



Does the introduction of newborn hearing screening improve vocabulary development in hearing-impaired children? A population-based study in Japan



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ABSTRACT

Objective: Permanent hearing impairment has a life-long impact on children and its early identification is important for language development. A newborn hearing screening (NHS) program has started in Okayama Prefecture, Japan, in 1999 to detect hearing impairment immediately after birth. We aim to examine the effect of this screening program on vocabulary development in pre-school children in a before and after comparative study design.

Methods: A total of 107 5-year-old children who graduated from Okayama Kanariya Gakuen (an auditory center for hearing-impaired children) between 1998 and 2011 were enrolled in this study. The pre-NHS group ($n = 40$) was defined as those who graduated between 1998 and 2003, while the post-NHS group ($n = 67$) was defined as those who graduated between 2004 and 2011. The primary outcome was receptive vocabulary, which was assessed by the Picture Vocabulary Test [score <18 (low) vs. score ≥ 18 (high)]. The secondary outcome was productive vocabulary, or the number of productive words, which was assessed by an original checklist [<1773 words (low) vs. ≥ 1773 (high)]. We calculated odds ratios and 95% confidence intervals for vocabulary development and compared both groups.

Results: The adjusted Picture Vocabulary Test score and number of productive words were significantly higher ($p < 0.01$) in the post-NHS group than the pre-NHS group. Odds ratios were 2.63 (95% confidence interval: 1.17–5.89) for receptive vocabulary and 4.17 (95% confidence interval: 1.69–10.29) for productive vocabulary.

Conclusions: The introduction of NHS in Okayama Prefecture significantly improved both receptive and productive vocabulary development in hearing-impaired children.

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1. Introduction

Permanent childhood hearing impairment (PCHI) has a lifelong influence on language development in hearing-impaired children

[1–3]. Late identification of PCHI in children results in language delay and consequently affects their performance in academics or other activities. Thus, the social and economic impact of PCHI is immeasurable and great effort has been put into its early identification. Recent progress in newborn hearing screening (NHS) has enabled hearing impairment to be identified just after birth and has therefore led to early diagnosis of PCHI [4–7].

Mishina et al. [8] organized an NHS research group in Japan in 1998 and in 2000, they developed practical guidelines for NHS.

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Although the official NHS program in Japan started in 2001, actual arrangements for implementing the program began in several districts, including Okayama where the first NHS system was prepared in 1999 [9]. Although more than a decade has passed since the program's initiation, no quantitative studies have been conducted to date to examine its effects.

Accordingly, we planned this study to examine the effects of NHS on language development in hearing-impaired children since its introduction in Okayama Prefecture. We examined vocabulary development as a measure of language because vocabulary is an important language domain that develops in children of all school ages, and it is one of the most fundamental abilities of language. Vocabulary consists of receptive vocabulary (i.e., how children understand the meaning of spoken words) and productive vocabulary (i.e., how children voluntarily speak words). We hypothesized that the introduction of NHS contributed to better development of both receptive and productive vocabulary in children with PCHI.

2. Participants and methods

2.1. Study design

Children who entered *Okayama Kanariya Gakuen* between the 1993 and 2011 academic years were enrolled in this study. *Okayama Kanariya Gakuen* is an auditory center for hearing-impaired children which was established in Okayama, Japan in 1969; this center uses the auditory-verbal method in children with hearing aids or cochlear implants from age 0 to 6 years [10]. The children received 60 min of individual and 60 min of group auditory-verbal training for two to three times a week using their

hearing devices (HA or CI). The training program was given by the speech–language–hearing therapists and teachers in a bottom-up approach. We applied a before and after comparative study design.

Five-year-old children who graduated from *Okayama Kanariya Gakuen* between the 1998 and 2003 academic years were defined as the pre-NHS group and the 5-year-old children who graduated between the 2004 and 2011 academic years were defined as the post-NHS group (Fig. 1). The children were divided into these two groups with 2003 as the dividing line because no graduates received NHS prior to 2003 while a considerable portion of the graduates after 2004 did receive NHS (Table 1). The aim of this study was to evaluate the total change in one particular district as a result of the introduction on NHS rather than a personal change after receiving NHS. Because the introduction of NHS is technically a political decision, changes were evaluated in a district-based study.

This study was approved by the Okayama University ethics commission on November 27, 2012.

2.2. Measures

As a primary outcome measure, we assessed receptive vocabulary by using the Picture Vocabulary Test (PVT), which is a localized version of the Peabody Picture Vocabulary Test for Japanese language users. In this test, the children were seated facing a speech language therapist in a sound-attenuated chamber and scored according to the test manual. Children receiving PVT-adjusted scores ≥ 18 were considered to have demonstrated age-appropriate vocabulary development according to the manufacturers' instructions for the test [11]. Thus, we divided the subjects into a lower receptive vocabulary group (scores < 18) and higher receptive vocabulary group (scores ≥ 18).

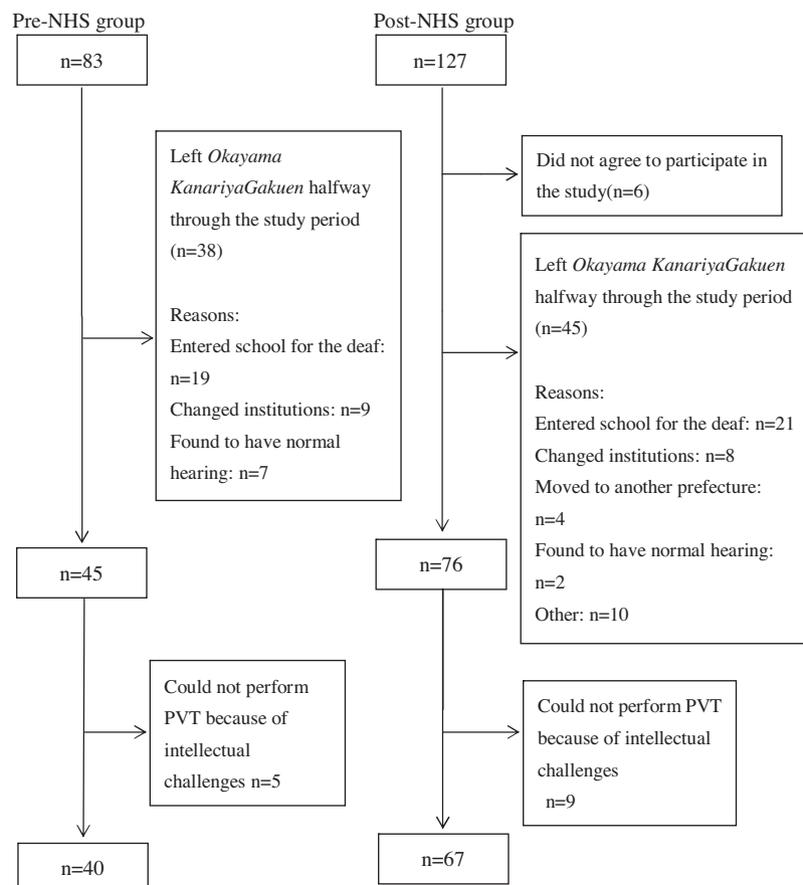


Fig. 1. Participant flow. PVT: Picture Vocabulary Test.

Table 1
NHS participation in the children in this study.

Academic year of graduation	Graduated children (n = 121)	Number of NHS participants	(%)
1998	4	0	0
1999	12	0	0
2000	9	0	0
2001	5	0	0
2002	8	0	0
2003	7	0	0
2004	7	5	71
2005	5	1	20
2006	6	1	17
2007	8	4	50
2008	11	9	82
2009	10	9	90
2010	16	15	94
2011	13	11	85

As a secondary outcome measure, productive vocabulary measured by an original vocabulary checklist created by *Okayama Kanariya Gakuen* in 1993 for 5-year-old children was used. It contains 4218 words normally spoken by 5-year-old children in daily life, including substantives (2887 nouns and 27 pronouns) [12], conjugated endings (823 verbs, 153 adjectives and 127 adjectival verbs), attachments (14 postpositional particles and four auxiliary verbs) and others (113 adverbs, 52 exclamations, 11 conjunctions and seven adjectival pronouns). Guardians were asked to check their children's spoken words according to the checklist three times a year (in March, August, December), document all of the productive vocabulary spoken during four months, and compare the number of the words in each category. The median number of words was 1773. Therefore, we divided the subjects into a lower productive vocabulary group (<1773 words) and a higher productive vocabulary group (≥ 1773 words).

Other clinical information, including sex, types of hearing devices, unaided/aided hearing level, NHS participation rate, age at identification of hearing loss, commencement of hearing aid use, duration of preschool training, aided hearing level, the results of speech perception test and WIPPSI were also obtained from the children's medical records.

2.3. Statistical analysis

First, Pearson's correlation coefficient was calculated to assess the correlation between the PVT-adjusted scores and the number of spoken words. Then, we used *t*-tests to compare the language data between the pre-NHS group and the post-NHS group. Subsequently, we calculated odds ratios and their 95% confidence intervals for higher vocabulary development to compare the post-NHS group with pre-NHS group by using chi-square analysis.

As a supplementary analysis, we collected language data for 83 children who left *Kanariya Gakuen* in the 1993 and 2011 academic years, halfway through the study period.

We considered *p* values <0.05 (two-sided) to be statistically significant. We used IBM SPSS version 19 software (IBM Corp., Armonk, NY, USA) for all analyses.

3. Results

3.1. Participants

A total of 210 children participated in this study. We restricted the participants to 5-year-old children who had graduated from *Okayama Kanariya Gakuen*, who could perform the PVT and agreed to participate in the study. As shown in Fig. 1,

107 children were included in the final analyses. Table 2 shows the characteristics of the participants. The male to female ratio was similar in the pre-NHS and post-NHS groups. Significantly earlier commencement of hearing aid use and eventually longer periods of preschool training were observed in the post-NHS group, reflecting the earlier identification of hearing loss after the introduction of NHS (Table 2).

3.2. Correlation between adjusted PVT scores and the vocabulary checklist

The vocabulary checklist results were significantly correlated with the adjusted PVT scores ($r = 0.747$, $p < 0.001$).

3.3. Comparisons between the pre-NHS and post-NHS groups

3.3.1. Primary outcome: receptive vocabulary

Fig. 2A and B shows the distribution of the adjusted PVT scores. The post-NHS group clearly demonstrated better scores (mean \pm standard deviation: 19.76 ± 8.31) than the pre-NHS group (14.83 ± 7.62) ($p = 0.003$). The proportion of children with extremely poor scores clearly decreased after 2004. The post-NHS group showed a significantly higher likelihood of higher receptive vocabulary development (odds ratio: 2.63; 95% confidence interval: 1.17–5.89) than the pre-NHS group.

3.3.2. Secondary outcome: productive vocabulary

Fig. 3A and B shows the distribution of productive vocabulary scores. The post-NHS group again showed better scores (1975.09 ± 642.26) than the pre-NHS group (1386.2 ± 839.08)

Table 2
Demographic factors of the children in this study.

	Pre-NHS group	Post-NHS group
Sex		
Male	21 (52.5%)	38 (56.7%)
Female	19 (47.5%)	29 (43.3%)
Hearing device		
CI user	6(15%)	32(47.8%)
Monolateral	6	30
Bilateral	0	2
HA user	34(85%)	35(52.2%)
Hearing impairment ^a		
Mild	8 (20%)	11 (16.4%)
Moderate	5 (12.5%)	13 (19.4%)
Severe	10 (25%)	20 (29.9%)
Profound	17 (42.5%)	23 (34.3%)
NHS		
Received	0 (0%)	48 (71.6%)
Not received	40 (100%)	19 (28.4%)
	Mean(SD)	Mean(SD)
Identification of hearing loss (age, months) ^b	23.70 (16.42)	12.34 (16.33)
Commencement of hearing aid use (age months) ^b	23.85 (16.10)	16.84 (17.94)
Period of preschool training (months) ^b	48.38 (17.68)	54.33 (20.74)
Aided hearing level (dB) ^a	49.50 (17.85)	32.89 (8.52)
Speech perception test WIPPSI	68.95(19.93)	80.45 (12.34)
IQ	89.46(21.42)	91.62(24.36)
PIQ	107.32(22.06)	105.65(25.32)
VIQ	75.06(24.36)	80.16(23.69)

^a Mild hearing loss: 26–40 dB, moderate hearing loss: 41–70 dB, severe hearing loss: 71–90 dB, profound hearing loss: ≥ 91 dB.

^b Values are mean and (standard deviation).

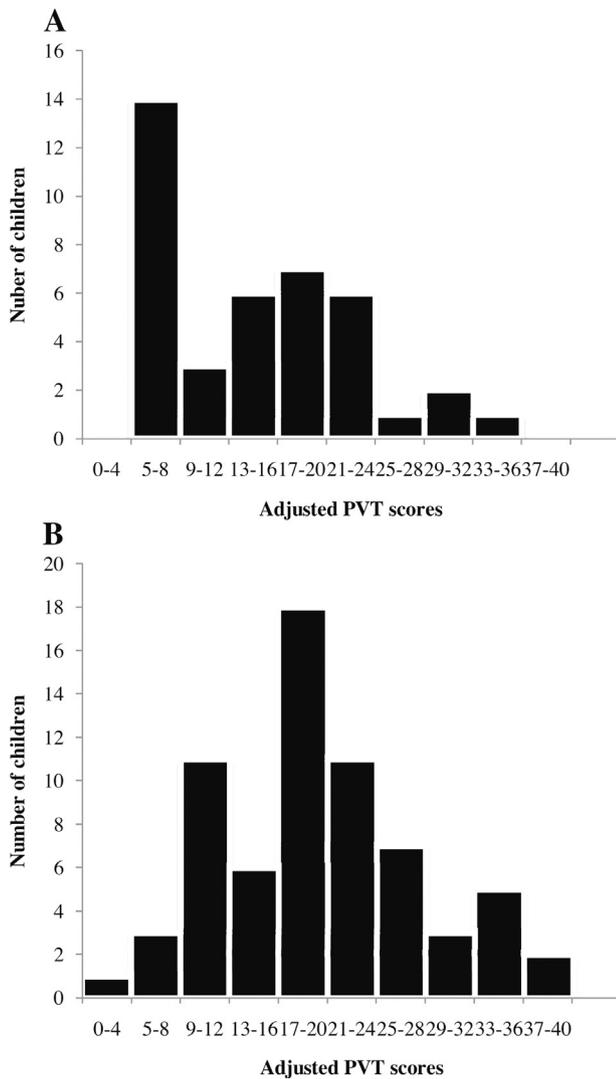


Fig. 2. Adjusted Picture Vocabulary Test scores for the pre-NHS group (A) and the post-NHS group (B).

($p < 0.001$). The proportion of children with extremely poor scores clearly decreased after 2004. The post-NHS group showed a significantly higher possibility for higher productive vocabulary development (odds ratio: 4.17; 95% confidence interval: 1.69–10.29) than the pre-NHS group.

3.4. Supplementary analysis: evaluation of children who left Okayama Kanariya Gakuen

Among the 83 children who left *Okayama Kanariya Gakuen* halfway through the study period, 40 children (48%) entered the Okayama School for the Deaf, 17 children (20%) changed institutions because they had other physical/intellectual challenges and required more intensive services than the *Okayama Kanariya Gakuen* could provide and nine children (11%) who were first suspected to have hearing impairment were later identified as having normal hearing. The other 17 children left for reasons such as moving to another prefecture. The children who left *Okayama Kanariya Gakuen* halfway through the study period were also divided into pre-NHS and post-NHS groups. Details are shown in Fig. 1.

Table 3 shows the details of early language development in the children who entered the Okayama School for the Deaf after leaving *Okayama Kanariya Gakuen*. There were no differences in the

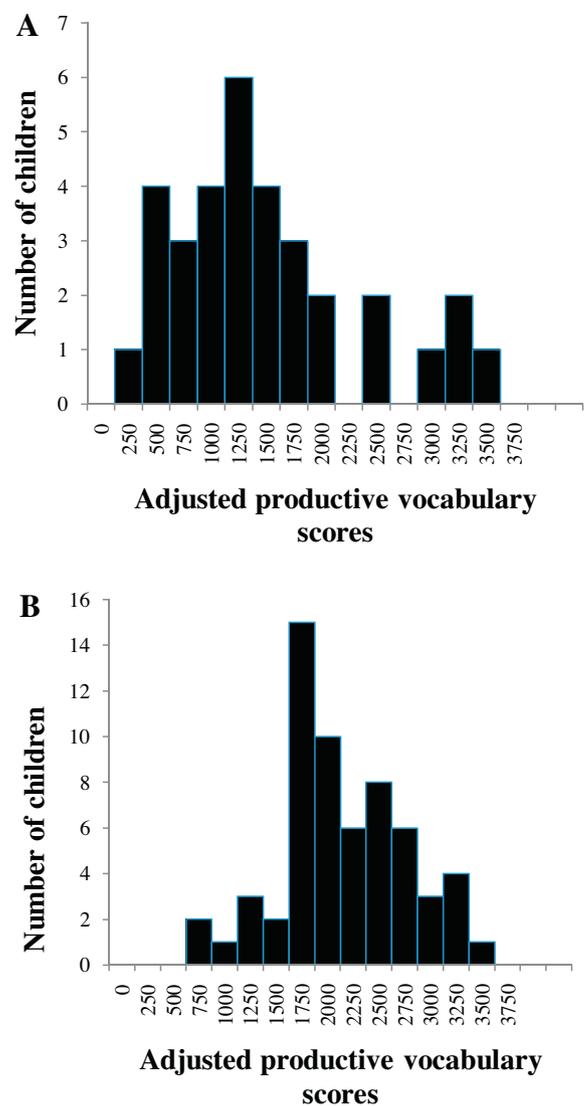


Fig. 3. Adjusted productive vocabulary scores in the pre-NHS group (A) and the post-NHS group (B).

Table 3
Early language development of children who left *Okayama Kanariya Gakuen* halfway through the study period.

Media	Language ability	Pre-NHS group		Post-NHS group	
		n	%	n	%
Reaction	Reaction to vocal sounds	5	13.2	6	13.3
Pointing Voice	Pointing	1	2.6	2	4.4
	Vocal imitation	6 (3)	15.8	7 (4)	15.6
Gesture	1–49 words	2 (1)	5.3	3 (3)	6.7
	50–99 words	0	0	3 (3)	6.7
	≥100 words	1 (1)	2.6	2 (1)	4.4
Productive language	1–49 words	5 (3)	13.2	7 (5)	15.6
	50–99 words	8 (6)	21.1	2	4.4
	100–199 words	3 (2)	7.9	2 (1)	4.4
	200–299 words	1 (1)	2.6	3 (1)	6.7
	300–399 words	1 (1)	2.6	2 (1)	4.4
	400–499 words	1 (1)	2.6	2 (1)	4.4
	≥500 words	4	10.5	4 (1)	9
Total		38 (19)	100	45 (21)	100

Numbers in parentheses are the numbers of children who transferred to Okayama school for the deaf and these are added to the total.

proportions of these children between the pre-NHS and post-NHS groups by chi-square test, and the language ability of these children showed quite similar tendencies.

4. Discussion

Although several factors affect language development in hearing-impaired children, the most important factors are the education principles (i.e., sign, auditory-oral or auditory-verbal), Performance Intelligence Quotient (PIQ), and hours of education and rehabilitation that are used for deaf education. Because the choice of education principles and hours of education/rehabilitation is at the discretion of the institute, it is very difficult to ensure consistent education among institutes. A single institute, before and after comparative study makes it possible to avoid the influence of different types of education. In addition, this type of study design can dramatically show educational development before and after the introduction of NHS in one particular district and the results can be useful to local health-care planners or government. Moreover, no difference in PIQ among the pre and post NHS group enabled to compare the change of vocabulary development equally in these two groups.

Our findings suggest that the introduction of NHS [13,14] significantly improved both receptive and productive vocabulary development in 5-year-old hearing-impaired children. Children in the post-NHS group showed twice the receptive vocabulary development and four times the productive vocabulary development than the pre-NHS group. The overall benefit may be primarily attributable to the decrease in the number of children with extremely poor vocabulary development, which may be the result of delayed commencement of education.

The introduction of NHS to a particular district can have a positive impact on total vocabulary development in the hearing-impaired children living there. The current results support the introduction of NHS by healthcare planners in other local districts. Needless to say, successful NHS depends on several factors including early diagnostic procedures by pediatric audiologists/otolaryngologists, an early intervention system that includes pediatric hearing aid fitting/cochlear implant indication, and continuous socioeconomic support for guardians. The proportion of children using CI has increased in post-NHS group, and this should have led to the improvement of the aided hearing level and the better results of speech perception test of this group, which is demonstrated in Table 2. Since earlier and more effective intervention by the CI is a positive factor to mediate the beneficial effect of NHS, as reported previously [15], the presence of NHS and the increased proportion of CI cannot be discussed separately. Moreover, the performance of both CI and HA are developing, we cannot compare the effect of the NHS or the language development only by the types of the hearing devices. Our data of increased proportion of CI in post-NHS group, thus support the hypothesis of beneficial effect of NHS on language development, which is documented in the present analysis.

However, we believe that political support for the introduction of NHS is the most important step for achieving better language development in hearing-impaired children.

A previous study by Yoshinaga-Itano [16] suggested that early identification of hearing impairment may improve language development in children since identification of hearing impairment by six months of age resulted in significantly better language scores. Pimperton et al. [17] reported that the introduction of universal newborn screening coupled with early intervention programs was significantly linked to positive language outcomes. McPhillips [18] reported that early intervention was conducted to improve language development in hearing-impaired children in the United States and that most hospitals and birthing

centers have commenced universal NHS programs. According to that report, the NHS is now conducted in up to 93% of the infants born in the United States

In Okayama, the NHS is now conducted up to 88%, and the present results suggest that the reduction in the number of children with poorer receptive and productive vocabulary development is the major advantage of NHS. Before the introduction of NHS, a fair number of children who potentially could have further developed their language skills were presumably mis-identified and might have lost their chance to be exposed to early intervention; however, these conditions were improved by the introduction of NHS. The main effect of NHS might be providing hearing-impaired children with the opportunity to develop their vocabulary to the best of their ability.

The vocabulary checklist results significantly correlated with the adjusted PVT scores. Understanding that receptive vocabulary is the basis of productive vocabulary is useful when interpreting the current results. Because there was good correlation between the vocabulary checklist currently used at *Okayama Kanariya Gakuen* and the standard PVT results, the vocabulary checklist approach can be considered reasonably eligible to evaluate productive vocabulary, even though this tool is not validated.

4.1. Limitations of this study

There are some limitations in this study. First, a number of children left *Kanariya Gakuen* before the age of 5. As remarked above, language development did not differ significantly between the pre-NHS and post-NHS groups in the children who left halfway through the study period. Therefore, we believe that the influence of those who left were relatively small.

Second, because the current results were essentially based upon a language test, children who had other physical/intellectual challenges such as cerebral palsy were not evaluated. If children with other physical/intellectual challenges had participated in the study, different results might have been obtained.

Third, as stated above, successful NHS depends on several factors and we cannot identify the factor most relevant to improve vocabulary acquisition in this study. We assume that earlier and proper interventions with hearing aids and cochlear implants may be the major factors that mediate the beneficial effects of NHS.

Fourth, language continues to develop throughout life, but the current study only demonstrated the improvement of vocabulary within the preschool period. Thus, a follow-up study including children of various ages is highly warranted.

Lastly, because other important domains of language including syntax, discourse and pragmatics were not evaluated in this study, further studies are needed in this area. We believe, however, that vocabulary is an important aspect of language.

5. Conclusions

Despite several limitations, the findings from our 14-year experience in Okayama suggest that the introduction of NHS significantly improved both receptive and productive vocabulary development in 5-year-old hearing-impaired children.

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