



Speech in adults treated for unilateral cleft lip and palate as rated by naïve listeners, speech-language pathologists, and patients

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Panel assessment

Summary Speech may be affected in patients with cleft lip and palate (CLP). Professional listeners, naïve listeners, and patients may perceive speech differently. The aim of the study was to assess speech among adults treated for unilateral CLP (UCLP) as rated by naïve listeners, speech-language pathologists (SLPs), and self-assessment and to evaluate how well these ratings correlate.

All patients with complete UCLP treated at the Uppsala University Hospital, Uppsala, Sweden, in 1960–1987 were invited. A total of 73 of 109 patients (67%) participated, with a mean of 35 years since the initiation of treatment. The noncleft control group consisted of 55 volunteers. All participants answered questionnaires for self-rating of speech, and their speech was audio-recorded digitally. Fourteen naïve listeners and four SLPs rated the speech individually from blinded recordings.

There were more speech abnormalities among patients compared to controls according to the ratings of naïve listeners and SLPs. In controls and patients, there were positive correlations between the speech ratings by naïve listeners and SLPs $r = 0.44$ to 0.71 , $p < 0.001$, Spearman). The patients were less satisfied and rated to have more speech abnormalities than controls ($p < 0.001$).

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Although adults treated for UCLP considered their speech as fairly good, they were less satisfied than noncleft controls. The agreement between ratings by naïve listeners and SLPs were good, while the agreement between these ratings and self-assessment varied widely. When assessing speech in adults treated for UCLP, differences in perception of speech abnormalities by professionals, laymen, and patients should be considered.

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Introduction

The quality of a person's speech is essential for verbal communication. Speech may be affected in patients with cleft lip and palate (CLP) and, as a consequence, may influence their quality of life (QoL).¹ Speech is one of the main outcome measures in the treatment of CLP.^{2,3} The way we speak determines how we are perceived and judged. It may affect interpersonal and social encounters, occupational possibilities, and QoL. A judgment of the personality and capabilities of a person can be formed after listening to 30 s of a voice recording.⁴ Hypernasality is one of the most characteristic speech deviations in individuals treated for CLP. Negative attitudes toward speakers with hypernasal resonance^{5,6} and other speech disorders⁷ have been reported. Previous studies report speech as the reason for being bullied in about one-third of bullied/teased children treated for CLP.^{8,9}

It is important that the outcome measures used in cleft care are representative, valid, and reproducible. Cleft palate speech is usually evaluated by speech-language pathologists (SLPs).¹⁰ These professionals are trained to identify specific speech abnormalities related to the cleft. However, it is unclear how well these assessments represent the people with whom the patients interact with in their daily life. Assessments of naïve listeners have been suggested to add validity to the traditional speech assessments of SLPs.^{11,12}

Several previous studies have evaluated cleft palate speech with naïve listeners and compared it to ratings of SLPs.^{11–18} However, there are no previous studies on the speech among adults treated for UCLP as rated by naïve listeners, SLPs, and the patients themselves.

The current study aims to assess speech among adults treated for UCLP as rated by naïve listeners, SLPs, and self-assessment and to evaluate how well these ratings correlate. The hypothesis is that the speech is assessed differently by naïve listeners, SLPs, and patients and that naïve listeners and self-ratings would correlate the most. Furthermore, the current study aims to assess the level of satisfaction/dissatisfaction with speech among adults treated for UCLP compared to adults with no cleft.

Materials and methods

Participants

All consecutive patients, born 1960–1987 with complete UCLP treated by the cleft-palate team at the Uppsala University Hospital, Uppsala, Sweden, were invited to partici-

pate in the study. Patients with incomplete clefts of the lip or palate and Simonart's band and/or syndromes were excluded. A total of 109 patients met the inclusion criteria, and 73 of them participated in the study (67%). The reasons for not participating were “do not have time/cannot get time off work”, “do not want to,” or “no reason”/did not answer the invitation. The mean follow-up time was 35 years (range 20–47 years) from the first operation to the participation date of the current study. An age and gender matched noncleft control group ($n = 55$) was recruited among employees of the hospital and students. The control and patient groups underwent the same assessments. The patients who participated in the study and the surgical methods have been described^{22,23} and²⁴. In short, the cleft palate had been repaired in one stage according to Veau and Wardill, as modified by Skoog, in patients born between 1960 and 1975.²⁵ For patients born between 1976 and 1987, a two-stage procedure had been performed with soft palate closure at the first stage, followed by closure of the residual cleft of the hard palate at the second stage.²³ Pharyngeal flap surgery had been performed on 11 of 73 patients (a total of 15%; 17% of one-stage and 12% of two-stage patients). Ninety-five percent of all the surgical procedures were performed by four different surgeons.

Audio recordings of speech

Speech was recorded during an outpatient visit that was especially arranged for the study. The recordings were made in a soundproof studio equipped with a microphone (Sennheiser mke 2-p, Sennheiser, Wedemark, Germany) at a constant distance from the mouth. A digital recording system (VoiceJournal™, Neovius Data och Signalsystem AB, Stockholm, Sweden) was used for the audio recordings in the study. At one time during the study, there were some technical problems with the studio, therefore eight of the recordings were made in a non-soundproof room.

The speech recordings consisted of the following three parts: 1: Sentences from the Swedish Articulation and Nasality Test (SVANTE) with six sentences including high pressure oral consonants, four sentences containing nasal and oral consonants, and one with only nasal consonants.²⁶ 2: A story (“The boy who wanted to borrow a spade”). 3: The story, retold with own words. A key for coding and randomization of the recordings was provided by a statistician. A person, who was not one of the raters, gave each recording (audio files) a nonidentifiable study-ID and transferred the audio files to a compact disk (CD) for computers in a randomized order. The CDs were used for the SLPs' speech ratings. The audio files were transferred to one USB memory stick for each naïve listener, with the order of the audio files being the same as

Table 1 Naïve listeners - age (years), sex and occupation.

ID	Age	Sex	Occupation
A	49	F	Pre-school teacher
B	32	F	Nanny
C	59	F	Pre-school teacher
D	31	F	Pre-school teacher
E	42	F	Student
F	71	F	Retired
G	13	F	Student
H	11	M	Student
I	33	M	Train conductor
J	35	F	Student
K	20	F	Student
L	31	F	Medical doctor
M	44	M	Medical doctor
N	19	M	Student

on the CD. Ten (14%) of the audio recordings of patients and five (9%) of the noncleft group were duplicated to make calculation of intra-rater agreement possible. The duplicated recordings were not directly adjacent to each other in the list of sound files on the USB sticks.

Assessment of speech by naïve listeners

Fourteen listeners without any listening training or education in assessing or rating cleft speech were recruited for the study through the authors (Table 1). Each listener received a USB memory stick containing the speech recordings and was instructed to listen to them with headphones connected to a computer in a silent environment and rate them one by one. Replay of any recording was allowed. The questions for speech assessment by naïve listeners were selected to match the SLPs speech rating format for comparison. The questions were chosen from the questionnaire of Brunnegard and co-workers¹⁸ (Table 2). The listeners were informed that “speak through the nose” can also mean “stuffy/obstructed nose” in this context and therefore represent both hypernasality and hyponasality in this context. The listeners received a small gift of appreciation of approximately 30 Euro.

Perceptual assessment of speech by SLPs

Four SLPs experienced in working with cleft palate teams assessed the recordings. Two of the SLPs worked at the Uppsala University Hospital, and the other two worked at other university hospitals. All SLPs listened to the recordings individually with headphones connected to a computer. The speech assessments performed by SLPs have been published previously.²² The following variables rated by SLPs were included in the current study and were rated on five point scales (Table 3). Grades 0 and 1 were considered acceptable. The median ratings of the four SLPs were used in the current study for analysis. The median ratings of the four SLPs, rounded to the closest integer scale step, were used for presentation in Figure 1.

Self-rating of speech by patients and controls

At the time of the speech recording, the participants answered a questionnaire regarding their own speech including the questions listed in Table 4. General data on patients' age, asthma/allergy, medication, smoking habits, etc., were retrieved from the dataset and patient notes and have been presented in previous studies.^{22,24,27}

Statistics

The computer software IBM Statistical Package for the Social Sciences statistics 24.0 (IBM Corporation, Route 100, Somers, NY 10589, USA) was used for statistical analysis. The Mann-Whitney U test was used to compare differences between ratings of speech between patients and controls, and the Wilcoxon-signed rank test was used to compare differences between naïve listeners' and SLPs' ratings. The McNemar's test was used to listener classification of a subject as a patient or a control. The level of significance was set at $p < 0.05$. Spearman's rank correlation was used for the calculation of correlations between the ratings of naïve listeners, SLPs, and self-ratings. Intraclass Correlation Coefficient (ICC) was applied for the calculation of inter-rater agreement and intra-rater agreement among listeners. The ICC was set to a mixed two-way model, consistency, and average measures. The cut-offs for inter-rater agreement

Table 2 Questions and the alternative answers included in the questionnaire used (Brunnegard and co-workers).

	0	1	2	3	4
Do you think the person speaks through the nose?	No	Yes, slightly	Yes a little	Yes, moderately	Yes, very much
“Does it sound like airflow noise/snort from the nose	No	Yes, once or twice	Yes, some occurrences	Yes, frequently occurring	Yes, always occurring
Do you notice any deviations in the articulation	No	Yes, once or twice	Yes, some occurrences	Yes, frequently occurring	Yes, always occurring
Is the speech intelligible	Normal	Mildly reduced	Much reduced		
Do you think the recording comes from an individual with cleft?;	Yes	No			

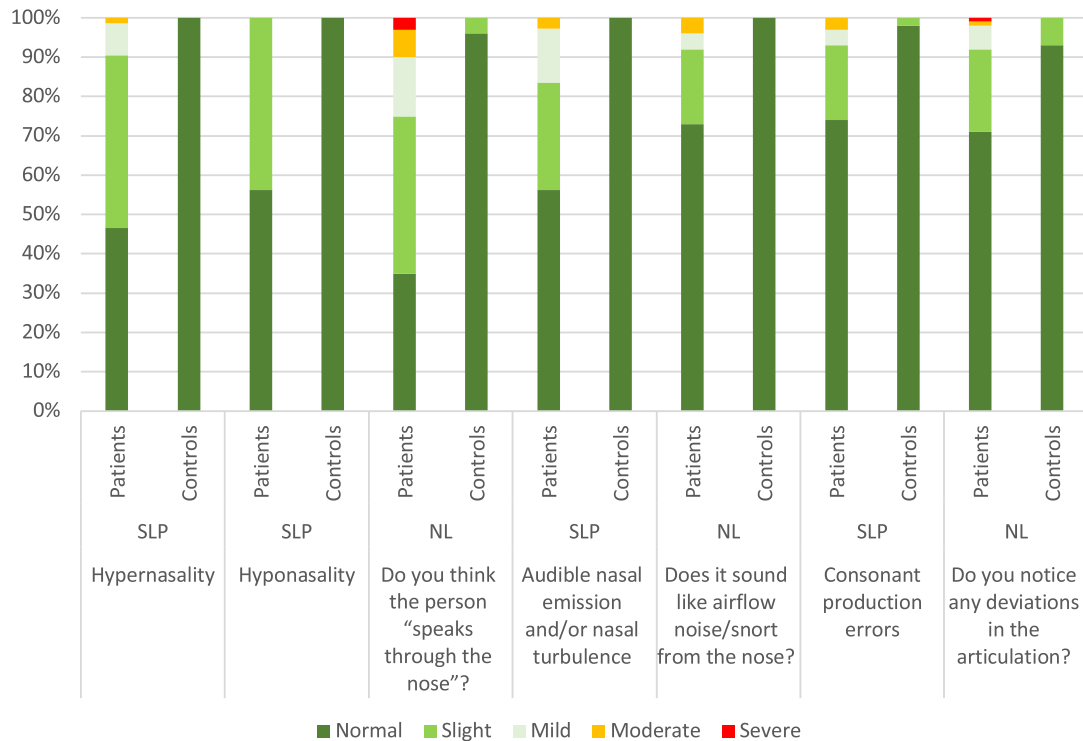


Figure 1 Ratings of speech in patients treated for UCLP by naïve listeners and SLPs.

Ratings of speech in 73 patients treated for UCLP and 55 non-cleft controls by 14 naïve listeners and 4 SLPs. Ratings on a five-point scale. Median values of all raters in each group respectively for every subject rounded to integers. There were significant differences between the ratings in patients and controls for all variables ($p < 0.001$, Mann Whitney U test). SLP- speech-language pathologists, NL- naïve listeners.

Table 3 Variables rated by SLPs included in the current study (rated on five point scales).

	0	1	2	3	4
Hypernasality or hyponasality	Normal	Slight	Mild	Moderate	Severe
Audible nasal emission and/or nasal turbulence and consonant production errors	Absent	Single occurrences	Some occurrences	Frequently occurring	Always occurring
Intelligibility	Normal	Mildly-moderately reduced	Severely reduced		
Do you think the recording comes from an individual with cleft?	No	Yes			

Table 4 Questions used for Self-rating of speech by patients and controls.

Questions:
Do you speak through your nose (leak)? (represents hypernasality and nasal airflow combined) (YES/NO)
Do you sound like you have a congested nose? (represents hyponasality) (YES/NO)
Do you have difficulties to articulate certain speech sounds? (YES/NO)
Do other people understand what you say? (YES/NO)
How satisfied are you with your speech? The questions were answered on a 100 mm visual analogue scale (VAS).
Do you experience your speech as abnormal? (YES/NO)

Table 5 Median ratings of Naïve listeners compared to SLPs with Spearman's rank correlation coefficient.

Naïve listeners variables	Speech-Language Pathologist variables	<i>r</i>	<i>p</i>
Do you think the person "speaks through the nose"?	Hypernasality	0.69	<0.001
Do you think the person "speaks through the nose"?	Hyponasality	0.58	<0.001
Does it sound like airflow noise from the nose?	Audible nasal emission and/or nasal turbulence	0.71	<0.001
Do you notice any deviations in the articulation?	Consonant production errors	0.52	<0.001
Is the speech understandable?	Intelligibility	0.44	<0.001

r = Spearman's rank correlation coefficient.

Table 6 Correlations between rating by naïve listeners and patients and controls self-ratings with Spearman's rank correlation coefficient.

Naïve listeners variables	Self-ratings variables	<i>r</i>	<i>p</i>
Do you think the person "speaks through the nose"?	Do you sound like you speak through your nose?	0.40	<0.001
Does it sound like airflow noise from the nose?	Do you sound like you speak through your nose?	0.34	<0.001
Do you think the person "speaks through the nose"?	Do you sound like you have a congested nose?	0.30	0.001
Do you notice any deviations in the articulation?	Do you have difficulties to articulate certain speech sounds?	0.13	0.16
Is the speech understandable?	Do other people understand what you say?	0.10	0.27

r = Spearman's rank correlation coefficient.

measures by ICC according to Cicchetti²⁸ are as follows: ICC <0.40 equals poor, ICC between 0.40 and 0.59 equals fair, ICC between 0.60 and 0.74 equals good, and 0.75-1.00 equals excellent.

Results

Inter- and intra-rater agreement

The inter-rater agreement for naïve listeners as ICC average measures ranged between 0.83 and 0.91. The inter-rater agreement for SLPs ICC average measures ranged between 0.65 and 0.89. The intra-rater agreement for naïve listeners as ICC in all five variables combined ranged from 0.3 to 1.0, and the mean values for the intra-rater agreement for each variable ranged from 0.5 to 0.7.

Correlations and differences between naïve listeners, SLPs, and self-ratings

The correlations between the ratings of naïve listeners and SLPs as Spearman's rank correlation coefficient ranged between 0.44 and 0.71 (Table 5). The correlations between ratings of naïve listeners and self-ratings as Spearman's rank correlation coefficient had a range from 0.10 to 0.40 (Table 6). The Spearman's rank correlation coefficient between ratings of SLP and self-ratings as had a range of -0.10 and 0.41 (Table 7). There were statistically significant differences between the ratings by naïve listeners and SLPs for all of the compared speech variables ($p = 0.039$ to $p < 0.001$, Wilcoxon-signed rank test).

Ratings of speech by naïve listeners and SLPs - patient vs. controls

The ratings of speech by naïve listeners and SLPs are shown in Figure 1. There were significant differences between the ratings of speech for patients and controls for all speech variables, as rated by naïve listeners and by SLPs ($p < 0.001$, Mann-Whitney U test). Intelligibility was rated to be mildly reduced in five patients by naïve listeners and mildly reduced in one patient by SLPs. None of the controls were rated to have "reduced intelligibility". Naïve listeners were able to correctly identify whether the recording was from a UCLP patient 28% of the time. The SLPs rated 56 (77%) of the patients' speech recordings as coming from a patient and 2 (4%) of the controls' speech recordings as coming from a patient. The difference was significant ($p < 0.001$, McNemar's test).

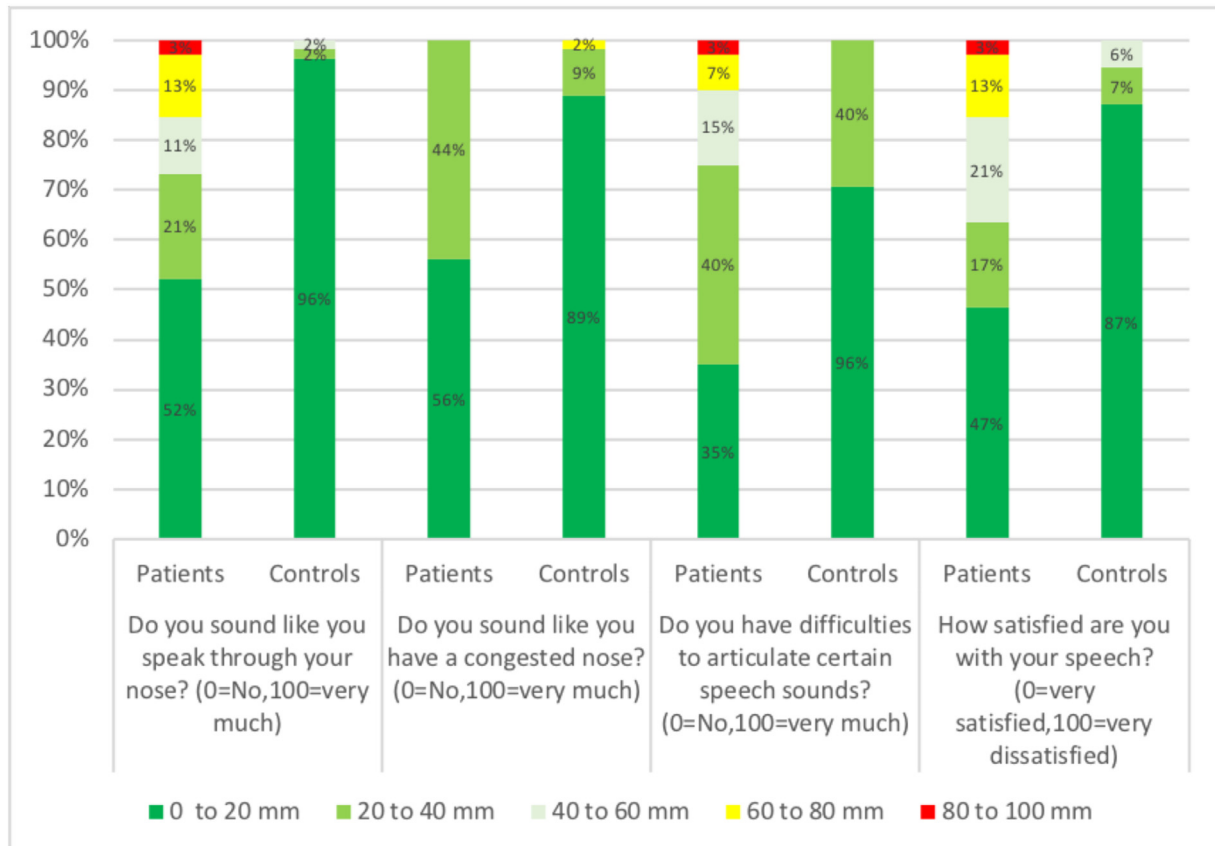
Self-ratings of speech by patients and controls

The self-ratings of speech by patients and controls are shown in Figure 2. There were significant differences between patients and controls for all the questions rated on VAS ($p < 0.001$, Mann-Whitney U test). The median rating for satisfaction with speech was 23 (interquartile range: 41) in patients and 3.5 (interquartile range: 12) in controls on the 100 mm VAS (0 = very satisfied, 100 = very dissatisfied). On the question: "Do you experience your speech as abnormal?" 16/70 (23%) patients answered "yes", and three of the patient answers were missing. None of the controls answered "yes" to this question.

Table 7 Correlations between rating by SLPs and patients' and controls' self-ratings, Spearman's rank correlation coefficient.

Speech-Language Pathologist variables	Self-ratings variables	r	p
Hypernasality	Do you sound like you speak through your nose?	0.41	<0.001
Hyponasality	Do you sound like you have a congested nose?	0.30	0.001
Audible nasal emission and/or nasal turbulence	Do you sound like you speak through your nose?	0.40	<0.001
Consonant production errors	Do you have difficulties to articulate certain speech sounds?	0.23	0.009
Intelligibility	Do other people understand what you say?	- 0.10	0.25

r = Spearman's rank correlation coefficient.

**Figure 2** Self-ratings of speech in patients and controls.

The figure shows the percentage of ratings on VAS within the following intervals: 0 to 20 mm, 20 to 40 mm, 40 to 60 mm, 60 to 80 mm and 80 to 100. VAS 100 mm (0 mm = Not at all, 100 mm = very much for all variables except “How satisfied are you with your speech”: 0 = very satisfied, 100 = very dissatisfied). There were significant differences between patients and controls for all the variables ($p < 0.001$, Mann-Whitney U test).

Discussion

The current study assessed speech among 73 adult patients treated for UCLP and 55 noncleft controls by 14 naïve listeners, 4 SLPs, and self-assessment. The possible correlations between the ratings of these different groups of raters were investigated. There were differences between ratings of naïve listeners and SLPs for all rated variables, and there were positive correlations between almost all combinations of ratings. In contrast to what was hypothesized, correlations were stronger between naïve listeners and SLPs as compared to correlations between self-ratings and ratings by the other groups. Self-ratings of satisfaction with speech were lower among patients than in controls. To the best of

our knowledge, the current study is the first study to compare speech assessments performed on adults treated for UCLP by naïve listeners, SLPs, and self-ratings by patients, and comparing the findings to a noncleft control group.

The assessment by SLPs of speech in adults treated for UCLP has previously been reported as good/fair for both one- and two-stage palate closure.²² In the current study, the patients were also assessed by naïve listeners who represent as people with whom the patients would interact in their daily lives. Naïve listeners were able to differentiate speech between cleft patients and noncleft controls, suggesting that the general population are able to discern cleft-related speech differences. On the other hand, only 38% of the cleft patients were identified as “cleft” by naïve listen-

ers, indicating that the cleft treatment is successful, as for 62% of the patients, where a cleft diagnoses was not picked up based on speech alone.

Differences in the ratings of speech performed by naïve listeners and SLPs were expected due to a difference in knowledge and experience of evaluating the characteristics of cleft palate speech. As an example, naïve listeners did use the highest score for “speak through the nose,” while the SLPs never used the highest score for “hypernasality”. This is in accordance with a study by Lewis and co-workers, where untrained listeners rated hypernasality higher than SLPs¹⁵ and may be explained by SLPs having more experience with the full spectrum of hypernasality. It is interesting to note that, even though the naïve listeners used the higher rating scores for “speak through the nose”, naïve listeners rated fewer of the patients’ recordings to actually come from a patient than SLPs. A reason for this may be that SLPs may perceive the more subtle speech abnormalities typical to the CLP population, and the naïve listener not necessarily contributing “speaking through the nose” to a cleft associated speech abnormality.

The positive correlation between ratings by naïve listeners and SLPs in the current study is in accordance with some previous studies that used assessments made by naïve listeners.^{11,16–18} In a study conducted by Starr and co-workers, speech of children with cleft palate was rated on 8-point equal appearing interval scales for the variables nasality and articulation by experts and untrained listeners.¹⁶ They found that ratings did not differ extensively between raters, but expert listeners were better at differentiating between nasality and articulation than naïve listeners. Two studies conducted by Witt and co-workers used different scales and variable analysis for naïve listeners and SLPs, complicating any comparison.^{13,14} Brunnegard and co-workers used speech variables for naïve listeners that were related to the variables nasality, nasal emission, and articulation rated on ordinal 5 point scales and a question about the need for speech therapy.¹⁸ It was found that untrained listeners’ ratings of speech in 10-year-old patients mainly confirmed the ratings performed by SLPs. This was also the method chosen in the current study to facilitate comparison. Nyberg and Havstam investigated how peers described the speech of 10-year-old children treated for cleft palate.¹⁷ In their study, SLPs and peers did not agree on minor signs of VPI, but they agreed on what was normal and severely impaired speech, respectively. This is in contrast to the study by Witt and co-workers found that naïve raters to be insensible to differences between the speech of cleft patients and controls.¹³ In the current study, the correlation between the ratings of the naïve listeners and the SLPs indicates that the experts’ assessments of speech may also be representative of the people the patients meet in their daily lives.

The correlations between self-ratings of speech and SLPs ratings were low in the current study. This is in concordance with earlier studies on self-ratings and experts ratings for cleft populations.^{2,29} In the current study, patients rated their satisfaction with speech at levels comparable to previous studies of patients treated for CLP.^{1,2,8,19,20,21} Additionally, patients rated lower on satisfaction with speech than noncleft people. This finding was similarly reported in a study by Hunt and co-workers, who reported on psychosocial functioning and self-ratings, including speech ratings,

in 8 to 21-year-old individuals treated for CL/P and a control group.⁸ In that study, 18% of the individuals treated for cleft and 4% of the noncleft individuals were dissatisfied with their speech. Contrasting results were presented in a study by Van Lierde and co-workers.¹⁹ They found that self-reported and parental satisfaction with speech was similar between 11 and 17-year-olds treated for UCLP ($n = 43$) and a control group ($n = 43$). Similarly, Oosterkamp and co-workers found no differences in quantitative measures of satisfaction with speech between patients and controls, but the patients expressed more concerns about their speech.²¹ Self-reported outcomes are considered important as these are known to be associated to QoL.^{1,30} Bickham and co-workers studied speech and QoL in 108 children treated for CP±L (age 5 to 19 years).¹ About 30% of the patients and parents expressed difficulties with their child’s speech. The same study showed that children treated for CLP who perceived more difficulty with speech also experienced worse scores on depression scales than those with less speech impairment. Similarly previous studies have shown correlation between satisfaction with speech and psychosocial difficulties among children treated for cleft malformation.³¹ Speech is one of the most important means to communicate, and one’s self-perception of speech may affect the ability and self-confidence to speak. As such, self-reported speech outcome could also be used as a basis for a dialog between the caregiver and the patient about the possibility of additional interventions for adult patients treated for UCLP.

The current study has certain limitations. Gender was not evenly distributed within the groups of raters, with the majority being female, though whether this affects the ratings is unknown. Secondly, rating forms used by the naïve listener and the questionnaires answered by the patients were not previously validated but were developed for the current study. The naïve listeners and the SLPs did not have any calibration or training sessions prior to the ratings. Calibration or training of listeners can increase rater agreement.³² For some of the variables and naïve listeners, an intra-rater reliability could not be calculated due to the absence of variability. This could have been avoided if more of the recordings had been rated twice by the raters, in addition to the 15 duplicated recordings in this study. The possible effect of the order of recordings presented to listeners was not evaluated; however, a previous study by Brunnegard and co-workers found that the order of recordings did not change the ratings significantly.¹⁸

It is challenging to compare assessments made by different groups of raters when the outcome cannot be on identical scales. In this study, we aimed to use similar variables assessing the speech outcomes for each group of raters. To achieve comparable rating scales, a translation of the professional terms and conditions was made into phrases expressing the conditions in laymen terms, as in the study by Brunnegard and co-workers.¹⁸

To accurately compare speech ratings of different groups of raters using different scales, the speech material should ideally include the full range of the rating-scale and an even distribution of different extents of speech abnormality, like in the study by Castick and co-workers.³³ This was not completely the case in the current study, where speech outcomes from consecutive cases of UCLP were evaluated, and it may have affected the ratings by naïve listeners. How-

ever, the current study had a relatively large group of study participants, which should partly compensate for this.

Conclusion

Although adult patients treated for UCLP during childhood considered their speech as fairly good, they were less satisfied than noncleft controls. The agreement between ratings by naïve listeners and SLPs were good, while the agreement between these ratings and self-assessment of speech varied widely. Differences in perception of speech abnormalities by professionals, laymen, and patients should be taken into account when assessing treatment outcomes in adult patients treated for UCLP.

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Declaration of Competing Interest

None declared.

Ethical approval

All participants gave their informed consent to the study. The study was approved by the Research Ethics Committee of Uppsala University (Reference number 2005:245).

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