚障害児の療育等により言語能力等の発達を確保する手段の研究」事業として平成21年から１年間に調査した症例対照研究のうち，人工内耳装用児の現状と語音明瞭度・言語発達に関する検討を行った。対象は幼稚園年中から小学校６年までの両耳聴力レベル70dB以上の言語習得期前の聴覚障害児で，124施設が参加した。言語検査が実施できた638名のうち人工内耳装用児は285名（44.7%）であり，言語発達検査の検討はハイリスク児を除外した190名であった。人工内耳＋補聴器併用児が69.5%，片側人工内耳のみが29.8%，両側人工内耳が0.7%を占めた。難聴発見年齢は平均11.7ヶ月，人工内耳装用開始年齢は平均３歳６ヶ月であった。人工内耳装用月齢と最高語音明瞭度とは高い相関を認め，人工内耳装用開始時期が24ヶ月前とその後で言語発達検査を比較するとすべての検査項目で早期人工内耳装用児群で高い値が得られ，早期人工内耳の有効性を支持する結果となった。

ーキーワードー

人工内耳，言語発達，語音明瞭度，コミュニケーション

はじめに

感覚器障害戦略研究「聴覚障害児の療育等により言語能力等の発達を確保する手段の研究」事業の一環として，聴覚障害児の日本語言語発達に影響を与える因子を明らかにし，発達を保障する手法を確立することを目的に平成21年から１年間言語発達検査，聴覚障害児の家族・医療・教育における背景を調査する症例対照研究が行われた。

我が国で人工内耳が行われてから約25年が経過し，聴覚障害児に対する人工内耳は本邦においても定着し，様々な施設からその有効性についての報告がなされている[1)]。しかし，全国規模かつ同じ基準でその有効性について検討されたことはこれまでにない。また人工内耳装用児を対象とした日本語の言語発達に関する詳細な検討は皆無に近い現状である。今回我々は，聴覚障害児に対する本症例対照研究のうち人工内耳装用児の難聴診断年齢や新生児聴覚スクリーニング受診率等の現状と伴に，これまで指摘されて来た語音弁別能への有効性について検討した。さらに，聴覚障害児への人工内耳の日本語による言語性コミュニケーションへの有効性についても検討したので報告する。

対象と方法

本戦略研究の対象は幼稚園年中から小学校６年生までの両耳が聴力レベル70dB以上の言語習得期前聴覚障害児であり，124施設（医療施設66，聴覚特

別支援学校34，通常教育環境施設（小学校）５，難聴幼児通園施設９，大学（医療施設以外）７，その他３）が参加し，770児が登録された。評価は医療機関，特別支援学校，通常教育環境施設（小学校），難聴幼児通園施設にて行った。施行した検査は語用的能力（音声コミュニケーション能力）を評価する質問応答関係検査（TQAID），教研式標準学力検査（国語と算数），統語（文法）の能力を評価する失語症構文検査（STA），語彙の理解能力を評価する改訂版絵画語彙発達検査（PVT-R）と標準抽象語理解力検査（SCTAW），語彙の産生能力を評価する語流暢性検査（WFT），読み書き能力を評価する読み書きスクリーニング検査（STRAW），行動面に関する評価を行う広汎性発達障害日本自閉症協会評定尺度（PARS），非言語性知能検査を評価するレーブン色彩マトリックス検査（RCPM）である。これらの言語発達の評価のための検査以外に，純音聴力検査，語音弁別能検査（67S語表），装用下音場閾値検査を実施した。

登録児の内，検査項目が揃っていた児は638名（83%）であり，その内人工内耳を使用している285名（44.7%）を対象とした。また，言語発達検査の検討には，定型発達児のみで比較を行うため，体重1800g未満の児，RCPMが平均の2SD以上低い児，PARSの得点が11点以上の児を除外した人工内耳装用児190名（66.7%）を対象とした。

なお，本研究の実施にあたっては，財団法人テクノエイド協会による倫理審査委員会にて承認を得たのちに実施し，また倫理委員会がある病院等においては更に各施設内にて承認を受けたのちに実施した。

統計解析にはIBM社のSPSS 18を用いた。「早期人工内耳装用の有効性に関して」の項は，対象児全体の得点の分布より回帰式を求め，平均的な言語発達の伸びを基に，全児が100ヶ月齢時の得点となるように補正を行った後に解析を行った。検定は等分散性のためのLeveneの検定およびｔ検定を行った。

結　果

１．対象者の構成に関して

人工内耳を装用している児に占める人工内耳＋補聴器（CI＋HA）併用児の割合は69.5%（198名：男児100名，女児98名），片側人工内耳のみ（CIのみ）の児は29.8%（85名：男児35名，女児50名），両側人工内耳装用児は0.7%（２名：男児１名，女児１名）であった。図１に各学年別の人工内耳装用児の数と，各学年に占める割合を示す。難聴発見年齢の平均はCIのみが12.5ヶ月で，CI＋HAが10.9ヶ月であった。新生児聴覚スクリーニング受診率はCIのみが23.5%，CI＋HAが48.0%であった。

純音聴力検査の４分法平均値はCI＋HAで108.5 dB，CIのみが110.8dBであった。装用下閾値の４分法平均値はCI＋HAで35.2dB，CIのみが36.6dBであった。最高語音明瞭度はCI＋HAで76.1%，CIのみが84.4%であった。

２．人工内耳の早期装用開始が言語発達に及ぼす影響

人工内耳を装用した月齢は平均で42ヶ月（３歳６ヶ月）で，分布は図２に示すように，20～50ヶ月前後に手術を受けた児が多い状況であった。人工内耳の早期装用は語音弁別能（単音節の聴き取り能力）と高い相関を認めた（図３：相関係数r＝0.52）。

人工内耳の早期装用が言語発達に及ぼす影響を詳細に検討するため，24ヶ月以前に人工内耳を開始した児（29名）と，24ヶ月以降に人工内耳を装用した児（161名）の日本語発達の比較検討を行った（表１）。その結果，今回実施したほぼ全ての検査項目

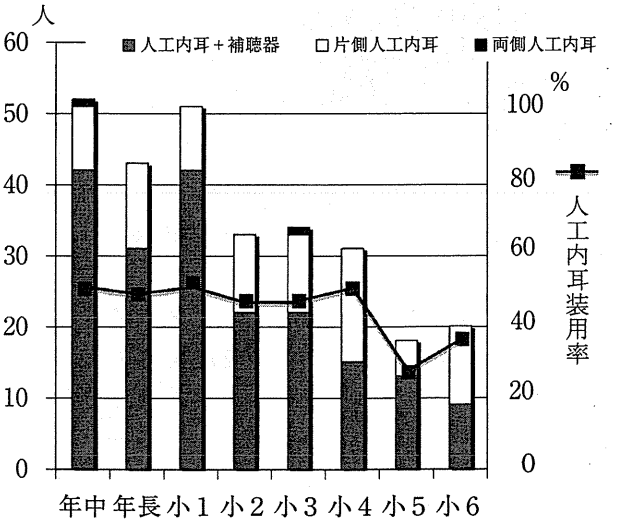

図１　各学年別の人工内耳装用者の数と，各学年に占める割合

で早期に人工内耳を装用した児のほうが平均点が高い結果が得られた。しかしながら，非言語性知能検査であるRCPMやPARSの点数には大きな違いは認められなかった。特に，WFTあかし合計，STA理解の合計点，STA産生の総得点では，早期に人工内耳を装用した児のほうが有意に得点が高い結果が得られた。

考　察

1．対象者の構成に関して

高度・重度難聴の聴覚障害児の中で人工内耳を使用している児は年中から小学4年生まで，各学年ともに50%弱であることが明らかとなった。詳細に見て行くと，小学5～6年の学年はやや少ない傾向がみられたが，これは，1998年から小児が人工内耳の適応に追加され，本邦で小児人工内耳が始まったころの学年であるためと考えられる[2]。また，小学4年，6年では片側人工内耳のみの児の割合が多かったのに対し，それより低い学年では人工内耳と補聴器を併用している割合（約70%）が多くなっていた。人工内耳がスタートした当初は，電気刺激による人工内耳と音響刺激による補聴器の中枢での統合は困難であり，かえって言語聴取は低下すると思われていた。しかし，その後電気的・音響的聴覚刺激は中枢で統合され，言語聴取成績の向上が得られるとの報告[3]が増え，本邦においても人工内耳と補聴器の併用が一般的になっている結果だと考えられる。今回の対象児の中に両側人工内耳は2名だけと少なかったが，両側人工内耳装用児の言語発達への有用性が最近多く報告[4]されるようになり，今後は両側人工内耳装用児が増えて行くと思われる。また，片側人工内耳のみの児（12.5ヶ月）よりも人工内耳と補聴器併用児（10.9ヶ月）の方が難聴発見年齢が早いことが明らかとなった。これは，新生児聴覚スクリーニング受診率が人工内耳と補聴器併用児（48.0%）で高かった事が1つの要因と考えられた。

人工内耳装用児は裸耳聴力レベルが高くても（平

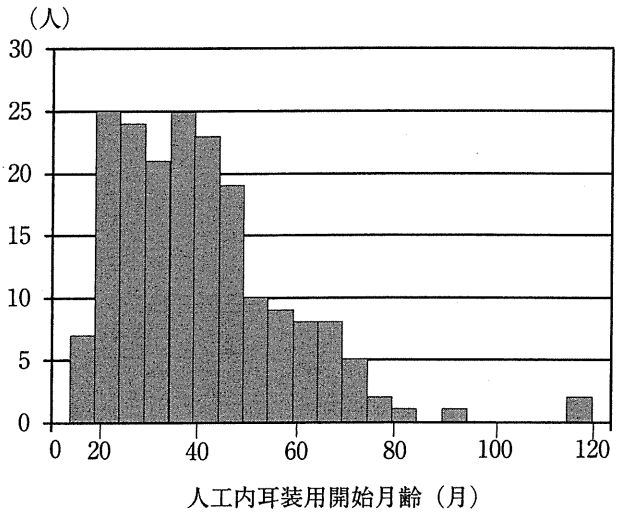

図2　対象者の人工内耳装用開始月齢

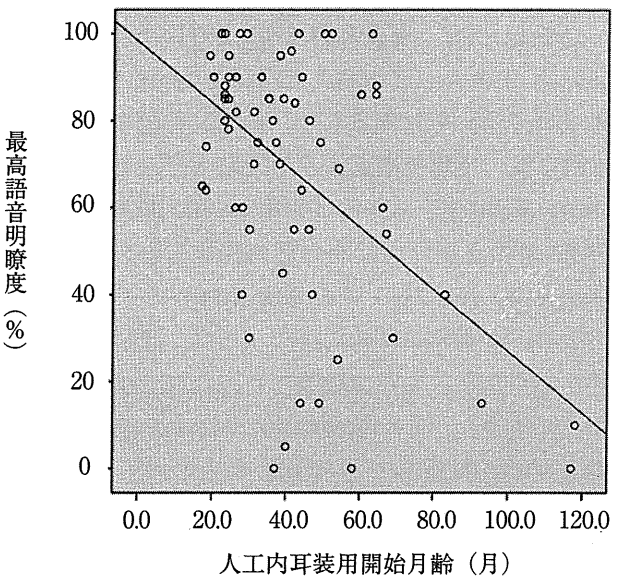

図3　人工内耳装用開始月齢と最高語音明瞭度の関係
相関係数 r＝0.52

表1　早期に人工内耳を装用した児と24ヶ月以降に人工内耳を装用した児の平均点
（回帰式を用いた補正により，両群とも100ヶ月齢時の得点に換算してある。）

	PVT-R 修正得点	WFT あかし計	WFT 動物	SCTAW 正答率	STA 理解 合計点	STA 産生 総得点	CRT-II 国語 合計点	CRT-II 算数 合計点	質問応答 関係検査	RCPM 合計点	PARS 現在得点
24ヶ月以降（29名）	29.5	12.9	9.5	13.7	23.1	34.1	46.1	57.0	209.4	27.5	5.3
24ヶ月前（161名）	32.2	15.6	10.6	13.8	26.2	39.0	52.3	59.8	229.7	28.1	4.5
有意確率（t検定）	0.19	0.02	0.07	0.99	0.04	0.04	0.09	0.39	0.06	0.77	0.30

均109.7dB)，装用下閾値は低く（平均35.9dB)，良好な語音明瞭度（平均80.3%）が得られていた。以前より低体重出生児は発達に何かしらの影響がみられやすいと言われており[5]，今回の言語関連の評価対象児を体重1800g未満の児，RCPMが平均の2SD以上低い児，PARSの得点が11点以上の児を除外した定型発達児に限定した事が，より明確な結果を導けた要因と考える。

2．早期人工内耳装用の有効性に関して

人工内耳の早期装用は，語音弁別能と高い相関を認めた。このことより，人工内耳を早期から装用することで，高い語音弁別能が得られる可能性が高いことが示唆された。海外でも早期人工内耳により，言語の理解のみならず産生にも有効であるとの報告[6]がみられるが，日本語の発達においても同様の事が確認できたと思われる。

今回の検査対象は，人工内耳の装用時期が20ヶ月～50ヶ月がピークで，平均は42ヶ月齢と，現在の小児人工内耳と比較するとやや装用時期が遅い傾向にあった。これは，前項と同様，1998年に本邦における人工内耳の適応に小児が追加され，徐々に適応年齢が低年齢化しているためだと考えられた。

今回行った24ヶ月未満に人工内耳を装用した早期装用の児と24ヶ月以降に人工内耳を装用した児の日本語言語発達の比較では，ほぼ全ての項目で早期人工内耳装用児の方が言語発達が良好であり，早期に人工内耳を装用することが，言語発達の全方面に有効であることが示唆された。それ以外に人工内耳後の言語発達には聴力レベル，難聴の期間，使用期間，IQ，ワーキングメモリー，家族の収入などが関与すると報告されている[7]。今後更に装用年齢による影響を含め，更に詳細なデータの解析を行い，人工内耳装用児の言語発達に影響する項目の関連を評価していきたいと考えている。

人工内耳を2歳代で受けた児より1歳代で受けた児の方が言語発達が早いとの報告も見られ，人工内耳の低年齢化が進んでいる[8]。人工内耳の早期装用開始のためにも早期診断が重要であり，今後，新生児聴覚スクリーニング受診率の向上に努めることが重要であることが改めて示されたと思われる。

ま と め

感覚器障害戦略研究「聴覚障害児の療育等により言語能力等の発達を確保する手段の研究」事業の一環として平成21年から1年間言語発達検査，聴覚障害児の家族・医療・教育における背景を調査する症例対照研究が行われたうち，人工内耳装用児の現状と日本語による言語性コミュニケーションへの有効性について検討した。聴覚障害児に対する早期人工内耳は言語・コミュニケーション能力の発達をもたらすことが確認され，難聴の早期発見に今後も取り組む必要がある。

謝　辞

感覚器障害戦略研究「聴覚障害児の療育等により言語能力等の発達を確保する手段の研究」に参加してくださった聴覚障害児とすべての施設のスタッフに心から感謝申し上げます。なお，本戦略研究実施団体である財団法人テクノエイド協会の支援を得て実施している。

Evaluation of the relation between speech development and the age at cochlear implantation

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The Research on Sensory and Communicative Disorders (RSCD) was originally planned as a nationwide research project to assess the effectiveness

of interventional methods for hearing-impaired children. Of the 638 hearing-impaired children who participated in the RSCD project in 2009, 282 (44.2 %) were cochlear implant users. Of these, 196 (69.5%) were both cochlear implant plus hearing aid (bimodal stimulation) users, 84 (29.8%) were unilateral-cochlear implant only users, and 2 children (0.7%) were bilateral cochlear implant users.

The average hearing loss level was 109.7dB. However, good outcomes of the hearing threshold (35.9dB) and maximum speech discrimination score (80.3%) were observed. A high correlation ($r=0.52$) was found between the age at cochlear implantation and the maximum speech discrimination score. The scores on the speech and language tests in the children who were under 24 months of age at the time of the cochlear implant surgery were better than those in the children who were over 24 months of age at the time of the surgery. The results of our study indicate that early implantation is beneficial for speech and language development.

参考文献

1) 福島邦博：人工内耳装用児の長期的な言語発達。JOHNS **21**：601-603, 2005
2) 伊藤壽一：人工内耳の適応に関する考察。耳鼻臨床 **104**：1-6, 2011
3) Potts LG, Skinner MW, Litovsky RY, et al: Recognition and localization of speech by adult cochlear implant recipients wearing a digital hearing aid in the nonimplanted ear (bimodal hearing). J Am Acad Audiol **20**: 353-373, 2009
4) Litovsky RY, Parkinson A, Arcaroli J: Spatial hearing and speech intelligibility in bilateral cochlear implant users. Ear Hear **30**: 419-431, 2009
5) Tamaru S, Kikuchi A, Takagi K, et al: Neurodevelopmental outcomes of very low birth weight and extremely low birth weight infants at 18 months of corrected age associated with prenatal risk factors. Early Hum Dev **87**: 55-59, 2011
6) Kirk KI, Miyamoto RT, Lento CL, et al: Effects of age at implantation in young children. Ann Otol Rhinol Laryngol Suppl **189**: 69-73, 2002
7) Moon IJ, Kim EY, Chu H, et al: A new measurement tool for speech development based on Ling's stages of speech acquisition in pediatric cochlear implant recipients. Int J Pediatr Otorhinolaryngol **75**: 495-499, 2011
8) Dettman SJ, Pinder D, Briggs RJS, et al: Communication development in children who receive the cochlear implant younger than 12 months: risks versus benefits. Ear Hear **28** (suppl): 11S-18S, 2007

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当院にて手術を施行した人工内耳装用児の言語発達評価

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要旨：本邦で人工内耳手術が開始された初期の難聴児は既に学齢期に達しており，その言語発達の評価が可能な年齢に達している。

当科で人工内耳埋め込み術を実施し，現在難聴幼児通園施設や小学校に在籍している26名の言語発達データを感覚器障害戦略研究聴覚分野にて収集された全国の人工内耳装用児184名と比較した。当科での実施例は新生児聴覚スクリーニング（NHS）の受検率が高く，生後６ヵ月以内の早期に補聴を開始できている割合が有意に高かった。また，言語検査結果を比較したところ，要素的な言語力では平均値で大きな違いはみられなかったが，当科群では，国語の学力試験結果が有意に良好であった。

システムとして整備された質の高い早期補聴から人工内耳に至るまでの道筋が，手術後の学力の伸びにつながる可能性が示唆された。

ーキーワードー
人工内耳，新生児聴覚スクリーニング，早期補聴，言語発達

はじめに

平成18年の適応基準の改訂以降，本邦では１歳半以上の難聴児に対する人工内耳手術が適応となった。それに伴い，当院における小児の人工内耳手術症例も年々増加する傾向にある。当院では小児の人工内耳手術を開始した平成４年以降，手術を施行された15歳以下の難聴児は154名で，さらに，平成18年以降に限ると，68名であった。特に５歳未満の低年齢の難聴児においては平成18年以降の手術件数が57例と，近年人工内耳手術の低年齢化が顕著である。

人工内耳手術が開始された初期の難聴児は学齢期に達しており，幼児期に人工内耳を装用した児童のその後の言語発達や学力についての評価が可能となっているが，これまで本邦で人工内耳装用児の言語発達が大規模に調査された例はなく，その実態は明らかではない。

感覚器障害戦略研究では，全国から781名のエントリーがあり，そのうちの約45％が人工内耳装用児であった[1)]。今回，当院にて人工内耳埋め込み術を実施し，現在難聴幼児通園施設および小学校に在学中の小児を対象に，言語発達や人口動態的背景を検討し，全国データとの違いを比較したので報告する。

対象と方法

１．対象

平成21年４月１日から平成22年３月31日までに感覚器障害戦略研究聴覚分野（以下，戦略研究）にお

いて書面で同意が得られた難聴児が781名であった。これらの難聴児に標準純音聴力検査，語音明瞭度検査，言語検査などの各検査を行い，保護者および療育者には質問紙調査を行った[1]。このうち，4歳から12歳までの出生時から高度の難聴を有する児（両耳の平均聴力が70dB以上）の基準を満たした難聴児は638名であった。また保護者に対する質問紙調査にて人工内耳の有無を問う項目から，人工内耳手術を受けている児が294名存在した。この中で，言語発達に影響を及ぼす可能性のある合併症を有すると考えられる児を解析から除外した。その内訳は出生時体重が1830g以下の児（n＝15），知的発達障害合併疑いの児（Raven's Coloured Progressive Matrices：以下，RCPMが学年の平均－2SD以下の児）（n＝4），および読み書き障害合併疑いの児（Screening Test of Reading and Writing for Japanese Primary School Children：以下，STRAWが学年の平均－1.5SD以下の児）（n＝65）である。その結果，今回の研究の対象者は210名となり，このうち当院で人工内耳手術を施行した児が26名でこれをA群，それ以外の児が184名でこれをB群と定義した。図1

尚，この研究は公益財団法人テクノエイド協会および岡山大学の倫理委員会により承認を受けた。

2．方法

対象となった児の背景因子（性別，新生児聴覚スクリーニング：Newborn hearing screening：以下NHSの有無，早期療育の有無）をPearsonのカイ二乗検定にて検討した。早期療育については，過去

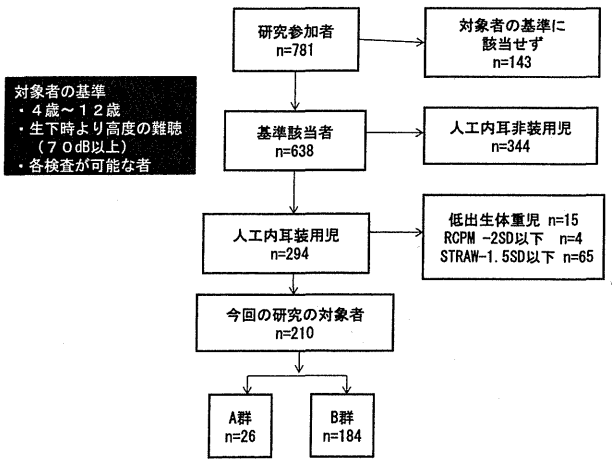

図1　研究参加者の内訳

の文献[2][3]より生後6ヵ月以内の補聴器装用開始とした。次に，2群の月齢，良聴耳の裸耳聴力，装用時聴力，補聴器または人工内耳装用下での自由音場による最高語音明瞭度，および人工内耳の装用期間をt検定にて検討した。また，言語検査の結果は年齢の違いによる得点の変化を調整するために偏差値化した上で，t検定にて解析を行った。偏差値化とは，今回の感覚器障害・戦略研究で得られた難聴児のデータについて学年ごとに各言語検査の平均値および標準偏差を求めた上で，各学年の平均値を50とし，標準偏差を10として算出した値である。この偏差値化を行うことにより，その難聴児が同学年の中でどのような位置に属するかが示されるため，各学年を一緒に解析することが可能である。尚，施行した言語検査は質問応答関係検査（Test of question-answer interaction development：以下TQAID），語流暢性検査（Word Fluency Test：以下WFT），絵画語彙検査（Peabody Picture Vocabulary Test-Revised：以下PVT-R），抽象語理解力検査（Screening Test for Abstract Words：SCTAW），失語症構文検査（Syntactic Processing Test for Aphasia：STA）および教研式学力検査（Criterion Referenced TestII：以下CRT-II）である。尚，SCTAWおよびCRT-IIについては就学以降の児にのみ実施し，いずれも有意水準は5％とした。

結　果

1．背景因子　表1

性別は，A群：男性13人（50％）・女性13人（50％）と同数，B群：男性85人（46.2％）・女性94人（51.1％）・不明5人（2.7％）であり，両群に有意な差があるとはいえなかった。

NHSの有無ではA群：受検15人（57.7％）・未受検11人（42.3％），B群：受検56人（30.4％）・未受検118人（64.1％）・不明10人（5.5％）であった。早期補聴の有無ではA群：6ヵ月以前11人（42.3％）・7ヵ月以降15人（57.7％），B群：6ヵ月以前38人（20.7％）・7ヵ月以降146人（79.3％）であり，A群ではNHSの受検率および早期の補聴開始率が有意に高い結果となった。（$p<0.05$）

月齢についてはA群：96.6ヵ月，B群：89.2ヵ月，良聴耳の裸耳の平均聴力（3分法）については

表1　A群とB群の背景因子

背景因子	A群	B群	p値
月齢	96.6ヵ月	89.2ヵ月	0.185
性別	♂13　♀13	♂85　♀94	0.836
NHS受検率	57.7%	30.4%	0.015
早期補聴の割合	42.3%	20.7%	0.024
良聴耳の裸耳聴力（3分法）	107.7dB	106.4dB	0.662
装用時の平均聴力（3分法）	38.0dB	32.0dB	0.001
装用下での最高語音明瞭度	80.6%	68.9%	0.037
人工内耳装用期間	56.7ヵ月	47.7ヵ月	0.100

表2　A群とB群の言語検査結果

言語検査結果	A群	B群	p値
TQAID	51.3	52.0	0.741
WFT	48.8	51.0	0.329
PVT-R	54.0	51.0	0.155
SCTAW	54.2	51.8	0.309
STA理解	53.2	51.4	0.418
STA産生	51.2	51.2	0.988
CRT-II国語	57.4	52.9	0.002
CRT-II算数	55.4	52.7	0.179

A群：107.7dB，B群：106.4dBでありA群とB群には有意な差があるとはいえなかった。装用時の平均聴力（3分法）についてはA群：38.0dB，B群：32.0dBであり，B群がA群より有意に良い結果となった。（$p<0.01$）一方で補聴器または人工内耳装用下での自由音場による最高語音明瞭度はA群：80.6%，B群：68.9%であり，A群がB群より有意に良好な結果となった。（$p<0.05$）また，人工内耳装用期間ではA群：56.7ヵ月，B群47.7ヵ月と両群に有意な差があるとはいえなかった。

2．言語検査結果　表2

PVT-R，SCTAW，STA理解，CRT-II算数ではA群がB群よりも良好な結果を示した。また，TQAID，WFTではB群がA群よりも良好な結果であったが，両群に有意な差があるとはいえなかった。一方で，CRT-IIの国語では平均値がA群：57.4（n=14），B群：52.9（n=79）でA群がB群よりも有意に良好な結果となった。（$p<0.01$）CRT-II国語の箱ひげ図を図2に示す。

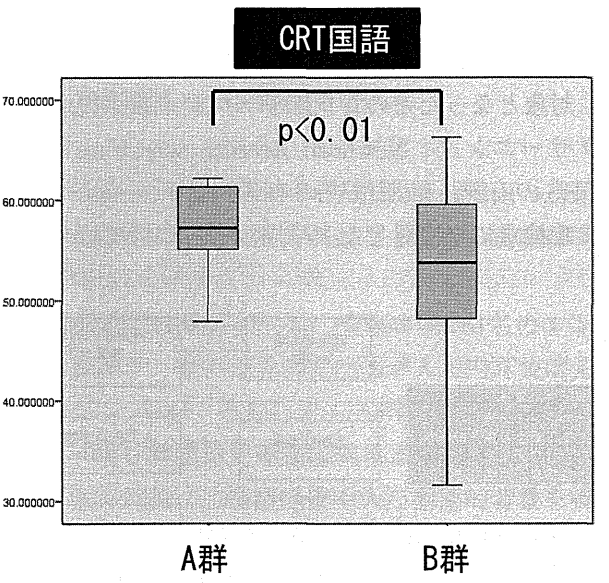

図2　A群とB群のCRT-II国語の箱ひげ図（A群が有意に良好な結果であった）

考　察

人工内耳装用児の言語発達についてはこれまで多くの議論がなされているが，人工内耳の手術時期，装用期間，術前の聴力，および補聴器の装用開始時期がその後の言語発達を予測する因子となりうると報告されている[4]。また，手術時年齢がより小さい方が言語発達が良好であるとされる報告は多い[5)6)]。しかし，多くの合併症のリスクを持つ児においては補聴効果の限界が明らかになりやすく，結果として人工内耳手術時期がより早くなる可能性があることや，逆に進行性・遅発性難聴の児で，当初補聴効果が見られた場合には必ずしも低年齢で手術を行わないことから，こうした個々の条件を統制した上での言語発達の比較は実際には困難を伴うことが多い。

今回の検討では，当科で人工内耳手術を施行した児の特徴を検討した。その結果，NHSにて難聴が早期に発見されている児の割合，および生後半年以内の早期に補聴を開始している児の割合が有意に高いことが明らかとなった。また，言語検査の結果を比較したところ，就学期以降における国語の学習習得度が有意に良好であることが示された。一方で，要素的言語力については有意な差は認められなかった。

NHSの有用性については多くの報告があるが，最近の報告ではNHSの有無のみでは言語性コミュニケーションに有意な差はみられないが，NHS受検により生後6ヵ月以内の早期補聴が可能となる確率が約20倍になるとされている[7)]。その他の報告[8)9)]でも，NHS導入により難聴発見時期や補聴開始時期が早くなったとされており，NHSが早期補聴にもたらす影響は大きいと考えられる。一方で，NHSの受検の有無と言語発達について明らかな関連を述べている文献は少ない。本邦ではNHSのモデル事業が開始されてから約10年が経過しているが，現在の小学校高学年以上はNHS未受検の児が大多数を占めることや，NHS受検がその後の補聴器装用開始に必ずしもつながっていない児の存在があること[7)]などから，NHSの有無とその後の言語発達の関連について議論するためには，今後長期的な視点での追跡調査が必要であると考える。

早期補聴と言語発達の関連についても多くの報告がみられるが，音声言語の入力には臨界期があるため，良好な音韻処理能力を獲得するためには生後6ヵ月を目安に補聴を開始することが推奨されている[10)]。笠井ら[7)]は，生後半年以内の早期に補聴を開始することにより，言語性コミュニケーションが約3倍良好となると述べている。当科で手術を施行した児は，人工内耳手術後の装用時聴力は全国の装用者より有意に不良であったが，語音明瞭度が有意に良好であった。これは早期に補聴を開始しているために音韻処理能力が獲得された結果と考えられた。このため，将来的にどのようなデバイスを用いることになったとしても，いわゆる「1-3-6rule」に基づいた難聴の早期発見および早期介入が重要であることが改めて認識されるべきである。

今回施行した言語検査において，当科での手術群が有意に良好であったのが国語力であったが，検査に用いたCRT-IIでは，国語力の中で読み書きの能力や言語についての知識・理解・技能を測定している。Geersら[11)]は就学前に人工内耳手術を施行された難聴児の小学校低学年の時点と高校生の時点での読み書き能力や音韻処理能力を追跡調査し，正常聴力の児と比較検討を行なっている。この報告では，人工内耳装用児は小学校低学年の時点では年齢相応の読み書き能力を有しているが，高校生になると書き取りや文章表現に困難をきたすと述べている。さらに，音韻処理能力が高校生の時点での読み書き能力の予測因子となりうると述べている。当科群で国語力が有意に良好であった要因としては，早期補聴の割合が有意に高く，良好な音韻処理能力が獲得された結果と考えられ，この報告とも一致する結果となった。

一方で，当科での手術群およびその他の群において言語性コミュニケーションや語彙・構文などの要素的言語力については有意な差があるとはいえなかった。これは，補聴器の装用開始時期が遅れたとしても，その後の適切な療育により要素的言語力はキャッチアップすることが可能であるためだと考えられる。また，特に構文検査であるSTAについて有意な差が認められなかった要因としては，構文の獲得には早期に獲得されるものと後期に獲得されるものの2つがあるとされること[12)]も一因と考えられる。このため，STAの結果をこういった構文の獲

得年齢における観点から解析することも必要であると考えられた。また，構文については，特に後期に獲得されるものの到達度を確認するため，就学以降での言語検査の実施を行うこと，また遅れがみられる難聴児には適切な介入を考慮すべきである。

NHSのモデル事業が本邦にて開始されて約10年が経過しており，難聴の早期発見のシステムは定着してきているが，その後の介入方法については地域により様々であり，一定の対策がとられていないのが現状である。岡山県では平成13年度よりNHSのモデル事業が開始され，現在では人口動態統計からみた県内の対象人口の約80％がスクリーニングを受けている。このように岡山県でNHSの受検率が高い要因としては，県内の47産科医療機関がNHSに参加しており，その中では100％に近い実施率がみられていることや，里帰り分娩などによるスクリーニング漏れを防止するために外来スクリーニングを導入していることなどが挙げられる。また，難聴の診断が確定した児の療育は，岡山かなりや学園に集約されている。難聴児の療育施設を統一することによって，県内在住の難聴児が均霑化（きんてんか）された療育が受けられること，また同園では岡山大学に所属する医師が定期的に診察を行なっており，大学との連携体制ができていることが，補聴器や人工内耳の導入が適切な時期になされることに寄与していると考えられる。

今回の検討からは，人工内耳を装用した上での言語発達および学力を論じるためには，人工内耳手術の有無だけではなく，NHSとその後に続く療育および教育などの人工内耳をサポートするさまざまな仕組みを含めたパッケージとして，対策を考える必要があると考えられた。このように，難聴児に対する療育の一体化されたシステムがさらに確立することを期待する。

結　語

当科において手術を施行した人工内耳装用児のデータでは，NHSの受検および生後6ヵ月以内の早期に補聴を開始している児が有意に多かった。また，国語力が有意に良好な結果を示した。難聴の早期発見とその後の早期補聴から人工内耳手術への一体化されたシステムが手術後の国語力の伸びに繋がる可能性が示唆された。

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Language development of prelingually deafened children with cochlear implant.

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With over 20 years of experience in the use of cochlear implants in Japan, many of the prelingual users have reached school age and are able to be subjected to tests for language development.

Language development was compared between 26 prelingually-deafened children who had received cochlear implants at Okayama University and 184 cochlear implant users enrolled in the Research on Sensory and Communicative Disorders（RSCD）project. The demographic background demonstrated a significantly wider prevalence of subjects who had undergone newborn hearing screening and earlier commencement of interventions in our group. Significantly better achievement of Japanese Language skills was observed in our group, although no significant differences were observed in the results of domain-based language tests.

Systematic approaches from early identification to cochlear implantation may allow better academic achievement among school-aged cochlear implant