Voice parameters in children with cochlear implants: a systematic review and meta-analysis

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ABSTRACT:

Introduction: An intact auditory system is essential for the development and maintenance of voice quality and speech prosody. On the contrary hearing loss affects the adjustments and appropriate use of organs involved in speech and voice production. Spectro-acoustic voice parameters have been evaluated in Cochlear Implant (CI) users, and the authors of previous systematic reviews on the topic concluded that Fundamental Frequency seemed preliminarily the most reliable parameter to evaluate voice alterations in adult CI users.

The main aim of this systematic review and meta-analysis was to clarify the vocal parameters and prosodic alterations of speech in pediatric CI users.

Materials and methods: The protocol of the systematic review was registered on the PROSPERO database, International prospective register of systematic reviews. We conducted a search of the English literature published in the period between January 1st 2005 and April 1st 2022 on the Pubmed and Scopus databases. A meta-analysis was conducted to compare the values of voice acoustic parameters in CI users and non-hearing-impaired controls. The analysis was conducted using the standardized mean difference as the outcome measure. A random-effects model was fitted to the data.

Results: A total of 1334 articles were initially evaluated using title and abstract screening. After applying inclusion/exclusion criteria, 20 articles were considered suitable for this review. The age of the cases ranged between 25 to 132 months at examination. The most studied parameters were F0, Jitter, Shimmer and Harmonic Noise Ratio (HNR); other parameters were seldom reported. A total of 11 studies were included in the meta-analysis of F0, with the majority of estimates being positive (75%); the estimated average standardized mean difference based on the random-effects model was 0.3033 (95% CI: 0.0605 to 0.5462; p = 0.0144). For Jitter (0.2229; 95% CI: -0.1862 to 0.7986; p = 0.2229) and shimmer (0.2540; 95% CI: -0.1404 to 0.6485; p = 0.2068) there was a trend toward positive values without reaching statistical significance.

Discussion and conclusions: This meta-analysis confirmed that higher F0 values have been observed in the pediatric population of CI users compared to age-matched normal hearing volunteers, whereas the parameters of voice noise were not significantly different between cases and controls. Prosodic aspects of language need further investigations. In longitudinal contexts, prolonged auditory experience with CI has brought voice parameters closer to the norm. Following the available evidence, we stress the utility of inclusion of vocal acoustic analysis in the clinical evaluation and follow-up of CI patients to optimize the rehabilitation process of pediatric patients with hearing loss.

Key words: Cochlear Implants; Jitter; Shimmer; Fundamental Frequency.

Abbreviations: ACE (Advanced Combination Encode); CAG (Case Group); CI (Cochlear Implant); COG (Control Group); EI (Early Implanted); FSP (Fine Structure Processing); F0 (Fundamental Frequency); F1-F2 (Formant’s 1 and 2); HL (Hearing Loss); HNR (Harmonic Noise Ratio); LI (Late Implanted); MDVP (Multi-Dimensional Voice Program); MPT (Mean Phonation Time); NA (Not Applicable); NOS (Newcastle Ottawa Scale); NR (Not Reported); PRAAT (Praat’s Voice Program); PTA (Pure Tone Average); SDF0 (Standard Deviation of Fundamental Frequency).

1. INTRODUCTION

The auditory system is essential for the development and maintenance of voice quality and speech prosody: normal hearing people have solid speech and voice control.[1] On the contrary, hearing loss affects adjustments and appropriate use of organs involved in speech and voice production, primarily because of the alteration of auditory feedback.[2–4]

Since the end of the 19th century, it has been known that an electrical stimulus can give an auditory sensation.[5] Dr. William House was the first to introduce a single electrode implant in the 1970s.[6] Since then, following technological development, the modern multi-channel implants became available.[5,7] Simultaneous bilateral cochlear implantation is currently the gold standard rehabilitation technique for patients with congenital severe-to-profound hearing loss.[8] This intervention provides significant benefits to speech perception, in most cases allowing proper language restoring communication functionality to be developed. Following adequate diagnosis, a cochlear implantation performed prior to 12 months of age offers the best opportunity to develop more typical auditory performances during late infancy and early childhood,[9] with functional outcomes of deaf children reaching that of normal hearing children before entering in primary school.[8] Moreover, it has been shown that auditory feedback cues have a crucial role for the Cochlear Implant (CI) users to monitor and fine-tune their speech articulation and make purposeful adjustments to their voicing,[4] even if subtle alterations can persist even through correct rehabilitation.[1,4]

Speech alterations can be quantified by computerized acoustic speech programs.[10] The main reported parameters are average fundamental frequency (FA0), jitter, shimmer and harmonic/noise ratio (HNR). Several studies examined the acoustic voice parameters in children with CI but, to the best of our knowledge, no normative data are available in this setting.[1,11] According to the few systematic reviews present in the international literature, preliminary Fundamental Frequency (F0) seemed to be the most reliable parameter to evaluate voice alterations in adult CI users.[1,11] Generally speaking, Medvedev et al. (2021)[1] and Coelho et al. (2012)[11], the authors of previous systematic reviews on the topic, concluded that there was not an effective number of studies with high levels of evidence that precisely demonstrate the effects of CI use on the spectro-acoustic voice parameters.[1,11]

The main aim of this systematic review and meta-analysis was to clarify the vocal parameters and prosodic alterations of speech in pediatric CI users. The secondary aim was to propose a spectro-acoustic tools panel with a clinical relevance.

2. MATERIAL AND METHODS

2.1 Protocol Registration

The protocol of the Systematic Review was registered on the PROSPERO database International prospective register of systematic reviews (Center for Reviews and Dissemination, University of York, York, UK). The ID number of the protocol is CRD42022345334.

2.2 Electronic database Search

We conducted a search of the English literature published in the period between January 1st 2005 and April 1st 2022 on the Pubmed and Scopus databases. We used the following keywords: “fundamental frequency cochlear”; “jitter cochlear”; “harmonic to noise ratio cochlear”; “shimmer cochlear”. MeSH terms and keywords were combined accordingly on the databases. The reference lists of all the included articles were accurately screened in order to identify other pertinent studies. The “Related articles” option present on the PubMed homepage was also considered.

2.3 Inclusion and Exclusion Criteria

A study was included only if the following general criteria were met: i) pediatric CI users evaluated before voice change (age ≤11 years old); ii) case-control study design; iii) detailed information about diagnosis and treatment of hearing loss; iv) detailed information about voice analysis.

Exclusion criteria were: i) study design of case report, editorial, survey, letter to the editor and review; ii) animal model study; (iii) non-English language study.

2.4 Data extraction and quality assessment

The authors analyzed the data from the available literature. Included studies were investigated to extract all available data and assure eligibility for all patients. The Newcastle-Ottawa Scale (NOS) was used to assess the quality of the included studies.[12] Disagreements about inclusion/exclusion of manuscripts were solved by a discussion among the study team members.

2.5 Statistical Analysis

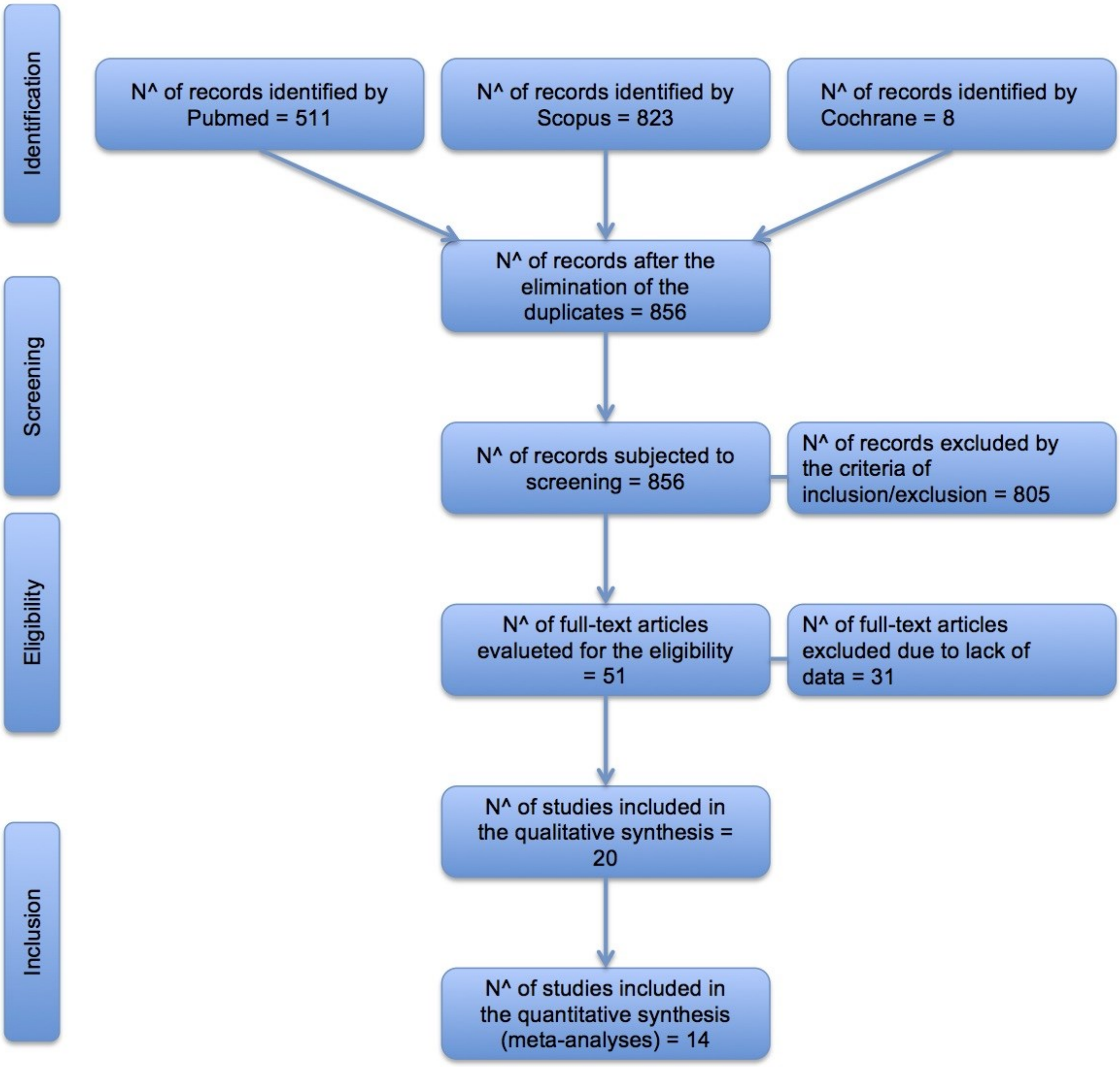
A meta-analysis was conducted to compare the values of voice acoustic parameters in cochlear implant users and non-hearing-impaired control groups. The analysis was performed using the standardized mean difference as outcome measure. A random-effects model was fitted to the data. The amount of heterogeneity was estimated using the restricted maximum-likelihood estimator, the Q-test for heterogeneity and the I^2 statistic.[13] An I^2 value of 0% indicates no heterogeneity, while values around 25%, 50% and 75% may be interpreted as low, moderate and high heterogeneity, respectively.[14] Studentized residuals and Cook's distances were used to examine whether studies may be outliers and/or influential in the context of the model. The rank correlation test and regression test, using the standard error of the observed outcomes as predictor, were used to check for funnel plot asymmetry. For the meta-analysis, Jamovi Computer Software Version 2.3 for MacOs Big Sur (Open Source available at https://www.jamovi.org/) was used.

3. RESULTS

3.1 General characteristics and overall quality of retrieved studies

A total of 1334 articles were initially evaluated in title and abstract screening. Fifty-one investigations were identified as potentially relevant to the topic. The full-text screening of those articles led to the exclusion of 31 original manuscripts, in agreement with the inclusion/exclusion criteria. The remaining 20 studies were considered suitable for this review.[15–34] A PRISMA flow diagram depicts the flow of information through the different literature review phases (**Figure 1**).

Figure 1. PRISMA diagram showing database search, from identification to inclusion.



Only three studies were prospective,[18,28,29] the majority being observational and retrospective.[15–17,19–27,30–34] According to the NOS scale, the quality of the included studies was considered as fair or high (≥6/9) for more than half of them,[18,22–27,29,31–33] and scarce or low

(<6/9) for 9 out of 20.[15–17,19–21,28,30,34] **Table 1** lists country, study designs and NOS quality scores of the included manuscripts.

Table 1. General information and Assessment of quality of included study according to Newcastle-Ottawa Scale.

First Author (Year)	Country	Type of study	NOS	Meta analysis
Poissant 2006[15]	USA	Observational	5	NA
Campisi 2006[16]	Canada	Observational	3	F0;Jitter;Shimmer
Hocevar-Boltezar 2005[17]	Slovenia	Longitudinal	2	NA
Hocevar-Boltezar 2006[18]	Slovenia	Observational	7	NA
De Castro Coelho 2009[19]	Brazil	Observational	3	NA
Allegro 2010[20]	Canada	Observational	3	F0;Jitter;Shimmer
Yun Suk An 2012[21]	Korea	Observational	3	NA
Souza 2012[22]	Brazil	Observational	8	F0;Jitter;Shimmer
Coelho 2015[23]	Brazil	Observational	6	F0;Jitter;Shimmer
Jafari 2016[24]	Iran	Observational	8	F0;Jitter;Shimmer
Joy 2017[25]	India	Observational	6	NA
Knight 2016[26]	South Africa	Observational	6	F0;Jitter;Shimmer
Moein 2017[27]	Iran	Observational	7	F0
Wang 2017[28]	China	Longitudinal	5	NA
Van de Velde 2018[29]	Netherland	Longitudinal	7	F0
Upadhyay 2019[30]	India	Observational	4	F0;Jitter;Shimmer
Delgado-Pinheiro 2020[31]	Brazil	Observational	7	F0;Jitter;Shimmer
Mao 2020[32]	China	Observational	7	NA
Umashankar 2021[33]	India	Observational	7	F0;Jitter
Xu 2021[34]	USA	Observational	5	NA

The literature search was updated to April 1st 2022.

3.2 Population and intervention

Most of the considered studies had control groups consisting of normal hearing volunteers.[22–34] In three investigations, the study group patients tested before undergoing CI implantation were used as controls.[17,18,21] In a different series, the children were tested in a CI-on condition for study group and in a CI-off condition for control one.[15] Allegro et al.[20] and Campisi et al.[16] used, the normal values of a previous study[35] as controls whereas De Castro et al.[19] used the normative values of the CSL/MDVP software for comparison (KayPENTAX, Lincoln Park, NJ, USA).

The study groups were composed of children with a cochlear implant[15-34]. The sample size ranged from 6[15,21] to 278[34] patients, as summarized in **Table 2**. The age of the cases at examination ranged between 25[29] and 132[34] months.

Table 2. Clinical characteristics of the population of included studies.

First Author (Year)	COG definition	COG number	COG age MEAN and SD (mts)	CAG number	CAG age MEAN (mts)	AGE AT CI (mts)	CI USE (mts)	HL Onset
Poissant 2006[15]	CI-off	6	85	6	85	26,4-90	33,6	5 pre, 1 post
Campisi 2006[16]	reference Campisi 2002[35]	NR	NR	21	124	36-211	NR	prelingual
Hocevar-Boltezar 2005[17]	Pre-CI	31	68,88±43,8	31*	80,88±43,8	74,88	6	prelingual
				31**	86,88±43,8	74,88	12	prelingual
				31***	98,88±43,8	74,88	24	prelingual
Hocevar-Boltezar 2006[18]	Pre-CI	NR	70,86±40,92	29	70,68±40,92	35,156	6,12	prelingual
De Castro Coelho 2009[19]	NR	NR	NR	25	NR	NR	NR	prelingual
Allegro 2010[20]	reference Campisi 2002[35]	NR	NR	10	86,4	NR	58	5 post; 5 pre
Yun Suk An 2012[21]	Pre-CI	6	71,04±21,84	6	71,04±21,84	71,04	NR	6 pre; 6 post
Souza 2012[22]	NH	25	61	36	51	20	28	prelingual
Coelho 2015[23]	NH	25	48	28 (ACE)	49,844±9,042	23,072	26,711	prelingual
				23 (FSP)	46,607±6,276	25,982	20,617	prelingual
Jafari 2016[24]	NH	15	77±18	15	72±14,64)	36	8	prelingual
Joy 2016[25]	NH	10	60,72	31*	60	43	6	prelingual
				31**	60	43	12	prelingual
				31***	60	43	24	prelingual
Knight 2016[26]	NH	10	51,36±1,14	5	80,76±1,01	24-48	36-72	Prelingual
Moein 2017[27]	NH	50	108±1,49	25	107,4±1,42	31.4	76,08	Prelingual
Wang 2017[28]	NH	15	68	30	62	62	24	prelingual
Van de Velde 2018[29]	NH	12	25,1±4,8	9 (EI)	25,2±8,4	36	11	prelingual
				9 (LI)	81,6±30	38,76	18	prelingual
Upadhyay 2019[30]	NH	42	48,24	42	48,12	35,64	12,48	prelingual
Delgado-Pinheiro 2020[31]	NH	20	124	13	124	24,05	nr	nr
Mao 2020[32]	NH	173	81,96±34,2	278	79,56±41,52	40,56	39,12	Prelingual
Umashankar 2021[33]	NH	43	44,4 ±7,2	44	41,76±0,45	6-60	nr	prelingual
Xu 2021[34]	NH	26	105,6±14,4	13 (Bimodal)	108±16,8	168	75,6	Prelingual
				31 (Sequential)	105,6±16,8	118,8	80,4	
				11 (Simultaneous)	132±13,2	115,2	87,6	

Legend: *evaluated at 6 months after CI; ** evaluated at 12 months after CI; *** evaluated at 24 months after CI.

Delgado-Pinheiro et al. reported data of 13 children with cochlear implant plus 7 with hearing aids; according to the objective of this review, we included only the data regarding the sub-group of 13 CI patients[31] in qualitative and quantitative analysis.

Hearing loss onset was pre-verbal in most series, except for three studies,[15,20,21] where a few patients with post-verbal hearing loss were also enrolled. The age at implantation ranged from 6[33] to 118 months.[34] The average time of CI use ranged from 6[17,18,25] to 87 months.[34]

Hearing performance with fitted CI were reported by four research groups as Pure Tone Average (PTA) at frequencies 500-1000-2000-4000Hz. PTA ranged from 23.55 to 47.25 dB;[28] PTA ranged from 21.25 to 46.75 dB;[18] mean PTA of 29±7.20dB;[31] PTA<30dB for included patients.[19] A different research group reported a mean value of voice detection threshold at speech audiometry: 23±4dB.[19] In half of the included studies, the patients underwent speech and hearing rehabilitation (treatment modalities were not being reported).[15,20,22,24–26,28–31] Objective evaluations of patients' larynges by endoscopy were reported only by two research groups.[16,17]

3.3 Spectro-acoustic parameters

Different softwares were used for voice analysis: PRAAT (Open Source available at www.praat.org);[22,24,27,29,33] MDVP software (KayPENTAX, Lincoln Park, NJ, USA)[15-21,23,26,28,31] and Dr. Speech software (Tiger Inc, Seattle, USA),[25,30] as summarized in **Table 3**. Two research groups did not report which software was used for acoustic analysis.[32,34] In most cases, the voice parameters were extracted from sustained /a/ vowel.[16–26,28,30,31] In one single investigation the /ə/ vocal was used[33]. Moreover, two research groups performed voice analysis on monosyllabic words[27,32]. On the contrary, Van de Velde et al.[29] and Poissant et al.[15] used spontaneous speech; Xu et al.[34] performed voice analysis on sang samples of "Happy Birthday".

3.3.1 Fundamental Frequency

The F0 refers to the approximate frequency of the periodic structure of voiced speech signals. It is defined as the average number of oscillations of the vocal folds per second, expressed in Hertz (normal value for pediatric population: 279.05±5.79Hz).[35] F0 was measured in the majority of included studies.[16-31,33] Significantly higher values of F0 in the case series were reported by 5[23,27,28,30,33] out of 17 studies. Two research groups reported significantly lower values in the CI group;[19,23] 10 others did not find significant differences (groups' characteristics are reported in **Table 2**, values are reported in **Table 3**).[16-18,20-22,24-26,29,31]

Table 3 Voice parameters analyses and significant results.

First Author (Year)	Software	Sample	F0 COG	F0 CAG	Jitter COG	Jitter CAG	Shimmer COG	Shimmer CAG	HNR COG	HNR CAG	OTHER PARAMETERS
Poissant 2006[15]	MDVP	Spontaneous speech	NR	NR	NR	NR	NR	NR	NR	NR	**word duration**
Campisi 2006[16]	MDVP	vowel |a|	279,1±5,8	267,76 ±40,75	1,24 ±0,07	0,88 ±0,45	3,35 ±0,12	3,51 ±1,32	NR	NR	vAm
Hocevar-Boltezar 2005[17]	MDVP	vowel |a|	286±58,22	295,52 ±57,09 274,46 ±75,31 294,54 ±70,92	2,89 ±1,87	**1,75*** ±1,312 **1,52*** ±1,25 **0,89*** ±0,52	6,27 ±3,78	**4,39*** ±1,8 **4,14*** ±1,98 **3,09*** ±1,25	0,18±0,11	0,18±0,23 0,15±0,04 **0,11***±0,02	NR
Hocevar-Boltezar 2006[18]	MDVP	vowel |a|	292,79±69,61	286,52 ±66,17	2,58 ±1,85	**1,54*** ±1,18	6,06 ±3,57	**4,21*** ±2,38	0,17±0,11	0,14±0,04	**vF0; vAm**
De Castro Coelho 2009[19]	MDVP	vowel |a|	279,05	**261,93***	1,24	1,32	3,35	3,73	0,11%	0,13%	NR
Allegro 2010[20]	MDVP	vowel |a|	279,05±56,16	293,93	1,24 ±0,68	0,95	3,35 ±1,16	3,6	NR	NR	NR
Yun Suk An 2012[21]	MDVP	vowel |a|	NR	270.8±30,9	NR	1,18 ±1,26	NR	**4,03*** ±1,9	NR	NR	NR
Souza 2012[22]	PRAAT	vowel |a|	281,92±50,04	300,72 ±49,03	0,38 ±0,16	1,28 ±5,27	3,42 ±1,67	3,42 ±1,81	21,08 ±3,94	20,83±3,93	**F0min; pitch range**
Coelho 2015[23]	MDVP	vowel |a|	279,53±39,00	**273,56***±49,56 **321,19***±51,63	1,23 ±0,9	4,51 ±15,13 6,58 ±3,43	3,54 ±1,52	6,01 ±4,55 4,51 ±2,29	0,12 ±0,02	2,1±0,24 2,06±0,23	**vAm; vF0**
Jafari 2016[24]	PRAAT	vowel |a|	250,74±34,22	270,16 ±32,03	0,662 ±0,23	0,779 ±0,376	5,063 ±0,865	6,104 ±1,863	19,737 ±2,498	18,43±3,04	NR
Joy 2016[25]	Dr. Speech	vowel |a|	NR	315,37 282,45 266,49	NR	0,65 1,11 **0,78***	NR	7,33 5,43 **5,61***	NR	NR NR NR	NR
Knight 2016[26]	MDVP	vowel |a|	297,74±58,01	265,82 ±26,66	1,23 ±0,45	1,79 ±1,09	5,72 ±1,93	5,1 ±2,23	NR	NR	NR
Moein 2017[27]	PRAAT	monosyllables	230,57±41,5	**256.65***±31,21	NR	NR	NR	NR	NR	NR	**pitch range**
Wang 2017[28]	MDVP	vowel |a|	283,23±9,51	**375,50***±11.19	0,59 ±0,13	**1,64*** ±0,23	3,24 ±0,42	6,62 ±0,36	NR	NR	**SDF0**
Van de Velde 2018[29]	PRAAT	Spontaneous speech	318.7±49,2	321,46 ±54,02 291,03 ±41,12	NR	NR NR	NR	NR NR	NR	NR NR	NR
Upadhyay 2019[30]	Dr. Speech	vowel |a|	289,95±19,13	**313,67***±35,28	0,39 ±0,13	**0,65*** ±0,44	1,65 ±0,4	**2,74*** ±1,04	21,4 ±3,31	**16,12***±3,15	**MPT**
Delgado-Pinheiro 2020[31]	MDVP	vowel |a|	221,2±51,7	252,2±52,8	1,015 ±0,733	0,836 ±0,485	2,711 ±0,81	2,638 ±1,045	0,121% ±0,019%	0,124%±0,014%	**vF0; vAm**
Mao 2020[32]	NR	monosyllables	NR	NR	NR	NR	NR	NR	NR	NR	NR
Umashankar 2021[33]	PRAAT	vowel /ə/	293,21±50,67	**310,36***±58,6	0,66 ±0,49	**1*** ±0,87	NR	NR	NR	NR	**F1; F2**
Xu 2021[34]	NR	Happy Birthday	NR	NR	NR	NR	NR	NR	NR	NR	**note deviation**

Legend * parameters reported as statistically significant by the authors at $p<0.05$; (SD) in squared the standard deviations are reported.

3.3.2 Jitter

Jitter (JITT%) is the cycle-to-cycle variability of the period duration of the acoustic signal coming from voice production; higher values may indicate irregularities in the frequency at which the vocal cords vibrate (normal value for pediatric population: 1.24±0.07).[35] Jitt% was measured by several research groups.[16-26,30,31,33] Significant differences were found in six[17,18,24,28,30,33] out of 14 investigations: higher value of Jitter in CI patients compared to normal hearing controls;[25,28,30,33] higher values of Jitter before CI implantation with significant reduction at 6,[17,18] 12[18] and 24[18] months after CI (group characteristics are reported in **Table 2**, values are reported in **Table 3**).

3.3.3 Shimmer

Shimmer (Shim%) expresses the average absolute difference between the amplitudes of consecutive periods divided by the average amplitude; higher values may indicate voice instability (normal value for the pediatric population: 3.35±0.12).[35] Shim% was measured in 13 studies.[16-26,30,31] Significant differences were found in 5 articles:[17,18,21,25,30] higher value of Shimmer in CI patients compared to normal hearing controls.[21,25,30] Higher values of Shimmer before CI implantation with significant reduction at 6,[17,18] 12[18] and 24[18] months after CI (group characteristics are reported in **Table 2**, values are reported in **Table 3**).

3.3.4 Harmonics-to-Noise ratio (HNR)

Various parameters were analyzed in the considered studies. The HNR is a measure of the proportion of harmonic sound to noise in the voice measured in dB: the lower the HNR, the more noise in the voice. Most of the included investigations measured the HNR without finding significant differences between cases and controls.[17-19,22,23,27,32] Only Upadhyay et al.[30] found significantly lower values in CI patients compared with controls: 16.1±3.15dB vs 21.4±3.31dB.[30]

3.3.5 Other significant parameters

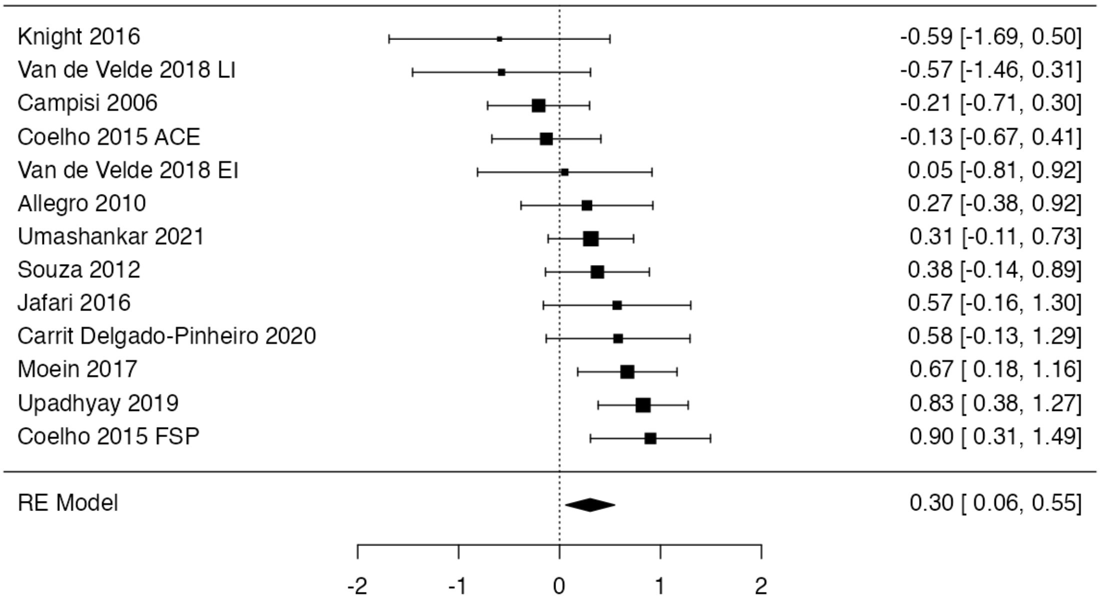
Other acoustic parameters were investigated. The variation of medium Amplitude (vAm) was significantly higher in the CI group compared to the normal hearing group in three investigations: 17.543% vs 13.013% (p=0.015);[23] 13.73% vs 11.13% (p=0.099);[31] 23.6% vs 15.1% (p=0.009).[16] The vAm was significantly decreased after CI compared to before surgery in one study (35.99% vs 29.13%; p=0.031).[18] Poissiant et al.[15] measured average mono-syllabic and bi-syllabic words duration in a group of 6 CI patients in a CI-on/CI-off condition and found significantly longer word duration in the former. Hocevar Boltezar et al.,[18] Coelho et al.[23] and Delgado-Pinheiro et al.[31] measured the variation of Fundamental frequency (vF0). Significantly higher values were found in the CI group compared to the normal one: 2.88% vs 1.93% (p=0.032) for Coelho et al.[23] and 2.59 vs 2.14 (p=0.011) for Delgado-Pinheiro et al..[31] Moreover, vF0 was also measured in the deaf children before surgery and compared to a period of 6-12 months after CI and there was a significant improvement: 8.26% vs 4.53% (p=0.014) for Hocevar Boltezar et al..[18] Wang et al.[28] found significantly higher values in standard deviation of F0 (sdF0) in the CI group than in the normal hearing group (16.76Hz vs 6.24Hz; p<0.05). Souza et al.[22] measured the minimum fundamental frequency (F0min) and the vocal extension in halftones in the CI group and normal hearing control group, with significantly different values: 213.69Hz vs 183.84Hz (p<0.020) and 6.80tones vs 8.40tones (p<0.006), respectively. Moein et al.[27] found a significant reduction in mean pitch ranges of speech in the CI group compared to the normal hearing group: 173.42Hz vs 201.39Hz (p<0.03). Upadhyay et al.[31] measured the maximum phonation time (MPT) and found significantly lower values in the CI group compared to normal hearing (4.83sec vs 5.86sec; p<0.001). Umashankar et al.[33] calculated the formants F1 and F2, with significant differences 899.5Hz vs 1103Hz and 1515hz vs 1652Hz (both values of p<0.01). In a study by Xu et al.[34] the mean note deviation from the original partiture singing the song "Happy Birthday" was considered, with significant values of 2.49 vs 1.39 (p<0.001) semitones in the CI group vs normal hearing group (group characteristics are reported in **Table 2**).[34]

3.4 Meta-analysis of voice parameters

According to the available data in the retrieved studies, a meta- analysis of results was possible for F0, Jitter and Shimmer by comparing values in CI groups vs age-sex matched normal hearing controls.

A total of 11 studies were included in the analysis of F0 values,[16,20,22-24,26,27,29-31,33] overall reporting 13 different cohorts due to subgroups presented in two investigations (**Figure 2**).[23,29] The observed standardized mean differences ranged from -0.5950 to 0.9007, with the majority of estimates being positive (75%). The estimated average standardized mean difference based on the random-effects model was 0.3033 (95% CI: 0.0605 to 0.5462), significantly different from 0 (p = 0.0144). The studies appeared to be moderately heterogeneous (I^2 = 52.3638%; p = 0.0127). An examination of the studentized residuals revealed no indication of outliers in the context of the model. According to Cook's distances, none of the studies could be considered to be overly influential. The regression test (Egger's) indicated funnel plot asymmetry (p = 0.0400) but not the rank correlation (Begg and Mazumdar) test (p = 0.0866).

Figure 2. Forest plots of Fundamental Frequency (F0)



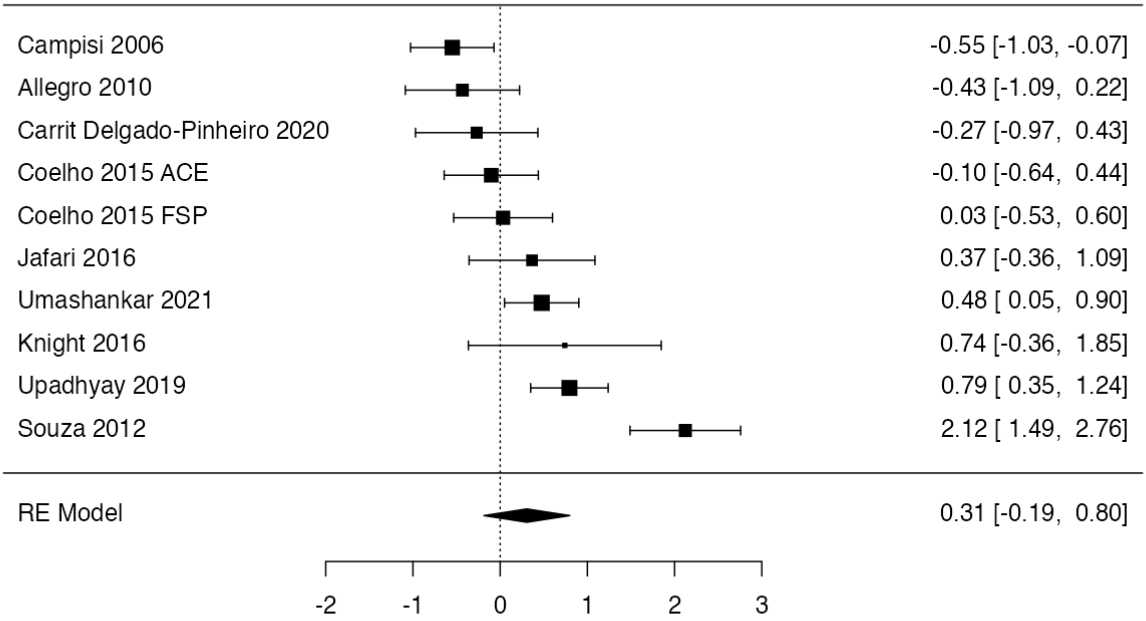
F0 Random-Effects Model (k = 13) and Heterogeneity Statistics

	Estimate	se	Z	p	CI Lower Bound	CI Upper Bound	
Intercept	0.303	0.124	2.45	0.014	0.060	0.546	
Tau	**Tau²**	**I²**	**H²**	**R²**	**df**	**Q**	**p**
0.315	0.0989 (SE=0.0793)	52.36%	2.099	-	12.000	25.475	0.013

Note. Tau² Estimator: Restricted Maximum-Likelihood

A total of 9 studies were included in the analysis of Jitter values,[16,20,22-24,26,30,31,33] overall reporting 10 cohorts due to subgroups presented in Coelho et al. (**Figure 3**).[23] The observed standardized mean differences ranged from -0.5490 to 2.1229, with the majority of estimates being positive (60%). The estimated average standardized mean difference based on the random-effects model was = 0.3062 (95% CI: -0.1862 to 0.7986). Therefore, the average outcome did not differ significantly from zero ($p = 0.2229$). The studies appeared to be highly heterogeneous ($p < 0.0001$, $I^2 = 86\%$). An examination of the studentized residuals revealed that one study (Souza et al.)[22] may be a potential outlier in the context of this model and, according to the Cook's distances, it could be considered to be overly influential. Neither the rank correlation nor the regression test indicated any funnel plot asymmetry ($p = 1.0000$ and $p = 0.7057$, respectively).

Figure 3. Forest plots of Jitter.



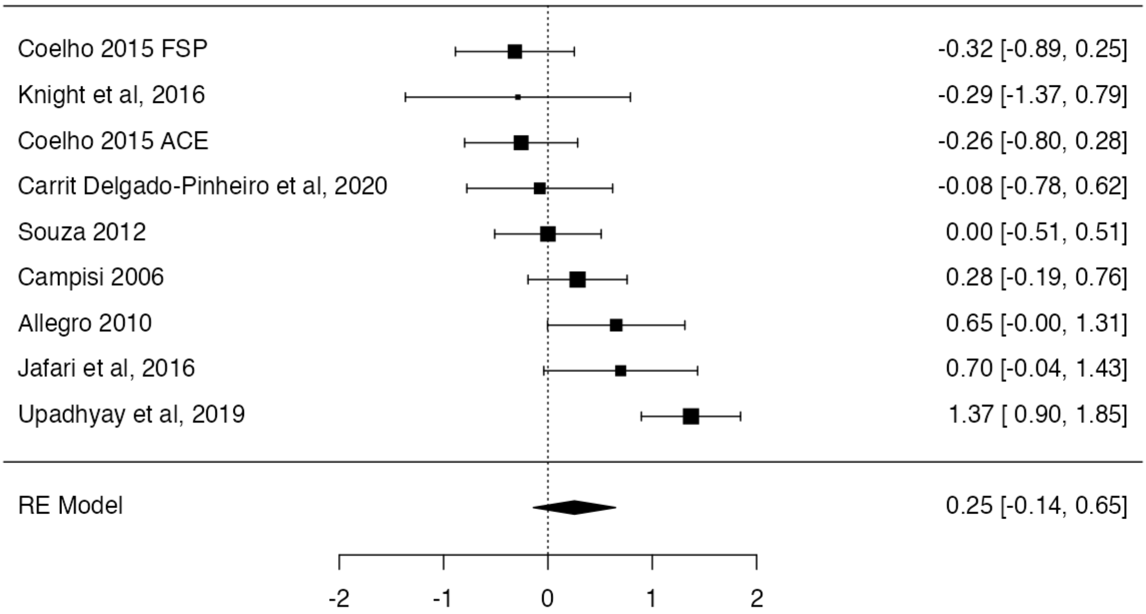
Jitter Random-Effects Model (k = 10) and Heterogeneity Statistics

	Estimate	se	Z	p	CI Lower Bound	CI Upper Bound	
Intercept	0.306	0.251	1.22	0.223	-0.186	0.799	
Tau	**Tau²**	**I²**	**H²**	**R²**	**df**	**Q**	**p**
0.726	0.5268 (SE=0.2965)	86.01%	7.146	-	9.000	60.201	< .001

Note. Tau² Estimator: Restricted Maximum-Likelihood

A total of 8 studies were included in the analysis of Shimmer values[16,20,22-24,26,30,31] overall reporting 9 cohorts due to subgroups presented in Coelho et al. (**Figure 4**).[23] The observed standardized mean differences ranged from -0.3169 to 1.3707, with the majority of estimates being negative (44%). The estimated average standardized mean difference based on the random-effects model was = 0.2540 (95% CI: -0.1404 to 0.6485). Therefore, the average outcome did not differ significantly from zero ($p = 0.2068$). The studies appeared to be heterogeneous ($p < 0.0001$, $I^2 = 74\%$). An examination of the studentized residuals revealed that one study (Upadhyay et al.)[30] may be a potential outlier in the context of this model and, according to the Cook's distances, it could be considered to be overly influential. Neither the rank correlation nor the regression test indicated any funnel plot asymmetry ($p = 0.9195$ and $p = 0.4762$, respectively).

Figure 4. Forest plots of Shimmer.



Shimmer Random-Effects Model (k = 9) and Heterogeneity Statistics

	Estimate	se	Z	p	CI Lower Bound	CI Upper Bound	
Intercept	0.254	0.201	1.26	0.207	-0.140	0.648	
Tau	**Tau²**	**I²**	**H²**	**R²**	**df**	**Q**	**p**
0.510	0.2606 (SE=0.1809)	74.11%	3.863	-	8.000	33.892	< .001

Note. Tau² Estimator: Restricted Maximum-Likelihood

4. DISCUSSION

The auditory system is essential for the development and maintenance of voice quality and speech prosody. Correct rehabilitation of hearing loss, including CI in appropriate cases, can greatly improve the hearing function.[7] Nonetheless, subtle alterations can last and affect voice control through auditory feedback, as revealed by appropriate tests[11]. The aim of this study was to quantify alterations of voice acoustic parameters in children with CI by a systematic review and meta-analysis approach.

The overall quality of the 20 original included studies was affected by the absence of a Randomized Control Trial, given the presence of few longitudinal studies and the great majority of retrospective studies (see **Table 1**). Nonetheless, the quality of the available investigations was rated as moderate to high for more than half of the studies, according to the NOS score. One main limit found in many study designs was that few research groups reported hearing performance with CI (five out of twenty);[18,19,28,31] objective laryngeal evaluation (two out of twenty)[16,18] and hearing rehabilitation strategies (none of the included manuscripts).

In fourteen investigations,[16-18,19-26,28,30,31] the voice parameters were extracted from sustained /a/ vowel. This experimental condition, far from being ecological to real-life speech communication, allows fair reproducibility of results and was therefore included by the authors of this review as a necessary criterion for a study to be considered in the quantitative meta-analysis.

The F0, resembling the periodic spectro-acoustic structure of the voice, was reported to be altered in adult CI users in a previous systematic review on the subject.[11] The results of our systematic review confirmed that F0 was found higher in the pediatric CI users than in the control group in most of the analyzed investigations (75%), with a pooled mean difference of 0.30 (Conf. Inter. 0.06-0.55; p=0.014). Elevation of F0 in hearing impaired patients is a well-known phenomenon probably due to reduced ability to control laryngeal vertical position. This neuromuscular control deficiency, revealed by high posture of the larynx, is often associated with increase in phonation attempts, difficulty to control subglottic pressure and tension elevation during glottis cycle.[24] These issues are mainly restored by auditory feedback guaranteed with CI but, according to the reviewed literature of the past 20 years, some alterations in the pediatric population are still present and need further attention in future studies, clinical indication and rehabilitations programs.

Regarding noise measurements (jitter, shimmer and HNR), a tendency toward worse parameters in CI children, without reaching statistical significance, was revealed in the meta-analysis: average mean of 0.31 (Conf. Inter. from -0.19 to 0.80; p=0.223) for Jitter and 0.25 (Conf. Inter. from -0.14 to 0.65; p=0.207) for shimmer. For HNR the meta-analysis was not possible due to different methods of reporting values, anyway most of the studies (seven out of eight included) did not find significant differences between the CI groups and normal hearing ones.[17-19,22,23,27,32] All these latter voice parameters are often altered in the presence of functional and/or organic disorders of the vocal folds. Abnormalities of the larynx were ruled out by laryngoscopy only in two of the included studies,[16,18] therefore we can only presume the integrity of vocal folds in most of the included studies. A degraded auditory feedback with consequent overload at the level of the phonation organs and augmented risk of functional pathology could be considered as the basis of altered noise measures in CI patients. In this sense, a recently published retrospective analysis claimed a positive correlation between hearing loss and increased speech discrimination, with perception of increased voice handicap measured with the Voice Handicap Index (VHI-10).[36] The results of our investigation did not find significant values of noise in sustained /a/ vowel, which could therefore be an indirect confirmation of positive results in terms of functional outcome of CI.

Other alterations of vocal parameters were reported (see **table 3**), such as the variation of Mean Amplitude (vAM),[16,18,23,31] the variation of F0 (vF0)[18,31] and Formant 1-2,[33] confirming the persistence of some modification of values in CI patients compared to normal hearing volunteers in experimental settings. The three longitudinal studies[18,28,29] showed that during follow-up appointments the alteration of vocal parameters tended to normalize, including decrease of F0. The research groups attributed this to the effects of prolonged hearing rehabilitation but it is necessary to remark that the lowering of the F0 can also be the result of the natural laryngeal growth as an effect of age.[18]

In linguistics, the patterns of accent and intonation that are crucial to communicate meanings and emotions are defined as prosody. Unfortunately, there are no standardized and widely-used clinical tests to evaluate prosodic alteration, especially in a cross-cultural context.[27] Nonetheless, researchers have set up various experimental settings to test alteration in these supra-segmental aspects of language. The included studies mainly reported reduction of intonation range,[22,27] lack of precision of intonation[34] and augmented word duration[15] in CI patients compared to normal hearing subjects. Taken together, these findings could indicate a possible decreased efficacy of communication secondary to an alteration of prosody in the pediatric CI users.[22] Nonetheless, the latter findings are not conclusive and further studies are needed with solid and reproducible methods including both objective (e.g. spectro-acoustic and audiometric measures) and subjective (e.g. Patient Reporting Outcomes measures) evaluations in a multidisciplinary context.

Previous studies conducted on the adult population found alteration in nasality components of voice in CI patients, similar studies in the pediatric population have not been conducted to the best of our knowledge but could play a role in evaluating an aspect that expresses quality and voice pleasantness.[37]

One last consideration is needed for the three longitudinal studies that assessed vocal acoustic parameters with repeated measures over a period of 24 months after CI implantation.[17,28,29] In all these studies, statistically significant improvements of spectro-acoustic parameters at follow-up appointments were reported. Therefore, included research groups agreed that after cochlear implantation, prelingually deaf children established auditory feedback and improved voice control and vowel production over time; constant auditory experience with the CI brought most voice parameters closer to the norm.[17,28,29]

Voice quality as outcome of CI rehabilitation can be considered as a neglected outcome in common clinical practice[11], even though its evaluation has been included in recent international guidelines.[38]

The rationale of the inclusion of voice evaluation and rehabilitation programs in the clinical management of CI patients takes place in the recently emerging concepts of advanced rehabilitation in hearing impaired subjects.[39] This can be stressed even more considering the limits of the available auditory programs disclosed in recent systematic reviews[40] and the results of the present manuscript.

The results of the present systematic review have some limitations: (i) the low quality of the evidence available mainly consisting of observational studies that are known to be susceptible to publication bias, despite the absence of funnel plot asimmetry in the present meta-analysis; (ii) the absence of standardized methods of studying vocal and prosodic alterations which was responsible for retrieving speared different values; (iii) the inability to segregate males and females in each study. The latter limitation is partially overcome by the reduced effect of gender in voice in the considered samples of pediatric patients in the prepubertal age.[41,42]

CONCLUSIONS

The present meta-analysis confirmed that higher values of F0 have been observed in the pediatric population of CI users compared to age matched normal hearing volunteers, whereas the parameters of voice noise were not significantly different between cases and controls. Prosodic aspects of language need further investigations. In longitudinal contexts, prolonged auditory experience with the CI have brought voice parameters closer to the norm.

Following the available evidence, we stress the utility of inclusion of vocal acoustic analysis in the clinical evaluation and follow-up of CI patients with the aim of improving rehabilitation and the functional outcomes of pediatric patients with hearing loss.

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