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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 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).

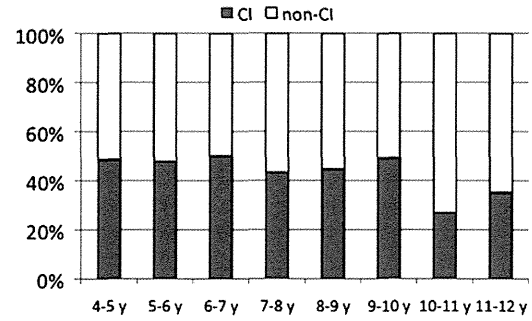

Fig. 1. The rate of CI-only users among the participating hearing-impaired children of each age group. About 45% of hearing-impaired children (>70 dB hearing level) in this study were CI-only users. CI: cochlear implant.

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.

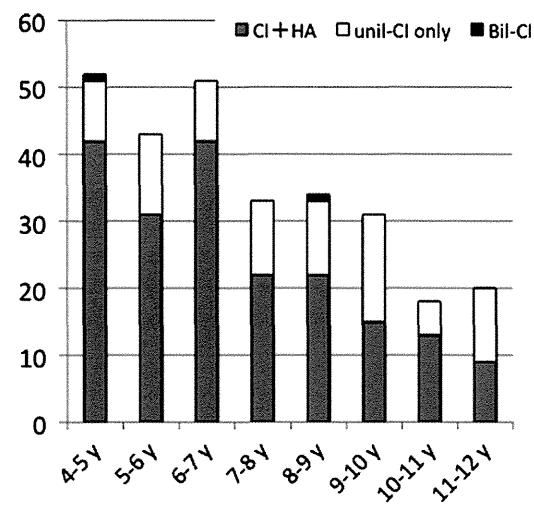

Fig. 2. Number of children in the CI–HA, unil CI-only, and bil-CI groups in each age range. CI plus HA users (bimodal stimulation) make up the majority of CI users. CI: cochlear implant; unil CI: unil CI-only users; bil-CI: bilateral CI users; CI–HA: HA and CI users.

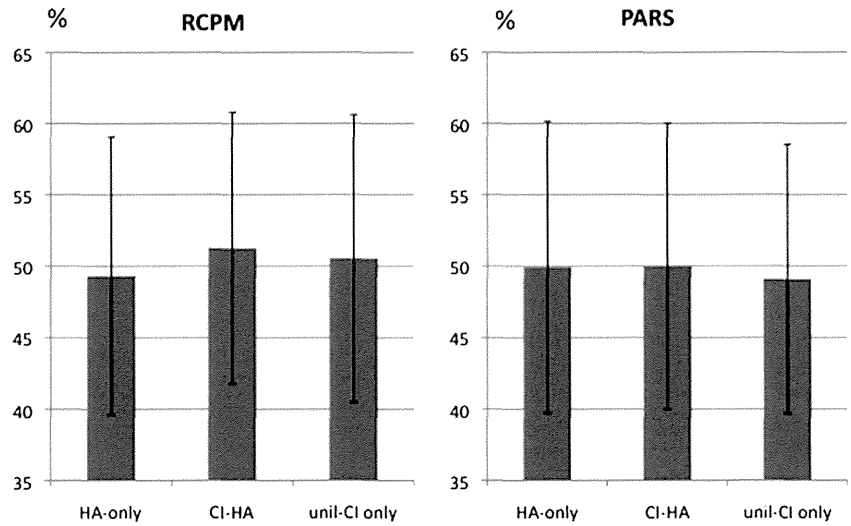

Fig. 3. The scores of PARS and RCPM tests in the HA-only, CI–HA, and unil CI-only groups. There were no significant differences in average scores among the groups. PARS: Pervasive Developmental Disorder ASJ Rating Scales for evaluating autistic tendency; RCPM: Raven's Colored Progressive Matrices test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

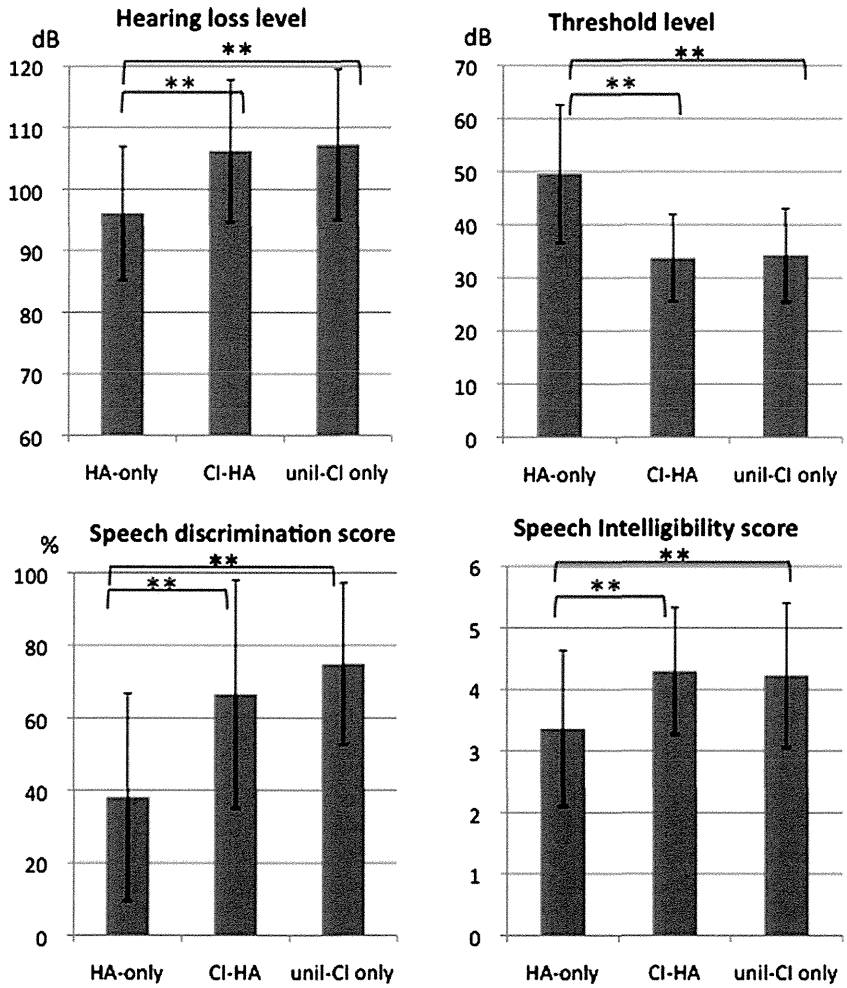

Fig. 4. Hearing loss levels, threshold levels, maximum speech discrimination scores, and speech intelligibility scores in the HA-only, CI–HA, and unil CI-only groups. There are significant differences ($p < 0.01$) in hearing levels, threshold levels, speech discrimination scores, and intelligibility 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,

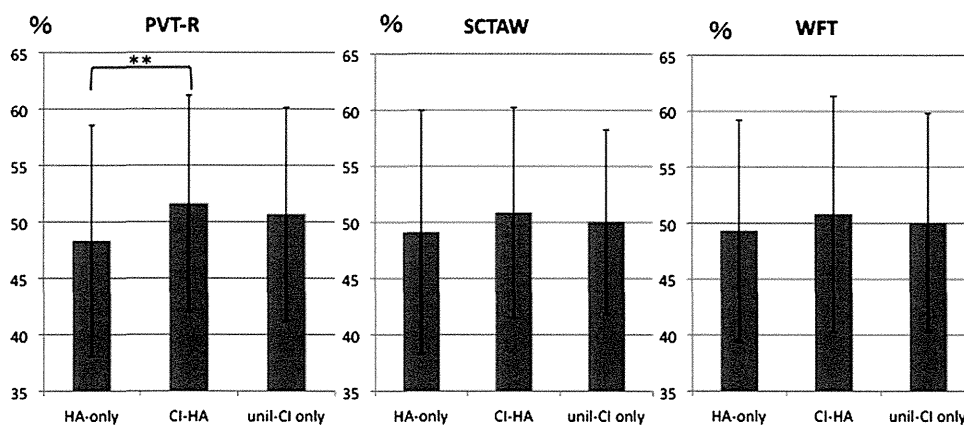

Fig. 5. Scores of the PVT-R, SCTAW, and WFT tests in the HA-only, CI–HA, and unil CI-only groups. Scores of the PVT-R, SCTAW, and WFT tests in the CI–HA and unil CI-only groups were better than those in the HA-only group. A significant difference ($p < 0.01$) was found in the scores of the PVT-R test. $^{**}p < 0.01$, 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.

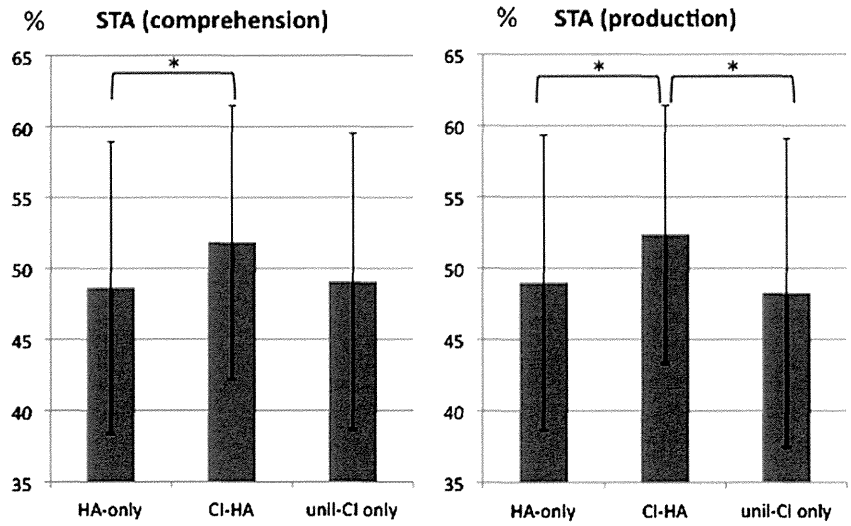

Fig. 6. Scores of the STA test (comprehension and production) in the HA-only, CI–HA, and unil CI-only groups. STA test scores (comprehension) in the CI–HA group were significantly higher ($p < 0.05$) than in the HA-only group. STA test scores (production) in the CI–HA group were significantly higher ($p < 0.05$) 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).

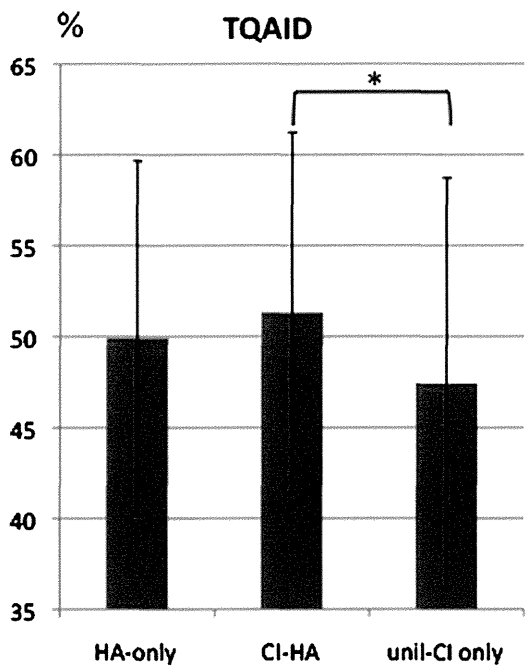

Fig. 7. The scores of TQAID test in the HA-only, CI–HA, and unil CI-only groups. The score of TQAID test in the CI–HA group is significantly ($p < 0.05$) better than that in the unil CI-only group. $^{*}p < 0.05$, TQAID: test for question-answer interaction development is for evaluating the IPCS (interpersonal 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).

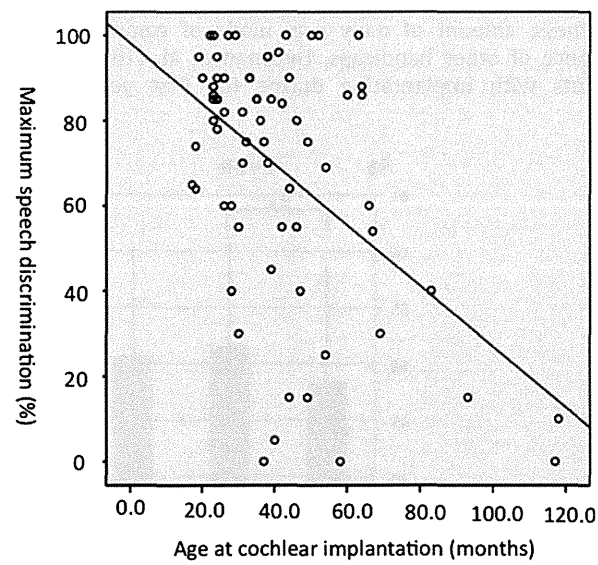

Fig. 8. The correlation between the age of cochlear implantation and maximum speech discrimination score. The high correlation ($r = 0.52$) has been found between the age of CI and maximum speech discrimination score.

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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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

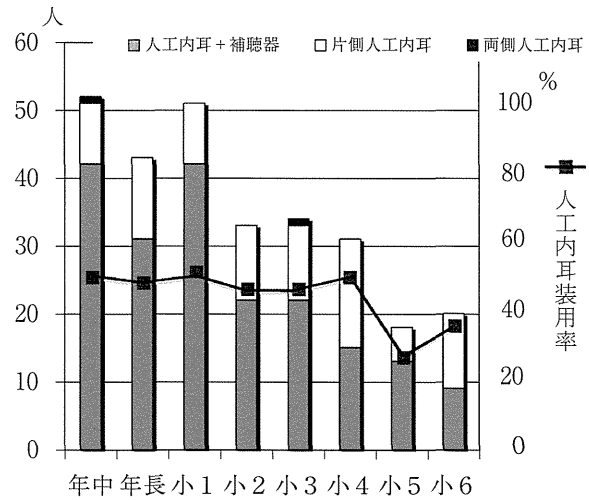

図１　各学年別の人工内耳装用者の数と，各学年に占める割合

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

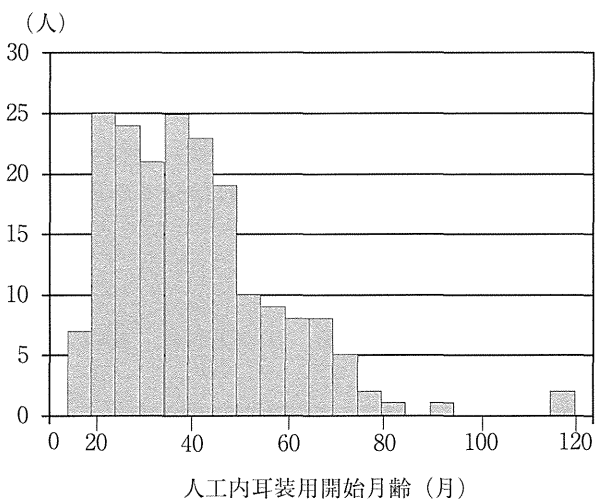

図2　対象者の人工内耳装用開始月齢

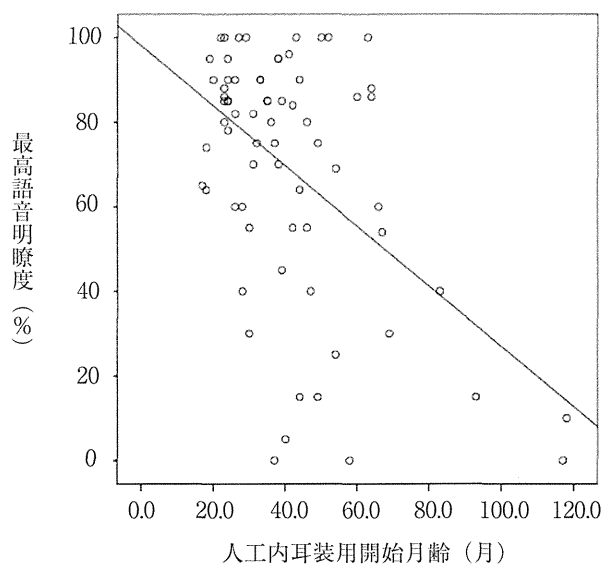

図3　人工内耳装用開始月齢と最高語音明瞭度の関係
相関係数 r＝0.52

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.

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人工内耳と補聴器の装用開始年齢による言語発達検査結果の検討

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要旨：平成21年から22年の間，聴覚障害児の日本語言語発達に影響を与える因子と，発達を保障する方法を考える目的で行われた感覚器障害戦略研究・症例対照研究のデータを元に，人工内耳手術年齢による言語発達の傾向について検討した。

対象は生下時から聴覚障害を持つ４歳から12歳までの平均聴力レベル 70dB 以上の638名のうち，聴覚障害のみを有すると考えられる人工内耳装用児182名を，補聴器の装用開始年齢，人工内耳の手術年齢のピークで４群に分け比較した。

結果を語彙，構文レベルの発達と，理解系，産生系課題とに分けてみると，語彙，構文共に，理解系の課題においては，補聴器装用早期群の成績が良好で，産生系の課題については，人工内耳手術年齢早期群の成績が良好であった。

このことから，早期に音を入れることが言語理解に，早期に十分弁別可能な補聴をすることが言語産生に影響を与える可能性があるという結果が得られた。

ーキーワードー

人工内耳，手術年齢，言語発達，感覚器障害戦略研究（聴覚分野）

はじめに

感覚器障害戦略研究（聴覚分野）症例対照研究は，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，発達を保障する方法を考える目的で平成21年４月～平成22年３月までの１年間124施設で行われた。実施内容は，家族・医療・教育を対象としたアンケートによる背景因子の調査と，語彙・構文・コミュニケーション力を見る日本語言語発達検査である。これらの結果から，聴覚障害児の言語発達の現況を捉え検討を行うことが可能となった。岩崎ら[1)]が人工内耳装用時期を24ヶ月前と後で言語発達検査結果を比較し，早期人工内耳の有効性を示したが，人工内耳を早期に装用している児は，補聴器の装用も早いことが予想される。そこで，人工内耳と補聴器の装用時期による言語発達の差異と傾向について検討したので報告する。

対　　象

生下時から聴覚障害を持つ４歳から12歳までの平均聴力レベル 70dB 以上の638名のうち，出生時の体重が 1800g 未満の児，広汎性発達障害日本自閉症協会評定尺度（PARS：スペクトラム出版社）の現在得点が11点以上の児，レーヴン色彩マトリック

ス検査（RCPM：日本文化科学社）において平均点－2SD以下の児，生下時に何らかの合併症（頭頸部奇形，髄膜炎，循環器疾患，呼吸器疾患など）を有する児および周産期リスク（先天風疹症候群，先天CMV感染症，胎児仮死，重症黄疸など）を有する児を除いた419名のうち人工内耳を装用している児182名（男児：83名，女児：99名）を対象とした。

方　法

上記児童を対象に，アンケートを実施して難聴発見時期，補聴器装用開始時の月齢および人工内耳手術時の月齢を調べた。また，言語発達検査として語彙レベルの発達を評価する事を目的に，改訂版絵画語い発達検査（PVT-R：日本文化科学社）および語流暢性検査（WFT）を，構文レベルの発達を評価する事を目的に失語症構文検査（STA：国際医療福祉大学大学院医療福祉学研究科）を実施した。また，コミュニケーション能力の発達を評価する事を目的に質問―応答関係検査（TQAID：エスコアール）を実施した。

PVT-Rでは修正得点を得点として用いた。また，WFTでは音韻課題の「あ」，「か」，「し」およびカテゴリー課題の「動物」の各1分間の産生語彙数の合計数を得点として用いた。STAでは，理解の合計得点をSTA理解の得点とし，産生の総得点をSTA産生の得点とした。またTQAIDでは総得点を採用した。上記の言語検査の成績は，全て素点を学年ごとに偏差値化したうえで集計し解析を行った。以下，言語発達検査結果の数値は全て偏差値を示す。平均点の比較における有意差検定は分散分析（Tukey HSD）により実施した。

結　果

対象児童の補聴器・人工内耳装用開始時期

対象児182名の難聴発見時期は，生下時から3歳9ヶ月であり，平均1歳0ヶ月であった。また補聴器装用開始月齢は，0歳2ヶ月から3歳11ヶ月であり，平均1歳3ヶ月であった。補聴器装用開始時期の分布については，図1に示すように16ヶ月前後を境界に2峰性の分布を示しており，6ヶ月前後と24ヶ月前後にピークが認められた。また，人工内耳手術時の月齢は，1歳5ヶ月～9歳10ヶ月，平均3歳6ヶ月であった。

一般的に，人工内耳を早期から装用している児は補聴器の装用開始も早いことが考えられるため，人工内耳の早期装用開始が言語発達に及ぼす影響について検討を行う上で，人工内耳を早期に実施した影響と，補聴器の早期装用による影響を区別して解析することが必要であると考えた。

そこで，本研究では，補聴器の装用開始月齢の分布（図1）を元に，補聴器の装用開始16ヶ月未満の児と16ヶ月以降の児の2群に分けた。さらに，それぞれの群の人工内耳手術時月齢の分布（図2）を元に，人工内耳手術年齢のピークの前後で2群に分け，合計4群での比較を行った。

補聴器装用開始年齢16ヶ月未満の群においては，人工内耳手術時月齢のピークが24ヶ月であった為，24ヶ月未満群と，24ヶ月以降群とに分けた。また補聴器装用開始年齢16ヶ月以降の群においては，人工内耳手術時月齢のピークが36ヶ月であった為，36ヶ月未満群と36ヶ月以降群に分けて比較を行った。以上の操作により，1）補聴器・人工内耳とも早期装用の群（HA・CI早期群），2）補聴器は早

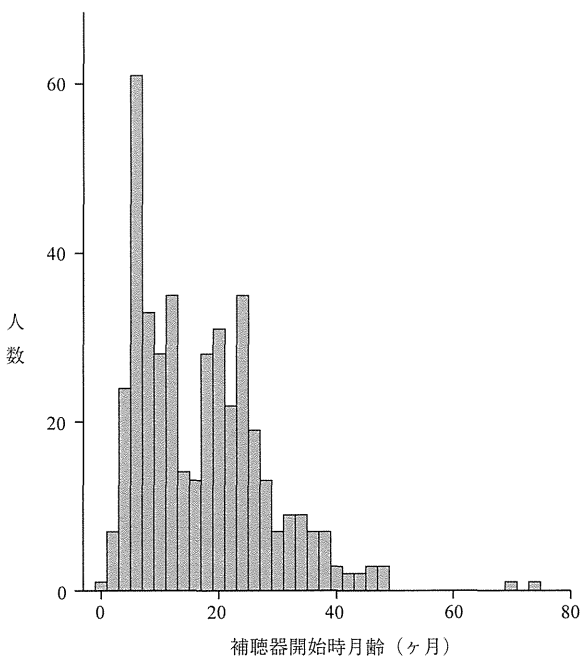

図1　補聴器装用開始時期（月齢）の分布
本研究に参加した児（638名）の補聴器装用開始時の月齢の分布を示す。16ヶ月前後を境界に6ヶ月前後と24ヶ月前後にピークを持つ2峰性の分布を示していることが分かる。

期装用だが，人工内耳はピーク以降の群(HA早期・CI非早期群)，3）補聴器装用開始は16ヶ月以降だが人工内耳は3歳未満と早期の群（HA非早期・CI早期群)，4）補聴器装用開始16ヶ月以降かつ人工内耳装用3歳以降の群（HA・CI非早期群）の4群で比較検討を行った。

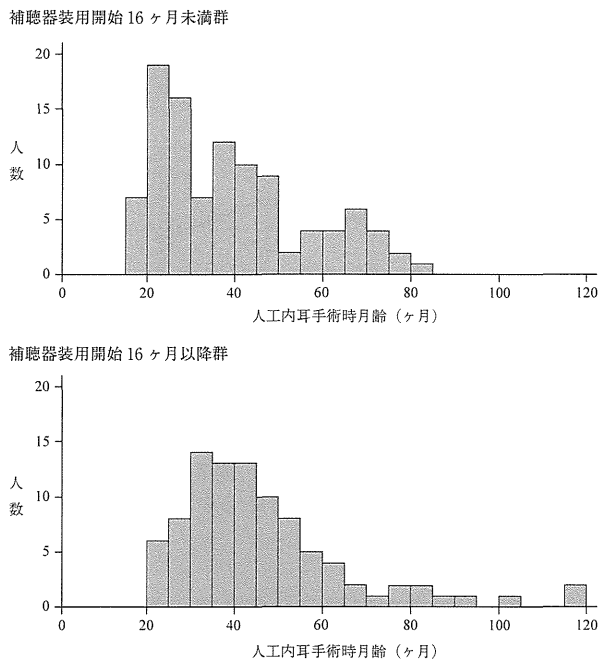

図2　人工内耳の手術時期（月齢）の分布
補聴器装用開始時月齢16ヶ月未満と16ヶ月以降の2群それぞれの人工内耳手術時の月齢の分布を示す。補聴器装用開始16ヶ月未満の群では24ヶ月が分布のピークであり，16ヶ月以降の群では36ヶ月がピークであった。

背景因子の比較

除外基準適用後のデータについて，4群間でその背景に差が無いかを検討した。その結果，新生児聴覚スクリーニングの受検率に関しては，補聴器を早期に装用開始した児では受診率が高く，新生児聴覚スクリーニングにより難聴が早期に発見され，早期補聴器装用に繋がることが改めて示された。また，検査実施時期に関してはHA・CI早期群がやや早く，HA・CI非早期群がやや遅い傾向が認められた。裸耳聴力および装用閾値に関しては大きな差を認めなかったが，最高語音明瞭度に関しては，人工内耳を早期に装用した群のほうが良好であった。

また，非言語性知能発達の指標であるRCPM（レーヴン色彩マトリックス検査）の偏差値では，HA・CI早期群が49.0，HA早期・CI非早期群が50.8，HA非早期・CI早期群が53.4，HA・CI非早期群が50.9であった。またPARS（広汎性発達障害日本自閉症協会評定尺度）得点の偏差値では，HA・CI早期群が52.3，HA早期・CI非早期群が48.3，HA非早期・CI早期群が50.7，HA・CI非早期群が50.1であり，いずれも大きな差は認められなかった。従って，4群間の言語発達検査の差は知的発達障害や広汎性発達障害による影響はないと推測される。

補聴器・人工内耳の装用開始時期が語彙の発達に及ぼす影響

語彙レベルの発達については，PVT-R（絵画語い発達検査）の修正得点の偏差値は，HA・CI早期

表1　対象児の背景因子

	HA・CI早期群	HA早期・CI非早期	HA非早期・CI早期	HA・CI非早期
新生児スクリーニング受検率	70%	59%	15%	7%
検査実施時年齢	5歳3ヶ月	6歳3ヶ月	6歳11ヶ月	7歳8ヶ月
裸耳聴力*	109.0dB	106.6dB	111.3dB	104.4dB
装用閾値*	33.9dB	34.7dB	31.2dB	33.7dB
最高語音明瞭度	82.1%	66.4%	82.4%	61.3%
RCPM**	49.0	50.8	53.4	50.9
PARS**	52.3	48.3	50.7	50.1

*裸耳聴力および装用閾値は4分法を示す。
**RCPM，PARSの得点は偏差値を示す。

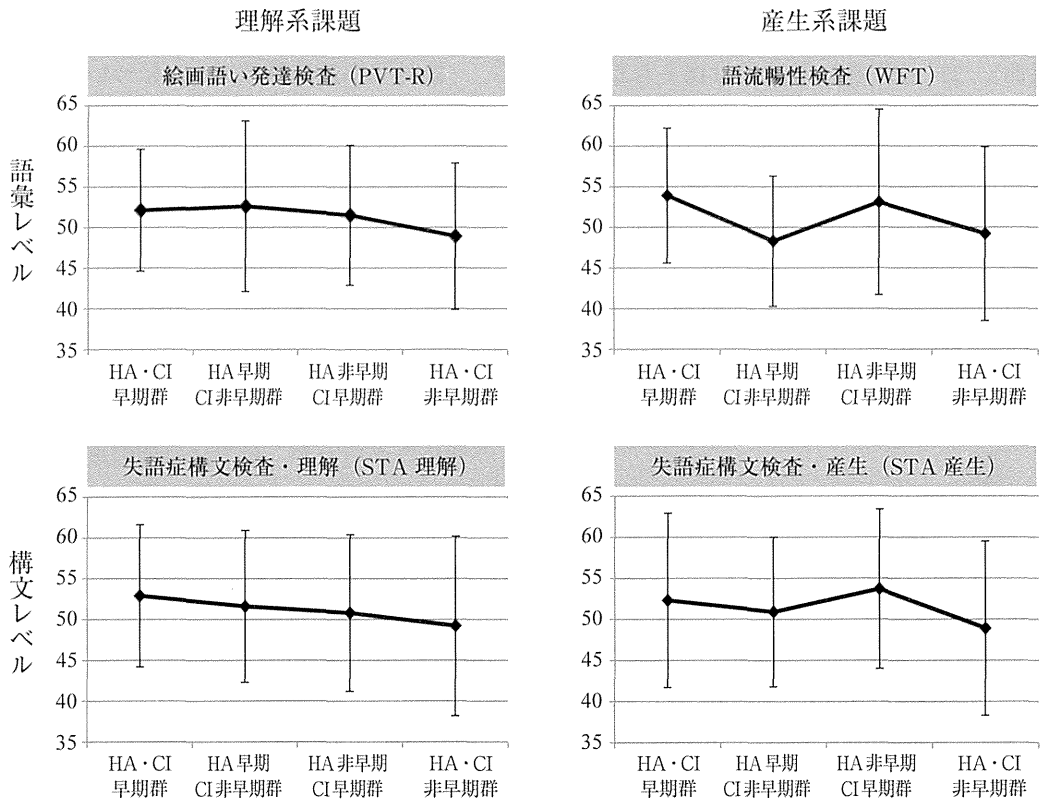

図3 各言語検査と補聴器・人工内耳装用時期との関係
4群の各言語評価検査結果（偏差値）を示す。理解系の課題（PVT-R および STA・理解）では補聴器を早期に装用した児の成績が良い傾向にあった。一方，産生系の課題（WFT および STA・産生）では，早期に人工内耳を装用した児の成績が良い傾向にあった。

群が52.1，HA早期・CI非早期群が52.6，HA非早期・CI早期群が51.5，HA・CI非早期群が47.9であり，有意差は認められなかったものの人工内耳の手術時期に関わらず，補聴器装用が早期の方がより良好な傾向にあった。一方，WFT（語流暢性検査）の偏差値については，HA・CI早期群が53.9，HA早期・CI非早期群が48.3，HA非早期・CI早期群が53.1，HA・CI非早期群が49.2であり，有意差は認められなかったものの補聴器の装用開始年齢に関わらず，人工内耳手術年齢が早い群の方が良好な傾向にあった（図3）。

補聴器・人工内耳の装用開始時期が構文の発達に及ぼす影響

構文レベルの発達については，STA（失語症構文検査）理解の合計得点の偏差値では，HA・CI早期群が52.9，HA早期・CI非早期群が51.6，HA非早期・CI早期群が50.8，HA・CI非早期群が49.2であり，有意差は認められなかったものの補聴器の装用開始年齢が早期の群が成績良好で，さらに，人工内耳の手術年齢の早い群の方が良好な傾向にあった。一方，STA産生の総得点の偏差値では，HA・CI早期群が52.3，HA早期・CI非早期群が50.9，HA非早期・CI早期群が53.7，HA・CI非早期群が48.9であり，有意差は認められなかったものの補聴器装用開始年齢に関わらず，人工内耳手術時の年齢が早い群の方が良好な傾向にあった（図3）。

補聴器・人工内耳の装用開始時期がコミュニケーション能力の発達に及ぼす影響

コミュニケーションレベルの発達をTQAIDの総得点の偏差値を用いて評価すると，HA・CI早期群が50.9，HA早期・CI非早期群が50.9，HA非早期・CI早期群が52，HA･CI非早期群が47.6であり，HA・CI非早期群が他の3群に比べ，成績が低いものの，他の3群の間に大きな差は認められなかった（図4）。

以上の結果をまとめると，語彙レベルにおいて

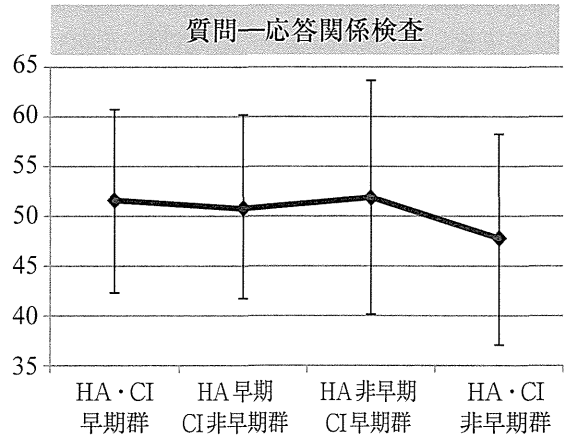

図4　コミュニケーションレベルの発達と補聴器・人工内耳装用時期との関係
4群のTQAID検査結果（偏差値）を示す。HA・CI非早期群が他の3群に比べ，成績が低いものの，他の3群の間に大きな差は認められなかった。

は，PVT-Rは補聴器の装用開始年齢が早い群の方が良好な成績であった。一方，WFTでは補聴器の装用年齢に関わらず，人工内耳の手術年齢が早い群の方が良好な成績であった。

構文レベルにおいては，STA理解に関しては補聴器の装用開始年齢が早い群が成績良好であり，STA産生については，人工内耳の手術年齢が早い群の方が成績良好であった。コミュニケーション力を見るTQAIDでは，補聴器の装用開始年齢，人工内耳の手術年齢における成績の差はない結果であった。

考　　察

従来より，人工内耳の早期装用が言語発達に及ぼす影響に関しては，複数の報告がなされているが，1歳代で人工内耳を受けた児は理解系と産生系の言語発達検査共に有意に発達が早いという報告[2]もあれば，1歳代と2歳代で差がなかったとの報告[3]も見られる。しかし，これまで，人工内耳早期装用効果と，補聴器の早期装用の効果を区別して日本語言語発達に及ぼす影響について検討を行った研究はない。

本研究の結果，語彙レベル，構文レベルを問わず，理解系の課題（PVT-RとSTA理解）については，人工内耳装用開始時期にかかわらず補聴器の装用開始年齢が早い群の成績が良好な傾向があった。一方，産生系の課題（WFTとSTA産生）については，補聴器の装用開始年齢に関わらず，人工内耳の手術年齢が早い群の成績が良好な傾向があった。

このことより，早期に補聴器装用を開始することにより，音韻認知とその処理の発達が促され，語彙および構文の理解能力を発達させる可能性が示唆された。本研究の対象児童は4歳までに70dB以上の高度難聴を有する児であり，なおかつ人工内耳を装用している児であることを考え合わせると，対象児の難聴の程度は非常に重度であり，補聴器による補聴では不十分な児が多数を占めていることが考えられる。しかしながら，本研究の結果からは，人工内耳装用開始時期にかかわらず補聴器の装用開始年齢が理解系言語発達に重要であることが示されたことより，仮に不十分であっても早期に補聴器装用し，早期から周囲の音声情報を認知させることが理解系の言語能力の発達に重要であることが示唆された。

一方，早期に人工内耳装用を開始することが産生系の言語能力の発達に影響を及ぼす結果となったことより，早期より語彙や構文を十分に弁別可能な聴取閾値を確保することが，おそらく正確な語彙の理解・助詞の使用などに良い影響をおよぼし，語彙及び構文の産生能力検査の結果を向上させたものと考えられた。

これまでの多くの研究から，早期に補聴器や人工内耳を装用することにより，良好な言語発達が期待できるという報告がなされている[4-15]。その時期については，新生児スクリーニング検査の導入や，2006年に改訂された日本耳鼻咽喉科学会による小児人工内耳適応基準により，年々低年齢化してきている状況である。

黄らによると，早期補聴，早期聴能教育による良い音声を獲得できる重要な時期として，0歳8ヶ月を補聴最適正年齢と呼び[12]，内山によれば，12ヶ月未満の時期から補聴器装用を含めた早期療育を開始することにより，言語発達が促進されるとの見解が示されている[13]。

本研究のデータについては，対象児の年齢が平成21年時に4歳から12歳ということから，補聴器，人工内耳それぞれの装用時期のピークが若干遅い印象があるものの，補聴器，人工内耳共に装用時期が早い群の方が成績良好な結果となり，先行研究を裏付