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A non-language-specific speech test to evaluate the speech of cleft patients from different language and cultural backgrounds – A pilot study

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ABSTRACT

Cleft speech tests are not universally available. We developed a tool to fill this gap, especially in the context of a cleft mission setting. We performed a pilot study to evaluate the test's ability to differentiate between the speech of cleft patients and healthy individuals from three different language backgrounds.

We used 78 made-up, nonsensical syllables to evaluate hypernasality, nasal emissions, and consonant errors. Cleft ($n = 41$) and non-cleft ($n = 39$) individuals from three countries were included in this study. Two speech and language pathologists, blinded to the examination, rated the audio recording independently.

Patients from Germany ($n = 12$; mean age 15.2), Iran ($n = 14$; mean age 7), and India ($n = 15$; mean age 14.7 years) were evaluated. We observed a significant difference in each category ($p < 0.05$) between patients and control subjects of the same language and cultural background. Hypernasality was affected the most.

The test proved to possess the correct phonetic characteristics to reveal and provoke relevant cleft speech pathologies independent of cultural and language backgrounds. The test sounds posed no articulatory difficulties to non-cleft individuals, with some exceptions regarding non-specific consonant errors. A comparison with other existing tests will further illuminate its value as a speech test.

1. Introduction

Cleft lip and palate is a complex condition affecting patients in different ways (Shaw et al., 2001). Cleft palate can result in significant anatomical and functional changes to the vocal tract, influencing speech production (Peterson-Falzone et al., 2001). One of the major treatment goals of modern cleft care is to achieve acceptable speech production (Bessell et al., 2013).

The assessment of a patient's speech plays an important role when evaluating the impact of different treatment strategies or the need for additional procedures. The spectrum of speech evaluation methods is broadly divided into non-invasive and invasive techniques. Nasometry, videofluoroscopy, and nasopharyngoscopy are effective but invasive techniques, and may necessitate exposure to

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radiation (Kummer, 2014). The perceptual assessment of speech is considered to be the gold standard, but requires complex and detailed speech evaluation tests that are not available in every setting, especially in developing countries (Sell, 2005). Our aim of offering high-quality cleft care in a cleft mission setting is limited by the language barrier, resulting in the need to develop a method for evaluating speech outcome independent of language. A test based on non-language-specific sounds allows speech evaluation in different regions with different language and cultural backgrounds while using the same set of test sounds. Henningsson et al. (2008) proposed a standardised method to evaluate speech in cleft patients by considering the aspects of hypernasality, hyponasality, nasal emissions and turbulence, consonant production errors, and voice disorder. Tests like the Swedish SVANTE (Klinto et al., 2011) or the British GOS.SP.ASS.'98 (Sell et al., 1999) make use of language-specific words and sentences selected to test the patient's speech abilities. Independent of the language, the test sound inventory is always based on the same specific phonetic characteristics to examine the patient's velopharyngeal function and competency (Brondsted et al., 1994; Kummer, 2013).

Our aim was to develop a non-invasive test based on made-up, nonsensical syllables to evaluate the speech of cleft patients without taking into account their cultural and language backgrounds. We conducted a pilot study to analyze the test's phonetic characteristics with respect to its ability to reveal and provoke the relevant cleft speech pathologies.

2. Material and methods

The ethics committee of RWTH Aachen University approved the study, which was conducted in accordance with the principles of the Helsinki declaration.

We examined 41 cleft patients and 39 healthy non-cleft individuals from Germany, Iran, and India (Table 1). The cleft patients varied with regard to the degree and extent of the cleft condition (Table 2). Syndromic patients or individuals with a history of hearing difficulties were excluded. The test included 78 test items, allowing the assessment of vowels and consonants in the initial, medial, and final sound position (Table 3). We devised a simple, easily accessible, and understandable binary scoring system. The rater evaluated every syllable in terms of signs of hypernasality, nasal emissions, and consonant errors. For each conspicuous sound, one point was given in the respective category; normal findings were rated with zero, resulting in a maximum of 78 points in each category and a possible 234 points in total. One senior surgeon and one senior medical student conducted the test. All test sounds were elicited by repetition using a standardised pattern. The session was digitally recorded using a microphone (SONY — Minato, Tokio, Japan — ECM-MS957) and an MD recorder (SONY — Minato, Tokio, Japan — MZ-B100). Syllables were repeated in case the test individual initially had problems understanding the examiner. Two speech pathologists, having previously received instructions on the study and the rating system, were blinded to the examination and evaluated the audio files independently. Both raters repeated the rating of eight randomly selected files 6 months after the initial scoring to assess intra-rater reliability. An exemplary calculation was performed for inter-rater reliability using the data from the German group.

2.1. Statistical analysis

The score totals are presented in a descriptive manner by median, 25%-quantile (Q1), 75%-quantile (Q3), minimum, and maximum. We conducted non-parametric Wilcoxon tests to compare speech scores, instead of reporting confidence intervals. One-sided Wilcoxon rank sum tests (unpaired) were performed to compare cleft patients with non-cleft individuals due to the expected higher scores of cleft patients. To compare the scores within the same patient group with regard to intra-rater reliability, we used a Wilcoxon signed rank test (paired, two-sided). We used exploratory data analysis and the p-values ≤ 0.05 were interpreted as statistically significant. To assess the degree of agreement, Lin's concordance coefficient (LCC) was determined and $\rho > 0.9$ was interpreted as concordance between the raters. The LCC takes the data variation (precision) as well as the distance from concordance (bisection line) into the account (accuracy). The precision of the agreement is reflected by the Pearson correlation coefficient (PCC), which is a factor of LCC. All statistical analyses were conducted using the SAS statistical analysis software package (SAS version 9.4; SAS Institute, Cary, NC, USA).

3. Results

The results of the statistical analyses of the points given by raters are presented in Table 4, divided by groups and countries. We observed significant differences between patients and control subjects for every category and language, represented by the Wilcoxon rank sum test ($p < 0.05$). It should be noted that the majority of errors in the control groups were registered in the category of consonant errors.

The results for inter-rater agreement are presented in Table 5. We selected the ratings for the German-speaking test individuals. While the precision shown by the PCC is acceptable, the LCC suggests only poor levels of agreement. The levels of percentage of identical scoring, on the other hand, suggest better levels of agreement.

Table 1
Demographic information representing gender and mean age (min–max).

	Germany ($n = 27$)	Iran ($n = 27$)	India ($n = 26$)
Patients ($n = 41$)	$n = 12$	$n = 14$	$n = 15$
M/F, mean (min–max)	6/6, 15.2 (5–33)	8/6, 7 (4–9)	11/4, 14.7 (5–24)
Controls ($n = 39$)	$n = 15$	$n = 13$	$n = 11$
M/F, mean (min–max)	9/6, 15.8 (5–58)	3/10, 7 (4–12)	5/6, 16.2 (5–30)
CP	4	7 (one with fistula)	2 (one with fistula)
UCLP	5	4 (one with fistula)	10 (three with fistula)
BCLP	3 (one with fistula)	3	2 (two with fistula)
Cleft lip[a]	–	–	1

F: female.
M: male.
CP: cleft palate.
UCLP: unilateral cleft lip and palate.
BCLP: bilateral cleft lip and palate.
[a] Due to incomplete clinical documentation during patient recruitment, this patient was suspected of suffering from UCLP, which later turned out be an isolated cleft lip.

Table 2
Detailed information on gender, age and cleft condition.

Germany ($n = 27$)					Iran ($n = 27$)					India ($n = 26$)				
Controls $n = 15$		Patients $n = 12$			Controls $n = 13$		Patients $n = 14$			Controls $n = 11$		Patients $n = 15$		
Sex	Age	Sex	Age	Cleft	Sex	Age	Sex	Age	Cleft	Sex	Age	Sex	Age	Cleft
M	5	M	5	CP	M	4	F	4	CP f	M	5	M	5	BCLP
F	5	F	5	CP	M	5	M	5	UCLP	F	8	M	7	BCLP f
M	5	F	7	UCLP	F	6	M	5	BCLP	F	12	M	7	UCLP
F	5	M	7	BCLP f	F	7	F	6	CP	F	13	M	8	UCLP f
M	6	F	7	CP	F	7	M	6	CP	M	13	F	11	CP
M	6	M	14	UCLP	F	7	M	6	CP	M	13	F	14	UCLP f
M	6	F	17	CP	F	8	M	6	UCLP	M	17	F	15	Lip[a]
F	7	M	20	BCLP	F	8	F	7	CP	F	21	M	16	UCLP
F	7	F	21	UCLP	F	8	F	7	CP	F	22	M	17	UCLP
M	8	M	22	BCLP	F	8	F	7	CP	F	24	M	18	UCLP
M	18	F	25	UCLP	F	9	M	7	BCLP	M	30	M	18	UCLP
M	22	M	33	UCLP	F	9	M	7	BCLP			M	18	UCLP
F	24				M	9	M	8	UCLP			M	20	UCLP f
M	56						F	12	UCLP f			M	22	UCLP f
F	58											F	24	CP f

F: female.
M: male.
CP: cleft palate.
UCLP: unilateral cleft lip and palate.
BCLP: bilateral cleft lip and palate.
f: fistula.
[a] Due to incomplete clinical documentation during the patient recruitment, this patient was suspected of suffering from UCLP, which later turned out be an isolated cleft lip.

Table 3
Test sound inventory used in the study.

Test sound	Phonetic transcription according to IPA	Test sound	Phonetic transcription according to IPA	Test sound	Phonetic transcription according to IPA	Test sound	Phonetic transcription according to IPA
pu	[pu:]	luk	[lu:k]	idi	[i:di:]	su	[su:]
upu	[u:pu:]	ki	[ki:]	lid	[li:d]	usu	[u:su:]
lup	[lu:p]	iki	[i:ki:]	da	[da:]	lus	[lu:s]
pi	[pi:]	lik	[li:k]	ada	[a:da:]	si	[si:]
ipi	[i:pi:]	ka	[ka:]	lad	[la:d]	isi	[i:si:]
lip	[li:p]	aka	[a:ka:]	gu	[gu:]	lis	[li:s]
pa	[pa:]	lak	[la:k]	ugu	[u:gu:]	schu	[ʃu:]
apa	[a:pa:]	bu	[bu:]	lug	[lu:g]	uschu	[u:ʃu:]
lap	[la:p]	ubu	[u:bu:]	gi	[gi:]	lusch	[lu:ʃ]
tu	[tu:]	lub	[lu:b]	igi	[i:gi:]	schi	[ʃi:]
utu	[u:tu:]	bi	[bi:]	lig	[li:g]	ischi	[i:ʃi:]
lut	[lu:t]	ibi	[i:bi:]	ga	[ga:]	lisch	[li:ʃ]
ti	[ti:]	lib	[li:b]	aga	[a:ga:]	nu	[nu:]
iti	[i:ti:]	ba	[ba:]	lag	[la:g]	unu	[u:nu:]
lit	[li:t]	aba	[a:ba:]	fu	[fu:]	lun	[lu:n]
ta	[ta:]	lab	[la:b]	ufu	[u:fu:]	mi	[mi:]
ata	[a:ta:]	du	[du:]	luf	[lu:f]	imi	[i:mi:]
lat	[la:t]	udu	[u:du:]	fi	[fi:]	lim	[li:m]
ku	[ku:]	lud	[lu:d]	ifi	[i:fi:]		
uku	[u:ku:]	di	[di:]	lif	[li:f]		

Regarding the intra-rater agreement, Table 5 shows that rater A achieved appropriate levels of agreement, whilst rater B struggled to reach acceptable scores on the LCC. Nevertheless, according to the Wilcoxon signed rank sum test, the difference in both rating cycles was not significant.

4. Discussion

The purpose of this pilot study was to evaluate whether a non-language-specific set of syllables and sounds could be used to detect mistakes specific to cleft speech and articulatory errors, independent of the spoken language. The age differences between the enrolled individuals from the three countries was not the primary interest of our study, as we did not aim to compare the results of the different countries. Additionally, we had a limited amount of time in each country to match the patients and control groups in terms of age, cleft deformities, and gender.

The test sound inventory was based on recommendations found in established publications (Henningsson et al., 2008; Lohmander et al., 2009; Kummer, 2013). We carefully selected certain high vowels and high-pressure consonants that stress the velopharyngeal sphincter and therefore lend themselves to assessing cleft speech. We focused on the high vowels/u/and/i/to assess the degree of hypernasality. They phonate for a fairly long time and cannot be substituted in most cases. They require a high position of the tongue to increase the oral sound pressure that provokes nasal escape and improper nasal resonance. To keep the number of test items manageable, the low vowel/a/was only used in context with the plosive sounds. The selected obstruents (/p/, /t/, /k/, /d/, /g/, /b/) were used to assess the degree of nasal turbulence and emissions

Table 4
Score comparison between patient and control groups.

	Germany[a]		Iran		India	
	Patients	Controls	Patients	Controls	Patients	Controls
Hypernasality						
median (Q1; Q3), min–max	2.0 (0.3; 49), 0–70.0 $p = 0.0003$*	All: 0	18.5 (0; 68), 0–75 $p = 0.0055$*	0 (0; 0), 0–4	23 (0; 59), 0–66 $p = 0.0064$*	0 (0; 0), 0–6
Nasal emissions						
median (Q1; Q3), min–max	0.0 (0; 12.8), 0–31.5 $p = 0.0065$*	All: 0	2.5 (0; 11), 0–22 $p = 0.0023$*	All: 0	4 (0; 20), 0–40 $p = 0.0031$*	All: 0
Consonant errors						
median (Q1; Q3), min–max	4.5 (0.3; 11.5), 0–42.0 $p = 0.0111$*	0 (0; 2.5), 0–7.5	5.5 (1; 9), 0–28 $p = 0.0043$*	0 (0; 1), 0–6	12 (8; 27), 2–49 $p = 0.0013$*	5 (2; 7), 0–9
Total score						
median (Q1; Q3), min–max	13.3 (2.3; 78), 0.5–129.0 $p = 0.0006$*	0 (0; 2.5), 0–7.5	29.5 (7; 87), 0–107 $p = 0.0011$*	0 (0; 1), 0–6	60 (10; 93), 2–140 $p = 0.0011$*	6 (2; 8), 0–9

Q1: 25% quantile.
Q3: 75% quantile.
min: minimum number of points registered.
max: maximum number of points registered.
*Wilcoxon signed rank tests (unpaired, one-sided) ($p < 0.05$).
[a] The mean represents the rating result from both raters.

due to the requirement for high pressure in the oral cavity making them suitable to test the function of the velopharyngeal sphincter. Voiceless sounds are considered the key element for testing nasal emissions and were included as voiceless fricatives (/f/, /s/, /ʃ/). The nasals (/m/, /n/) were also included. Even though we selected common German vowels and consonants, these were also used by a multi-center, cross-linguistic study known as the Scandcleft project (Lohmander et al., 2009).

We conducted the speech test in three different countries and settings. Although the examiners were not fluent in the local languages, they could easily perform the tests. The test individuals quickly understood the concept of listen-and-repeat without extensive explanation of the procedure. We did not require a translator — the basic instructions of 'listen' and 'repeat' were given in the local language. The subjects did not require a specific level of education or alphabetization.

Table 5
Inter- and Intra-rater agreement (showcased by the German results).

	Inter-rater agreement			Intra-rater agreement		
	Patient group			Rater A		
	PCC	LCC	PIS (%)	PCC	LCC	*p*-value
Hypernasality	0.86	0.81	25%	0.87	0.86	0.88[#]
Nasal emissions	0.82	0.72	58%	0.96	0.95	0.5[#]
Consonant errors	0.88	0.85	25%	0.99	0.99	1[#]
Total score	0.88	0.85	0%	0.95	0.95	0.7[#]
	Control group			Rater B		
	PCC	LCC	PIS (%)	PCC	LCC	*p*-value
Hypernasality	–[a]	–[a]	100%	0.90	0.88	0.88[#]
Nasal emissions	–[a]	–[a]	100%	0.83	0.82	1[#]
Consonant errors	0.78	0.51	66%	0.79	0.73	0.31[#]
Total score	0.78	0.51	66%	0.90	0.89	0.69[#]

PCC: Pearson correlation coefficient.
LCC: Lin correlation coefficient.
PIS: Percentage of identical scoring.
[#] Wilcoxon signed rank sum test ($p < 0.05$).
[a] Due to complete agreement in the categories of hypernasality and nasal emissions, the correlation coefficients cannot be determined and the test cannot be conducted; therefore, values for PCC and LCC cannot be presented.

In general, the selected test sounds posed no major articulatory difficulties to healthy individuals (Table 4). The majority of the few mistakes in the control group were consonant errors. We suspected unspecific mistakes such as lisp in most cases. General aspects of speech development processes have to be taken into consideration as well, especially in younger individuals. A closer matching of patients and control individuals in terms of age probably would have eliminated this variable even further.

These general observations were made for all three languages. Statistical analysis revealed significant differences in the scoring results between patients and control groups. For the three languages tested, the selected test sounds therefore seemed to be useful in assessing the existence of hypernasality, nasal emissions, and consonant errors in cleft patients, due to their specific phonetic characteristics. However, a small degree of registered consonant errors should be considered as non-specific to the cleft condition, as these mistakes were also identified in the control groups.

The assessment of intra-rater agreement revealed differences between both raters. Rater A, as the more experienced speech and language pathologist, achieved good levels of agreement according to LCC, with the exception of hypernasality. Comparably low levels of intra-rater agreement with regard to hypernasality were observed, as has been reported before (Brunnegård and Lohmander, 2007; Lohmander et al., 2012). The poor levels of agreement achieved by rater B may have been caused by the lower level of experience in the evaluation of cleft speech. We suspect that a steeper learning curve influenced the ratings over repeated listening, which impacted the results and led to poor levels of agreement. Nevertheless, according to the Wilcoxon analysis, the results were not significantly different, showing a consistency in the scorings. Overall, the different levels of experience in the assessment of cleft speech, as well as the rather small sample size, might have had an effect on the results. A third rating cycle, or a greater number of assessed sound samples, may have further improved results.

Assessing the results for inter-rater reliability showed some discrepancies. Whilst LCC suggested only poor levels of agreement, the level of identical scores was comparatively high. In the category of hypernasality and nasal emissions in the control group, complete agreement between both raters was documented. The LCC results did not reflect this concordance properly. Further research is required to find out if the results on agreement could be improved by transforming the 78-point scale into a categorized 4–5-point scale.

The pilot project character of this study, using a new test and scoring system, should be considered by assessing the results. It was noticeable that despite poor LCC, both raters could detect significant differences between the two groups. Both raters arrived at the same overall results independently. They were able to identify the heavily affected individuals within the cleft group. Overall, the rating system was easy to understand and apply. The translation of points into different categories, or a search for specific cut-offs, would probably simplify the interpretation of the results in terms of possible consequences for the patient. However, whether the test results can be used for decision-making on future treatment plans requires further evaluation.

In recent years, there has been a lot of controversy regarding cleft speech in the clinical as well as in the scientific context (Lohmander and Olsson, 2004). The ultimate goal is to create economical and standardized methods to improve cleft care for everyone. The majority of contributions originate from medically highly advanced countries, usually requiring a sophisticated, trained, and specialist team to perform high-quality cleft care. Perceptual assessment and narrow transcription are considered the gold standard of speech assessment, requiring a well-trained and experienced speech and language pathologist (Sell, 2005). However, we still face medical, educational, and social challenges in many developing countries (Shaw, 2004). We needed a reliable, low-tech, time-sensitive, cross-linguistic, and appropriate assessment of cleft speech for developing regions. Consequently, some of the complex requirements had to be sacrificed.

The idea of using nonsensical syllables to assess cleft speech has been used in other contexts before. Forner (1983) compared the duration of speech segments between cleft and non-cleft children using spectral analysis. They used five nonsensical syllables in the CVC configuration with five different consonants in the initial position, each combined with/i/in the medium and/p/in the final edition. Eshghi et al. (2013) used CVC nonsensical words, with the targets/t/and/k/, to perform a spectral analysis of words with initial alveolar and velar plosives in Iranian cleft children. To compare the nasalance between cleft and non-cleft patients, Nandurkar (2002) used CVC syllables with selected consonants in the initial position, each combined with an/a/in the middle and an/l/in the final position, and all creating existing words in Marathi.

We do not claim to be replacing the established language-specific speech tests; nor was our main focus on evaluating and distinguishing between highly specific consonant errors. In fact, we aimed to create a reasonable addition to the existing diagnostic tools. Language is still the important background variable in assessing speech outcome. This becomes even more important when comparing patients from different language and cultural backgrounds (Hutters and Henningsson, 2004). We were aware that increasing the usability, simplicity, and universality of a test tool was only possible at the expense of other aspects. Voice disorder and intelligibility, as defined by Henningsson et al. (2008), cannot be assessed by means of the proposed test due to the lack of connected speech or sentences. Nevertheless, it is debatable whether the assessment of speech intelligibility will be possible in the context of a cleft mission trip to a country, where the examiner neither speaks nor understands the local language. The greater the variety in words and sounds, the more precise the test's assessment of the patient's speaking ability will be. This is the great advantage of the existing language-specific speech evaluation tests. On the other hand, the test in this study provides an estimation of the patient's speech abilities and main speech pathologies, and can be performed within a reasonable time scope and with acceptable effort. In the context of a cleft mission, this can be of great help where there is no access to language-specific tests or trained native speakers. Using the same set of test sounds independent of the spoken language would clearly reduce a trip's preparation time and improve applicability. This approach may therefore represent a valid alternative to the Sri Lankan cleft project's method described by Sell and Grunwell (1990), since research on the local languages is often limited.

5. Conclusion

The employed test sound inventory based on non-language-specific syllables possesses the appropriate phonetic and linguistic characteristics, however with some restrictions. We were able to distinguish the speech of cleft patients clearly from non-cleft individuals from three different language and cultural backgrounds. The binary scoring system could identify and quantify the existence of hypernasality, nasal emissions, and consonant errors in cleft patients. However, we need additional research to make a statement on the qualitative aspect of the cleft speech pathologies and possible implications for further treatment planning. We should consider aspects like matching the patient and control groups in terms of age, gender, and diagnosis to add to the value of the test. Additionally, the evaluation by other speech-language pathologists from different language backgrounds, as well as a comparison with the results obtained by established examinations, would further prove the test's precision and validity.

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Conflicts of interest

None.

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