

ORIGINAL RESEARCH

Unimodal versus bimodal auditory stimulation in inner ear malformations: Cognitive, language, and motor performance

Filiz Aslan PhD¹  | Gorkem Ertugrul PhD¹  | Gonca Sennaroglu¹  |
Levent Sennaroglu² 

¹Department of Audiology, Faculty of Health Sciences, Hacettepe University, Ankara, Turkey

²Department of Otorhinolaryngology, Faculty of Medicine, Hacettepe University, Ankara, Turkey

Correspondence

Filiz Aslan, Department of Audiology, Faculty of Health Sciences, Hacettepe University, Ankara, Turkey.

Email: filizaslan@hacettepe.edu.tr

Abstract

Purpose: New perspectives on rehabilitation options for inner ear malformations have still been studied in the literature. This study investigated the cognitive, language, and motor skills of auditory brainstem implant (ABI) users in unimodal and bimodal groups.

Methods: The motor competency of the participants was assessed with Bruininks-Oseretsky Motor Proficiency Test-2 Short Form (BOT2 SF). Language performance was evaluated by the test of Early Language Development-3 and Speech Intelligibility Rating. Word identification, sentence recognition tests, and Categories of Auditory Performance were used to assess auditory perception skills. To examine the cognitive performance, Cancellation Test and Gesell Copy Form were administered. All the tests were conducted in a quiet environment without any distractions.

Results: The participants were divided into two groups: (1) 17 children in the unimodal group and (2) 11 children in the bimodal (who used a cochlear implant on one side and ABI on the other side) group. There were significant correlations between the chronological age of participants and BOT2 SF total score, cancellation tests, auditory perception tests, and language performance. Similarly, there were significant correlations between the duration of ABI use and auditory perception tests, language performance, cancellation test, and some BOT2 SF subtests ($r = -0.47$ to -0.60 , $p < .001$). There was no significant difference between the unimodal and bimodal groups in any task ($p > .05$). However, there were moderate-to-strong correlations among the auditory perception tests, cancellation test, language test, and BOT2 SF total score and subtests ($r = 0.40$ to 0.55 , $p < .05$).

Conclusion: Although there were no significant differences between bimodal and unimodal groups, a holistic approach, which indicates that hearing and balance issues can have broader impacts on a person's physical, emotional, social, and psychological aspects, should be used in the assessment process.

Level of Evidence: Level 4.

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KEYWORDS

auditory brainstem implantation, cochlear implantation, cognitive skills, inner ear malformations, motor competency

1 | INTRODUCTION

Inner ear malformations (IEMs) account for around 10%–20% of congenital sensorineural hearing loss.^{1,2} Sennaroglu¹ classified IEMs as complete labyrinthine aplasia (CLA), rudimentary otocyst (RO), cochlear aplasia (CA), common cavity (CC), incomplete partition type I (IP-I), type II (IP-II), type III (IP-III), cochlear hypoplasia type I (CH-I), type II (CH-II), type III (CH-III), type IV (CH-IV), enlarged vestibular aqueduct (EVA), and cochlear aperture abnormalities (CAA). Moreover, Sennaroglu¹ described the definite and possible indications for auditory brainstem implant (ABI): Definite indications of ABI are accepted as CLA, RO, CA, CAA, cochlear nerve aplasia, and IP-I, CC, CH with no cochlear nerve.

Bimodal stimulation is recommended for cochlear implant (CI) users to enhance their speech perception skills.^{3,4} The most widely used approach is one ear with a CI and a contralateral ear with a hearing aid. However, children with severe IEMs (such as cochlear nerve or cochleovestibular nerve hypoplasia) may have limited benefits with CIs and the application of ABI can be an option for their contralateral ear.⁵ Bimodal stimulation is suggested/performed in these cases who use ABI on the one ear and CI on the contralateral ear.⁵ These cases are described as bimodal users in this study.

The cerebellum and basal ganglia-brainstem pathways are crucial for postural control, while the cortical and cerebellar areas are vital for cognitive performance such as executive functions and learning.⁶ Children who are detected to have IEMs late are considered to have developmental delays in the early stages.^{7,8} Especially, parents of children with severe IEMs first apply to developmental pediatrics and child neurology clinics because their gross motor skills such as holding their head, sitting, and walking are quite delayed compared to their peers.⁷ Most of these studies reported the negative effect of additional cognitive impairments in IEMs or ABI users.^{9,10} As hypothesized that poor cognitive and speech perception, and speech production abilities can be associated.¹¹ However, in the literature, the authors did not find any comprehensive publication on cognitive, language, and motor skills in bimodal ABI users.

The aims of this study were to investigate the cognitive, language, and motor skills of unimodal and bimodal users. We hypothesized that the bimodal auditory stimulation group would have better performance on the cognitive, language, and motor skills than the unimodal group.

2 | MATERIALS AND METHODS

The non-Interventional Ethics Board of Hacettepe University approved this study (GO 21/640), which followed the Declaration of Helsinki's requirements. The parents of the study subjects provided their written and verbal agreement.

2.1 | Study sample

In this study, there were two study groups in this comparative cross-sectional study. To date, there are 139 pediatric ABI users in our clinic. In our clinic, there are 61 children using unilateral ABI for at least 2 years and 24 children using CI and ABI. A total of 41 out of 61 were between 4 and 15 years old, because of the cut-age of the auditory perception test battery is 15 years old. Rest of the children and families were not volunteered to attend the study various reasons (social distance rules, hygiene conditions for Pandemic, time-restrictions etc). The first study group ($\bar{X}_{\text{chronological age}} 10.09 \pm 3.52$) consisted of 17 children (6M:11F) who have been using an ABI (unimodal group) on one side for at least 2 years. At present, there are 12 children with bimodal implants who are over 4 years old in our clinic. One child's family refused to participate in the study due to the COVID-19 pandemic. Thus, the second study group ($\bar{X}_{\text{chronological age}} 8.56 \pm 2.54$) consisted of 11 children (3M:8F) who used a CI on one side and ABI on the other side (bimodal group), as shown in Table 1. The mean duration of ABI was 6.89 ± 3.25 years in the unimodal group and 4.02 ± 2.47 years in the bimodal group. The mean duration of CI was 5.30 ± 2.67 in the bimodal group, as shown in Table 1. Additional needs were determined four children (1 attention deficits, 1 CHARGE syndrome, and 2 learning disorders) in the unimodal group and four children (2 attention deficits, 1 social communication disorders, and 1 Down syndrome) in the bimodal group. Eight unimodal users have been using the auditory-verbal communication method, while 8 of them have been using the total communication approach, which indicates simultaneous use of multiple methods (such as oral, manual, and auditory). However, most of the bimodal users ($n=9$) have communicated with the auditory-verbal approach. Only one participant in each group had been using sign language. In this study, there was no control group including children with typical development.

2.2 | Data tools

2.2.1 | Motor performance

Bruininks-Oseretsky Motor Proficiency Test-2 Short Form

This test is a performance-based screening test that evaluates the motor development and coordination skills of individuals between the ages of 4 and 21.¹² It consists of eight main categories and 14 items: fine manual precision (drawing lines through paths—crooked, folding paper), fine motor integration (copying a square, copying a star), manual dexterity (transferring pennies), bilateral coordination (jumping in place—same sides synchronized, tapping feet and fingers—the same side synchronized), balance (walking forward on a

TABLE 1 Demographic characteristics of the participants.

| Hearing status | Groups | | | |
|------------------------|-------------------------|------------|------------------------|------------|
| | Unimodal users (n = 17) | | Bimodal users (n = 11) | |
| | Mean ± SD | Min-max | Mean ± SD | Min-max |
| Age (year) | 10.09 ± 3.52 | 4.00-16.60 | 8.56 ± 2.54 | 4.90-12.50 |
| Age of ABI (year) | 3.37 ± 1.70 | 1.25-6.45 | 4.37 ± 1.49 | 1.72-6.67 |
| Age of CI (year) | - | - | 3.10 ± 2.30 | 1.03-8.08 |
| Duration of ABI (year) | 6.89 ± 3.25 | 2.30-13.60 | 4.02 ± 2.47 | 0.35-8.46 |
| Duration of CI (year) | - | - | 5.30 ± 2.67 | 0.27-9.87 |

| Communication method | Groups | | | |
|----------------------|-------------------------|-------|------------------------|-------|
| | Unimodal users (n = 17) | | Bimodal users (n = 11) | |
| | n | % | n | % |
| Total communication | 8 | 47.06 | 1 | 9.10 |
| Sign language | 1 | 5.88 | 1 | 9.10 |
| Auditory-verbal | 8 | 47.06 | 9 | 81.80 |

| Inner ear malformation types | Groups | | | |
|------------------------------|-------------------------|-----------------------|------------------------|------------------|
| | Unimodal users (n = 17) | | Bimodal users (n = 11) | |
| | ABI side (n = 17) | Non-ABI side (n = 17) | ABI side (n = 11) | CI side (n = 11) |
| Cochlear aplasia | 2 | 1 | 1 | 1 |
| Cochlear hypoplasia | 6 | 7 | 5 | 6 |
| Common cavity | 4 | 4 | 2 | 2 |
| Incomplete partition Type I | 3 | 3 | 1 | 1 |
| Labyrinthine aplasia | 1 | 1 | 1 | 0 |
| Cochlear aperture stenosis | 1 | 1 | 1 | 1 |

line, standing on one leg on a balance beam—eyes open), running speed and agility (one-legged stationary hop), upper-limb coordination (dropping and catching a ball—both hands, dribbling a ball—alternating hands), and strength (knee push-ups, sit-ups). All raw scores of Bruininks–Oseretsky Motor Proficiency Test-2 Short Form (BOT2 SF) (BOT-2 SF) are converted to the point scores and the total point score of short-form (maximum = 88) is calculated. The total BOT-2 SF point scores for each subject are then converted to standard scores based on age and gender using the BOT-2 manual's normative data. The standard scores may be described into five categories as “well-above average, above average, average, below average, and well-below average.”

2.2.2 | Language development

The Test of Early Language Development-3 Edition

The language performance of the participants was assessed with the Test of Early Language Development-3 Edition (TELD-3): Turkish version.¹³ The test has two subtests: receptive language and expressive language. The TELD-3 evaluates spoken language performance of

children aged 2;0 and 7;11 years old, focusing on semantics, syntax, and morphology. The age ranges of the participants are not represented in the norm scores, so the standard scores do not apply to the current study. The age-equivalent scores were determined in both the receptive and expressive language subtests.

Speech Intelligibility Rating

Speech Intelligibility Rating (SIR) was conducted to examine speech intelligibility in children with ABI implants by quantifying spontaneous speech for clinical comparisons.¹⁴ After listening to each child's utterances, an experienced clinician rated his or her speech. This was used to assess the child's speech intelligibility in real-life situations.

2.2.3 | Speech perception performance

Categories of Auditory Performance-II

Categories of Auditory Performance (CAP) was developed as a rating scale to assess the auditory performance of CI users in everyday situations.¹⁵ The CAP-II is a modified version, and it covers different ranges of auditory performance. The categories are ranked between

0 (no awareness to environmental sounds) and 9 (use of the phone with an unknown speaker in an unpredictable context).

Word Identification Test

To assess the ability to identify the three-syllable words in the closed set condition.¹⁶ The test contained 12 pictures of common objects. During the test, the child was asked to show a picture of a word. Each word is presented twice randomly. The total score ranged between 0 (no response) and 24 (all correct responses).

The Sentence Recognition Test

To evaluate the sentence recognition performance of the participants, a list of 10 sentences was presented.¹⁶ The children were asked to repeat these words/sentences. Each sentence was presented only in an auditory, using a live voice. The total score is the reported percentage of sentences repeated correctly.

2.2.4 | Cognitive development performance

Cancellation Test

To measure the general attention and visuospatial abilities.¹⁷ The participant is given two verbal and two nonverbal symbol sheets in Mesulam and Weintraub's Cancellation Test. In the symbol cancellation test, on an 8.5 × 1.0-inch page, a range of known and unfamiliar forms (e.g., circles, triangles, and stars) are presented in both randomized and organized formats in the symbol component of this test. Participants are asked to draw a line through all of the target shapes they can find on this page. Sixty target stimuli (15 targets in each quadrant) are embedded in a background of more than 300 distractor stimuli in the random symbol version. In the letter cancellation task, the participants are asked to draw a line on all "A" symbol, under similar conditions. The number of omission errors calculated as a task performance. This test assesses executive functions such as visual attention, interference control, cognitive flexibility, and response inhibitions.

Gesell Copy Form Test

Gesell Developmental Test, developed by Arnold Gesell, to assess children will be handled and evaluated within the framework of developmental stages, depending on the comprehension of physical object and space relations.¹⁸ The developmental figures test is a subtest of the Gesell Development Test. An A4 size paper, rearranged for use in clinical practice and containing all the figures side by side, is placed in front of the child and the child is asked to draw each figure. It included nine figures. The total score is obtained by summing the shape distortion, combining and rotation errors separately for each figure.

2.3 | Procedure

The assessments were explained to the parents and children and informed consent was obtained from the parents. Then, participants

were assessed in a quiet therapy room without any distraction. The data tools were completed in two sessions on the same day. In the first session, the cognitive, speech perception, and language tests were administered by the first author. In the second session, the motor performance of the child was assessed by the second author. If the children needed it, they were given 10-min intervals between tests. All tests were performed for each participant with ABI or CI and ABI device-on mode, to obtain the optimal performance of the children.

2.4 | Statistical analysis

The Kolmogorov-Smirnov test was used to determine the normality of data distribution for numerical variables. Descriptive statistics are reported as mean, standard deviation, median, and minimum-maximum values. The categorical variables are presented as frequencies and percentages. Mann-Whitney *U* test was used to compare the differences between the groups as a non-parametric test. The correlations between the qualitative data were analyzed using Fisher's Exact chi-square test. Spearman's correlation coefficient was used to determine the correlation between motor scores, language skill performance, auditory perception performance, and cognitive task scores. The significance level was accepted as $p < .05$, and an effect size of >0.50 was considered.¹⁹ All statistical analyses were performed using the IBM SPSS Statistics 23.0 software.²⁰

3 | RESULTS

In this study, cochlear hypoplasia (13 ears in the unimodal group, 11 ears in the bimodal group) was the most common IEM in both groups. This is followed by the common cavity IEM (8 ears in the unimodal group, 4 ears in the bimodal group), as shown in Table 1. In Table 1, the minimum values of CI and ABI durations were less than 6 months because two children in the bimodal group had recently started to use their second auditory implants.

There was no significant correlation between the chronological age of the participants and their BOT2 SF total performance scores. However, there was a moderately significant correlation between the duration of ABI use and BOT2—fine motor integration ($r = 0.43$, $p < .05$), manual dexterity ($r = 0.53$, $p < .001$), upper limb coordination ($r = 0.42$, $p < .05$), and strength ($r = 0.58$, $p < .001$). In the cognitive tasks, there was a strong negative correlation between chronological age and cancellation tasks ($r = -0.61$ to -0.72 , $p < .001$). Similarly, there was a moderate negative correlation between the duration of ABI use and cancellation tasks ($r = -0.47$ to -0.60 , $p < .001$).

Additionally, there was a moderate positive correlation between chronological age and auditory perception tasks (CAP $r = 0.53$, word identification $r = 0.62$, sentence recognition $r = 0.58$, $p < .001$). There were strong correlations between chronological age and language scores (receptive $r = 0.86$, expressive $r = 0.65$, $p < .001$). According to the duration of ABI use, there was a moderate correlation between

TABLE 2 The comparisons between groups in all tasks.

| | Unilateral ABI | | Bimodal (CI + ABI) | | p |
|---|----------------|----|--------------------|----|-------------------|
| | Mean ± SD | n | Mean ± SD | n | |
| BOT-2 SF Standard Score | 33.71 ± 5.07 | 17 | 40.45 ± 16.39 | 11 | .12 ^a |
| Gesell | 3.65 ± 3.571 | 17 | 4.64 ± 3.72 | 11 | .48 ^b |
| Cancellation Letter random error | 16 ± 4 | 13 | 3 ± 4 | 7 | .27 ^b |
| Cancellation Letter structured error | 12 ± 4 | 13 | 4 ± 4 | 7 | .48 ^b |
| Cancellation Symbol random error | 15 ± 5 | 13 | 2 ± 3 | 7 | .18 ^b |
| Cancellation Symbol structured error | 77 ± 21 | 13 | 34 ± 81 | 7 | .81 ^b |
| CAP | 3.94 ± 1.24 | 17 | 4.36 ± 1.50 | 11 | .40 ^b |
| SIR | 2.29 ± 1.49 | 17 | 2.82 ± 1.72 | 11 | .51 ^b |
| Word identification | 71.18 ± 37.06 | 17 | 69.09 ± 41.09 | 11 | .96 ^b |
| Sentence recognition | 29.29 ± 32.22 | 17 | 31.82 ± 30.92 | 11 | .96 ^b |
| Receptive language | 51.76 ± 19.15 | 17 | 45.82 ± 20.83 | 11 | .35 ^b |
| Expressive language | 34.59 ± 20.62 | 17 | 37.09 ± 24.27 | 11 | 1.00 ^b |

Abbreviations: BOT-2 SF, Bruininks–Oseretsky Motor Proficiency Test-2 Short Form; CAP, categories of auditory performance; SD, standard deviation; SIR, speech intelligibility rate.

^aIndependent Sample t-test.

^bMann–Whitney U Test.

TABLE 3 The correlations between the BOT-2 SF subtests and cognitive tasks (n = 28).

| | BOT-2 Short form | | | | | | | |
|---|----------------------------|--------|------------------------------|--------|------------------------|--------|----------------------|--------|
| | Fine Motor Precision Score | | Fine Motor Integration Score | | Manual Dexterity Score | | Total Standard Score | |
| | <i>r_s</i> | p | <i>r_s</i> | p | <i>r_s</i> | p | <i>r_s</i> | p |
| Cognitive tasks | | | | | | | | |
| Gesell figures | −0.562 | .002* | −0.569 | .002* | −0.537 | .003* | −0.248 | .203 |
| Cancellation Letter random error | −0.799 | <.001* | −0.718 | <.001* | −0.866 | <.001* | −0.769 | <.001* |
| Cancellation Letter structured error | −0.755 | <.001* | −0.710 | <.001* | −0.770 | <.001* | −0.602 | <.001* |
| Cancellation Symbol random error | −0.743 | <.001* | −0.676 | <.001* | −0.755 | <.001* | −0.512 | <.001* |
| Cancellation Symbol structured error | −0.715 | <.001* | −0.649 | <.001* | −0.755 | <.001* | −0.512 | .005* |
| Speech perception tasks | | | | | | | | |
| Word identification | 0.502 | .006* | 0.427 | .024* | 0.513 | .005* | 0.321 | .096 |
| Sentence recognition | 0.501 | .007* | 0.448 | .017* | 0.463 | .013* | 0.403 | .034* |
| CAP | 0.605 | .001* | 0.424 | .025* | 0.485 | .009* | 0.556 | .002* |
| Language performance tasks | | | | | | | | |
| Receptive language performance | 0.653 | <.001* | 0.585 | .001* | 0.664 | <.001* | 0.361 | .059 |
| Expressive language performance | 0.653 | <.001* | 0.585 | .001* | 0.489 | .008* | 0.556 | .002* |
| SIR | 0.569 | .002* | 0.505 | .006* | 0.462 | .13* | 0.529 | .004* |

Abbreviations: BOT-2 SF, Bruininks–Oseretsky Motor Proficiency Test-2; CAP, Categories of Auditory Performance; *r_s*, Spearman's Correlation Coefficient; SIR, Speech Intelligibility Rate.

*p < .05 statistically significant.

auditory perception tasks and language scores. There was also a moderate negative correlation between the duration of ABI use and cancellation tasks.

There were no significant differences between the groups in terms of motor performance screening (BOT-2 SF scores), cognitive tasks (Gesell figures and Cancellation tests), speech perception tasks (word identification, sentence recognition, and CAP), and language performances (receptive language, expressive language, and SIR), as shown in Table 2 ($p > .05$). However, significant negative correlations were found between the BOT-2 SF standard scores and errors of the cancellation test assessing cognitive abilities (letter random error $r = -0.76$, $p < .001$; letter structured error $r = -0.60$, $p < .001$; symbol random error $r = -0.51$, $p < .001$; symbol structured error $r = -0.51$, $p < .05$; Table 3). Moreover, there were significant positive correlations between the BOT-2 SF standard scores and speech perception tasks (sentence recognition $r = 0.40$, $p < .05$; CAP $r = 0.55$, $p < .05$) and language performance tasks (expressive language $r = 0.55$, $p < .05$; SIR $r = 0.52$, $p < .05$; Table 3). In particular, only fine motor precision, fine motor integration, and manual dexterity subtest of the BOT-2 SF were significantly associated with the cognitive tasks, speech perception, and language performance, as shown in Table 3 ($p < .05$).

In the cognitive tasks, all participants completed drawing the Gesell figures in the copy form test. Thirteen children in the unilateral group were delayed in drawing Gesell figures, and six children in the bimodal group showed poor performance. Four children in the unilateral group and five children in the bimodal group successfully completed the task. In the cancellation tasks, four children in the unilateral ABI group, and four children in the bimodal group did not complete the tasks. Therefore, in Table 4, the comparisons between the groups in terms of BOT-2 SF subtests were analyzed among 20 participants ($n = 13$ in the unimodal group, $n = 7$ in the bimodal group). According to the BOT-2 SF subtests, there was a significant difference between the performances of the unimodal and bimodal groups in balance and running speed and agility subtests ($p < .05$; Table 4). The bimodal group showed better performance in these subtests. There was no significant difference the cognitive and speech perception performance between groups. There was no significant correlation between the other subtests of the BOT-2 SF and cognitive and speech-language performance ($p > .05$).

In this study, the scores of Gesell (delayed, normal) and BOT-2 SF (below average, average, and above-average) were divided into two categories. Only children who completed the cognitive tasks ($n = 20$) were included in this analysis. There was a significant correlation between both categories ($p < .05$; Table 5). According to this finding, participants who had delayed Gesell scores performed below average on the BOT-2 SF test.

4 | DISCUSSION

This study investigated the cognitive, language, and motor skills of unimodal and bimodal users who had IEMs. The main findings to emerge from the analysis was that manual dexterity, fine motor precision, and integration were associated with all cognitive, speech, and

TABLE 4 The comparisons of BOT-2 Short Form subtests between the groups.^a

| Groups | Fine motor precision | | Fine motor integration | | Manual dexterity | | Bilateral coordination | | Balance | | Running speed and agility | | Upper-limb coordination | | Strength | |
|-----------------------|----------------------|--|------------------------|--|------------------|--|------------------------|--|-------------|--|---------------------------|--|-------------------------|--|-------------|--|
| | Mean ± SD | | Mean ± SD | | Mean ± SD | | Mean ± SD | | Mean ± SD | | Mean ± SD | | Mean ± SD | | Mean ± SD | |
| Unimodal ($n = 13$) | 9.15 ± 2.37 | | 9.54 ± 1.39 | | 5.69 ± 1.97 | | 6.54 ± 0.96 | | 4.23 ± 1.92 | | 4.23 ± 1.87 | | 7.85 ± 3.13 | | 8.85 ± 3.36 | |
| Bimodal ($n = 7$) | 10.86 ± 3.28 | | 9.57 ± 0.78 | | 5.71 ± 2.43 | | 7.00 ± 0.00 | | 5.86 ± 2.03 | | 7.29 ± 1.25 | | 9.00 ± 3.36 | | 9.57 ± 3.25 | |
| p | .19 | | .53 | | .87 | | .18 | | .05* | | <.001* | | .35 | | .66 | |

Abbreviations: BOT-2 SF, Bruininks-Oseretsky Motor Proficiency Test-2; SD, standard deviation.

^aMann-Whitney U Test.

* $p < .05$ statistically significant.

TABLE 5 Comparison between BOT-2 SF and Gesell performance groups.

| Chi-square | BOT-2 SF Total Score | | | | | | <i>p</i> ^a |
|------------|---------------------------|----|---------------|----|----------|-----|-----------------------|
| | Average and above average | | Below average | | Total | | |
| | <i>n</i> | % | <i>n</i> | % | <i>n</i> | % | |
| Gesell | | | | | | | |
| Delayed | 3 | 15 | 14 | 70 | 17 | 85 | .01* |
| Normal | 3 | 15 | 0 | 0 | 3 | 15 | |
| Total | 6 | 30 | 14 | 70 | 20 | 100 | |

Abbreviation: BOT-2 SF, Bruininks–Oseretsky Motor Proficiency Test-2 Short Form.

^aFisher's exact chi-square test.

**p* < .05 statically significant.

language performances. These findings are consistent with our previous research.⁷ In children with ABI, Ertugrul et al.²¹ found a linear correlation among manual control, balance, speech perception, and language skills. In the literature, there were several studies indicating correlations between fine- and gross motor development and language (e.g., vocabulary) performance in children.^{22,23} According to a systematic review study by van der Fels and colleagues,²⁴ weak to strong associations were found between some motor (fine manual control, manual dexterity) and cognitive (visual processing, short and long-term memory) skills in children with typical development. As an interesting finding, there is no significant correlation between the bilateral coordination, balance, running speed and agility, upper-limb coordination, and strength. According to Luz et al.,⁶ the poor association between the balance and cognitive skills may be explained by the fact that balance is not regulated by a high level of cognitive processing. In contrast to the current study, Higashionna et al.²⁵ has reported a significant but poor correlation between the motor coordination ability, cognitive ability, and academic achievement of Japanese children with neurodevelopmental disorders. Also, in the same study, balance and manual dexterity were associated with both cognitive and academic skills.²⁵ It was determined that as the chronological age increased, the performance of the tasks in terms of motor skills (bilateral coordination, balance, running speed and agility), cognitive skills (attention and reasoning), language skills and auditory perception skills (word identification and sentence recognition) increased. This suggests that the effect of the developmental maturation of the participants over time on the results of the tests should be taken into account.

Moreover, it was found that as the duration of ABI use increased fine motor integration, manual dexterity, upper limb coordination, and strength scores were increased. However, there were no significant correlation between the duration of ABI use and balance, bilateral coordination, and running speed and agility subtests. In contrast to our study, Cushing et al.²⁶ has reported that the duration of CI use was also found to be a major factor in balance ability as in the BOT-2 complete balance subtest. These discrepancies may be explained by several factors: (1) The BOT-2 SF contains only two items in the balance subtest rather than the BOT-2 complete balance subtest. (2) All participants had severe IEMs, which are definite indication for ABI in this study. In this study, when the duration of ABI use increased, the participants' visual attention and reasoning performances were getting

better. The observed relationship between the duration of ABI use and cancellation tasks could be interpreted in this study: As the children gain more experience with ABI, their attention and reasoning abilities will probably improve. Our results supported Colletti and Zocante's²⁷ findings that after at least 1 year of experience with ABI, visuospatial attention and fluid reasoning performances of these children were improved significantly. Moreover, we should keep in mind that cognitive skills, such as reasoning can be improved also with age. Consistent with the literature, this study found that auditory perception and language skills enhance with the duration of ABI use.^{28–30}

In this study, there was no significant difference between the unimodal and bimodal groups in terms of cognitive, language, and motor competence. There are several possible explanations for these results. First, even though the mean ages of both groups were matching, the age of the children varied. Second, their experience with their ABIs and CIs were not matching.

In the recent literature, a strong relationship has been reported between the cognitive and motor skills of children with typical development.^{31–33} According to a comprehensive systematic review³² on the motor competence of typically developing children, physical exercise during childhood significantly improves and supports their motor skills, skeletal development, and communication skills. Zeng et al.³⁴ has stated in a comprehensive review study that physical activity has a positive effect on motor skills and cognitive development in children with typical development aged 4–6. Hudson et al.³³ indicate that participating in activities including cognitive-motor skills may increase not only motor competence but also computational skills in children with typical development. McClelland and Cameron³¹ claim that executive functions and motor skills develop synergistically. Dominance in one could compensate for a shortcoming in the other. Also, children have a stronger correlation between the two skills.³¹ Consistently, we found that as the motor competence improved, the participants' cognitive skills, such as general attention and visuospatial abilities were also progressed. According to this finding, if the unimodal or bimodal users have poor motor competence, the probability of making errors on cognitive tasks, including attention and visuospatial perception, may increase in these children.

There were no significant differences between the unimodal and bimodal groups when all participants were included in the current study. When the participants with additional needs did not

complete the cognitive tasks and were excluded from the study, bimodal users had significantly better performance on balance and running speed and agility tasks compared with unimodal users. As stated in previous studies,^{35,36} the sense of hearing and CI may help improve postural stability while standing or walking and to minimize the probability of falling. Unfortunately, the effect of ABI on balance and postural stability is still unknown. Additionally, we speculate that cognitive skills may play an important role in performing these motor activities. Similarly, some studies have shown that there was a positive correlation between motor development and cognitive development in typically developing children or children in the low risks.³⁷⁻³⁹

According to our categorical data of motor and visuospatial perception performances, 70% (14/20) participants who performed below average in BOT-2 SF total scores also lagged behind their peers in Gesell scores. This finding may help us understand whether there can be a relation between the motor competence and cognitive abilities such as visuospatial perception, in these children.

In this study, we emphasized the importance of vestibular, speech perception, and cognitive assessment of children with IEMs. The authors suggest that multidimensional and comprehensive test batteries and rehabilitation programs are needed to determine the benefits of bimodal stimulation in these groups of children. One of the study's limitations was that the sample size was limited in both groups. Due to the limited number of patients followed in the clinic, the low number of patients in both groups is an expected result. As a result, the scarcity of our case series limits the generalization of findings. However, it should not be underestimated in demonstrating the necessity of including both auditory and balance assessment in the assessment process. In clinical settings, the findings should be interpreted with caution. Second, there were no preoperative scores of children because the age at implantation of children with IEMs is usually under 4-year-old in our clinic. Additionally, all participants must be older than 4 years old as a requirement of the data tools.

Modern technologies (TV, tablet, smartphone, etc.) have a negative effect on motor competence and visual perception, fine motor precision and integration, manual dexterity, and strength.³² Ling et al.⁴⁰ have shown that the children who spent more time with modern technologies have poor performance on these tasks compared with children who spent less time with these tools. There are still many unanswered questions about the factors affecting motor skills, cognitive performance, and speech perception skills of children with IEM due to unimodal and bimodal stimulation.

In summary, there was no statistically significant difference between unimodal and bimodal groups, the moderate-to-strong correlation between motor tasks, cognitive tasks, and speech and language performance. In the future studies, larger sample size and comprehensive test battery could provide more information on these groups.

5 | CONCLUSION

This study has shown that the motor competence, cognitive skills, speech perception and language performance of children with IEMs are necessary to determine their holistic improvement of them with

both unimodal and bimodal auditory stimulation. In the clinical settings, professionals benefit from the holistic approach during their assessments and follow up process of ABI users. To the authors' knowledge, this is the first study to investigate the relationships between cognitive, motor, and language skills, as well as speech perception, in children with severe IEMs. We believe that this study will guide the future researches in the field.

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CONFLICT OF INTEREST STATEMENT

The authors declare no conflicts of interest.

ORCID

Filiz Aslan  <https://orcid.org/0000-0002-4267-2126>

Gorkem Ertugrul  <https://orcid.org/0000-0003-2567-2711>

Gonca Sennaroglu  <https://orcid.org/0000-0002-3337-2391>

Levent Sennaroglu  <https://orcid.org/0000-0001-8429-2431>

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