Acoustical and Perceptual Voice Characteristics in Adults With Early- and Late-Onset Auditory Neuropathy Spectrum Disorder

Caractéristiques acoustiques et perceptuelles de la voix d'adultes atteints d'un trouble du spectre de la neuropathie auditive apparu de façon précoce et tardive

Kavassery Venkateswaran Nisha
Prateek Lokwani
Prashanth Prabhu

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Kavassery Venkateswaran Nisha, Prateek Lokwani, & Prashanth Prabhu

Department of Audiology, All India Institute of Speech and Hearing, Manasagangothri, Mysuru, INDIA

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Abstract

This study aimed to describe onset-related differences in vocal characteristics (acoustic and perceptual) of individuals with early- and late-onset auditory neuropathy spectrum disorder, and it is the first of its kind in the literature. The study included 31 participants (15 early- and 16 late-onset) aged 15 to 30 years diagnosed with the disorder. The voice samples (sustained phonation) recorded by the participants using Android smartphones of selected configuration were sent by email to the experimenter. Acoustic parameters (fundamental frequency, formant frequencies, jitter, shimmer, and harmonic-to-noise ratio) were assessed using Praat software. This was supplemented by perceptual evaluations (consensus auditory perceptual evaluation of voice) by five speech-language pathologists. Results revealed significantly ($p < .05$) increased fundamental frequencies for all vowels along with decreased second and third formant frequencies of /i/ in early-onset participants compared to the late-onset group, which can be explained based on differences in pathophysiology of the disorder. There was no statistically significant difference between the mean perturbations (jitter and shimmer) and harmonic-to-noise ratio of the two groups. These differences were also complemented by perceptual evaluation findings: greater severity of pitch, breathiness, strain, hoarseness, and overall severity in the early-onset group. The findings from this study highlight the role of acoustical and perceptual voice evaluation in verifying the onset of auditory neuropathy spectrum disorder, which otherwise can only be retrospectively inspected from patient medical reports. The insights from the onset-based voice characteristics can help audiologists choose appropriate management options.

Abrégé

L'objectif de la présente étude était de décrire les différences observées sur le plan de la voix (caractéristiques acoustiques et perceptuelles) entre les personnes atteintes d'un trouble du spectre de la neuropathie auditive apparu de façon précoce et celles atteintes d'un trouble du spectre de la neuropathie auditive apparu de façon tardive. Il s'agit de la première étude en son genre recensée dans la littérature. Trente et un participants âgés de 15 à 30 ans et atteints d'un trouble du spectre de la neuropathie auditive (15 pour qui le trouble est apparu de façon précoce et 16 pour qui le trouble est apparu de façon tardive) ont été inclus dans l'étude. Des échantillons de voix (voyelles soutenues) ont été enregistrés par les participants au moyen de téléphones Android respectant des critères prédéterminés et ont été envoyés par courriel à la personne responsable de l'étude. Les paramètres acoustiques de la voix (fréquence fondamentale, formants, *jitter, shimmer* et rapport harmoniques/bruit) ont été analysés à l'aide du logiciel Praat. Des évaluations perceptuelles complémentaires de la voix (*Consensus Auditory-Perceptual Evaluation of Voice*) ont été réalisées par cinq orthophonistes. Les résultats ont révélé une augmentation significative ($p < 0,05$) de la fréquence fondamentale lors de la production de toutes les voyelles, ainsi qu'une diminution des fréquences des deuxième et troisième formants lors de la production de la voyelle /i/, chez les participants atteints d'un trouble du spectre de la neuropathie auditive apparu de façon précoce, lorsque comparés aux participants atteints d'un trouble du spectre de la neuropathie auditive apparu de façon tardive. Ces différences peuvent être expliquées par des différences pathophysiologiques associées au trouble. Les résultats ont également révélé une absence de différence statistiquement significative entre les participants des deux groupes pour ce qui est des moyennes des mesures de perturbation de la voix (*jitter* et *shimmer*) et du rapport harmoniques/bruit. Les différences observées sur le plan acoustique étaient également supportées par les résultats des évaluations perceptuelles. Spécifiquement, la sévérité globale du trouble de la voix et la sévérité de quatre paramètres de qualité vocale (tonalité, voix rauque, voix éteinte, voix forcée) ont été jugées supérieures pour les participants atteints d'un trouble du spectre de la neuropathie auditive apparu de façon précoce. Les résultats de cette étude soulignent le rôle des évaluations acoustique et perceptuelle de la voix lorsqu'on cherche à déterminer le moment d'apparition d'un trouble du spectre de la neuropathie auditive, moment qui ne peut autrement qu'être vérifié de façon rétrospective à partir des dossiers médicaux des patients. L'information que procurent les caractéristiques de la voix sur le moment d'apparition d'un trouble du spectre de la neuropathie auditive peut aider les audiologistes à choisir des options de prise en charge appropriées.

Auditory neuropathy spectrum disorder (ANSD) is a retro-cochlear pathology in which outer hair-cell functioning is normal but the auditory nerve is abnormal (Rance, 2005). Individuals with the disorder present with severely abnormal or absent auditory brainstem responses and typical otoacoustic emissions results (Berlin et al., 2010). Since its first description by Starr et al. (1996), ANSD has captivated the attention of audiologists worldwide due to its heterogeneity. Every aspect of the disorder presents an array of heterogeneous manifestations, including its onset (Berlin et al., 2010; De Siati et al., 2020; Jijo & Yathiraj, 2012; Kumar & Jayaram, 2006; Shivashankar et al., 2003), prevalence (Kumar & Jayaram, 2006; Mittal et al., 2012; Rance, 2005), aetiology (Berlin et al., 2003; Draper & Bamiou, 2009; Prabhu et al., 2012; Rance et al., 1999), pathophysiology (Nikolopoulos, 2014; Rance & Starr, 2015), symptomatology (Berlin et al., 2010; Prabhu et al., 2012; Rance, 2005), and rehabilitative options (Nikolopoulos, 2014; Rance & Starr, 2015).

A stark distinction in ANSD onset is an alluring heterogeneic manifestation of the disorder. A run-through of literature (Berlin et al., 2003, 2010; Jijo & Yathiraj, 2012; Kumar & Jayaram, 2006; Mittal et al., 2012; Rance & Starr, 2015) pointed at varied evidence in the pathophysiology based on the onset of the disorder. Literature reports in Western countries like Belgium (Boudewyns et al., 2016), cited childhood onset of the disorder, particularly under the age of 10 years. In contrast, studies in the Eastern world reported a late onset, with symptoms beginning between the first and second decades of life (Chandan et al., 2013; Jijo & Yathiraj, 2012; Kumar & Jayaram, 2006; Narne et al., 2014; Shivashankar et al., 2003). Although rare, scanty reports on late-onset ANSD were reported in Western literature too (Berlin et al., 2010; De Siati et al., 2020).

Other onset-based distinctions in ANSD can be traced to heterogeneity in its aetiology, symptoms, and pathophysiology. Early-onset ANSD is usually secondary to hyperbilirubinemia (Berlin et al., 2010; Rance et al., 1999), ototoxic drugs, low birth weight, low APGAR scores, anoxia, and positive family history (Berlin et al., 2003). In contrast, Prabhu et al. (2012) reported that late-onset ANSD cases did not have any pre-, peri-, or postnatal causes; instead, there were some predisposing factors associated with those individuals. These factors included exposure to toxic chemicals (pesticides) and toxic solvents (xylene), low socioeconomic status, and hormonal variations, which were present soon after puberty. Draper and Bamiou (2009) reported that exposure to xylene was noted in late-onset auditory dyssynchrony. Other aetiologies associated with late-onset ANSD are temperature-dependent changes, hereditary sensory and motor neuropathy, Charcot-Marie tooth disease, and mutations in genes such as autosomal dominant auditory neuropathy-1, protocadherin 9, otoferlin, and gap junction beta protein 2 (Cianfrone et al., 2006; Manchaiah et al., 2011; Rance et al., 2012). The clinical symptoms seen in late-onset patients are vertigo, headache, tinnitus, impaired vision, and difficulty in understanding speech (Prabhu et al., 2012) whereas early-onset patients exhibit difficulty in understanding speech which is disproportionate to the degree of hearing loss, difficulty hearing in noise (Starr et al., 1996), tinnitus (Narne et al., 2014) and vestibular problems (Prabhu & Jamuar, 2017). Late-onset patients tend to show a rising configuration of hearing loss which could be pathophysiologically linked to having more affected apical nerve fibres (Jijo & Yathiraj, 2012; Kumar & Jayaram, 2006). In contrast, early-onset ANSD patients show a flat loss, with pathophysiological bearings related to the degradation of apical fibres followed by the basilar region (Kumar & Jayaram, 2006).

Although the onset-based heterogeneity in ANSD patients is usually explored using the above-cited manifestations, studies on late onset were primarily conducted retrospectively using only the target (late-onset) population, limiting the scope of any comparisons with early-onset-related manifestations. Although onset related distinctions were often described for explanatory purposes in these studies, a direct inference could not be made as they lacked the experimental control that can only be made in prospective designs. The retrospective nature of case reports or studies fundamentally limits direct comparisons of population characteristics of late-onset and early-onset groups. Late-onset diagnosis in these cases is also dependent on the patient complaints documented in their case histories (Berlin et al., 2010). However, when patients report onset of symptoms in late adulthood, lack of audiological reports from childhood substantiating normal auditory functions in earlier years cannot be ruled out. Further, questions regarding the efficacy of newborn hearing screening and primary infrastructure for audiological testing in developing countries (Gupta & Venkatesan, 2018) where the late-onset cases are reported makes the research strides (comparison of late- vs. early-onset characteristics) in this direction even more challenging.

To date, there has been no study that systematically explored such group differences (late- vs. early-onset). Thus, the present study aims to establish onset-based differences in a relatively understudied dimension (voice characteristics) of ANSD patients. The motivation for the study is derived from Maruthy et al.'s (2019) research findings on deviant voice characteristics in long-standing late-onset adult (17–30 years) ANSD patients. They reported increased roughness, breathiness, and strain, along with increased pitch and reduced loudness in the voice of adults with late-onset ANSD when compared to normal age-matched individuals. In contrast, studies reporting childhood-onset of the hearing problem show high variability of the fundamental frequency, excessive intonation and pitch variation, increased loudness, and irregularities in resonance (Evans & Deliyski, 2007). In addition, it is widely agreed that 68% to 90% of ANSD patients with early-onset experience significant hearing loss (≥ 41 dB HL; Rance et al., 1999; Sininger & Oba, 2001), so the use of top-down compensatory mechanisms for phonemic restoration are usually compromised (Başkent, 2010). Perhaps the distortions occurring at the neural level along with suprathreshold changes that typically accompany moderate to severe hearing loss reduce the efficacy of top-down processing in them. This argument can also hold well for a hearing-severity-matched late-onset group, however the onset delay in this group could have effectively reduced signal distortions occurring at the auditory neural level, which in turn translates to partially intact top-down phonemic restoration in them. The partially intact phonemic restoration helps the latter group in processing acoustic and linguistic redundancy in speech signals, which not only contributes to improved speech perception but also to speech production. A number of studies have elaborately explained the voice characteristics in individuals with early-onset hearing loss (Campisi et al., 2006; Evans & Deliyski, 2007; Wirz et al., 1981), but direct generalization of these findings to the ANSD group cannot be done due to different pathophysiology and duration of the disorders.

Based on this evidence, we hypothesize that late-onset ANSD patients are likely to show fewer deviant voice characteristics compared to the early-onset ANSD group. This study is the first of its kind aimed at describing the ANSD onset-related vocal manifestations in early- and late-onset ANSD patients. This will indirectly help in deciding the management options for ANSD patients. Although applications of cochlear implants in patients with early onset may be advisable (Kontorinis et al., 2014), the utility of hearing aids (Barman et al., 2016; Jijo & Yathiraj, 2013) or assistive listening devices in the late-onset group can be advocated as the first line of rehabilitation. The specific objectives of the study are to compare the differences (if any) in acoustic (fundamental frequency, formants, harmonic-to-noise ratio, jitter, and shimmer) and perceptual voice characteristics between early- and late-onset groups with ANSD.

Methods

Participants

A total of 31 participants aged 15 to 30 years and diagnosed with bilateral ANSD by Rehabilitation Council of India-certified audiologists were considered for the study. All the subjects had Kannada as their native language. The criteria adopted to diagnose ANSD in the audiology clinic were those recommended by Starr et al. (2000): absent or abnormal auditory brainstem responses (delayed in latency or attenuated in amplitude), presence (average or robust amplitude) of otoacoustic emissions, and absent middle ear reflexes. Based on the clinical record, the diagnosis of ANSD was confirmed by a neurologist using clinical examination and computerized axial tomography/magnetic resonance imaging.

The participants were divided in two groups based on the onset of the ANSD symptoms: early-onset (*n* = 15; 11 females, four males; mean age = 22.33 ± 4.18) and late-onset (*n* = 16; 12 females, four males; mean age = 22.78 ± 4.20). A cut-off criterion of 12 years of age for the group segregation was used in the study, based on the recommendations of the Centers for Disease Control and Prevention (n.d.). Participants who were diagnosed with ANSD in childhood (6.0–10.2 years) or with the problem reported at birth (reported and assessed at the Department of Audiology, All India Institute of Speech and Hearing, Mysore, India between 2005 and 2011) were considered for the former group, and the latter group comprised adults who were diagnosed with ANSD over the age of 12 years, with no complaints of auditory deficits in childhood (reported and assessed between 2013 and 2020). Caution was taken to include only participants with onset less than 5 years duration in the late-onset groups, as long-standing ANSD adversely affects vocal characteristics (Maruthy et al., 2019). Also, to rule out any language problems, the Clinical Evaluation of Language Fundamentals (Semel & Wiig, 1980) was administered in a telephone interview with late-onset patients, who were included only if their language skills were age-appropriate. The waveforms/data recorded from three female participants in the early-onset group were pruned out as they did not fulfill the noise-free criterion for inclusion due to noisy waveforms (more background noise). **Table 1** shows the demographic and audiological details, comprising the degree (Clark, 1980; Goodman, 1965) and configuration (Pittman & Stelmachowicz, 2003) of hearing loss of all participants included in the study.

Informed Consent and Ethical Considerations

Informed consent was collected from all participants, with each participant informed about the objective of the

Table 1

Demographic and Audiological Details of All Participants

P	Early-onset group					Late-onset group			
	Ear	Age (years)	Gender	Degree of hearing loss	Hearing loss configuration	Age (years)	Gender	Degree of hearing loss	Hearing loss configuration
1	Right	17.2	Female	Severe	Flat	20.6	Female	Moderately severe	Rising
	Left			Severe	Flat			Moderate	Rising
2	Right	15.5	Female	Moderately severe	Flat	20.5	Male	Moderate	Irregular
	Left			Moderately severe	Flat			Moderate	Rising
3	Right	21.4	Male	Moderately severe	Rising	16.2	Male	Minimal	Rising
	Left			Moderately severe	Irregular			Minimal	Rising
4	Right	28.5	Female	Severe	Flat	18.4	Female	Minimal	Rising
	Left			Severe	Flat			Normal	
5	Right	24.4	Female	Moderately severe	Flat	20.1	Male	Normal	
	Left			Severe	Flat			Minimal	Rising
6	Right	21.5	Female	Severe	Flat	21.8	Female	Minimal	Flat
	Left			Severe	Flat			Minimal	Rising
7	Right	23.0	Female	Moderately severe	Flat	27.1	Female	Moderate	Rising
	Left			Moderately severe	Flat			Moderate	Rising
8	Right	26.4	Male	Moderate	Rising	27.2	Female	Moderate	Rising
	Left			Moderate	Rising			Moderate	Rising
9	Right	22.7	Female	Moderately severe	Flat	16.4	Female	Mild	Flat
	Left			Moderately severe	Flat			Moderate	Rising
10	Right	21.8	Female	Severe	Flat	26.8	Female	Minimal	Flat
	Left			Severe	Flat			Mild	Flat
11	Right	24.8	Male	Severe	Flat	23.5	Female	Minimal	Rising
	Left			Severe	Flat			Normal	
12	Right	22.1	Male	Moderately severe	Rising	21.5	Female	Moderate	Rising
	Left			Moderately severe	Rising			Moderate	Rising
13	Right	23.6 [a]	Female	Profound	Flat	29.9	Female	Mild	Rising
	Left			Severe	Sloping			Moderate	Rising
14	Right	20.5 [a]	Female	Moderate	Flat	26.8	Female	Moderately Severe	Flat
	Left			Moderately severe	Flat			Moderately Severe	Rising

Table 1 (continue)

15	Right	21.6 [a]	Female	Severe	Flat	26.0	Male	Mild	Flat
	Left			Profound	Flat			Minimal	Rising
16	Right					21.5	Female	Normal	
	Left							Moderate	Rising

Note. P = participant number.
[a] Excluded participants due to noisy recordings.

study and its need in brief. The anonymity of the participants was maintained throughout the study. The willingness of any patient to participate in the study did not affect their routine audiological assessment or other evaluations. All procedures performed in this study adhered to the bio-behavioural research standards (Venkatesan & Basavaraj, 2009) framed by the institutional ethical review board, whose permission was obtained for the study (SH/ACA/19AUD028/2020-21).

Procedure

The short-listed participants, after the screening of medical records, were contacted by telephone interview to assess their language skills (as discussed in inclusion criteria) and voice characteristics. The participants were asked to record sustained phonation of vowels /a/, /i/, and /u/ for a duration of at least 5 s, with three trials per vowel, and send the recorded voice samples over email. To facilitate the understanding of the task, a recorded video of the instructions along with a sustained phonation sample enacted by an Indian male speaker was sent for participant viewing. The participants were asked to keep the microphones of their smartphones 6 cm away from their mouths (or two thirds the length of their index fingers, for better understanding by the participants). Smartphones above specific configurations (Android 4, CPU frequency > 1.3 GHz) were used for recording purpose (Manfredi et al., 2017). Uloza et al. (2015) showed that smartphones are reliable in recording and assessing acoustic voice parameters. The reason for choosing sustained phonation rather than connected speech is because connected speech may show more variations while being recorded from smartphones when compared to using a standard microphone for recording. Also, previous research (Manfredi et al., 2017) done to assess the quality of voice used sustained vowel phonation rather than connected speech.

The rationale for the inclusion of the online-based data collection stemmed from the need for social distancing and alternative assessment procedures during the COVID-19 crisis. The use of alternative methods rather than conventional voice assessment in the COVID-19 pandemic for voice assessment (Lin et al., 2012) has become increasingly efficacious as they offer both accessibility and safety. To further validate the utility of the online-based recordings to the conventional voice sample recordings, a pilot study comprising voice samples of five normal adults (aged 18–25) was carried out using both methods. The adults were asked to phonate /a/. An Android 8 smartphone with a CPU frequency of 2.05 GHz was used for online recording, and the offline analyses were carried out using Audacity software (version 3.0.1; Audacity, 2020). The vocal parameters used in the current study were compared between the two recording modes using the Mann-Whitney U test, which showed no statistically significant difference ($p > .05$) between the recordings on all the parameters considered (fundamental frequency [F_0]: $|z| = 0.31$, $p = .75$; formant frequency [F_1]: $|z| = 0.10$, p = .92; F_2: $|z| = 0.32$, $p = .75$; F_3: $|z| = 0.94$, $p = .35$; jitter: $|z| = 0.11$, $p = .92$; shimmer: $|z| = 1.05$, $p = .29$; harmonic-to-noise ratio [HNR]: $|z| = 0.73$, $p = .47$).

In order to monitor the environmental noise, an Android-based application, Sound Meter, developed by Smart Tools Company (Ibekwe et al., 2016), was used at the participants' end. Live monitoring of the online recording session was supervised by the experimenter through an online video call. The participants were also asked to send the environmental noise data throughout the recording, which were further analysed by the experimenter before the inclusion of the voice sample. Samples with environmental noise less than 45 dB SPL were included for analysis.

Voice Analyses

The voice samples with less noise (< 45 dB SPL) were subjected to both acoustic and perceptual analyses. The window of analysis was kept constant at 2 s for all participants. The acoustic parameters of voice were assessed using Praat software (Boersma & Weenink, 2010). The samples recorded via smartphones (.mp3 format) were converted into formats usable by Praat (.wav format) using an online converter available free on the internet. The segment of recording which looked to have the most stable waveform was extrapolated from the recording and analysis was done. F_0 and F_1, F_2, & F_3 were computed for each recording.

Jitter (local) percentage, shimmer (local) percentage, and HNR ratio (in dB) were also calculated using the point-process option in Praat. Burris et al. (2014) concluded that fundamental frequencies and formants generated by Praat were reliable and accurate and were comparable to the values obtained in acoustic analysis using other software packages such as WaveSurfer (Sjölander & Beskow, 2000), and Computerized Speech Lab (Pentax Medical, n.d.). For generation of spectrograms, MATLAB audio toolbar was used, where the resolution of generated figures was found to be better than Praat software. Use of spectrogram measure in MATLAB has also been validated and proven effective in previous studies (Wang et al., 2019).

The computation of fundamental and formant frequencies was done to explore the difference in vocal characteristics of hearing-impaired (Campisi et al., 2006; Evans & Deliyski, 2007) and individuals with ANSD (current study). Jitter, the parameter of frequency variation from cycle to cycle, and shimmer, which relates to the amplitude variation of sound wave, were computed to quantify the perturbations in pitch and amplitude, possibly due to reduced auditory feedback. The HNR was computed to quantify and explore the difference in aperiodicity between the groups.

Subjective voice quality ratings were obtained from five certified speech-language pathologists (S-LPs), who were asked to perceptually rate the voice samples using a standardized voice assessment scale, the Consensus Auditory Perceptual Evaluation of Voice (CAPE-V; Kempster et al., 2009). The S-LPs listened to the voice recordings with Sennheiser HDA 200 circumaural headphones (Wedenmark, Germany) and rated the loudness, pitch, breathiness, strain, and roughness of voice on a 100 mm visual analog scale, with 0 indicating *normal voice* and 100 indicating *severely affected voice*. The individual ratings were then compared for interrater reliability and were factored into the statistical analyses.

Although auditory-perceptual ratings are considered the gold standard for evaluating voice disorders and assessing treatment progress (Oates, 2009; Selby et al., 2003), the current study considered joint analyses (acoustic plus perceptual) of the data for two reasons. First, the sensitivity of detection of certain voice disorders is higher for perceptual judgments, and its specificity is higher when using acoustic analyses (Heikkinen et al., 2021), Second, perceptual analysis requires specific expertise (S-LPs) which might not always be available in audiologic clinics, whereas acoustic analysis can be available and it releases clinicians from making subjective descriptions. The latter, however, can have drawbacks of high time-consumption and requiring procedural knowledge to carry out the analyses. Hence a combination was considered optimal in analyzing differences in vocal characteristics between the late-onset and early-onset ANSD groups.

Statistical Analyses

The data obtained were subjected to statistical analyses using SPSS version 25.0. A Shapiro-Wilk test of normality was done to check for the normal distribution of the data. A multivariate analysis of variance (MANOVA) test was carried out for the parametric data, and the Mann-Whitney U test was done to compare the differences (if any) in vocal characteristics between the groups when the data followed nonnormal distribution. The measure of effect size $r = Z/\sqrt{N}$, where Z is the nonparametric statistic and N is the population size, was computed for parameters where significant differences were observed in nonparametric tests. Similarly, partial eta squared (η_p^2) was noted wherever significant differences were observed in parametric tests. Modified Bland-Altman plot and single-rater type of interclass correlation were also computed to check for the interjudge agreement for perceptual ratings. The utility of interclass correlation over Pearson's correlation coefficient as a measure of interjudge reliability is empirically proven to be reliable for data where the order of the two measurements is unimportant (as in the present study, perceptual ratings of each judge did not follow any temporal order and were made independently of each other).

Results

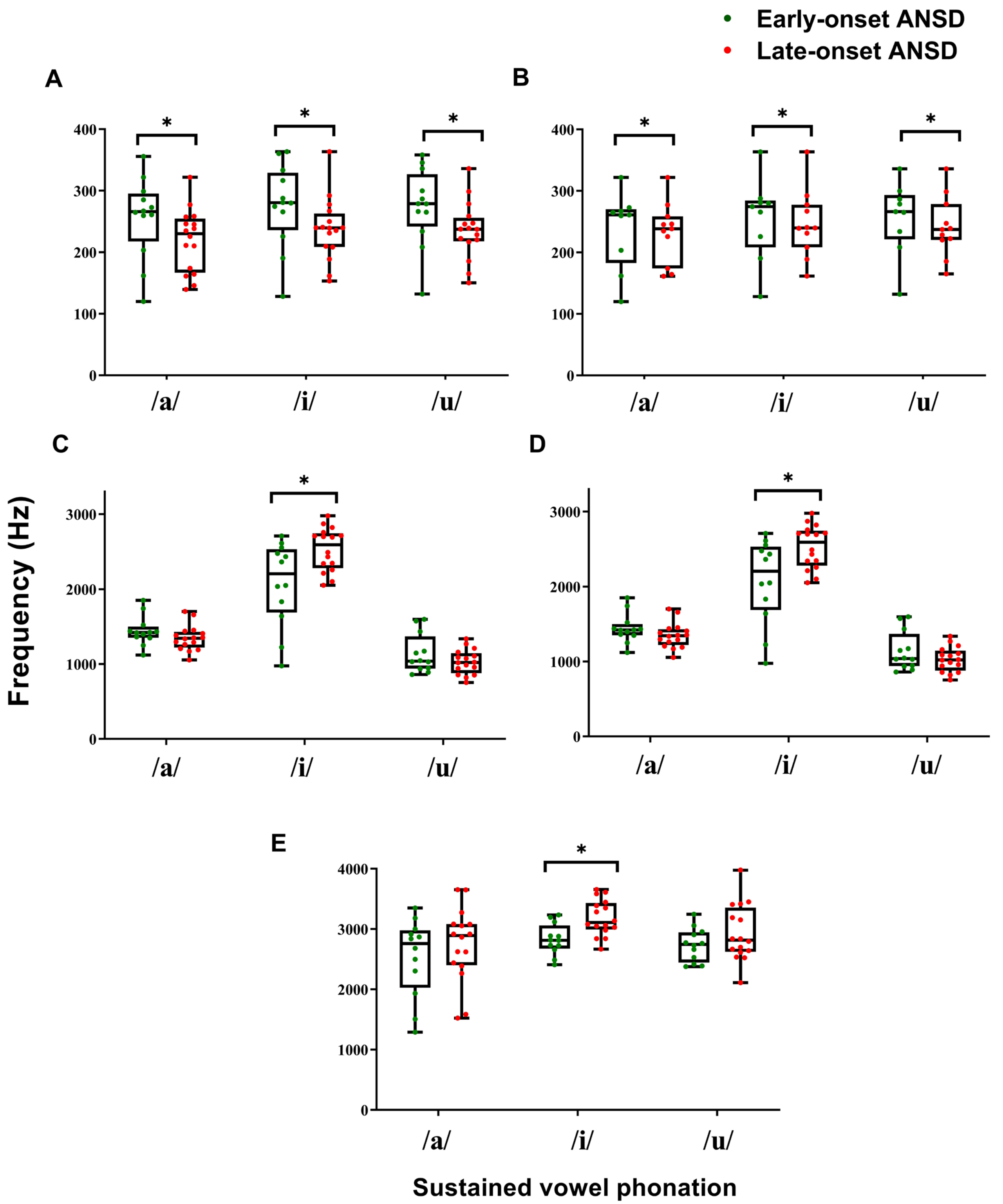
Although the voice samples were obtained from 31 participants, 3 samples (participants 13, 14, and 15 in the early-onset group, as listed in **Table 1**) were excluded due to excessive background noise (> 45 dB SPL). Hence, the vocal characteristics from the recorded waveforms with clear waveforms were analyzed. This comprised 12 individuals with early-onset and 16 individuals with late-onset ANSD. The statistical differences in the voice characteristics on the acoustic and perceptual parameters are highlighted in this section.

Group Differences in Acoustical Parameters of Voice

Fundamental Frequency and Formant Frequencies

The Shapiro-Wilk test showed the acoustical data (fundamental and formant frequencies) of all the vowels (/a/, /i/, & /u/) adhered to normal distribution ($p > .05$). The descriptive statistics comprising the mean for fundamental and formant frequencies along with standard deviation for the three vowels are shown in **Figure 1**. The results of inferential statistics (MANOVA) for the significant group differences are marked by asterisks in **Figure 1**. On visual

Figure 1



Sustained Vowel Phonation Frequencies in Early-Onset and Late-Onset ANSD Groups

Note. ANSD = auditory neuropathy spectrum disorder. Box plots depict median (centre line) and interquartile range (error bars). Panel A: Fundamental frequency (F_0). Panel B: F_0 for only female participants. Panel C: First formant (F_1). Panel D: Second formant (F_2). Panel E: Third formant (F_3). The individual data points for F_0 and the first three formants are also indicated on the corresponding plots.
$^*p < .05$

examination, the late-onset ANSD group, in general, exhibited higher formant frequencies compared to the early-onset group.

The results of the MANOVA for vocal pitch analyses showed main effect of ANSD onset for fundamental frequencies of all three vowels, as shown in **Table 2**. The results showed that the F_0 of the early-onset group was significantly higher (/a/: $p = .03$ /i/: $p = .02$, /u/: $p = .02$) than for the late-onset group for all vowels reported in the study. As the samples were not normalized, the females of both the groups were analysed separately (due to higher proportion of females in the overall sample), where the statistical results revealed higher F_0 of females in the early-onset group than the in the late-onset one.

Additionally, the main effect of ANSD onset was also seen for F_2 and F_3 of the vowel /i/, with the late-onset group demonstrating significantly higher F_2 ($p = .02$) and F_3 ($p = .01$) compared to the early-onset group.

The parameters in which onset-based group differences are seen to be significant (F_0 and F_2 & F_3 of /i/) for phonation samples obtained from two female participants (participants number 6 in both early-onset and late-onset groups, **Table 1**) are shown in **Figure 2** (for F_0) and **Figure 3** (for F_2 & F_3 of /i/) respectively. The early-onset participant had bilateral severe hearing loss and the late-onset patient had bilateral minimal hearing loss. The group differences are visually appreciable in their spectrograms as indicated in the figures. The colour-coded bands in the spectrogram correspond to bands of acoustic energy. On visual inspection, it is clear that F_0 is distantly located for the two groups (**Figure 2**). It is also seen that the energy bands depicting the portions of F_2 and F_3 are located differently for the two samples (**Figure 3**). Further, the mean F_2 and F_3 for the early-onset group were 2108 Hz and 2875 Hz respectively, whereas the same were higher (2529 Hz and 3187 Hz) for the late-onset group. However, no statistical differences were observed in F_1 of the vowel /i/.

Perturbations and Harmonic-to-Noise Ratio

The Shapiro-Wilk test revealed nonnormal distribution for perturbations (jitter and shimmer) of all three vowels ($p < .05$), and the data corresponding to the HNR were normally distributed ($p > .05$). The median values (centre line), along with the interquartile range (errors bars) of these three measures are shown in **Figure 4**. The early-onset group had higher perturbations for all sustained phonation. On the other hand, a relatively lower HNR was seen in the voice samples of the early-onset group, especially for /a/ phonation. However, none of the above cited differences withstood statistical verification, as shown in **Table 3**.

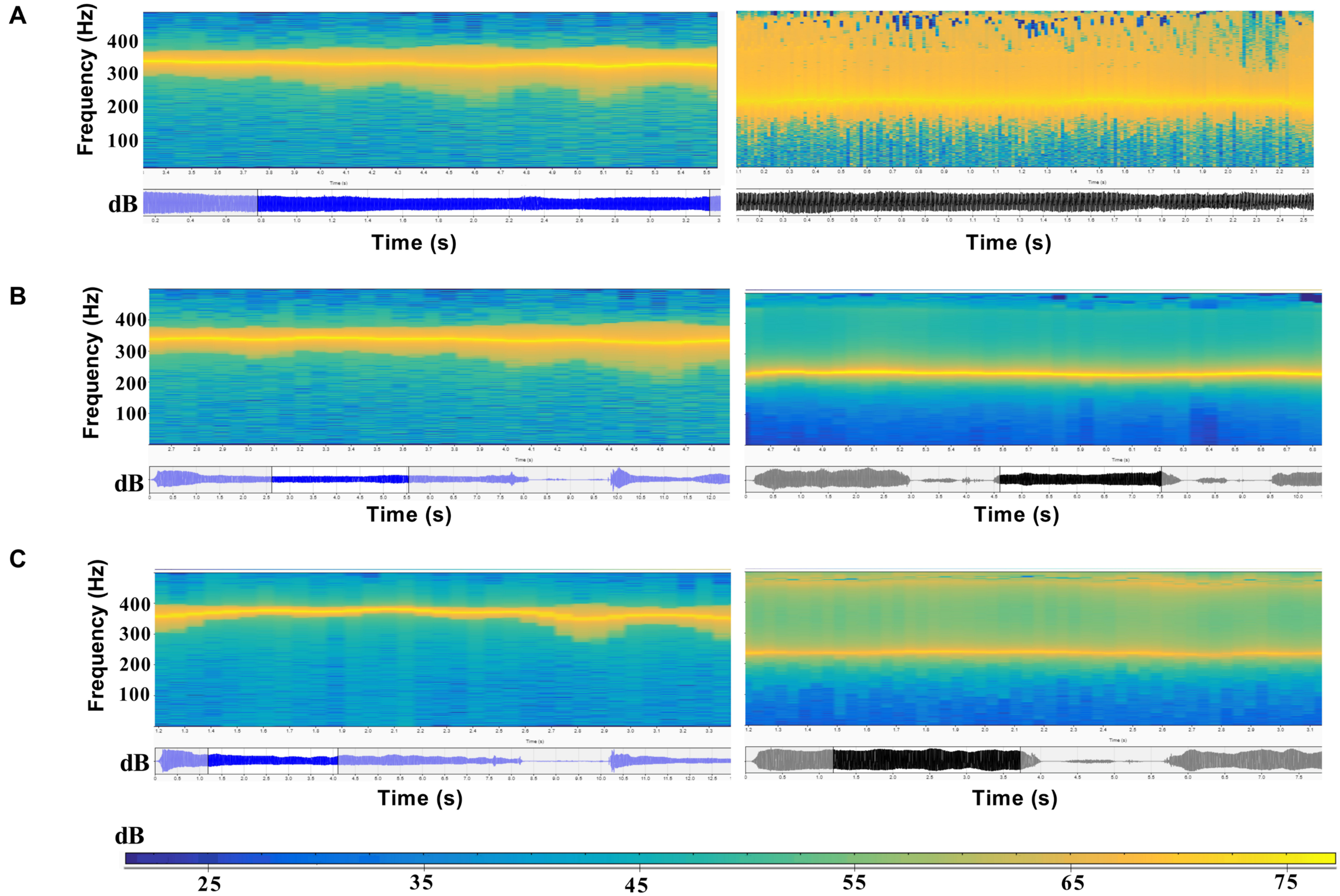
Table 2

Results of MANOVA Showing the Main Effect of Onset on Fundamental and Formant Frequencies of Sustained Phonation of Vowels /a/, /i/, and /u/

Acoustic frequency parameters	Vowels		
	/a/	/i/	/u/
F_0	**$F(1, 26) = 4.96$**	**$F(1, 26) = 5.96$**	**$F(1, 26) = 6.35$**
	$p = .03^*, \eta_p^2 = 0.16$	**$p = .02^*, \eta_p^2 = 0.19$**	**$p = .02^*, \eta_p^2 = 0.20$**
F_0	**$F(1, 18) = 4.89$**	**$F(1, 18) = 5.05$**	**$F(1, 18) = 4.23$**
(only female participants)	**$p = .02^*, \eta_p^2 = 0.18$**	**$p = .02^*, \eta_p^2 = 0.16$**	**$p = .03^*, \eta_p^2 = 0.19$**
F_1	$F(1, 26) = 3.79$	$F(1, 26) = 1.89$	$F(1, 26) = 0.27$
	$p = .06, \eta_p^2 = 0.13$	$p = .44, \eta_p^2 = 0.07$	$p = .61, \eta_p^2 = 0.01$
F_2	$F(1, 26) = 1.92$	**$F(1, 26) = 6.10$**	$F(1, 26) = 1.89$
	$p = .18, \eta_p^2 = 0.07$	**$p = .02^*, \eta_p^2 = 0.20$**	$p = .18, \eta_p^2 = 0.07$
F_3	$F(1, 26) = 0.80$	**$F(1, 26) = 8.02$**	$F(1, 26) = 1.06$
	$p = .38, \eta_p^2 = 0.30$	**$p = .01^*, \eta_p^2 = 0.24$**	$p = .31, \eta_p^2 = 0.04$

Note. F_0 = fundamental frequency; F_1, F_2, and F_3 = first, second, and third formant frequencies, respectively; MANOVA = multivariate analysis of variance. Bolded frames represent parameters with significant main effect of group.
*$p < .05$

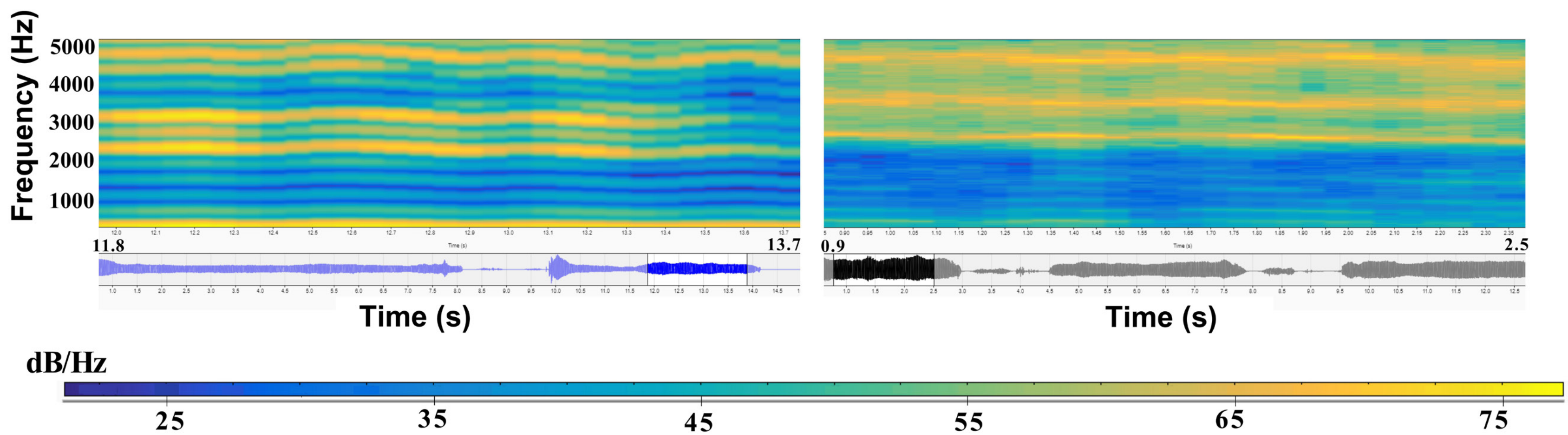
Figure 2



Spectrograms of Sustained Phonation (for F_0 Differences) of Female Patients With Early-Onset and Late-Onset ANSD

Note. ANSD = auditory neuropathy spectrum disorder; F_0 = fundamental frequency. Results from the patient with early-onset ANSD are presented on the left and the patient with late-onset ANSD on the right. Panel A: /a/. Panel B: /i/. Panel C: /u/.

Figure 3



Spectrograms of /i/ Sustained Phonation (for F_2 and F_3 Differences) of Female Patients With Early-Onset and Late-Onset ANSD

Note. ANSD = auditory neuropathy spectrum disorder; F_2, F_3 = second and third formant frequencies, respectively. Results from the patient with early-onset ANSD are presented on the left and the patient with late-onset ANSD on the right.

Group Differences in Perceptual Parameters of Voice Quality

The interjudge reliability of perceptual ratings of voice quality using a modified Bland Altman plot is shown in **Figure 5A**. The average perceptual agreement scores of all five S-LPs are shown on the *x*-axis, and the difference in perceptual agreement score is shown on the *y*-axis. On visual inspection of the Bland Altman plot, it is apparent that most perceptual judgments (262 out of 280) of the S-LPs were within the limits of agreement (± 1.96 *SD*, blue shaded area in **Figure 5A**). The analyses of the outliers in the modified Bland Altman plot showed that 18 out of 280 judgments did not correlate well, accounting for an error rate of 6.43%. The overall percentage of agreement between judges was approximately 93.57%, indicative of high interjudge reliability.

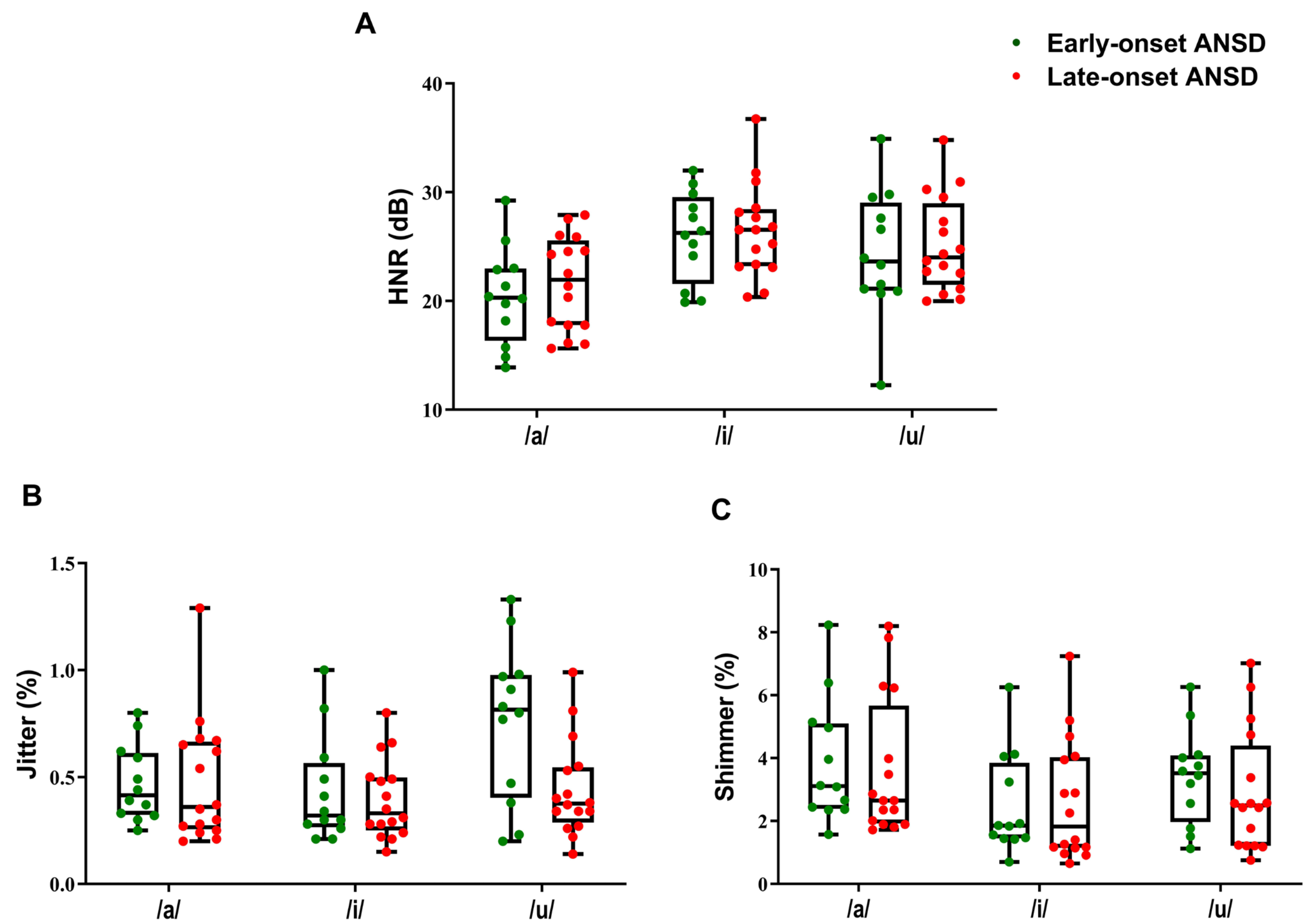
These observations were further complemented by the results of interclass correlation analyses which revealed a moderate to high degree of agreement amongst the perceptual judgments of the five S-LPs, as shown in **Table 4**.

The Shapiro-Wilk test revealed normality ($p > .05$) in average perceptual ratings of pitch, loudness, strain, breathiness, and roughness. The descriptive statistics with mean scores, along with standard deviations, of perceptual ratings are shown in **Figure 5B**, with the S-LPs' ratings for the early-onset group being more affected (greater pitch, breathiness, strain, and roughness) than for the late-onset group. The perceptual parameters with significant statistical difference (MANOVA test) are marked with asterisks in **Figure 5B**. MANOVA revealed the main effect of ANSD onset on all perceptual vocal parameters (pitch, F [1, 26] = 7.77, $p = .01$, $\eta_p^2 = 0.23$; breathiness, F [1, 26] = 5.68, $p = .03$, $\eta_p^2 = 0.18$; roughness, F [1, 26] = 9.24, $p = .01$, $\eta_p^2 = 0.22$; strain, F [1, 26] = 7.29, $p = .01$, $\eta_p^2 = 0.22$; and overall severity, F [1, 26] = 4.77, $p = .04$, $\eta_p^2 = 0.16$) except for one parameter (loudness, F [1, 26] = 1.05, $p = .31$, $\eta_p^2 = 0.04$].

Discussion

The present study aimed to delineate the differences in vocal characteristics of early- and late-onset ANSD using acoustical and perceptual measures. The findings of the study point at key indicators (**Figures 1 & 5**) in the vocal characteristics that can segregate the two onset-based ANSD groups. Amongst the few available retrospective studies, the existence of late-onset ANSD was

Figure 4



Sustained Vowel Phonation HNR, Jitter, and Shimmer in Early-Onset and Late-Onset ANSD Groups

Note. ANSD = auditory neuropathy spectrum disorder. Box plots depict median (centre line) and interquartile range (error bars). Panel A: Harmonic-to-noise ratio (HNR). Panel B: Frequency perturbation (jitter). Panel C: Perturbations (jitter and shimmer, respectively).

documented in only case studies by a few researchers (Berlin et al., 2010; De Siati et al., 2020; Jijo & Yathiraj, 2012; Kumar & Jayaram, 2006; Narne et al., 2014). Thus, the findings from the current study are the first of their kind in research design, plausibly explaining the onset-based group differences in vocal characteristics in a prospective research design. The strength of the study is the precise control of variables at the start of the study. The participants of the study were age matched between the groups to reduce the effect of any age-related changes in voice. All the subjects passed language screening in the late-onset ANSD group, which in turn helped the experimenters to understand the aptness of the participant inclusion, as the presence of early ANSD (even if of milder degree) is known to adversely affect language outcomes (Rance et al., 2012). The control was also exercised on the recording of voice samples, with prior succinct segregation of environmental noise using mobile applications. The check between Android-based voice recording and the conventional voice recording using the Computerized Speech Lab software during the pilot is another strength of the study. The combination of these experiment-based controls further consolidates the results obtained in the study, as well as providing flexibility to conduct such studies during a COVID-19 pandemic.

The results of the MANOVA showed that the fundamental frequency of all three vowels were increased in the early-onset group. These differences are unlikely

Table 3

Results of Inferential Statistical Tests (Mann-Whitney U and Independent *t* Test) for Comparison of Group Differences in Acoustical Measures of Voice

Parameter and vowel	Test statistic	*p*
HNR		
/a/	*t* (26) = −0.64	.53
/i/	*t* (26) = 0.17	.86
/u/	*t* (26) = −1.08	.29
Jitter		
/a/	/*z*/ = 1.21	.22
/i/	/*z*/ = 0.33	.74
/u/	/*z*/ = 1.86	.06
Shimmer		
/a/	/*z*/ = 0.84	.40
/i/	/*z*/ = 0.19	.85
/u/	/*z*/ = 1.67	.10

Note. HNR = harmonic-to-noise ratio.

related to the gender of the participants because the effect remains for early- versus late-onset ANSD within the female group too. This finding is on par with previous studies of acoustic features in vocalizations of people with long-standing hearing loss (Campisi et al., 2006; Evans & Deliyski, 2007). These results are attributed to poor laryngeal control, greater laryngeal muscular tension, or impaired internal auditory feedback. The fundamental frequency is the acoustic correlate of pitch which, when affected, impacts the social well-being of the individual and can be detected perceptually with voice quality rating scales.

The results of the MANOVA also showed that the second and third formants of the vowel /i/ in the late-onset ANSD group were significantly higher than in the early-onset group. This finding is suggestive of ANSD onset-based differences in the production of sounds with high-frequency formants. A comparison of the normative data (Sreedevi, 2000) for formant frequencies in Kannada speakers revealed that the late-onset sustained phonation characteristics (mean and *SD*) of the ANSD group for vowels /a/, /i/, and /u/ were similar to the adults in the same age range. However, the F_2 and F_3 in sustained phonation of /i/ for the early-onset ANSD group were relatively lower than the normatives. The finding could be related to the group differences in the pathophysiology of the disorder, which gets manifested as a production deficit in voice.

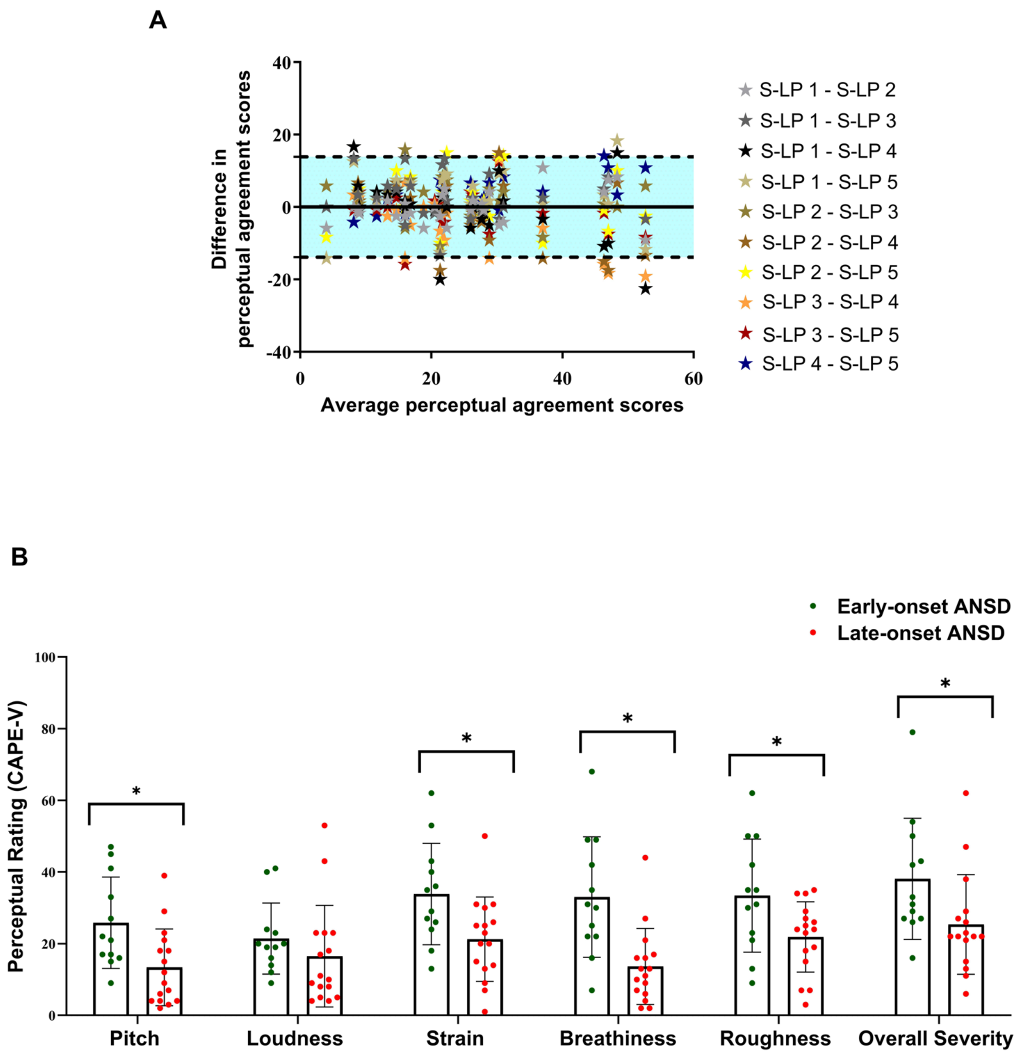
Pathophysiologically, patients with early-onset ANSD present flat audiograms (equally impaired perception across all frequencies), whereas those with late-onset ANSD exhibit a rising type of hearing loss with less impaired high-frequency perception (Kumar & Jayaram, 2006). The pathological limitation of impaired high-frequency perception in the early-onset ANSD group, which occurs at a relatively young age, places them at a disadvantage in the perception of F_2 and F_3 formants of /i/. However, the vowels /a/ and /u/ have relatively lower F_1 and F_2 (though F_3 is high), which might have resulted in lesser perceptual deficits.

The perception-related disadvantage in the early-onset group can be postulated to transfer to the production-related aspect as well. The production-related deficits originating from the perceptual disadvantage can be explained based on behavioural learning theory (Watson, 1913), which advocates that the learning of vocal sound productions occurs by environmental conditioning, feedback reaction, and strengthening behaviour through repeated actions. According to the theory, the feedback received on the perception of the sound gets strengthened through repeated productions. The altered/distorted feedback in individuals with ANSD (Maruthy et al., 2019) during childhood (early-onset) can lead to a deficit in the precise relay of vocal production to the auditory cortex. Thus, high-frequency sound productions, though normal at the early stages, get strengthened by a long-term vicious loop of feedback and altered perception in the early-onset ANSD group, resulting in altered recalibration of high frequency perception. The perception of high-frequency sounds in early-onset groups like those with even mild to moderate sensorineural hearing loss is documented in the literature (Evans & Deliyski, 2007). The relative lack of frequency shifts in late-onset ANSD (compared to the early-onset group) is indicative of the very nature of delayed onset in this group, which otherwise would have affected their voice characteristics, especially the higher formant frequencies.

The group differences noticeable in the acoustic characteristics of voice were also noticed in the CAPE-V ratings by S-LPs, indicative of perceptual voice manifestations of onset-related differences in voice quality between the two groups. This high degree of reliability in voice quality ratings between the five S-LPs in the study ensured similarity and effective perceptual analyses of the voice samples.

The findings of the MANOVA for perceptual parameters suggest that participants in the early-onset group have significantly greater roughness and breathiness of

Figure 5



Perceptual Ratings of Voice Quality

Note. ANSD = auditory neuropathy spectrum disorder; CAPE-V = Consensus Auditory Perceptual Evaluation of Voice. Panel A: Bland-Altman plots for interjudge agreement. Coloured stars show the agreement between two judges. The blue shaded area represents the limits of agreement (± 1.96 *SD*) of the observations. Panel B: Bar graphs depicting *M* and *SD* (error bars) of perceptual ratings of five parameters of voice quality in early-onset and late-onset ANSD groups.
* *p* < .05

Table 4

Objective Measure of Agreement in Perceptual Judgments of Five Speech-Language Pathologists

Parameter	Average interclass correlation coefficient	Range of interclass correlation coefficient	*p* value
Pitch	.67	.47–.78	< .001
Loudness	.59	.43–.75	< .001
Strain	.82	.72–.90	< .001
Roughness	.71	.57–.83	< .001
Breathiness	.69	.54–.81	< .001
Overall severity	.88	.80–.99	< .001

voice than those in the late-onset group. This finding is in consensus with the literature reports of the voice of adults with early-onset hearing loss (i.e., the onset of hearing loss in childhood), who invariably demonstrated greater hoarseness, breathiness, and monotonous voices compared to patients who showed symptoms of hearing loss at a later age (Campisi et al., 2006; Wirz et al., 1981). The reflections of disruptive neural firing in childhood itself in early-onset ANSD can be postulated to limit their auditory feedback (Maruthy et al., 2019), which in turn manifests as difficulties in monitoring their own speech. As a long-standing disruption in auditory neuronal firing, the early-onset group might have been at a serious disadvantage of poor auditory feedback for a long time. Similar findings of reduced vocal loudness in late-onset ANSD, with a long-standing duration (> 5 years) of the disorder, is reported in the literature (Maruthy et al., 2019). In contradiction to these findings, the present study revealed no significant difference in loudness across the groups. This could be because the earlier study compared long-standing ANSD patients with normal controls, whereas the present study included late-onset patients where loudness could be affected due to reduced self-confidence, rather than any physiological basis. Thus, significant difference was not found for this parameter in the present study.

Apart from its strengths, the research also had a few limitations: inclusion of a control group (age and gender matched normal hearing) along with the different onset experimental groups would have given better representation of results. The research, bring preliminary in this topic, used visual analyses to locate the most stable parts of the phonation, however more automatic methods for voice detection, such as considering successive 1 s time intervals of the samples (Lechien et al., 2017), could have provided useful information, and comparisons of these methods can be promising avenues for future research in determining ANSD onset-based vocal differences. As ANSD patients are typically described as having monotonous voices, the use of variability as well as mean tendencies would give better representation of the data sets and the extent of deviation present in the sample set. Confounding variables such as history of smoking, time of recording, etc., were not taken into consideration in the present research. Acoustic measures such as smoothened cepstral peak prominence, acoustic voice quality index, and dysphonia severity index have been proven to be more effective in estimating the extent of dysphonia and are linked better with perceived voice quality. Thus, future work can take these considerations and strengthen the link between voice and onset of ANSD.

Conclusions

The present study highlights the acoustic and perceptual differences in vocal characteristics of individuals with early- and late-onset ANSD. The findings from the study add diagnostic value to voice evaluation in individuals with ANSD, which is largely ignored in current clinical practice. The perceptual and acoustical voice evaluation results can be used as tools to verify the onset of the disorder, which is often retrospectively reported by the patient (especially in early-onset ANSD). This can indirectly help in the management of ANSD, with other management options such as compensatory strategies (anticipatory and repair strategies), and environmental modification which would be beneficial for late-onset ANSD patients: applications of cochlear implants in the early-onset group and utility of hearing aids or assistive listening in the late-onset group, as cochlear implants can help in better perception of high frequencies.

Despite the study being a preliminary attempt in the direction of onset-based ANSD differentiation, the generalization of study findings to the ANSD population as a whole warrants caution. A thorough profiling of other voice parameters such as rapid amplitude perturbation, amplitude perturbation quotient, soft phonation index, voice turbulence index, degree and number of voice breaks, and subharmonics will add valuable metrics and help in thorough understanding of onset-based group differences in voice. The study also opens the scope for promising new research on correlation of voice and audiological characteristics, apart from delineating the measures which best predict such onset-based differences.

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Authors' Note

Correspondence concerning the article should be addressed to Prashanth Prabhu, Assistant Professor in Audiology, Department of Audiology, All India Institute of Speech and Hearing, Manasagangothri, Mysuru- 570 006, INDIA. Email: **prashanthprabhu@aiishmysore.in**

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