The relationship between health-related quality of life and speech in patients with cleft palate

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Abstract

Introduction

In health-care, current efforts focus on providing patient-centered care. Specifically for patients with velopharyngeal insufficiency, and by extent patients with cleft palate, the Velopharyngeal Insufficiency (VPI) Effects on Life Outcomes (VELO) questionnaire (Skirko et al., 2012; 2013) allows the clinician to map the impact of speech and swallowing difficulties on the patient's health-related quality of life (HRQoL). The current study evaluated the hypothesized association between this speech-related HRQoL measure and perceptually and instrumentally assessed speech variables, to provide evidence for the construct validity of the Dutch version of the VELO questionnaire.

Materials and methods

Thirty participants, twenty-five patients with cleft palate and five controls, were enrolled. Perceptual speech assessment was conducted following the recently developed Dutch outcome tool for perceptual speech assessment in patients with cleft palate. In addition, nasalance values and the Nasality Severity Index (NSI) 2.0 were determined. The relationship between these speech outcomes and the scores on the VELO parent report was determined using Spearman rank-order correlation coefficients.

Results

Moderate to strong correlations were found between the total score on the VELO parent report and five speech variables: the VPC-SUM score (r_s= -0.476), speech understandability (r_s= -0.657), passive CSC's (r_s= -0.654), speech acceptability (r_s= -0.591) and the need for

C(L)P-related speech therapy (r_s= -0.711). Furthermore, these variables were associated with at least one subscale of the VELO questionnaire.

Discussion and conclusion

Correlations between speech outcomes and the Dutch version of the VELO questionnaire provide evidence for the construct validity of this version of the instrument. Furthermore, insights in these associations may lead the way to efficient therapy approaches, targeting speech features with the greatest impact on the patient's health-related quality of life.

Keywords

Cleft palate; health-related quality of life; speech

1. Introduction

In health care, current efforts focus on providing patient-centered care. One way of contributing to patient-centered care is to take into account the patient's perspective in clinical practice, research and policy making [1]. The patient's perspective can be captured in a standardized way by means of patient-reported outcome measures (PROM). Such disease-specific PROM was developed for patients with velopharyngeal insufficiency (VPI) to evaluate their health-related quality of life (HRQoL), namely the VPI Effects on Life Outcomes (VELO) questionnaire [2, 3]. Velopharyngeal insufficiency refers to the inability to achieve adequate velopharyngeal closure due to an anatomical or structural defect, most frequently caused by a cleft (lip and) palate (CP±L), resulting in speech and swallowing difficulties [4]. The VELO questionnaire evaluates the impact of these speech and swallowing difficulties on HRQoL based on the responses of the parents (parent report) and the patients themselves (youth report) on five subscales: speech limitations (7 items), swallowing problems (3 items), situational difficulties (5 items), emotional impact (4 items) and perception by others (4 items). Additionally, the parent report comprises a subscale that evaluates the impact on the caregiver by means of three more items. Originally, the items of these subscales were selected following focus groups with VPI patients and their parents, and clinician panels that were set up for the development of the Velopharyngeal Insufficiency Quality-of-Life (VPIQL) instrument [5]. The VELO questionnaire was then derived from the VPIQL questionnaire following item reduction [2]. In several subsequent studies, the validity and reliability of this English questionnaire were evaluated [2, 3, 6, 7]. Recently, the VELO questionnaire was translated to Dutch and showed good internal consistency, discriminant validity, validity of the proxy assessment by the parents, construct validity based on a hypothesized age effect, and reproducibility [8, 9]. These validation studies were conducted in patients with cleft palate, without taking into account the presence of VPI, as the authors argued the items of the questionnaire to be applicable for in all patients with cleft palate.

Given that the VELO questionnaire captures speech-related HRQoL, an association between VELO scores and speech outcomes can be hypothesized. By providing evidence for this association, the construct validity of the VELO questionnaire can be demonstrated, as construct validity refers “to the extent to which scores on a particular instrument relate to other measures in a manner that is consistent with theoretically derived hypotheses concerning the concepts that are being measured” (Terwee et al., 2017, p. 36) [3, 10]. Nonetheless, previous studies in English speaking participants only limitedly evaluated this relationship. Skirko et al. [3] reported a significant correlation (r = -0.37) between VELO scores on the parent report (n = 84) and the perceptual evaluation of speech intelligibility using a five-point scale. No association between the VPI severity, perceptually evaluated on a five-point scale as well, and VELO scores could be demonstrated. A more recent study by Bhuskute et al. [7] reported significant correlations between total VELO scores and speech intelligibility, evaluated using the ordinal scale proposed by Henningsson et al. [11], before (r = -0.71) and after (r = -0.64) speech surgery in twenty-four patients with VPI. In both studies, no information regarding the speech pathologists conducting the speech analysis, including their intra- and inter-rater reliability, was provided. Furthermore, these studies used speech intelligibility as the only speech outcome, providing a general insight in the patient’s speech adequacy. The perceptual evaluation of speech intelligibility can be influenced by several factors that should be controlled for as much as possible [12, 13]. Moreover, there is controversy on using this parameter as the sole outcome measure. Therefore, Witzel [14] recommended to report speech intelligibility in conjunction with other speech outcomes, to complement the interpretation of speech results.

Indeed, most internationally accepted speech outcome tools for patients with cleft palate include a perceptual evaluation of resonance, nasal airflow and consonant production in combination with variables providing an overall insight in the patient's speech performance, such as speech understandability and/or speech acceptability [11]. Recently, a Belgian Dutch speech outcome tool for the perceptual evaluation of speech was developed and validated [15]. This outcome tool includes the perceptual evaluation of global speech variables (speech understandability, speech acceptability, and the need for SLT intervention, either CP±L-related or non-CP±L-related), and specific speech variables: categorized consonant production errors (anterior, posterior, non-oral and passive), hypernasality, hyponasality, nasal emission, nasal turbulence, voice and nasal grimace. Although speech evaluation by a well-trained ear is considered the gold standard [16, 17], the reliability of such evaluation is susceptible for several influencing factors, including the listener's characteristics, task factors and the interaction between these listener and task factors [18]. Consequently, instrumental assessment of the velopharyngeal function, resonance and/or nasal airflow to support clinical decision-making, is often included in the speech assessment protocol. At the Ghent University Hospital, speech evaluation generally includes instrumental assessment of resonance by means of the Nasometer^{TM} and the calculation of the Nasality Severity Index (NSI) 2.0, in addition to the perceptual assessment. In the case of suspected VPI, a multiview videofluoroscopy is performed.

An even more holistic approach would be the inclusion of the VELO questionnaire to assess the impact of speech (and swallowing) difficulties on functioning and well-being. Therefore, it would be interesting to evaluate to what extent these VELO scores are related to speech variables. Moreover, evidence for such associations would support the construct validity of the Dutch VELO questionnaire. Hence, the aim of this study was to assess the relationship between perceptually and instrumentally assessed speech variables, evaluated using a comprehensive test battery as performed at the Ghent University Hospital, and the results of the VELO questionnaire. A negatively directed relationship showing lower, thus poorer, VELO scores with higher (poorer) ratings on the ordinal scale of the perceptual assessment, higher nasalance values and lower NSI 2.0 values was hypothesized, and considered evidence to support the questionnaire's construct validity.

2. Methods

The ethical committee of the Ghent University Hospital approved this study (2016/0338). All parents of the participants signed an informed consent.

2.1. Participants

Twenty-five children with a CP±L and five controls without CP±L or any other craniofacial malformation were included in the analysis. The mean age of the participants was 6.7 years (standard deviation (*SD*) = 1.95 years (y)). All participants had Dutch as their native language. Participants were between 3 and 10 years old. Speech samples were selected to represent a range of severity regarding speech understandability, resonance, nasal airflow and articulation, decided based on clinical records. Patients with cleft palate were followed by the Craniofacial Centre of the Ghent University Hospital and presented with an isolated CP±L. Six patients had a cleft palate only, eleven patients had a unilateral cleft lip and palate, seven patients had a bilateral cleft lip and palate, and one patient presented with a submucosal cleft palate. Of all patients with CP±L, two received secondary palatoplasty: one patient with a submucosal cleft had a re-repair following Sommerlad's procedure, and in one patient closure of a palatal fistula was performed. Nineteen of the patients with CP±L followed speech therapy at the moment of the assessment or in the past.

The control group consisted of children without speech disorders, velopharyngeal or other craniofacial deformities, known syndromes, developmental disorders or general disability.

2.2. Perceptual speech assessment

Speech samples and the listening protocol were set up following the recently developed Dutch outcome tool for perceptual speech assessment in patients with cleft palate [15]. Speech variables were evaluated using ordinal scales. Hypernasality was rated on a five-point scale, whereas speech understandability and speech acceptability were evaluated on a four-point scale. Anterior, posterior, non-oral and passive cleft speech characteristics (CSC's), hyponasality, nasal emission and nasal turbulence were scored on a three-point scale. A dichotomous scale was used to evaluate the need for CP±L-related and non- CP±L related SLT intervention, voice and grimace.

Speech understandability was evaluated based on an audio sample of spontaneous speech without utterances of the conversation partner. This speech sample was edited using Praat software version 6.0.14 [19], also ensuring a similar length of the speech samples of approximately 60 syllables. For the evaluation of other speech variables, speech samples of spontaneous speech, reciting automatic sequences and repetition of sentences were available in an audio and audio/video format. Furthermore, the speech sample for speech understandability could only be played once, whereas all other speech samples could be replayed as much as needed. Hypernasality, hyponasality, nasal emission, nasal turbulence and CSC's were evaluated based on the audio and video recordings of the sentences only. CP±L-related and non- CP±L related SLT intervention and speech acceptability were scored based on the audio and audio/video recordings of spontaneous speech, automatic sequences and the repetition of the sentences. Based on the audio recordings of these samples the variable voice was scored, whereas only video recordings were evaluated to score the presence of grimace. For a further overview of the structured listening protocol and a detailed description of the Belgian Dutch speech sample we refer to Bruneel et al. [15]. Audio samples were recorded using a unidirectional condenser microphone (Samson C01U) to ensure sufficient sound quality. Audiovisual recordings were made using a Sony Handycam HDR-CQ280E, framing the participant's head and shoulders.

Samples were rated by two speech language pathologists with experience in speech assessment in patients with CP±L. Ratings were conducted independently using over-ear headphones (Sennheiser EH 150 and Sennheiser Momentum). The ratings of rater 1, who was blinded to the study purposes, were used for further analysis. To determine the intra-rater reliability, 20% of the speech samples (6/30) was re-rated. During randomization, the order of the samples was controlled carefully so that none of the samples that were rated twice succeeded immediately.

Several speech variables of the Dutch speech outcome tool were based on translated definitions and scales of the English CAPS-A outcome tool [20, 21], translated and adapted to Dutch [15]. Consequently, the guidelines by Pereira et al. [22] to calculate velopharyngeal composite scores based on ratings using the CAPS-A tool, could be applied. Composite scores were derived from the perceptual evaluation of the speech variables 'hypernasality', 'nasal emission' and 'nasal turbulence', 'non-oral CSC's' and 'passive CSC's'. Based on these composite scores, the Velopharyngeal composite score-summary CAPS-A (VPC-SUM) was determined, indicating the presence of VPI. VPC-SUM scores were interpreted as follows: score 0-1: sufficient velopharyngeal function; score 2: borderline deficit and score 3-4: insufficient velopharyngeal function [22, 23].

2.3. Instrumental speech assessment

In addition to the perceptual speech evaluation, nasalance values and the Nasality Severity Index 2.0 (NSI 2.0; [24]) were determined. The mean nasalance values of the repetition of a Dutch oral, oronasal and nasal text [25] were determined using the Nasometer™ II model 6450 (Kay Pentax, NJ, Lincoln Park). The oral text contains no nasal consonants, whereas the nasal text is loaded with nasal consonants (57%). In the oronasal text, 11.76% of the consonants are nasal, approximately the same oronasal balance as was found in standard Dutch speech (11.63%) [26]. The NSI 2.0 includes three parameters: the nasalance values of the vowel [u:] and the oral text, and the voice low tone to high tone ratio (VLHR) with a cutoff frequency of $4.47*F_0$Hz of the sustained production of the vowel [i:]. The VLHR and the NSI 2.0 were determined using Praat software version 6.0.14 [19]. The resulting NSI value can be either negative or positive, with a negative value indicating the presence of hypernasality.

2.4. VELO questionnaire

One of the parents of the participants completed the VELO parent report at the same time as the speech sample collection. When the participant was eight years or older, they completed the youth report themselves. However, as only nine youth reports were completed, these responses were not further analyzed. The parent report consists of 26 items each being scored on a Likert-type scale ranging from zero (never) to four (almost always). The sum of these responses was converted to a scale from zero to hundred, with hundred being the best possible HRQoL.

2.5. Statistical analysis

Statistical analyses were performed using the SPSS version 25 software package (SPSS Inc., Chicago, Illinois). Inter- and intra-rater reliability were determined using quadratic weighted kappa [27, 28] and interpreted following the classification described by Altman [29] adapted from Landis and Koch [30]. The relationship between speech variables and the total score on the parent report was analyzed using Spearman rank-order correlation coefficients (r_S). Size of the correlation coefficient (either positive or negative) was interpreted following the classification described by Mukaka [31]: 0.00 to 0.30: negligible correlation; 0.30 to 0.50: low correlation; 0.50 to 0.70: moderate correlation; 0.70 to 0.90: high correlation; 0.90 to 1.00: very high correlation. Because of the multiple correlations, the significance level α was set at 0.01. Results between 0.01 and 0.05 were considered trends. At this stage, Bonferroni correction was not applied as this conservative method lowers the probability of finding significant results with higher numbers of comparisons [32]. Furthermore, in order to enhance the interpretation of the correlations and their significance level, 99% confidence intervals were calculated using a bias corrected and accelerated bootstrap method (BCa CI). For post-hoc analyses of the correlations between speech variables and the subscales of the VELO questionnaire, a Bonferroni correction was applied, setting the significance level at 0.001 (0.01/6).

3. Results

3.1. Perceptual speech assessment – inter- and intra-rater reliability

For most speech variables, good to very good inter-rater reliability was established, except for posterior CSC’s, non-CP±L-related SLT intervention, hyponasality and nasal emission (table 1). Similar results were found for the intra-rater reliability: for all variables good to very good intra-rater reliability was found, with the exception of anterior oral CSC’s, hyponasality and nasal emission (table 2).

Table 1: Inter-rater reliability of the perceptual speech assessment using weighted quadratic kappa

Speech parameter	**κ**	**95% CI κ**	**Interpretation***
VPC-SUM	0.82	0.66 – 0.91	Very good
Speech understandability	0.86	0.73 – 0.93	Very good
Anterior oral CSC's	0.68	0.42 – 0.83	Good
Posterior oral CSC's	0.51	0.19 – 0.73	Moderate
Non-oral CSC's	0.86	0.72 – 0.93	Very good
Passive CSC's	0.75	0.54 – 0.88	Good
Non-C(L)P speech errors	0.60	0.31 - 0.79	Moderate
Hypernasality	0.85	0.71 – 0.93	Very good
Hyponasality	0.52	0.20 – 0.74	Moderate
Nasal emission	0.52	0.21 – 0.74	Moderate
Nasal turbulence	0.70	0.45 – 0.84	Good
Voice	0.61	0.33 – 0.80	Good
Nasal grimace	0.77	0.58 – 0.89	Good
Speech acceptability	0.85	0.71 – 0.93	Very good
Speech therapy C(L)P	0.94	0.87 - 0.97	Very good

*[29, 30]

Table 2: Intra-rater reliability of the perceptual speech assessment using weighted quadratic kappa

Speech parameter	**κ**	**95% CI κ**	**Interpretation***
VPC-SUM	0.88	0.38 – 0.98	Very good
Speech understandability	0.83	0.20 – 0.97	Very good
Anterior oral CSC's**	-	-	-
Posterior oral CSC's	1.00	1.00 – 1.00	Very good
Non-oral CSC's	0.91	0.48 – 0.99	Very good
Passive CSC's	0.88	0.37 – 0.98	Very good
Non- C(L)P speech errors	0.71	(-0.11) – 0.95	Good
Hypernasality	0.92	0.54 – 0.99	Very good
Hyponasality**	-	-	-
Nasal emission	0.19	(-0.67) – 0.82	Poor
Nasal turbulence	0.62	(-0.26) – 0.94	Good
Voice	1.00	1.00 – 1.00	Very good
Nasal grimace	0.62	(-0.26) – 0.94	Good
Speech acceptability	1.00	1.00 – 1.00	Very good
Speech therapy C(L)P	1.00	1.00 – 1.00	Very good

*[29, 30]

** Not possible to calculate a kappa value due to negative covariance (anterior oral CSC's) or zero variance (hyponasality)

3.2. The relationship between perceptually evaluated speech variables and the VELO parent report

An overview of the correlations between the total score on the VELO parent report and perceptually evaluated speech variables is provided in table 3. Significantly negative correlations with the total VELO score were found for five speech variables: the VPC-SUM score (r_s(28)= -0.476), speech understandability (r_s(28)= -0.657), passive CSC's (r_s(28)= -0.654), speech acceptability (r_s(28)= -0.591) and the need for CP±L-related SLT intervention (r_s(28)= -0.711). Post-hoc analyses revealed a significant correlation between all five speech variables and at least one VELO subscale (table 4). The VPC-SUM was significantly correlated with the subscale 'speech limitations' (r_s(28)= -0.711). For speech understandability, analyses showed moderate to high correlation with the subscales 'speech limitations' (r_s(28)= -0.612) and 'situational difficulties' (r_s(28)= -0.764). Furthermore, passive CSC's were moderately associated with the subscales 'speech limitations' (r_s(28)= -0.675), 'situational difficulties' (r_s(28)= -0.632) and 'caregiver impact' (r_s(28)= -0.571). Speech acceptability was correlated with only one subscale, namely 'situational difficulties' (r_s(28)= -0.621). Lastly, the need for CP±L-related SLT intervention was associated with four subscales: 'speech limitations' (r_s(28)= -0.704), 'situational difficulties' (r_s(28)= -0.627), 'emotional impact' (r_s(28)= -0.600) and 'perception by others' (r_s(28)= -0.689).

3.3. The relationship between nasalance values and the NSI 2.0, and the VELO parent report

Table 5 presents the results of Spearman rank-order correlations between nasalance values and the NSI 2.0, and VELO scores. Analyses revealed two trends: for the nasalance value of the oral text (r_s(25)= -0.471), and the NSI 2.0 (r_s(23)= -0.436). However, 99% BCa CI's

Table 3: Spearman rank-order correlations between perceptually assessed speech variables and the total score on the VELO parent report

Speech parameter	***n***	r_s	**BCa 99% CI**	***p***
VPC Sum score	30	-0.476	[-0.709, -0.128]	0.008*
Speech understandability	30	-0.657	[-0.817, -0.380]	<0.001*
Anterior oral CSC's	30	-0.418	[-0.685,-0.102]	0.022**
Posterior oral CSC's	30	-0.210	[-0.480,0.054]	0.266
Non-oral CSC's	30	-0.242	[-0.544,0.128]	0.198
Passive CSC's	30	-0.654	[-0.812,-0.349]	<0.001*
Non-C(L)P speech errors	30	-0.240	[-0.605,0.196]	0.202
Hypernasality	30	-0.337	[-0.638,0.111]	0.068
Hyponasality	30	-0.284	[-0.598,0.187]	0.129
Nasal emission	30	-0.199	[-0.526,0.155]	0.292
Nasal turbulence	30	-0.349	[-0.623,0.001]	0.059
Voice	30	0.187	[-0.059,0.425]	0.322
Nasal grimace	30	-0.164	[-0.427,0.088]	0.387
Speech acceptability	30	-0.591	[-0.799,-0.272]	0.001*
Speech therapy C(L)P	30	-0.711	[-0.848, -0.454]	<0.001*

*Statistically significant, $p \leq 0.01$

** Trend, $0.01 < p < 0.05$

Table 4: Post-hoc Spearman rank-order correlations between perceptually assessed speech variables and the subscales of the VELO parent report

Post-hoc correlations between the VPC-SUM and the VELO subscales			
	r_s	**BCa 99% CI**	***p***
Speech limitations	-0.558	[-0.823,-0.055]	0.001*
Swallowing problems	-0.072	[-0.588,0.424]	0.705
Situational difficulty	-0.474	[-0.788,0.135]	0.008
Emotional impact	-0.342	[-0.756,0.231]	0.065
Perception by others	-0.352	[-0.728,0.101]	0.057
Caregiver impact	-0.342	[-0.748,0.177]	0.064
Post-hoc correlations between speech understandability and the VELO subscales			
	r_s	**BCa 99% CI**	***p***
Speech limitations	-0.612	[-0.841, -0.168]	<0.001*
Swallowing problems	-0.011	[-0.563,0.462]	0.952
Situational difficulty	-0.764	[-0.910,-0.428]	<0.001*
Emotional impact	-0.537	[-0.829,-0.150]	0.002
Perception by others	-0.543	[-0.805,-0.176]	0.002
Caregiver impact	-0.362	[-0.833,0.165]	0.049
Post-hoc correlations between passive CSC's and the VELO subscales			
	r_s	**BCa 99% CI**	***p***
Speech limitations	-0.675	[-0.885,-0.274]	<0.001*
Swallowing problems	-0.206	[-0.683,0.347]	0.274
Situational difficulty	-0.632	[-0.851,-0.206]	<0.001*
Emotional impact	-0.501	[-0.810, -.101]	0.005
Perception by others	-0.502	[-0.838,-0.179]	0.005
Caregiver impact	-0.571	[-0.856,-0.203]	0.001*
Post-hoc correlations between speech acceptability and the VELO subscales			
	r_s	**BCa 99% CI**	***p***
Speech limitations	-0.520	[-0.823,0.061]	0.003
Swallowing problems	-0.207	[-0.634,0.255]	0.272
Situational difficulty	-0.621	[-0.896,-0.170]	<0.001*
Emotional impact	-0.508	[-0.834,-0.104]	0.004
Perception by others	-0.524	[-0.808,-0.136]	0.003
Caregiver impact	-0.345	[-0.770,0.159]	0.062
Post-hoc correlations between the need for C(L)P-related speech therapy and the VELO subscales			
	r_s	**BCa 99% CI**	***p***
Speech limitations	-0.704	[-0.867,-0.351]	<0.001*
Swallowing problems	-0.297	[-0.765,0.177]	0.111
Situational difficulty	-0.627	[-0.895,-0.139]	<0.001*
Emotional impact	-0.600	[-0.903,-0.184]	<0.001*
Perception by others	-0.689	[-0.911,-0.407]	<0.001*
Caregiver impact	-0.414	[-0.795,0.111]	0.023

*Statistically significant, $p \leq 0.001$

Table 5: Spearman rank-order correlations between nasalance and NSI 2.0 values and the total score on the VELO parent report

Speech parameter	***n***	r_s	**BCa 99% CI ***	***p***
Nasalance value oral text	27	-0.471	[-0.743,0.037]	0.013**
Nasalance value oronasal text	26	-0.227	[-0.634,0.433]	0.265
Nasalance value nasal text	25	-0.115	[-0.704,0.446]	0.585
NSI 2.0	25	+0.436	[0.064,0.664]	0.029**

** Trend, $0.01 < p < 0.05$

of all these correlations crossed zero, hence no clear associations between nasalance values and the NSI 2.0 and VELO scores can be stated.

4. Discussion

The aim of this study was to evaluate the association between speech variables and results on the VELO questionnaire. A negative association showing poorer HRQoL with poorer speech results was hypothesized. By testing this hypothesis, the construct validity of the Dutch version of the VELO questionnaire [8] was assessed. Within expectation, the majority of the variables showed a negatively directed correlation between speech and VELO scores.

Correlations were mostly significant for perceptually assessed variables providing a more general insight in speech adequacy, namely speech understandability, speech acceptability, the VPC-SUM and the need for CP±L-related SLT intervention. This may be explained by the influence of specific speech variables on more general variables. For example, variables such as hypernasality and CSC's probably attribute to the perception of speech understandability and speech acceptability. On the other hand, the need for CP±L-related SLT intervention will be decided based on general speech performance, such as speech understandability and speech acceptability, and specific speech features such as the presence of CSC's. Based on this rationale, it may be assumed that general speech variables worsen with a higher number of poor specific speech variables, hence resulting in stronger negative correlations between general speech variables and VELO scores. The finding that none of the instrumentally assessed parameters was significantly associated with the VELO scores is in accordance with this explanation. These instrumental assessments focused on resonance disorders, only part of the spectrum of specific speech disorders contributing to the overall speech adequacy.

The correlation between speech understandability and HRQoL as measured by the VELO questionnaire, confirms the findings by Skirko et al. [3] and Bhuskute et al. [7] who also reported a significantly negative relationship between these two measures. More specifically, Bhuskute et al. [7] described the association between speech understandability and the subscales 'situational difficulties' and 'perception by others'. In this study, speech understandability was significantly associated to the subscales 'speech limitations' and 'situational difficulties'. This can be explained by the content of these subscales, as the subscale 'speech limitations' maps the specific speech difficulties the patient experiences, and the subscale 'situational difficulties' explicitly questions the patient's speech understandability for listeners with a varying familiarity with the child's speech (family, friends, and strangers). Interestingly, results also suggested a relationship between the concepts of speech understandability and speech acceptability, as the parameter speech acceptability was only significantly associated with the subscale 'situational difficulties', which, as stated above, quantifies the child's understandability. Following the Dutch speech outcome tool, speech understandability was defined as the degree to which the speech is understandable for a listener unfamiliar with the child's speech, whilst speech acceptability was defined as the degree to which the speech draws the attention, independent of the communicative message. Given these findings, it would be interesting in future studies to identify the components contributing to the perception of acceptable or inacceptable speech, and their influence on the perception of speech understandability. Moreover, analyses revealed a borderline significant correlation between speech understandability and the subscales 'emotional impact' and 'perception by others'. Studies by Lee et al. [33] and

Bettens [34] showed that attitudes of peers, questioned in the VELO subscale ‘perception by others’, deteriorated with poorer speech understandability, providing an explanation for the association found in this study. Furthermore, negative attitudes may result in emotional distress for the patient, and thus poorer results on the subscale ‘emotional impact’.

The VPC-SUM and one component of this summary parameter, namely passive CSC’s, showed a significant association with the total VELO questionnaire and the subscale ‘speech limitations’. Additionally, passive CSC’s were correlated with the subscales ‘situational difficulties’ and ‘caregiver impact’. Passive CSCs are characteristic speech features resulting from the presence of VPI [35]. Hence, these correlations can be explained by the effects of VPI on speech and consequently speech understandability, assessed by the subscales ‘speech limitations’ and ‘situational difficulties’. On the other hand, it can be hypothesized that patients with VPI, and thus passive CSC’s, have more severe speech problems in comparison to patients with cleft palate without VPI, resulting in a greater impact on the caregiver. Originally, the VELO questionnaire was designed for patients with VPI. Hence these findings strengthen the evidence for the questionnaire’s construct validity. However, as the correlation between the VPC-SUM and the VELO questionnaire was rather low (r_s= -0.476), this suggests the VELO questionnaire to be sensitive for a broader range of speech disorders and speech-related distress than only those resulting from VPI. Consequently, together with the results of previous validation studies including patients with cleft palate without taking into account VPI [8, 9], this justifies the use of the questionnaire in all patients with cleft palate.

The need for CP±L-related SLT intervention was correlated to all VELO subscales with the exception of the subscales ‘swallowing problems’ and ‘caregiver impact’. The nonexisting relationship between the subscale ‘swallowing problems’ and the need for speech therapy does not come as a surprise, as they measure other concepts. In contrast, a relationship between the need for speech therapy and the subscale ‘caregiver impact’ was expected, given that a large outcome study by Sell et al. [36] highlighted the predictive value of the parent’s concerns for poorer speech outcomes. The fact that the caregiver impact was not significantly related to the need for speech therapy, might contribute to the suggestion proposed by Bruneel et al. [9]. These authors suggested that factors other than those specifically related to speech, might play also role in the perceived HRQoL. In the analysis, only the perception of the parents on their child’s HRQoL was included. As such, contextual factors might play a role, such as the parent’s expectations, concerns and coping style, and to a broader extent the environment of the patient including their family and peers. Nevertheless, as is shown by this study, speech characteristics contribute to a major extent to the perceived HRQoL. From this perspective, new insights in how to increase speech therapy effectiveness could be explored. As the majority of the significantly correlated variables were general speech variables (speech understandability, speech acceptability and the need for C(L)P-related speech therapy) or summary variables (VPC-SUM), future research could identify which specific speech features, e.g. articulation, should be targeted in therapy to improve general speech variables and thus to improve HRQoL.

Speech variables were perceptually assessed using ordinal scales following the Dutch speech outcome tool. Although these ordinal scales allow for straightforward communication between members of the cleft team, the use of visual-analogue scaling (VAS) might have resulted in even stronger correlations between VELO scores and speech variables. Furthermore, evidence for more valid perceptual evaluations of speech variables such as hypernasality using VAS is growing [17, 37-39]. To meet this need, Bruneel et al. [15]

stated to explore the use of VAS as part of the Dutch speech outcome tool in future studies. Additionally, the inclusion of a greater number of participants might have resulted in stronger correlations as well, possibly revealing more subtle associations than those described in this study. However, perceptual speech evaluations were rather time consuming, given the inclusion of a phonetic transcription to identify CSC's, and therefore time constraints were considered when determining the sample size. Despite this room for improvement, this study provided evidence for moderate to strong correlations between HRQoL measures and speech variables evaluated using an exhaustive test battery.

5. Conclusion

The results on the parent report of the VELO questionnaire were significantly associated with speech variables providing a global insight in the patient's speech adequacy, namely speech understandability, speech acceptability, the VPC-SUM and the need for C(L)P-related speech therapy, confirming the construct validity of the Dutch version of the VELO questionnaire. Furthermore, insights in these correlations can lead the way to efficient therapy approaches, targeting speech features that have the greatest impact on the patient's health-related quality of life.

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7. References

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