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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

Heiko Cornelis Kleinfeld [a, 1], Ann Christina Foldenauer [b, 1], Mehrangiz Ghassemi [c], Ali Modabber [d], Bijan Movahedian Attar [e], Syed Sayeed Ahmed [f], Christiane Neuschaefer-Rube [g], Alireza Ghassemi [h, i, *]

[a] Department of Urology, Kliniken Maria Hilf, Teaching Hospital University RWTH-Aachen, Viersener Straße 450, 41063 Mönchengladbach, Germany
[b] Department of Medical Statistics, University Hospital RWTH-Aachen, Pauwelsstraße 30, 52074 Aachen, Germany
[c] Department of Orthodontics, University Hospital RWTH-Aachen, Pauwelsstraße 30, 52074 Aachen, Germany
[d] Department of Oral and Maxillofacial Surgery, University Hospital RWTH-Aachen, Pauwelsstraße 30, 52074 Aachen, Germany
[e] Department of Maxillofacial Surgery, School of Medicine, Isfahan University of Medical Sciences, Isfahan, Iran
[f] Department of Oral and Maxillofacial Surgery, Dr. A. Z. Dental College, Aligarh Muslim University (AMU), Aligarh, 202002, India
[g] Department of Phoniatrics, Pedaudiology and Communication Disorders, University Hospital RWTH-Aachen, Pauwelsstraße 30, 52074 Aachen, Germany
[h] Oral and Maxillofacial Surgery, Klinikum-Lippe, Röntgenstr. 18, 32756 Detmold, Teaching Hospital, Georg-August-University Göttingen, Germany
[i] Medical Faculty University RWTH-Aachen, Pauwelsstraße 30, 52074 Aachen, Germany

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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

* Corresponding author. Teaching Hospital Klinikum Lippe, Oral and Maxillofacial Surgery, Röntgenstr. 18, 32756 Detmold, Germany. Fax: +49 5231 721403.
E-mail address: aghassemi@ukaachen.de (A. Ghassemi).
[1] Both authors contributed equally to the manuscript.

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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.

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