

Original Article

Language–articulation development dissociation in a child with asymmetric hearing loss due to unilateral cochlear nerve deficiency: a case study

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Purpose Unilateral or asymmetric hearing loss (UHL/AHL) is often under-managed despite its potential effects on articulation accuracy. This longitudinal single-case study aimed to describe language and articulation development in a child with AHL caused by unilateral cochlear nerve deficiency (CND).

Methods Clinical records of a child who began using hearing aids and receiving speech–language therapy at age 2 were retrospectively reviewed. Standardized assessments were conducted between ages 1 and 3, and the results were converted to z-scores.

Results At age 3, language development was within the normal range, whereas articulation accuracy was markedly below expectations, indicating a marked dissociation.

Conclusions This case suggests a marked dissociation between language and articulation development in a child with AHL. This observation underscores the importance of monitoring articulation development, even when language outcomes appear typical.

Keywords cochlear nerve deficiency, asymmetric hearing loss, language–articulation dissociation

INTRODUCTION

Cochlear nerve deficiency (CND), which encompasses cochlear nerve aplasia and hypoplasia, is one of the major causes of unilateral sensorineural hearing loss (HL) [1,2]. CND is present in 25%–48% of patients with congenital unilateral HL and 15.4% of those with profound HL [3].

Asymmetric HL (AHL) occurs when one ear has severe HL that cannot be effectively managed with conventional amplification devices, whereas the contralateral ear has only mild-to-moderate HL. In a previous study [2], CND had a prevalence ranging from 0% to 28% among pedia-

tric patients with AHL, and among 87 pediatric patients with AHL, cochlear nerve aplasia was identified in nine and hypoplasia in one.

Unilateral and asymmetric HL can lead to impaired sound localization and speech perception in noisy environments [4,5]. Despite these challenges, these conditions have not historically been considered primary targets for active intervention compared with bilateral profound HL. However, recent studies have indicated a shift toward cochlear implantation (CI) in the poorer ear to restore bilateral auditory input in individuals with unilateral profound HL [5,6].

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These studies have also suggested that unilateral profound HL during early childhood may negatively affect language development and emphasize that early intervention before age 3 is critical for optimal auditory and language outcomes. Furthermore, a study revealed that compared with their normal-hearing peers, children with unilateral HL demonstrated significantly lower performance in receptive and expressive language, verbal intelligence, and school adjustment during the school-age period (6–12 years) [7]. These findings suggest that, without early intervention, difficulties across multiple domains can persist into later developmental stages.

Compared with unilateral HL, AHL may impose greater functional limitations on listening performance, potentially leading to an increased developmental burden.

CI is an effective intervention for children with profound HL [8]. However, in previous discussions regarding interventions for unilateral CND in children, CI has often not been considered a viable option or has been suggested to have limited effectiveness, possibly due to insufficient neural responses to electrical stimulation resulting from CND [9,10].

Nevertheless, studies tracking speech perception development following CI in children aged < 4 with unilateral or bilateral CND have reported different outcomes across individuals [3]. These findings suggest that CI outcomes in children with unilateral CND may vary widely across speech and language domains.

To date, studies examining language and articulation development in children with unilateral CND remain limited. A recent study investigated receptive language and articulation development in preschool children in Korea diagnosed with CND-induced unilateral HL [11]. Using standardized norm-referenced assessments, the study found markedly reduced articulation accuracy in 25 preschool children with unilateral CND, whereas receptive language abilities were within the normal range in 23. These findings suggest a dissociation between language and articulation development in children with uni-

lateral CND and highlight the need for more detailed analyses of their developmental profiles as well as long-term follow-up.

This study was conducted to retrospectively describe the pattern of dissociation between language and articulation development in a child with AHL caused by unilateral CND who was recommended for CI. Accordingly, this study aimed to present an empirical case supporting the need for long-term follow-up and intervention in children with HL resulting from rare etiologies, such as the present case. In this case, the child underwent language and articulation assessments from age 1 to age 3, and progress was evaluated following hearing aid use and speech-language therapy. Accordingly, longitudinal analysis was possible based on data obtained from four assessment time points.

CASE REPORT

Case Description

The patient was a 3-year-old child who presented with abnormal findings on newborn hearing screening at birth. Subsequent magnetic resonance imaging revealed hypoplasia of the left cochlear nerve. Auditory brainstem response testing indicated moderate HL (41–55 dB) in the right ear, with no evaluable response in the left ear. Auditory steady-state response (ASSR) testing demonstrated moderate HL in the right ear and severe HL in the left ear, and no significant changes were observed in follow-up assessments. Specifically, ASSR thresholds in the right ear were 60-dB HL at 500 and 1000 Hz and 50-dB HL at 2000 and 4000 Hz. In the left ear, thresholds were 80-dB HL at 500, 1000, and 2000 Hz and 100-dB HL at 4000 Hz. Tympanometry results were within normal limits bilaterally.

The child used bilateral hearing aids consistently from age 2. Real-ear measurements indicated that the left ear

generally met the Desired Sensation Level v5.0a target output. However, the speech intelligibility index (SII), a measure of speech audibility, was 40% and 43% at input levels of 55 and 70 dB sound pressure level (SPL), respectively, indicating limited access to speech.

In contrast, the right ear did not fully reach the target output in the 2,000–4,000-Hz range. Nevertheless, SII values were 70% and 80% at the same input levels, indicating relatively better speech access than those in the left ear.

The child wore hearing aids for approximately 7–9 h per day. During the same period, the child received speech-language therapy 1–2 times per week at a private clinic, focusing on the development of language and articulation rather than auditory rehabilitation. Therapy incorporated visual cues, such as the clinician's mouth movements, along with visual materials that matched the child's preferences.

Results from the Korean Developmental Screening Test for Infants and Children, conducted during routine health check-ups up to age 2, indicated typical development across gross and fine motor, cognitive, social, and self-help domains. After being diagnosed with CND and AHL, the child was recommended for CI at a university hospital. Longitudinal follow-up of language and articulation development was conducted through four assessments between ages 1 and 3.

This study was approved by the Institutional Review Board of Seoul National University Bundang Hospital (No. B-2602-1028-701).

Assessments

Following standardized administration procedures, all

assessments at each time point were performed by a certified speech-language pathologist with experience in evaluating children with HL.

Phoneme-level speech perception was assessed using the Ling six sound test. This test evaluates a child's ability to detect, discriminate, and identify speech sounds /m/, /a/, /u/, /i/, /j/, and /s/, which represent the frequency range of connected speech (250–8,000 Hz), presented verbally by the examiner [12]. Receptive and expressive language development was assessed using the sequenced language scale for infants (SELSI). It is a standardized tool for children aged 5–36 months that enables early identification of delays in receptive and expressive language development. It is based on a combination of caregiver report and direct observation by the examiner and provides normative developmental data [13]. The Urimal test of articulation and phonology (U-TAP) was used to assess articulation accuracy. This test assesses consonant accuracy at the word level in children aged 2–6 years through a picture-naming task, allowing comparison with standardized norms [13]. Auditory performance was evaluated using the categories of auditory performance (CAP). CAP is an eight-level hierarchical rating scale (0–7) based on caregiver reports that reflects functional auditory performance in everyday situations [12,14]. Overall speech intelligibility was assessed using the speech intelligibility rating scale. This five-level scale allows clinicians to rate the intelligibility of a child's spontaneous speech based on defined criteria [12].

A summary of the assessment tools and domains evaluated in this study is presented in Table 1.

Table 1. Assessment tools and corresponding domains

Assessment tool	Assessment domain
Ling six sound test	Speech perception (phoneme level)
Sequenced language scale for infants	Receptive and expressive language
Urimal test of articulation and phonology	Articulation and phonology
Categories of auditory performance	Auditory performance
Speech intelligibility rating	Speech intelligibility

RESULTS

Follow-up assessments for each domain were conducted at intervals of 3–7 months. To ensure participant deidentification, age at each assessment was categorized into early and late periods within each age year. To examine differences from typically developing peers, SELSI and U-TAP scores were converted to z-scores based on the normative data provided in the respective manuals and compared (Figure 1). Z-scores were calculated as (observed score - normative mean)/normative standard deviation. The analytical procedure using z-scores was adapted from previous studies [11,15]. This approach enabled the examination of longitudinal changes in language and articulation development.

Speech Perception

Closed-set phoneme perception tasks using meaningful words are generally applicable to children aged 3 years or older [12]. However, in the present case, reliable assessment using word- and sentence-level speech perception tests was not feasible because of inconsistent responses

and reduced articulation accuracy at this developmental stage. Therefore, phoneme-level speech perception was longitudinally monitored using the Ling six sound test.

The sounds were presented using live voice from approximately 1 m while visual speech information was blocked. Detection was scored as correct when the child showed a clear behavioral response to the sound, such as orienting toward the sound source or attempting to imitate it. For discrimination, the examiner determined whether the child produced distinguishable imitative responses to different sounds; accurate production of the target sounds was not required. Identification was scored when the child correctly repeated the presented sound.

During the early and late phases of age 2, the child could detect all Ling six sounds under the auditory-only condition while wearing bilateral hearing aids. Discrimination and identification were possible for all sounds, except for the high-frequency phonemes /ʃ/ and /s/ (>2000 Hz).

At age 3, the child demonstrated immediate identification and accurate imitation of all six Ling sounds under the same conditions. Speech perception performance for the Ling six sounds at each assessment point is summarized in Table 2.

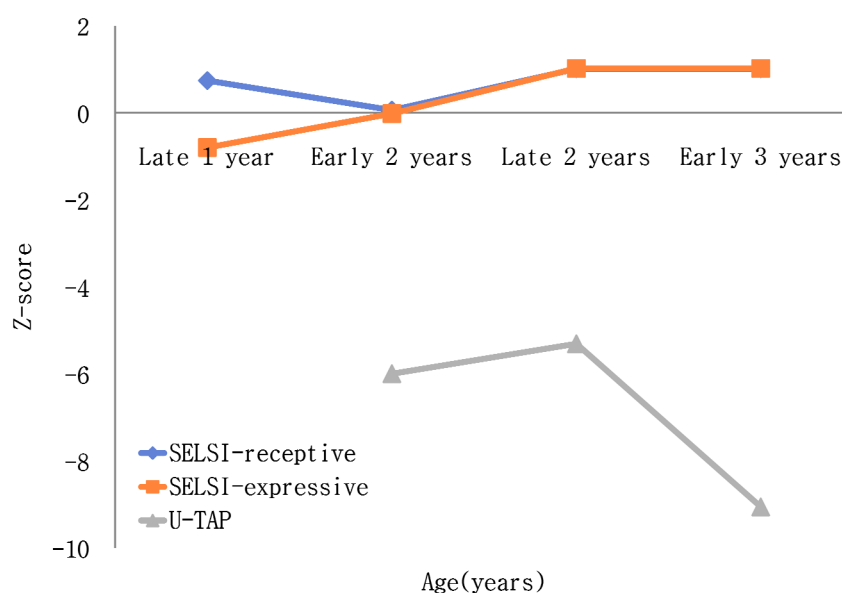


Figure 1. Changes in SELSI and U-TAP z-scores across assessment points

Table 2. Ling six sound detection, discrimination, and identification across ages

Age (years)	Auditory level	/m/	/a/	/u/	/i/	/ɔ/	/s/
Early phase of age 2	Detection	+	+	+	+	+	+
	Discrimination	+	+	+	+	-	-
	Identification	+	+	+	+	-	-
Late phase of age 2	Detection	+	+	+	+	+	+
	Discrimination	+	+	+	+	-	-
	Identification	+	+	+	+	-	-
Early phase of age 3	Detection	+	+	+	+	+	+
	Discrimination	+	+	+	+	+	+
	Identification	+	+	+	+	+	+

+ correct response; - incorrect response.

Language Development

Z-scores were calculated based on normative data provided in the SELSI manual [16]. According to the SELSI criteria, scores within -1 SD are classified as “normal development,” between -2 SD and -1 SD as “mild delay,” and below -2 SD as “language delay” [16]. At the final assessment, the child was 36 months of age. Although SELSI is clinically administered through 36 months, its normative tables extend through 35 months [16]. Therefore, the final z-scores were calculated using the nearest available 35-month norms and interpreted cautiously together with the raw scores.

Receptive language development was classified as “normal development” at all assessment points. Z-scores remained above 0 throughout the observation period, with a value of 0.74 at age 1, a slight decrease to 0.06 during the early phase of age 2, followed by an increase to 1.01 in the late phase of age 2, which was maintained at age 3. Expressive language development was classified as “mild delay” at age 1 but fell within the normal range from the early phase of age 2 onward. The z-score increased from -0.80 at age 1 to -0.02 in the early phase of age 2 and further to 1.02 in the late phase of age 2, which was maintained at age 3.

At age 3, the child demonstrated age-appropriate receptive and expressive language abilities during assessment. The child responded appropriately to questions about tools (e.g., “What should we use to color?”), demonstrated an

understanding of spatial concepts, and followed comparative instructions (e.g., selecting larger or smaller items). The child also demonstrated comprehension of color names and responded appropriately to sentence-level everyday questions (e.g., “Who did you come here with?”).

Regarding expressive language, the child could describe personal experiences concisely and use the interrogative ‘why’ appropriately. The child produced simple sentences with modifiers. Spontaneous speech was frequent and included onomatopoeic expressions. The child could produce two- to four-word utterances without difficulty (e.g., ‘나무 다섯개야.’ [‘Five trees.’], ‘이렇게, 이렇게 해요?’ [‘Do it like this?’], ‘엄마 나 책상 있지.’ [‘Mom, I have a desk.’]).

Articulation Development

Beginning at age 2, when the U-TAP became applicable, consonant accuracy at the word level was assessed using 43 target consonants. Following the standardized procedure, the child was asked to name each pictured item. The percentage of consonants correct (PCC) was calculated by dividing the number of correctly produced target consonants by 43 and multiplying by 100. The PCC values were converted into z-scores based on age-matched normative data [17]. Children with HL experience greater difficulty in perceiving and producing consonants than vowels [18], suggesting that consonant production

errors may sensitively reflect deficits in auditory input. Accordingly, word-level consonant accuracy was selected as the primary outcome measure in this study.

According to the U-TAP criteria, scores above -1 SD are classified as “normal or requires monitoring,” between -2 SD and -1 SD as “consider intervention,” and below -2 SD as “intervention required” [17]. The child’s consonant accuracy at all assessment points from the early phase of age 2 to age 3 fell below -2 SD, corresponding to the “intervention required” category.

In the early phase of age 2, 8 of 43 target consonants were produced correctly (PCC = 18.60%; $z = -6.00$). In the late phase of age 2, 11 of 43 consonants were produced correctly (PCC = 25.58%; $z = -5.31$). At age 3, 10 of 43 consonants were produced correctly (PCC = 23.26%). Based on the normative mean of 91.75 and SD of 7.56, the z -score was calculated as $(23.26 - 91.75)/7.56 = -9.06$. This more extreme z -score largely reflects the higher normative mean at age 3 rather than a clear decline in articulation ability, because the child’s PCC remained relatively stable between the late phase of age 2 and age 3.

At age 3, 33 consonant errors were identified among the 43 target consonants. The percentage of each error type was calculated using the 33 consonant errors as the denominator. The child often replaced multiple initial consonants with the tense alveolar stop /t=/ (Korean /ㅌ/) and omitted final consonants. In addition, fronting errors involving some velar consonants were observed.

Notably, articulation accuracy improved when target words were presented more slowly or when the child imitated the examiner’s articulatory movements. Oral motor tasks (e.g., tongue protrusion and lateral movement) were normal when following verbal or gestural instructions. Prosody at both the word and sentence levels was relatively preserved.

Table 3 summarizes the child’s predominant error patterns and representative examples. Overall, tensification, final consonant omission, stopping, and combined stopping and tensification accounted for approximately 70%

Table 3. Phonological process analysis of consonant error patterns

Error type	n/33 (%)	Representative examples
Tensification	7 (21.2)	<i>tokki</i> → <i>ttakki</i> <i>pungseon</i> → <i>tteudeon</i>
Final consonant omission	6 (18.2)	<i>mot</i> → <i>mo</i>
Stopping	5 (15.2)	<i>jadongcha</i> → <i>dajojja</i> <i>jjaekjjaek</i> → <i>ttette</i>
Combined stopping and tensification	5 (15.2)	<i>danchu</i> → <i>dattu</i>

of all consonant errors.

Auditory Performance

To indirectly examine the influence of functional auditory performance on language and articulation development, the CAP scale was administered.

At age 1, the child was classified as CAP 3, indicating the ability to follow simple instructions presented at conversational speech levels in proximity and to respond consistently to environmental sounds [12]. After starting to use hearing aids, the child reached CAP 4 in the early and late phases of age 2, demonstrating the ability to discriminate at least two of the five Ling sounds (/m/, /u/, /i/, /j/, and /s/) at conversational speech levels. At age 3, the child achieved CAP 5, indicating the ability to understand questions such as, “Where did your sister go?” and “What did you eat today?” and to respond appropriately under auditory-only conditions. Because attainment of CAP 5 typically presupposes language abilities at approximately the 3-year level [12], this finding supports age-appropriate language development.

Speech Intelligibility

Overall speech intelligibility was assessed using the speech intelligibility rating scale. The child consistently remained at category 2 across all assessment points from age 1 to 3, showing no notable improvement. Speech intelligibility reflects the extent to which a listener can understand a speaker’s utterances.

According to the SIR criteria [12], category 2 corre-

Table 4. Age-related results across assessment tools

Age (years)	SELSI (z-score)		U-TAP PCC (z)	CAP	SIR
	Receptive raw (z)	Expressive raw (z)			
Late 1 year	43 (0.74)	31 (−0.80)	—	3	2
Early 2 years	48 (0.06)	45 (−0.02)	18.60% (−6.00)	4	2
Late 2 years	56 (1.01)	56 (1.02)	25.58% (−5.31)	4	2
Early 3 years	56 (1.01)	56 (1.02)	23.26% (−9.06)	5	2

CAP, categories of auditory performance; SELSI, sequenced language scale for infants; SIR, speech intelligibility rating; U-TAP, Urmal test of articulation and phonology.

sponds to a level at which the intelligibility of connected speech is limited, and even word-level utterances are difficult for unfamiliar listeners to understand. At age 3, the child's speech remained difficult to understand at both the word and sentence levels without contextual or visual cues (e.g., lip movements). This finding was consistent with the markedly reduced consonant articulation accuracy observed in this case, given the reported association between consonant articulation accuracy and speech intelligibility [14].

A summary of the assessment results across all domains is presented in Table 4.

Overall, speech perception, language development, and functional auditory performance improved with age. In contrast, improvements in articulation accuracy and speech intelligibility were limited.

By age 3, a clear dissociation between language and articulation development was observed (Figure 1). In this case, marked articulation delay persisted despite language performance within the normal range.

DISCUSSION

Early childhood, particularly the period from birth to age 3 years, has been described as a critical “window of opportunity” during which CI can maximize developmental outcomes [6]. In the present case, the child was within this critical period at age 3 and demonstrated age-appropriate language development based on standardized as-

sessments, as well as progressive improvement in speech perception and functional auditory performance.

Despite improvements in auditory performance and age-appropriate language development, articulation accuracy and speech intelligibility remained markedly low with limited improvement over time. In this case, the observed dissociation highlights the need to monitor articulation development separately, even when auditory and language development appear to progress favorably.

Similar patterns of developmental dissociation have been reported in previous studies. In a study of preschool children diagnosed with unilateral HL due to CND [11], five of six children who underwent repeated assessments showed persistently low articulation accuracy despite having normal receptive language abilities. Consistent findings have also been reported in children with unilateral congenital aural atresia (CAA) [15]. Although CAA primarily causes conductive HL with preserved bone conduction, this group demonstrated age-appropriate language abilities but significantly reduced articulation accuracy. These previous findings provide context for the present case.

Speech sound disorder (SSD) has been reported in children who demonstrate age-appropriate receptive and expressive language abilities but exhibit unexplained articulation difficulties. These children may experience deficits in phonological working memory compared with their typically developing peers [19,20]. Increased listening effort is commonly reported in individuals with hearing impairment, and even those with unilateral HL require greater cognitive resources than individuals with normal

hearing when performing working memory tasks or engaging in daily communication [21]. Such cognitive demands may place additional constraints on articulation, which requires continuous monitoring and rapid feedback based on phonological working memory [22–24]. In the present case, asymmetric auditory input may have increased phonological processing demands, potentially contributing to the observed articulation difficulties.

Conversely, the preservation of language development within the normal range in the present case may be explained by the availability of compensatory strategies in the language domain. Compared with articulation, language processing can rely on contextual and visual cues, allowing children to compensate for incomplete auditory input [20]. A previous study suggested that speech perception is influenced not only by peripheral auditory input but also by contextual information, which may help individuals with HL fill in gaps in linguistic input [25]. Consistent with this account, the child in this study utilized visual cues, such as lip movements and picture cards, during speech-language therapy. The child also demonstrated compensatory communicative behaviors, such as the use of gestures or circumlocution when target words were not readily accessible.

In this case, the child exhibited profound HL in one ear caused by CND in combination with moderate HL in the contralateral ear, which may have resulted in greater listening demands. In a previous study of children aged 2–6 years with unilateral CND [11], the mean z-scores for receptive language and consonant accuracy were 0.03 and –2.90, respectively. At age 3, the corresponding z-scores in the present case were 1.01 and –9.06, respectively, indicating a more pronounced language–articulation dissociation in this case than the mean pattern reported for children with unilateral CND.

Previous studies have reported that children with HL may exhibit developmental error patterns, such as stopping, final consonant omission, and fronting of velar consonants [26]. A study on speech production in young chil-

dren with HL who use hearing aids documented long-term atypical development, particularly involving fricatives and affricates [18]. In this case, the child demonstrated predominant patterns of stopping affecting fricatives and affricates, frequent tensification, and syllable structure simplification by omitting final consonants, which are broadly consistent with phonological characteristics reported in children with HL.

In Korean, tense consonants are distinguished by acoustic features, such as shorter voice onset time (VOT), higher fundamental frequency (F0), and stronger burst energy [27]. Because consonant contrasts rely substantially on acoustic cues such as VOT [28], the acoustic salience of Korean tense consonants may offer one possible context for the frequent tensification observed in this case.

In cases where isolated SSD of unknown etiology is present, atypical articulatory movements, abnormal tongue movement, and swallowing patterns have frequently been reported. However, these features were not observed in the present case. Furthermore, articulation accuracy improved with visual cues, and clinical screening indicated that nonverbal oral motor performance and prosodic features of spontaneous speech were within normal limits. These findings supported the clinical differentiation of this case from other motor speech disorders [26].

A previous study classified sound preference substitution as one subtype of atypical speech sound errors and reported lower phonological working memory in children with atypical errors [29]. In the present case, the frequent substitution of multiple word-initial consonants with the tense alveolar stop /t̚/ (Korean /ㅌ/) was consistent with this error pattern. However, the underlying perceptual or phonological mechanism could not be determined from the present data.

This study is based on a single case; therefore, the findings cannot be generalized and do not support causal conclusions. In addition, phonological working memory and listening effort were not directly measured; therefore, the proposed explanations involving these factors remain

tentative. Future studies should include larger samples and incorporate quantitative measures of auditory input efficiency, working memory, and listening effort to better understand the influence of these factors on articulation development. Thus, even when language development falls within the normal range, significant articulation difficulties may warrant continued monitoring and comprehensive individualized assessment.

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