# Characteristic of Sound Localisation of Unilateral Microtia and Atresia with and without a Non-surgical Bone Conduction Device

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#### Abstract

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# ABSTRACT

**Objective** : To determine the characteristics of sound localisation in children with unilateral microtia and atresia (UMA) when unaided and aided with a non-surgical bone conduction device (BCD).

**Design:**Retrospective data analysis.

Setting : Tertiary referral centre.

Participants: Eleven children with UMA and 11 age-matched children with NH as control.

Intervention: A non-surgical BCD.

Main outcomes and measures: The sound field hearing threshold, word recognition score (WRS), speech reception threshold, subjective questionnaire of the international outcome inventory for hearing aids, and sound localisation tests.

**Results** : The average unaided WRS and speech-to-noise ratio (SNR) for all UMA patients was 18.27  $\pm$  14.63 % and -5  $\pm$  1.18 dB sound pressure level (SPL), while the average aided WRS and SNR conspicuously changed to 85.45  $\pm$  7.38 % and -7.73  $\pm$  1.42 dB SPL, respectively. Compared with the stimulated UHL, some children with UMA were able to detect sound sources and this sound localisation ability did not deteriorate with a BCD.

**Conclusions** : For children with UMA, the non-surgical BCD provided a definite benefit on speech recognition and high participant satisfaction without deteriorating their sound localisation abilities. It is an efficient and safe solution for the early hearing intervention of these patients.

#### Key points

- 1. The sound localisation ability was disrupted both in children with UMA and stimulated UHL.
- 2. In children with UMA, an inter-subject variability of the directional hearing was observed. Some of them could use the remaining distorted binaural cues to detect sound sources, unlike the children with stimulated UHL.
- 3. The non-surgical BCD provided a definite benefit on speech recognition and high participant satisfaction to children with UMA.
- 4. The non-surgical BCD did not improve or deteriorate the original sound localisation ability of children with UMA.
- 5. The non-surgical BCD is a valid and safe device for children with UMA and is recommended as an interventional device from an early age.

**Keywords** : Unilateral; Microtia and Atresia; Conductive Hearing Loss; Speech Perception; Sound Localisation; Adhesive Bone Conduction Device

#### INTRODUCTION

Microtia and atresia, developmental malformations of the external and middle ear, is a common cause of congenital unilateral conductive hearing loss  $(\text{UCHL})^{1, 2}$ . Disability in speech recognition in noisy backgrounds and inaccuracy of sound localisation are the two main functional deficits experienced by these patients <sup>3, 4</sup>.

Common treatment options for patients with unilateral microtia and atresia (UMA) include traditional canaloplasty, active middle ear implants, bone conduction implants, and non-surgical bone conduction devices (BCDs). For children with UMA, who are not willing to undergo surgery or who have not reached the age for surgery, non-surgical BCDs represent an important transition intervention <sup>5</sup>. However, whether these patients can achieve more accurate sound localisation after receiving such interventions remains disputed, as diverse audiological results are reported. Some studies have shown remarkable improvements in horizontal spatial hearing in patients with UCHL using a BCD<sup>6, 7</sup>. In contrast, other studies have suggested that patients with congenital UCHL had adapted to their hearing impairment through the use of spectral shape cues and ambiguous monaural head shadow effect (HSE) cues from the normal hearing ear that they had acquired through long-term unilateral hearing deprivation <sup>10, 11</sup>. Using a BCD may cause such listening cues to be distorted, thereby jeopardizing directional hearing.

Given the uncertain benefits of hearing amplification and unesthetic appearance, studies concerning the sound localisation ability of children with UMA are limited by heterogeneous patient populations, varying in study design and audiological test results. Currently, there is no research investigating the characteristics of sound localisation and the effects of non-surgical BCDs in school-aged children with UMA. To provide a theoretical basis for early hearing intervention of non-surgical BCDs for children with UMA, two primary objectives were addressed in this study. First, to compare characteristics of sound localisation in children with congenital UMA and stimulated UCHL. Second, to detect the effect of a BCD on speech perception and sound localisation in children with congenital UMA.

## METHODS

## 2.1 Participants

Eleven children (mean: 7.45 years, standard deviation [SD]: 1.81 years) who had UMA and experienced congenital UCHL were included. All patients had normal hearing (NH) in one ear (hearing thresholds of [?] 25 dB HL across 0.5–4 kHz) and pure conductive hearing loss in the impaired ear (air-bone gap of [?] 25 dB HL, with bone conduction thresholds of [?] 25 dB HL across 0.25–4 kHz). Eleven children who had bilateral NH were recruited as controls. Detailed demographic data are summarized in Table 1.

#### 2.2 Setup and stimuli

All tests were conducted in a double-walled soundproof laboratory. Participants sat in a chair placed 1 m in front of seven loudspeakers. Sound field hearing thresholds were obtained by warble tones for octave frequencies across 0.25-4 kHz in dB hearing level (HL). Speech perception under quiet was measured by the word recognition score (WRS [%]) of the Mandarin speech test materials (MSTMs) <sup>12</sup> at 65 dB speech presentation level (SPL). Speech perception in noise was measured by the speech reception threshold (SRT) of the MSTMs. The spectrum-shaped noise (SSN) was set at 65 dB SPL, and the speech signal started at 0 dB speech-to-noise ratio (SNR), with the following disyllables changing adaptively in 2 dB SPL steps as the participants responded. The SRT was defined as the speech signal level presented when a participant identified 50% of the words correctly. The SNR was calculated as the difference between the SRT and SSN. The sound localisation test (Supplementary Fig. 1a) consisted of seven audiometric loudspeakers placed at 30deg intervals in a semicircle within a horizontal plane (+- 90deg, azimuth). Broadband noise (0.5–20 kHz) was randomly played at three different sound levels (65-, 70-, and 75-dB SPL). Each time the loudspeakers finished a presentation, participants indicated which loudspeaker they believed to be the source of the burst.

#### 2.3 Device and listening conditions

An adhesive BCD (DHEAR, MED-EL, Innsbruck, Austria) was used in the current study. For all experimental conditions, the devices were set to omnidirectional mode, and the volume was adjusted according to patients' preferences.

Children with UMA were tested with the BCD off (unaided condition) and on (aided condition) (Supplementary Fig. 1b). Children with NH were measured with both ears unplugged (UP condition) as normal controls. When measuring the sound localisation, control listeners were tested with plugging (P condition) to stimulate an acquired UCHL to reveal the characteristics of directional hearing of acquired UCHL. Plugging was performed by covering an ear with an earmuff (Peltor X5A; 3M Company, MN, USA), along with a foam earplug (E-A-R soft; 3M Company) inserted into the external auditory canal. For the UMA group, unaided audiological tests were performed on the day when receiving the BCD and aided audiological tests were measured after a mean period of 9.27 +- 1.85 weeks. For the NH group, all tests were performed in one visit.

#### 2.4 Subject satisfaction

The Chinese version of the international outcome inventory for hearing aids (IOI-HA) (IOI-HA)<sup>13</sup> was handed out to all patients at the end of the follow-up. The IOI-HA consists of seven items: daily use, benefit, residual activity limitations, satisfaction, residual participation restrictions, impact on others, and quality of life. Each item has a five-point rating scale, with higher ratings reflecting better outcomes (or fewer residual difficulties).

#### 2.5 Data analysis

 $MAE = \sum_{i=1}^{n} |\alpha|$  $RESP^{-\alpha}$ 

 $\operatorname{TARG}^{i}_{\overline{n}}(1)$ 

 $\alpha_{\text{RESP}} = g \bullet \alpha_{\text{TARG}} + b \ (2)$ 

The mean absolute error (MAE) was calculated using Equation 1 to assess the sound localisation accuracy under different conditions. The  $a_{\text{RESP}}$  and  $a_{\text{TARG}}$  referred to the response azimuth (in degrees) and target azimuth (in degrees), respectively. Additionally, the best linear fit of the target–response relationship for each participant was also computed using Equation 2, where g is the response gain (slope, dimensionless), and b is the response bias (offset in degrees). In this study, the right side was defined as the impaired side; therefore, azimuth coordinates for patients with left ear impairment and controls with NH with left ears plugged were inverted.

Paired and independent t- tests were conducted to evaluate differences under different test conditions. The p-values of <0.05 and <0.001 were considered statistically significant. SPSS version 26.0 and GraphPad Prism version 8.0 were used to analyse the data and draw diagrams.

#### RESULTS

#### 3.1 Hearing thresholds

For patients with UMA, the mean  $\pm$  SD of the unaided hearing threshold was 51.36  $\pm$  5.02 dB HL, which significantly improved to 27.64 $\pm$ 2.38 dB HL with a mean FHG of 23.73  $\pm$  3.47 dB HL (p < 0.001). For the NH comparison group, the mean hearing threshold was 15.36  $\pm$  3.88 dB HL. The aided mean hearing threshold of the UMA group was still higher (worse) than the mean hearing threshold of the NH group (p < 0.001). Detailed outcomes across 0.5–4 kHz are depicted in Fig. 1a.

#### 3.2 Speech perception abilities

In Fig.1b-c, the average unaided WRS and SNR for patients with UMA were  $18.27 \pm 14.63$  % and  $-5 \pm 1.18$  dB SPL, respectively, while the average aided WRS and SNR conspicuously changed to  $85.45\pm7.38$  % and  $-7.73 \pm 1.42$  dB SPL, respectively (WRS: p < 0.001; SNR: p < 0.05). The mean WRS and SNR of the comparison group were  $99.27 \pm 1.35$  % and  $-10.55 \pm 2.77$  dB SPL, respectively. Fig. 1b-c also indicates significant differences in the speech levels between the patients aided with BCDs and their peers with NH (WRS: p < 0.001; SNR: p < 0.05).

#### 3.3 Sound localisation in patients with UMA and stimulated UHL

Fig. 2 shows the individual sound localisation target-response plots for two children with UMA and one control (P6, P7, and N2) under monaural (unaided and P, left column) and binaural (aided and UP, right column) listening conditions. Under the unaided condition, P6 showed a poor localisation ability and perceived most stimuli from the healthy ear side. However, P7 exhibited relatively better sound localisation accuracy than P6. When aided with the BCD, the sound localisation accuracy improved in P6 (delta gain = 0.422, delta MAE =  $-14.28^{\circ}$ ) and worsened in P7 (delta gain = -0.167, delta MAE =  $18.58^{\circ}$ ). Under the P condition, all data points of N2 fell along the diagonal dotted line, indicating a sharply deteriorated localisation performance (gain = 0.12, MAE =  $65^{\circ}$ ).

Individual data on gender, age, MAE, response gain, bias, and r<sup>2</sup> for all participants are presented in Table 2. The MAE and response gain under monaural listening conditions (unaided and P) are plotted against those under binaural listening conditions (aided and UP) in Fig. 3a–b. Varying sound localisation performance was observed in the 11 children with UMA under the unaided condition. When the mean gain and MAE of all children with UMA were compared between the unaided and aided conditions, no significant differences were identified (gain: p = 0.104, MAE: p = 0.436). Control listeners showed good sound localisation performance in the UP condition. All of them exhibited considerable deterioration after being plugged (gain: p < 0.001; MAE: p < 0.001), with most of them unable to localize the stimuli presented from the plugged side. Although no significant difference was observed between the unaided and P conditions (gain: p = 0.073; MAE: p = 0.073), the results indicated that children with UMA showed better sound localisation performance (smaller MAE and gain) than the stimulated UHL listeners. This phenomenon might be related to the adaptation to congenital unilateral asymmetric hearing loss; however, the benefit of this adaptation was insufficient for children with UMA to localize sound as accurately as the normal controls did.

### 3.4 Influence of a BCD on sound localisation accuracy

Fig. 3c–d shows the localisation accuracy of the patients with UMA and the NH controls on the impaired (the atretic and plugged) side and the contralateral (the healthy and unplugged) side, respectively. A better sound localisation accuracy was observed in children with UMA ( $43.18 \pm 30.58^{\circ}$  vs.  $83.18 \pm 37.82^{\circ}$ , p < 0.05) on the impaired side than in controls in the P condition (Fig. 3c). When aided with a BCD, there was no

difference in the MAE between the unaided and aided conditions on the impaired side (43.18  $\pm$  30.58° vs. 34.14  $\pm$  17.9°, p = 0.303) or the contralateral side (26.97  $\pm$  24.68° vs. 27.42  $\pm$  14.52°, p = 0.79), indicating that the BCD use was not detrimental to the original sound localisation ability of the patients with UMA.

According to the criterion in a previous study, 11 children with UMA were divided into two subgroups: good performers (n = 5; gain > 0.75) and poor performers (n = 6; gain [?] 0.75) <sup>14</sup>. When the MAE outcomes were separately compared bilaterally, a significantly better sound localisation accuracy on the atretic side was observed in good performers (15.67deg +- 10.71deg vs. 66.11deg +- 19.77deg, p < 0.05, Fig. 4a). Linear regression was further conducted to explore the predictive effect of gain on the benefits of sound localisation accuracy by fitting BCDs (delta MAE, MAE<sub>aided</sub> - MAE<sub>unaided</sub>). The results revealed an evident relationship between response gain and delta MAE (r<sup>2</sup> = 0.553, p < 0.05), indicating that children with UMA who have poor sound localisation performance (lower gain) show more improvement after being fitted with BCDs (Fig. 4b).

#### 3.6 Participant satisfaction

An IOI-HA survey was administered to all BCD users. The mean overall IOI-HA score was 4.57 + 0.73. A score > 3 per item, defined as a benefit from the BCD, was found for nearly all participants. Mean scores for items 1–7 were 3.86 + 0.31, 3.57 + 0.25, 4.29 + 0.19, 3.79 + 0.24, 4.14 + 0.14, 4.64 + 0.13, and 4 + 0.23, respectively.

#### DISCUSSION

#### 4.1 Hearing benefits

Here, the ADHEAR system remarkably improved the hearing thresholds and speech perception under quiet and noisy conditions in patients with UMA. Patients with UMA had a mean FHG of 23.73 +- 3.47 dB HL over the range of 0.5–4 kHz; this result lies in the middle of the range of the previously published data of children wearing the ADHEAR system (17–35.6 dB HL) <sup>15, 16</sup>. Better speech perception abilities were also achieved in quiet and noisy conditions with high participant satisfaction post-BCD-use.

#### 4.2 Sound localisation

The results of this study showed evident declines in directional hearing in both children with UMA and stimulated UHL; however, the group of UMA children performed better than those with stimulated UHL. Consistent with previous reports, good performers among children with congenital UCHL may have learned to localize sound sources using monaural cues and residual binaural difference cues after a long period of adaption. Vogt et al. <sup>11</sup> confirmed that patients with congenital UCHL rely on monaural spectral cues to detect high-frequency sound sources by comparing localisation accuracy with and without moulding the normal hearing ear pinna. Van et al.<sup>10</sup> evaluated nine listeners with chronic unilateral hearing loss through a group of broadband sound stimuli fixed at 60 dB; the results indicated a strong reliance on the ambiguous HSE in familiar acoustic environments.

Our study revealed no significant improvement in sound localisation accuracy between the BCD-unaided and aided conditions. Similar results have also been obtained in previous studies <sup>8, 9</sup>regarding the application of bone-anchored hearing aids and Bonebridge (Med-EL, Innsbruck, Austria) in congenital UCHL. The inability to achieve binaural hearing may be a consequence of two factors. First, hearing symmetry still exists, as the BCD is not able to provide the same hearing threshold as that of the normal hearing ear, and the insufficient intensity input also disrupts binaural hearing. Second, the processing time delay and inconsistent stimulation are inherent characteristics of the BCD signals; Additionally, the bone conduction signals with less reliable and constant cues may also prevent children with congenital UCHL from having a restored binaural hearing<sup>17.</sup> However, other studies showed improvement in sound localisation accuracy when BCDs were used in patients with congenital UCHL <sup>6, 7, 18</sup>. Potential explanations for these conflicting observations may be the age gap of the enrolled patients and methodological differences in the procedure. Besides, it is favourable that the sound localisation abilities of the intact ear did not deteriorate owing to cross-hearing induced by the BCD, likely due to insufficient high-frequency sound transmission of the BCD<sup>19</sup>, which did not interfere with the use of spectral cues by the healthy ear. Furthermore, the original sound localisation performance was a good predictor of sound localisation accuracy under the BCD-aided conditions. Moreover, there is a high need for early hearing intervention in poor performers who cannot make good use of asymmetric binaural cues to localize sound sources.

#### CONCLUSIONS

Some children with UMA were able to compensate using the remaining distorted binaural cues to detect sound sources, unlike the children with acutely stimulated UHL; however, this compensating ability was still far worse than children with NH and varied across individuals. As the application of BCD provided a definite benefit on speech recognition abilities and high participant satisfaction, it is recommended that children, particularly those with poor sound localisation performance, should be fitted with non-surgical BCDs at an early age.

#### Tables

Table 1. The demographic data of 11 patients with UMA and 11 children with NH.

Table 2. The MAE, response gain, bias, and  $r^2$  for all participants.

#### **Figure Legends**

Fig. 1 (a) Hearing thresholds, (b) WRS in quiet, and (c) SRT in the noise of patients with UMA in unaided and aided conditions, as well as the controls. Group means are presented as mean +- SD. Significant differences are labelled with \*(p < 0.05) and \*\*(p < 0.01) and ns, not significant. WRS, word recognition score; SRT, speech reception threshold; SNR, speech-to-noise ratio; HSE, head shadow effect; NH, normal hearing; BCD, bone conduction device; SD, standard deviation.

Fig. 2 Sound-localisation target-response plots of two patients (P6 and P7) and one control (N2) in monaural (unaided and P, left column) and binaural (aided and UP, right column) listening conditions. Stimulus sound levels are indicated by black circle, white square, and cross data points (65-, 70-, and 75-dB SPL, respectively). Best-fit linear regression is indicated by a black line. For participants with an ideal optimal localisation ability, g is 1, whereas MAE and b are 0. MAE, mean absolute error; g, response gain; b, bias;  $r^2$ , R square.

**Fig. 3** (a) The MAE and (b) Gain under monaural listening conditions (unaided and P, Y-axis) are plotted against those under binaural listening conditions (aided and UP, x-axis). Black circle data points indicate eleven UMA children, and white circle data points indicate the controls. The two UMA children and one control depicted in Fig. 2 are marked in this figure (P6, P7, and N2). An MAE near 0 and a gain near 1 demonstrate a close-to-normal sound localisation performance. Data of participants with the same sound localisation performance when listening monaurally and binaurally are displayed on the grey dotted diagonal. A data point below the diagonal in Fig. 3a and above the diagonal in Fig. 3b represents a better sound localisation performance when listening under binaural conditions than under monaural conditions. (c-d) The mean MAE of patients with UMA and the controls, respectively, on the impaired (including the atretic side of UMA and the plugged side of controls) side and the contralateral (the healthy side of UMA and the unplugged side of controls) side. Error bars represent mean +- SD. UMA, unilateral microtia and atresia; MAE, mean absolute error; Gain, response gain; Unaided, the unaided condition of patients with UMA; Aided, the aided condition of patients; SD, standard deviation.

Fig. 4 (a) MAE outcomes of two subgroups of good performers (n = 5; gain > 0.75) and poor performers (n = 6; gain [?] 0.75) were separately compared on the attrict and healthy sides. (b) Individual data of response

gain of UMA children in unaided conditions are plotted as a function of delta MAE between unaided and aided conditions. The linear regression was conducted to explore the predictive effect of gain on the benefits of sound localisation accuracy by fitting BCDs. P6 and P7 depicted in Fig. 2 are marked in this figure. Delta MAE = MAE<sub>aided</sub> – MAE <sub>unaided</sub>. Significant differences are labelled with \*(p < 0.05) and \*\*(p < 0.01), and ns, not significant. MAE, mean absolute error.

**Supplementary Fig. 1** Test setup and listening conditions. (a) Seven loudspeakers were placed at 30deg intervals in a semicircle in a double-walled, soundproof laboratory. (b) The monaural (the left column) and binaural listening (the right column) conditions are designed for UMA patients: unaided and aided conditions and for controls: P and UP conditions. P, the plugged condition; UP, the unplugged condition.

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