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Ambulatory Phonation Monitoring in Prelingual and Postlingual Deaf Patients after Cochlear Implantation

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Keywords

Cochlear implants · Prelingual deafness · Postlingual deafness · Speech · Voice parameters

Abstract

Introduction: Hearing loss is known to play a fundamental role in voice production due to a lack of auditory feedback. In this study, we evaluated both fundamental frequency (F_0) and loudness of voice on adult deaf patients subjected to cochlear implantation, and we analyzed these results according to the prelingual or postlingual onset of the deafness. **Methods:** The study population, balanced in terms of sex, consisted of 32 adults who had undergone cochlear implantation due to severe or profound bilateral hearing loss (16 with prelingual deafness and 16 with postlingual deafness) and their outcomes were compared with a control group of 32 normal hearing (NH) subjects. All subjects were asked to utter the sustained vowel /a/ for at least 5 s and then to read an Italian phonetically balanced text. Voice recordings were performed by means of an ambulatory phonation monitoring (APM 3200). Measurements were performed without cochlear implant (CI), then with CI switched on, both in quiet condition and with background noise. **Results:** Compared to NH subjects, deaf individuals were overall charac-

terized by higher F_0 and loudness values, especially in the vowel task than the reading. In the sustained vowel task, no patients demonstrated significant voice changes after switching on the CI; contrarily, in the reading task, the use of the CI reduced both loudness and F_0 up to values comparable to NH subjects, although only in males. There was no significant difference in speech parameters between prelingual and postlingual deafness, although overall lower values were evident in case of postlingual deafness. The use of the CI showed a significant reduction of F_0 in males with postlingual deafness and of loudness, both for patients with prelingual and postlingual deafness. Finally, there was a positive correlation between postoperative hearing thresholds and overall speech loudness, highlighting how subjects with better hearing outcomes after CI positioning generally speak with a lower loudness and therefore a reduced vocal effort and load. **Discussion/Conclusion:** We found similar speech performances between prelingual and postlingual deafness, both in the vowel /a/ phonation and in the reading, providing a further suggestion that prelingual adult patients may benefit from cochlear implantation in phonation as well, in addition to the known excellent hearing outcomes. Overall, these results highlight the ability of the CI to adjust in everyday speech certain phonatory aspects such as F_0 and loudness by restoring the auditory feedback.

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Introduction

People with hearing loss are more likely to suffer from voice and speech disorders than those with normal hearing (NH) due to their poor auditory feedback mechanisms. Auditory feedback is an internal communication loop that helps speakers, using the sensory information acquired while the task is in progress, to self-monitor and adjust their voice during phonation [Ubrig et al., 2019]. NH individuals commonly exhibit robust control of speech and adapt their vocal production to compensate for competitive acoustic scenarios, such as in presence of background noise where the Lombard effect happens, and speakers raise vocal loudness to be heard and intelligible [Lee et al., 2017]. In case of severe hearing loss, the poor auditory feedback mechanisms may determine vocal alterations, such as increased pitch and loudness variability, as well as problems in managing speech intensities and intelligibility, thus compromising social interactions. Extensive literature demonstrates that the use of a cochlear implant (CI), i.e., an electronic device that is surgically implanted in the inner ear directly stimulating the auditory nerve fibers to provide sound sensation, in addition to all the hearing benefits, provides advantages for voice production by restoring the auditory feedback [Wilson et al., 1991; Coelho et al., 2009]. In particular, the main findings in adults are related to the reduction of vocal pitch/fundamental frequency (F_0) and speech loudness (sound pressure level, SPL) [Schenck et al., 2003; Perkell et al., 2007; Gautam et al., 2019; Ubrig et al., 2019], which in turn imply a reduced effort, as well as a variable decreasing of both jitter (pitch variability) [Evans and Deliyski, 2007; Gautam et al., 2019] and shimmer (amplitude variability) [Hocevar-Boltezar et al., 2006; Gautam et al., 2019]. Other parameters investigated were related to the improved phonatory control of vowels and consonants by reducing variability [Langereis et al., 1997; Schenck et al., 2003] and the decreased speech timing duration [Gautam et al., 2019]. However, evidence so far is limited in considering mainly speech production with CI in postlingually adult deaf patients or prelingually deaf children. To the authors' knowledge, only Evans reported phonatory data about prelingual adult deaf [Evans and Deliyski, 2007].

In addition, most of the studies focusing on speech production in CI patients evaluated phonation with only simple vocal tasks and in quiet condition, although they confirm how strongly the latter is influenced by the restoration of auditory feedback [Schenck et al., 2003; Wang et al., 2017; Gautam et al., 2019; Ubrig et al., 2019]. This

approach does not provide sufficient scientific understanding about speech production in real communication scenarios such as noisy environments. Again, to the authors' knowledge only Lee reported the effect of background noise on speech modifications after cochlear implantation, although only in postlingually deaf patients [Lee et al., 2017].

Furthermore, despite many authors have analyzed voice quality modifications in subjects with profound hearing loss treated with CI, all studies evaluated only a short-lasting phonation consisting in the repetition of single words or vowels protracted for few seconds at a comfortable pitch and constant amplitude [Hocevar-Boltezar et al., 2006; Lee et al., 2017; Upadhyay, 2019]. The only authors who implemented the reading of sentences or short texts in his vocal assessments were Ubrig, although limited to postlingually deaf adults [Ubrig et al., 2019], and Ruff, who evaluated text's reading both in adults and children but only focusing on the evaluation of the reading difficulty and words recognition after cochlear implantation [Ruff et al., 2017].

The abovementioned studies carried out voice recordings through unidirectional or multidirectional microphones, normally positioned from 4 cm to 8 cm from the speaker's labial commissure and at an angle of 45°, with the participants remaining seated during recordings [Hocevar-Boltezar et al., 2006; Ubrig et al., 2019; Upadhyay, 2019]. Possible drawbacks of such kind of evaluations consist in the potential for picking up unwanted environmental sounds, including the speech of others or no volitional voice use such as throat-clearing or coughing, and the alteration of the speech signal due to the influence of supraglottal vocal tract resonances [Cheyne et al., 2003]. Moreover, the inevitable variability of the instruments used for the analysis makes it difficult to interpret and perfectly match the data.

The purpose of the present study was thus to track changes of phonatory parameters in adult patients with CI with the high accuracy of a portable vocal dosimeter as the ambulatory phonation monitoring (APM) [Hillman et al., 2006; Cantarella et al., 2014]. This instrument, although not specifically designed for this purpose, has proven indeed to be insensitive to background noise and to provide reliable data on vocal parameters such as F_0 and SPL, rather than those acquired at common unidirectional or multidirectional air microphones included in previous studies [Svec et al., 2005; Mozzanica et al., 2019]. Another strength of this study, which differentiates it from any other similar in literature, concerns the inclusion in the group under examination of postlingual deaf,

who have been poorly evaluated so far, in addition to prelingual deafened adults and, above all, the assessment of reading a full text besides to the simple sustained vowel emission. Finally, the analysis of the CI effect was performed by measuring different listening conditions (i.e., quiet condition and in presence of background noise) allowing for the speculation on the usefulness of phonation measurements as a tool for evaluating the success of the cochlear implantation in speech production in relation to the time of onset of the hearing loss.

Materials and Methods

An observational cross-sectional study was conducted in a tertiary care center with a regular CI program. The study was conducted from January 2020 to December 2021 and all clinical data were taken from the CI registry maintained at the institution. The study was carried out according to the Declaration of Helsinki and it was previously approved by the Institutional Review Board (clinical trial No. 3546).

Population

The study population, balanced in terms of sex, consisted of adults who had undergone cochlear implantation due to severe or profound bilateral hearing loss as per the institute's candidacy criteria (pure-tone average hearing threshold >75 dB HL at 500 Hz, 1,000 Hz, and 2,000 Hz and a free-field speech perception threshold equal to or lower than 50% with the best possible amplification through hearing aid in the ear to be implanted) [Quaranta et al., 2009]. Patients were divided based on the prelingual and postlingual onset of the deafness. Exclusion criteria were reading limitation of any origin, speech disorders due to malformation, acquired damages to the speech organ, motor speech disorders, voice disorders of any origin besides deafness, difficulties in auditory rehabilitation or CI fitting, associated disabilities.

A cohort of 32 patients with CI has been thus included in the study: 16 males (8 prelingual and 8 postlingual) and 16 females (8 prelingual and 8 postlingual). Mean age of the patients was 49.7 ± 6 years (range 19–81 years of age). Mean preoperative pure tone average (PTA), evaluated in free field at speech frequencies (0.5–1–2–4 kHz), resulted to be equal to 78.5 ± 7 dB HL, whereas mean post-implantation PTA resulted to be equal to 27.3 ± 8 dB HL. Six patients (19%) underwent bilateral cochlear implantation and, among them, 4 had congenital profound bilateral deafness. As for patients with prelingual deafness, all of them were implanted for the first time at a later age (mean age 30.8 years) and all of them still had, at the time of surgery, hearing residues at low frequencies (82 dB at 250 Hz, 86 dB at 500 Hz, and 110 dB at 1 kHz on average). For this reason, despite profound deafness, all these patients have used hearing aids since childhood (average length of use: 32 years), although some of them achieved very low gains in speech recognition, and all have undergone a lot of speech therapy rehabilitation so that everyone had developed a fully or partially structured oral language.

As for patients with postlingual deafness, only 4 out of the 16 enrolled in the study had an auditory deprivation before cochlear

implantation (27.5 years on average): in accordance with recent literature, we defined those “auditory deprived” patients with severe to profound hearing loss that did not use hearing aids for a period longer than 15 years before CI surgery [Canale et al., 2016]. All surgeries were performed by the same senior surgeon. Among the patients with unilateral CI, four of them had a bimodal hearing restoration (CI and contralateral hearing aid). The manufacturers of the CIs implanted were Advanced Bionics (4 subjects, 13%), Cochlear (18 subjects, 56%), and Med-El (10 subjects, 31%). All the patients with CI underwent auditory rehabilitation after cochlear implantation, had at least 2 years of regular CI mapping after processor's activation, and were therefore considered stable from a hearing rehabilitation point of view.

A control group composed by 32 NH subjects (16 males and 16 females), aged between 20 and 64 years (mean 29.7 ± 3 years), was enrolled. All the NH subjects demonstrated a PTA ≤ 15 dB HL (mean 9.18 ± 4 dB HL). Each subject enrolled in the study gave his/her written informed consent.

Measurement Procedure

Preliminary room acoustic measurements were carried out aiming at assessing whether the reverberation time (RT_{60}) of the selected space, namely, the time taken for a signal to decay the full 60 dB from its initial level, was suitable for the administration of the test. The evaluations were performed in compliance with the EN ISO 3382-1 standard [ISO, 2009], applying the interrupted noise method through a sound level meter (Acoustilyzer AL1) and a pink noise generator (Minirator MR-1) connected to the main speaker. As the testing room was acoustically treated and had a volume below 45 m^3 , the measured RT_{60} was below 0.5 s at medium frequencies and thus the environment was considered acoustically suitable for the purpose of the study.

In order to evaluate the spectral and loudness modification of voice in terms of F_0 and SPL, respectively, according to different hearing conditions, NH subjects and patients with CI were asked to utter the sustained vowel /a/ for at least 5 s and to read a brief text in Italian named “Il ramarro della zia,” which is a phonetically balanced content created by Vernerio and Schindler in 1998 for speech therapy purposes [Vernerio et al., 1998]. NH subjects performed these tasks both in a quiet condition and with a background energetic masking noise of 50 dBA. Similarly, patients with CI performed these tasks twice, both in a quiet condition and with the same background noise of 50 dBA. First, they were asked to switch off their CI; second, they were asked to switch on their CI. Background noise was artificially added using three calibrated loudspeakers, controlled by an audiometer and placed at a standard ear height (1 m from the floor) and at the same distance from the receiver (2 m) in order to obtain the maximum possible masking (one loudspeaker at 0° and the lateral ones placed with an angle of 110°).

CI patients and NH subjects were sat in a comfortable position. Among CI patients wearing processors in which it was possible to adjust the direction of the microphone, a fixed orientation stimulating the pinna was chosen, which is the most similar condition to NH. Furthermore, the adaptive microphone adjustment function of the CIs, capable of suppressing background noise, has never been selected to avoid any facilitation in the intelligibility of the patient's voice. In addition, in the 4 patients who had a bimodal hearing restoration, the hearing aid was always removed during the recordings.

Voice Recording

In order to provide an objective measurement of voice characteristics, the APM used in the study was the APM model 3200 (KayPENTAX, Lincoln Park, NJ, USA). It consists of an accelerometer, placed adhesively along the anterior part of the neck, which measures the vibrations from the vocal folds through the tissues of the neck and converts them into SPLs (in dB) of speech. The APM gathers acoustic voice raw data at a rate of 20 samples per second and these data are transferred to a microprocessor unit worn in a waist pack. Among the multiple parameters acquired by the APM, it was decided to collect the following:

- Average F_0 (in Hz) expresses the mean frequency at which the vocal folds vibrate.
- Average loudness in terms of emitted SPL (in dB) expresses the mean value of the amount of energy of the voice sound wave.

Phonation measured in this way has been shown to be relatively insensitive to surrounding sounds and to differentiate volitional voice from other behaviors, such as throat clearing or coughing [Hillman et al., 2006; Mozzanica et al., 2019]. Before starting the real voice monitoring, a calibration of the acquisition system was needed subject-by-subject. As the contact sensor placed at the jugular notch needs to provide referred SPL values, in fact, a comparison calibration with respect to an air microphone (placed exactly 15 cm from the speaker's mouth) was thus performed. In this way, after acquiring together referred SPL values from the air microphone and voltage levels from the contact sensor due to the skin acceleration generated by the vocal folds' vibration, a calibration function containing subject-related constants could be obtained and then applied while monitoring the real voice. All 64 participants were thus initially asked to perform such calibration procedure, which in practice consisted in the vocalization of a sustained vowel /a/ at increasing loudness levels, from whispers to screams in order to produce all the possible loudness levels produced in the subsequent monitoring. The time required to calibrate the APM never exceeded 5 min and all the patients well tolerated the APM device during the evaluations.

Statistical Analysis

Statistical analysis was performed using SPSS 24.0 statistical software for Microsoft Windows (SPSS, Inc., Chicago, IL, USA). Preliminary analyses were performed to ensure any violation of the assumptions of normality, linearity, and homoscedasticity. Variables were compared by means of nonparametric tests due to non-normally distributed data, in particular the Wilcoxon signed-rank test and the Mann-Whitney U test for nonindependent and independent samples, respectively. Analysis of variance was performed with Kruskal-Wallis test and correlations were assessed by means of Spearman's rank-order test. Two-sided exact tests were used and p values <0.05 were considered significant.

Results

A Mann-Whitney U test was conducted to compare the post-implantation PTA scores according to the gender, the laterality of the CI, and the onset of the deafness. There was no significant difference in postoperative PTA values between males and females ($p = 0.138$), between

Table 1. Phonatory outcomes of NH subjects

NH subjects		
Male F_0 ($n = 16$)	Vowel	112.8±15 Hz
	Reading	122.9±12 Hz
	Reading + noise	129.3±13 Hz
Female F_0 ($n = 16$)	Vowel	202.6±27 Hz
	Reading	202.8±22 Hz
	Reading + noise	210.7±21 Hz
Loudness ($n = 32$)	Vowel	77.0±8 dB
	Reading	76.1±6 dB
	Reading + noise	79.9±7 dB

F_0 , fundamental frequency; noise, background noise at 50 dBA.

unilateral and bilateral cochlear implantation ($p = 0.524$), and between prelingual and postlingual deafness ($p = 0.491$). Based on these similarities between groups in terms of postoperative auditory results, we found it appropriate to consider all patients like each other and therefore valid and significant the outcomes of the phonatory tests. Similarly, there were no significant differences between males and females concerning the age, as well as between unilateral and bilateral CI ($p < 0.05$); on the contrary, patients with prelingual deafness resulted significantly younger (mean 42.5 years old, $n = 16$) compared to postlingual deafness (mean 62.5 years old, $n = 16$) ($p < 0.001$). The speech F_0 and loudness values obtained from both control subjects and CI recipients are reported in Tables 1–3. The Kruskal-Wallis test did not reveal any statistically significant difference between speech characteristics of the CIs belonging to the three different CI companies (Advance Bionics, $n = 4$; Cochlear, $n = 10$; Med-EL, $n = 18$; $p > 0.05$), neither as regards the speech F_0 values nor for the loudness.

Sustained Vowel Task

The Kruskal-Wallis test revealed a statistically significant difference in F_0 values across NH male subjects ($n = 16$), deaf males without CI ($n = 16$), and deaf males with CI on ($n = 16$) ($p = 0.001$). The deaf males with CI switched off demonstrated higher F_0 scores than the other two groups. A similar difference across these three groups was also demonstrated for females ($p = 0.001$), with significantly higher F_0 values in patients with CI switched off compared to women with CI on and NH women. A statistically significant difference at Kruskal-Wallis test was also demonstrated concerning the vowel /a/ loudness values between NH subjects ($n = 32$), patients with CI switched off ($n = 32$), and patients with CI turned on ($n = 32$).

Table 2. Phonatory outcomes of deaf patients with CI in the sustained vowel task

Vowel task – deaf patients		
Male F ₀ (n = 16)	CI off	156.5±40 Hz
	CI on	150.8±42 Hz
Female F ₀ (n = 16)	CI off	251.2±54 Hz
	CI on	218.4±52 Hz
Loudness (n = 32)	CI off	82.5±11 dB
	CI on	80.9±13 dB

F₀, fundamental frequency; CI off, cochlear implant switched off; CI on, cochlear implant turned on.

= 32) ($p = 0.031$). Deaf patients without the use of the CI demonstrated higher loudness values as compared to the other two groups. Among deaf patients, the Wilcoxon signed-rank test revealed a slight decrease of F₀ values, although not statistically significant, following the activation of the CI, both in males ($p = 0.278$) and females ($p = 0.352$). Likewise, there were no significant differences in loudness values in the vowel task after CI activation ($p = 0.286$).

The Mann-Whitney U test was furthermore used to compare both F₀ and loudness of the vowel task between prelingual and postlingual deafness. In particular, males with prelingual deafness showed lower F₀ values, although not statistically significant, than males with postlingual deafness, both with CI off ($p = 0.781$) and with CI on ($p = 0.486$). Contrarily, females with prelingual deafness demonstrated higher F₀ values, although not statistically significant, than females with postlingual deafness, both with CI off ($p = 0.376$) and with CI on ($p = 0.133$). As regards the loudness, higher though not significantly different values were reported in prelingual patients compared to postlingual ones, both with CI off ($p = 0.174$) and with CI on ($p = 0.250$). The switching on and therefore the use of the CI has not shown, at paired-samples *t* test, to significantly modify the values of F₀ and loudness in the vowel task, both in case of prelingual and postlingual deafness ($p > 0.05$) (Table 4).

Reading Task

Concerning the NH subjects, a statistically significant increase in speech loudness was reported following the addition of background noise at 50 dBA of intensity when reading the text “Il ramarro della zia” ($p < 0.001$). Similarly, a significant increase of the F₀ scores in the reading with background noise was shown in both NH males and

Table 3. Phonatory outcomes of patients with CI in the reading task

Reading task – deaf patients		
Male F ₀ (n = 16)	CI off	143.8±31 Hz
	CI on	136.8±34 Hz
	CI on + noise	143.0±31 Hz
Female F ₀ (n = 16)	CI off	222.2±50 Hz
	CI on	218.4±52 Hz
	CI on + noise	226.7±55 Hz
Loudness (n = 32)	CI off	76.7±8.7 dB
	CI on	73.2±9.4 dB
	CI on + noise	76.2±8.9 dB

F₀, fundamental frequency; CI off, cochlear implant switched off; CI on, cochlear implant turned on; noise, background noise at 50 dBA.

Table 4. Phonatory differences between prelingual and postlingual deafness on deaf patients in the vowel task

Vowel task – deaf patients	CI off	CI on	<i>p</i> value
Prelingual deafness			
Females – F ₀	263.6±61 Hz	266.6±59 Hz	0.821
Males – F ₀	153.6±49 Hz	143.0±39 Hz	0.240
Loudness	85.2±12 dB	83.6±14 dB	0.360
Postlingual deafness			
Females – F ₀	238.7±47 Hz	223.9±47 Hz	0.486
Males – F ₀	159.5±31 Hz	158.5±48 Hz	0.927
Loudness	79.8±10 dB	78.2±12 dB	0.427

F₀, fundamental frequency; CI off, cochlear implant switched off; CI on, cochlear implant turned on.

females ($p < 0.001$ at Wilcoxon signed-rank test). Similarly, deaf patients’ speech evaluation with CI on demonstrated a significant increase of the F₀ values when a background noise was added, both in males and females ($p = 0.007$ and $p = 0.008$, respectively), and a similar significant increase of values was also shown for loudness with respect to the assessment in quiet conditions ($p < 0.001$).

The Mann-Whitney U test showed, in males and in quiet conditions, significantly higher F₀ values in deaf patients with CI off than in NH subjects ($p = 0.035$) and subsequent activation of CI highlighted a significant reduction in these same values ($p = 0.023$ at Wilcoxon signed-rank test), with outcomes that have become comparable to the F₀ of NH subjects ($p = 0.184$). In contrast, there was no significant difference between female NH subjects and female deaf with CI switched off ($p = 0.402$), and the further switching on of the CI did not significantly affect the

Table 5. Phonatory differences between prelingual and postlingual deafness on deaf patients in the reading task

Reading task – deaf patients	CI off	CI on	<i>p</i> value
Prelingual deafness			
Females – F ₀	246.7±58 Hz	242.0±59 Hz	0.629
Males – F ₀	147.2±40 Hz	141.3±44 Hz	0.455
Loudness	78.3±9 dB	75.2±8 dB	0.006*
Postlingual deafness			
Females – F ₀	197.6±25 Hz	194.8±33 Hz	0.614
Males – F ₀	140.6±21 Hz	132.5±24 Hz	0.011*
Loudness	75.1±8 dB	71.1±10 dB	0.001*

F₀, fundamental frequency; CI off, cochlear implant switched off; CI on, cochlear implant turned on. * Significant value <0.05.

F₀ in female patients ($p = 0.717$). As regards the speech loudness in quiet condition, there was no significant difference in values between NH subjects and deaf patients with CI switched off ($p = 0.989$), whereas a statistically significant reduction of the values was demonstrated in the same deaf patients after CI activation ($p < 0.001$).

NH subjects showed similar values between the sustained vowel task and the reading task as for loudness ($p = 0.640$) and the F₀ in females ($p = 0.717$), while in NH men the average F₀ value resulted significantly lower in the phonation of the vowel /a/ ($p = 0.008$). Conversely, deaf patients with CI off showed significantly higher F₀ values ($p = 0.003$ for females and $p = 0.026$ for males) and loudness values ($p < 0.001$) in the vowel task than in the reading task.

The relationship between PTA values and speech characteristics of deaf patients was investigated using Spearman correlation coefficient. By analyzing the reading task with and without CI, there was no significant correlation between mean post-implantation PTA thresholds and F₀ values, both for males and for females ($p > 0.05$); similar results were also obtained by assessing the vowel task ($p > 0.05$). On the contrary, there was a positive correlation between mean PTA thresholds and speech loudness, both with CI off ($r = 0.36$, $p < 0.05$) and CI on ($r = 0.35$, $p < 0.05$): higher speech loudness values resulted associated with higher PTA thresholds.

Furthermore, in the reading task, there was a negative correlation between the age of deaf patients and their mean F₀ scores, in both genders and with CI on ($r = -0.31$, $p < 0.05$), with higher F₀ scores detected in younger patients. Contrarily, any other correlation between speech characteristics and patients' age was found as they all resulted to be not significant ($p > 0.05$).

Further comparative analyses carried out on the reading task between prelingual and postlingual subgroups showed lower F₀ values in all patients with postlingual deafness, both male and female, both with and without CI, although this difference was only statistically significant in deaf women, without the use of the CI ($p = 0.047$). Lower though not statistically significant values were also demonstrated in case of postlingual deafness concerning the speech loudness, both with CI off and CI on ($p > 0.05$). Furthermore, we did not report any significant difference in speech characteristics between prelingual or postlingual deafness when speech was assessed with background noise ($p > 0.05$).

The switching on of the CI showed to significantly reduce the F₀ values only in males with postlingual deafness ($p = 0.011$), whereas there were no differences among males with prelingual deafness or in females after CI activation ($p > 0.05$). On the contrary, the use of the CI demonstrated a significant decrease in the speech loudness values in all patients ($p < 0.05$), both in cases of prelingual and postlingual deafness (Table 5).

Discussion

The aim of the present study was to evaluate the voice modifications in adults with profound hearing loss following cochlear implantation, particularly focused on differences between prelingual and postlingual deafness. Our study group consisted of 32 profoundly deaf adults who underwent cochlear implantation, equally distributed between males and females, and between prelingual and postlingual deafness. A control group composed by 16 NH females and 16 NH males was also involved. Both groups undergone voice recordings consisting in the reading a phonetically balanced passage while being equipped with a contact-sensor-based voice monitoring device (i.e., the APM device by KayPENTAX). From the monitoring, mean fundamental frequency and SPL were extracted for each participant, both in quiet and in noise conditions.

Role of Cochlear Implantation and Subjective Features in Voice Production

It is well recognized how hearing loss plays a fundamental role in vocal production. Patients with congenital deafness, although submitted to cochlear implantation, frequently manifest pronunciation errors, vowel substitutions, and difficulties in intonation, resulting in very unintelligible speech [Hocevar-Boltezar et al., 2006; Len-

den and Flipsen, 2007]. Similarly, even those subjects who experience the occurrence of deafness as adults demonstrate a degradation of the speech over time and the restoration of the auditory feedback by CI has been shown to induce adjustments in speech production, particularly in the reduction of the fundamental frequency and the speech loudness [Ubrig et al., 2011, 2019; Coelho et al., 2012; Gautam et al., 2019; Boisvert et al., 2020]. However, as stated by Coelho in her systematic review of the literature, controversial results and the heterogeneity of the methods used in most studies make it difficult to understand the real effect of the CI on deaf patient's speech [Coelho et al., 2012]. To the authors' knowledge, only Ubrig analyzed a large case series, comparable to the one considered in the present study, although he took in consideration exclusively adults with postlingual deafness [Ubrig et al., 2011].

Consistent with the congenital onset of deafness and the related need to restore the auditory feedback earlier, the mean age of the prelingual deaf group was significantly lower (42 years old) than patients with postlingual deafness (62 years old). Nonetheless, a very satisfactory mean postoperative PTA threshold (27.3 dB HL in free-field assessment) was achieved in all patients, with no significant differences in hearing thresholds depending on gender, unilateral or bilateral implantation, and between prelingual or postlingual deafness. In fact, although the literature suggests that early cochlear implantation plays an important role in the hearing outcomes of prelingually deafened patients, recent studies did not report significant differences in the electrically evoked compound action potential of the auditory nerve in CI recipients between prelingual and postlingual deafness [Harrison et al., 2005]. Furthermore, confirming our good results on patients with prelingual deafness, Canale reported no differences in perceived quality of life and in the benefit from CI between prelingually and postlingually deafened groups, suggesting that hearing outcomes obtainable in subjects with congenital hearing loss implanted in adulthood not only depends on the duration of auditory deprivation but also on the extent of rehabilitation with hearing aids and speech therapy carried out in childhood [Canale et al., 2016, 2019]. Unfortunately, unlike the good hearing results, there were no data in literature on the variations in phonation of adults with prelingual deafness so far.

Hillman showed that a vocal accelerometer provides superimposable data of F_0 , vocal loudness, and phonation time to those recorded by a traditional microphone, both in control subjects and in individuals with mild and se-

vere dysphonia [Hillman et al., 2006]. Furthermore, Švec demonstrated that the APM can provide the average SPL value of soft, comfortable, or strong voices with an accuracy higher than ± 2.8 dB in 95% of cases, even more accurate than microphones [Švec et al., 2005]. This agrees with Astolfi et al. who found, for other contact-sensor-based devices, a significant advantage in using a contact microphone despite its higher uncertainty [Astolfi et al., 2018]. Indeed, although a headworn air microphone provides an uncertainty of up to 2 dB and a contact-sensor-based device of up to 3 dB, the latter neglects the presence of background noise – even of high magnitudes – and allows for long-term, accurate, and repeated monitoring. To date, only Mozzanica included the APM in voice production assessment after cochlear implantation, although related to the registration of a 24-h working day and limited to postlingual deafness [Mozzanica et al., 2019]. Our voice recordings included the prolonged emission of the vowel /a/ at habitual pitch and loudness, which was chosen because mainly dependent on acoustic rather than orosensitive control [Svirsky and Tobey, 1991]. However, with the aim of evaluating the speech in a condition as close as possible to everyday life, we also included the reading of a phonetically balanced text, both in quiet conditions and with a background noise of 50 dBA.

To date, except for a study by Lee et al., 2019, the speech characteristics of deafs with CI have never been evaluated in competitive acoustic conditions but always only with simple vocal tasks and in quiet condition [Hoccevar-Boltezar et al., 2006; Evans and Deliyski, 2007; Wang et al., 2017; Ubrig et al., 2019; Upadhyay et al., 2019], therefore not providing a sufficient understanding about speech production in real communication conditions and noisy environments. Our results showed, as predictable, a significant increase of both F_0 and loudness in the reading task with background noise, which was evident in both NH subjects and deaf patients with CI on. Similar outcomes, although limited to postlingual deafness, were confirmed by Lee as patients with CI seem to respond to background noise by adjusting speech production accordingly, as a potential perceptual benefit of the Lombard effect which works regularly in NH subjects, and which is properly restored with CI turned on [Lee et al., 2017].

In the comparison between the vowel and the reading tasks, NH females were shown to maintain both F_0 and loudness relatively steady, whereas NH males showed similar loudness but significantly lower F_0 values in the vowel task. As far as the steadiness of voice loudness is concerned, and assuming that the vowel uttering and the

text reading are two successive voice production tasks, the obtained results corroborate a study by Castellana et al. who found that NH subjects exhibit a low intra-speaker variability within 1 dB for equivalent and mean SPLs and below 2 dB for mode SPL [Castellana et al., 2017]. On the contrary, all deaf patients demonstrated higher F_0 and loudness values in the vowel task compared to the reading. A very useful review of the literature by Borden suggests that a very short auditory information is not sufficient for motor control centers to simultaneously regulate speech production [Borden, 1979]. Otherwise, a reading, lasting about 1 min, allows the subject more time to analyze his speech and possibly make a correction of its parameters.

Role of CI Activation

Similar results were also found in relation to CI activation, highlighting its role in bringing a change in the way voice is handled by patients. After switching on the CI in the sustained vowel task, despite a slight but not significant reduction in F_0 and loudness values, the whole sample of deaf patients did not show the expected voice modifications presumably due to the sudden change in auditory feedback. As mentioned by Gautam, indeed, vocal control may not be sometimes dependent on moment-to-moment feedback but over longer time scales, thus not allowing sufficient vocal adaptation in case the CI is switched on and off within a few minutes and in case the task is too short [Gautam et al., 2019]. In this regard, we highlighted heterogeneous and discordant results in literature: Monini reported a significantly reduced F_0 in the voice samples of the Italian vowel /a/ at an early stage after cochlear implantation, although adults and children were assessed together [Monini et al., 1997]. Differently, Kirk and Edgerton reported, in the vowel /a/ assessment, lower F_0 values and a reduced variability of loudness level only on male patients, whereas females showed higher F_0 and an increasingly variable loudness with CI on [Kirk and Edgerton, 1983].

As for the reading of the text, the switching on of the CI seems able to significantly reduce both loudness and F_0 in deaf men, up to values comparable to NH subjects: this result is consistent with the observations of Hamzavi et al. whose CI patients tended to have lower F_0 postoperatively approaching the normal range of F_0 [Hamzavi et al., 2000]. Leder, in this regard, demonstrated that when adequate auditory feedback is restored with cochlear implantation, the F_0 is the first acoustic characteristic to approximate normal values again and that was particularly evident in men [Leder et al., 1987]. Conversely, the

CI activation caused overall no significant changes of the F_0 values in deaf women during the reading task. Such a great variability of frequency among deaf subjects can be found in all the very few works proposed so far in the literature on the subject, approximately all discordant with each other in the results and mostly focused on pediatric population [Borden, 1979; Kirk and Edgerton, 1983; Hamzavi et al., 2000; Coelho et al., 2009].

The analysis of the vocal characteristics of the patients did not allow to highlight any significant difference in the phonatory outcome between CI recipients from different manufacturers. Since the hearing perceived by any type of hearing aid is certainly also characterized by a relevant subjective component, it is very complex to compare the hearing outcomes between two different CI companies; however, as in our study, Withers previously found no differences in PTA and speech perception in a case of bilateral cochlear implantation using different devices, although patients' opinions on perceived sound quality significantly differed [Withers et al., 2011]. In fact, although any CI of each company has unique technical features and heterogeneous hearing outcomes have been frequently described in literature depending on CI specific features, any device, if properly implanted and correctly functioning, is able to improve hearing and thus determine a restoration of the auditory feedback. Therefore, we can conclude that the previously described speech modifications in terms of F_0 and loudness are exclusively related to the simple use of the device and not to the model or the brand of the CI adopted.

Role of Prelingual and Postlingual Deafness

The period of onset of the deafness is known to affect speech as early deprivation of auditory feedback affects F_0 control and articulation accuracy, just as people with prelingual deafness have difficulty learning to speak intelligibly [Ruff et al., 2017]. Nonetheless, although there were lower values of both F_0 and loudness in postlingual deafness, we had no significant differences between speech characteristics of prelingual and postlingual deaf patients, both in the sustained vowel task and in the reading task, as also the speech quality of postlingual deaf patients decreases due to a lack of adequate auditory feedback. The only exception was reported for females, whose subjects with postlingual deafness showed significantly lower F_0 values than deaf females with prelingual deafness.

Similar results were also reported after CI activation, both in the vowel phonation and in the reading, with no differences between prelingual and postlingual deafness. We can therefore affirm that, although different postop-

erative auditory results are reported in the literature depending on the period of onset of the hearing loss, almost all deaf patients behave in a similar way from the phonatory point of view, whatever the nature (prelingual or postlingual) of their deafness. Moreover, the further addition of background noise to speech assessments performed on CI recipients did not demonstrate significant differences in their phonatory characteristics, both in case of prelingual and postlingual deafness.

The analysis of how the patients' speech parameters changed after switching on the CI showed an important reduction in loudness values when reading the passage, both for patients with prelingual and postlingual deafness. Similarly, we found that the application of the CI also plays a decisive role in modifying the F_0 in patients with postlingual deafness, although this only happens in males. Different outcomes were reported by Smoorenburg in the evaluation of speech samples before and one to 4 years after cochlear implantation: although analyzing only postlingual deafness, he noticed that abnormally high pitches of deafs decreased after CI in some of the implanted women but not in men [Smoorenburg et al., 1994].

The significant positive correlation that emerged between postoperative hearing thresholds and speech loudness confirmed that subjects with better hearing outcomes after CI activation generally speak with a lower loudness, which literature has shown to turn in a reduced vocal effort and load [Bottalico and Astolfi, 2012; Puglisi et al., 2017]. Furthermore, the negative correlation found between overall patients' age and speech F_0 values highlighted how older deaf patients, whether males or females, generally speak with a lower F_0 when the CI is on, both in quiet conditions and in the presence of background noise. This result agrees with past studies, although conducted only on NH listeners, as F_0 tended to decrease markedly in association with aging [Nishio and Niimi, 2008]. Such correlation could be explained not only by the simple application of the CI but also by the reduced speed of cognitive processing with advancing age: a slowdown of specific executive cognitive resources, such as working memory, is known to influence several top-down mechanisms, one of which could also be phonation [Zucca et al., 2022].

Conclusions

The aim of the present study was to evaluate the voice modifications in a cohort of adults submitted to cochlear implantation, balanced between prelingual and postlin-

gual deafness. All patients were subjected to speech recordings consisting of both vowels and the reading a phonetically balanced passage while equipped with a contact-sensor-based voice monitoring device (i.e., KayPENTAX's APM device). Overall, we highlighted the ability of the CI to adjust certain phonatory aspects such as fundamental frequency and loudness in most deaf patients simply by restoring auditory feedback, thus improving their vocal experience in whatever acoustic conditions they wish to communicate. Particularly, we found similar speech performances between prelingual and postlingual groups, both in the vowel /a/ phonation and in the reading. Moreover, although poorer auditory outcomes with CI have been commonly demonstrated in adults with prelingual deafness due to the longer history of sound deprivation, our results provide a further suggestion that patients with congenital prelingual deafness may absolutely benefit from cochlear implantation, even at a later age. A future development of this study will be the analysis of further qualitative aspects of voice production after CI application such as pitch strength, cepstral peak prominence smoothed, acoustic voice quality index, jitter, shimmer, and harmonics-to-noise ratio.

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Statement of Ethics

The study was approved by the bioethics Institutional Review Board of the University of Turin (approval number 3546). The study was conducted in accordance with the ethical standards of our institution and the principles expressed in the Declaration of Helsinki. Written informed consent to participate in the study was obtained from all participants.

Conflict of Interest Statement

The authors have no conflicts of interest to declare.

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Author Contributions

Andrea Albera, Giuseppina Emma Puglisi, and Andrea Canale performed measurements, analyzed data, and wrote the paper; Arianna Astolfi and Francesco Mozzanica designed the study; and Giuseppe Riva and Claudia Cassandro provided statistical analysis and critical revision.

Data Availability Statement

All data generated or analyzed during this study are included in this article. Further inquiries can be directed to the corresponding author.

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