

**Health-related quality of life in patients with cleft palate: reproducibility, responsiveness
and construct validity of the Dutch version of the VELO questionnaire**

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Funding and declaration of interest

This work was supported by a PhD fellowship of the Research Foundation Flanders (L.B.).

The authors declare that there is no conflict of interest.

Abstract

Objectives: Recently, the Velopharyngeal Insufficiency (VPI) Effects on Life Outcomes (VELO) questionnaire, which evaluates the impact of speech and swallowing difficulties on health-related quality of life (HRQoL) in patients with VPI (Skirko et al., 2012), was translated to Dutch (Bruneel et al., 2017). The purpose of this study was to evaluate the reproducibility, responsiveness and construct validity of this Dutch version of the questionnaire.

Methods: To evaluate the reproducibility, 50 parents and 14 children with cleft palate re-completed the questionnaire after two weeks. Thirty-five parents and 8 children with cleft palate completed the VELO questionnaire after one year for the evaluation of the responsiveness. The correlation between age and the VELO questionnaire (construct validity), and the internal consistency (Cronbach's α) were re-determined based on the responses of 73 parents and 24 children.

Results: Based on descriptive statistics, results of the Wilcoxon signed rank-test, and the absolute (SEM) and relative (ICC) consistency, the questionnaire showed good reproducibility. VELO scores did not significantly differ after one year, neither when performing separate analyses for the intervention (speech therapy) and the non-intervention group. Correlations indicated higher HRQoL, as perceived by the parents, with increasing age. The reverse was the case for the report. Cronbach's α showed excellent internal consistency for both reports.

Conclusions: The VELO questionnaire showed good reproducibility and internal consistency. Moreover, results reconfirmed the age effect on VELO scores. To understand the implications of the results regarding the questionnaire's responsiveness, future research should focus on the identification of factors influencing the patient's evolution in HRQoL.

Keywords: cleft palate, health-related quality of life, speech, questionnaire

1. Introduction

To date, the call to take into account the patient's perspective is omnipresent in health care services. The assessment of the patient's point of view can serve several purposes, such as the evaluation of care and outcomes, and decision making regarding the patient's treatment options [1]. A standardized way to understand and evaluate the patient's perspective are patient-reported outcome measures (PROMs), that can be either generic or disease specific [2]. An example of a disease specific PROM is a questionnaire evaluating the patient's health related quality of life (HRQoL), defined as the perceived impact of a certain condition on several aspects of health, including physical, mental and social aspects [3].

Specifically for patients with cleft palate, Hall et al. [4] and Alansari et al. [5] advocated the need to incorporate the patient's perspective as an outcome variable in the treatment process, in service evaluations, and in research. A qualitative study by Wong Riff et al. [6] identified three concepts of interest to these patients: facial functions (speech, eating and drinking, and other facial functions), appearance and health-related quality of life (psychological and social aspects, school, and speech-related distress). As illustrated by these results, this congenital condition affects several aspects of the patient's functioning and well-being. Hence, a multidisciplinary team should cover care. An important member of this team is the speech pathologist, who is responsible for the diagnosis and treatment of communication problems, including speech and language difficulties, and feeding difficulties. In general, speech of patients with cleft palate can be characterized by resonance disorders, abnormal nasal airflow, and passive and active articulation disorders, resulting in reduced speech intelligibility and/or speech acceptability [7]. Regarding feeding, some of the most reported difficulties in this population are sucking difficulties and nasal regurgitation [8, 9]. Often, these speech and feeding difficulties are the result of velopharyngeal insufficiency (VPI) caused by the structural changes of a cleft palate [10]. Other anatomical deformities, e.g. oronasal fistula

after palatal closure, or the presence of a (craniofacial) syndrome and/or other conditions, can affect speech and feeding additionally [10, 11].

The English VPI Effects on Life Outcomes (VELO) questionnaire assess the HRQoL of patients with velopharyngeal insufficiency, including the parent's and the patient's view [12]. This questionnaire was constructed following item reduction of the Velopharyngeal Insufficiency Quality-of-Life (VPIQL) questionnaire by Barr et al. [13], and has been validated rigorously in several subsequent studies [12, 14-16]. The VELO questionnaire consists of a parent report and a youth report, completed by children aged 8 years and older. Both reports have the same subscales: speech limitations (7 items), swallowing problems (3 items), situational difficulties (5 items), emotional impact (4 items) and perception by others (4 items). These last three subscales are related specifically to speech distress. Additionally, the parent report has three items assessing the impact on the caregiver. Each item is scored from zero (never) to four (almost always). The eventual score on the VELO questionnaire is generated by fitting the scores on a scale from zero to 100, with 100 representing the highest quality of life [12].

Recently, the VELO questionnaire has been translated and validated in other languages such as Spanish [17], Norwegian [18] and Dutch (Flemish) [19]. Specifically for the Dutch version, Bruneel et al. [19] described the translation procedure and the first results regarding the translated questionnaire's validity and reliability. The Dutch VELO questionnaire showed good internal consistency, linguistic validity, discriminant validity and validity of the parent proxy assessment. Furthermore, the results on the parent report and the age of the child were positively correlated. A priori a positive evolution of HRQoL with increasing age was hypothesized, and therefore results possibly attributed to the questionnaire's construct validity. These findings were based on the responses of patients with an isolated cleft (lip and)

palate and their parents, without taking into account the presence of VPI. The majority of the items of the VELO questionnaire concerns the impact of speech disorders or deviant speech on functioning and well-being, without specifying the type of speech disorders. Hence, in addition to the results by Bruneel et al. (2018), the authors argued the VELO questionnaire to be applicable in all patients with cleft palate, irrespective of the presence of VPI. However, the sample size in the study by Bruneel et al. [19] was rather small (parent report: $n = 39$; youth report: $n = 14$) and consequently no specific statements regarding the construct validity and the overall internal consistency could be made. Moreover, the questionnaire's reproducibility and responsiveness were not evaluated yet. Therefore, the aims of the current study were to evaluate the reproducibility and responsiveness, and to re-evaluate the internal consistency and construct validity of the Dutch version of the VELO questionnaire using a greater sample size of patients with an isolated cleft (lip and) palate.

2. Methods

This study was approved by the ethical committee of the Ghent University Hospital (2016/0338; 2017/0999; 2017/1000). All patients and their parents participated voluntarily and signed an informed consent.

2.1. Evaluated measures

Reproducibility was considered the ability of the questionnaire to provide similar results for measurements with a two-week interval in stable participants [20]. Responsiveness was defined as the questionnaire's ability to capture clinically important changes over time [20, 21], namely one year. To evaluate the latter measure of longitudinal validity the following results were hypothesized: (1) all patients with cleft palate have a positive evolution in VELO scores after one year, and (2) participants who received intervention (i.e. speech therapy) have a greater positive evolution in VELO scores after one year compared to participants who did not receive intervention. To provide stronger evidence for the VELO's construct validity, the

correlation between age and VELO scores was re-evaluated. Lastly, the internal consistency, defined as the extent to which all items of the questionnaire measure the same concept, was re-evaluated.

2.2. Participants and data collection

Participants enrolled in the study by Bruneel et al. [19] were re-contacted by phone, e-mail or during the annual consultation of the craniofacial team, to complete the VELO questionnaire one year after their first response for the assessment of the questionnaire's responsiveness. Complete responses with a one-year interval were collected in 35 participants, with 8 additional responses on the youth report. For evaluation of the questionnaire's reproducibility, all these 35 participants were asked to complete the questionnaire twice after one year, with the second time being two weeks later. Additionally, new participants were recruited who also completed the questionnaire twice with a two-week interval. Eventually, these responses with a two-week interval for the evaluation of the reproducibility were complete in 50 participants, together with 14 responses on the youth report. Of all participants described above, the first response ever, i.e. either the response included in the study by Bruneel et al. [19] or the first response for the evaluation of reproducibility, was analyzed for the re-evaluation of the internal consistency and the age effect (construct validity). Eventually 73 responses on the parent report and 24 responses on the youth report were included in the analysis of the internal consistency and the age effect.

Only Flemish, Dutch-speaking patients with a cleft (lip and) palate followed by the multidisciplinary craniofacial team of the Ghent University Hospital were included. Participants were excluded when the questionnaire was incomplete, when the parents and/or the child had a native language other than Dutch, when the cleft palate was part of a syndrome, when the participant had an associated condition known to affect psychological

Table 1: overview of the participants for the different analyses: reproducibility, responsiveness, internal consistency and age effect

	Reproducibility		Responsiveness		Internal consistency and age effect	
	Parent report (<i>n</i> = 50)	Youth report (<i>n</i> = 14)	Parent report (<i>n</i> = 35)	Youth report (<i>n</i> = 8)	Parent report (<i>n</i> = 73)	Youth report (<i>n</i> = 24)
Sex	26 M ; 24 F	8 M ; 6 F	14 M ; 21 F	2 M ; 6 F	33 M ; 40 F	11 M ; 13 F
Type CP	18 CP ; 32 CL + P	6 CP ; 8 CL + P	13 CP ; 22 CL + P	5 CP ; 3 CL + P	31 CP ; 42 CL + P	13 CP ; 11 CL + P
Oronasal fistula	<i>n</i> = 17	<i>n</i> = 1	<i>n</i> = 12	<i>n</i> = 0	<i>n</i> = 23	<i>n</i> = 1
Mean age of the child at first response	7,5 y (<i>SD</i> = 2.56)	10.9 y (<i>SD</i> = 1.27)	6.3 y (<i>SD</i> = 2.23)	9.1 y (<i>SD</i> = 0.99)	7.0 y (<i>SD</i> = 2.46)	9.6 y (<i>SD</i> = 1.28)
Mean time interval first and second response	18.8 d (<i>SD</i> = 7.91)	17.6 d (<i>SD</i> = 7.04)	11.9 m (<i>SD</i> = 2.43)	13.0 m (<i>SD</i> = 2.00)	N.A.	N.A.

Abbreviations: M = male; F = female; CP = cleft palate; CL + P = cleft lip and palate; y = years; m= months; d=days; SD = standard deviation; N.A. = not applicable

gnitive functioning (e.g. attention deficit/hyperactivity disorder), when the had a moderate to severe hearing loss (subjectively reported by the parents), and participant received special education. Indications of VPI were not taken into r the inclusion of the participants. None of the participants received surgery during of this study. Concerning the response for the evaluation of the responsiveness, n regarding speech therapy during the one-year interval was collected. An overview ic details of the participants for each analysis can be found in table 1.

.Statistical analysis

analysis was performed using SPSS version 24.0 (SPSS Inc., Chicago, Illinois). ificance level α was set at ≤ 0.05 for all analyses, with the exception of post-hoc r which a Bonferroni correction of the significance level was applied.

2.3.1. Reproducibility

at both time points (two-week interval) were compared using the Wilcoxon signed dditionally, differences of the total VELO scores were calculated by subtracting n the first time point from the score on the second time point. Based on these ulative tables (raw/%) with the absolute differences were composed. Furthermore, orrelation coefficients (ICC) (one-way random, single measures) were determined, eted following the classification by Cicchetti [22] (ICC < 0.40: poor; ICC between .59: fair; ICC between 0.60 – 0.74: good ; ICC ≥ 75 : excellent). ICC measures e relative consistency of the responses between both time points. As such, an reliability parameter evaluating the absolute consistency of the scores, the standard asurement (SEM), was calculated following the formula $SEM = SD \sqrt{1 - ICC}$, with

SD representing the standard deviation of the scores of all subjects [23]. Additionally, the minimal detectable differences (MDD) to reveal differences that can be considered as a real observation, not due to personal variation or measurement error, were calculated by determining the 95% confidence interval (CI) around the SEM following the formula $MDD = 1.95 SEM * \sqrt{2}$ [23].

2.3.2. Responsiveness

Responses of all participants were compared between both time points (one-year interval) using the Wilcoxon signed rank-test. In addition, absolute differences were calculated and cumulative tables (raw/%) of these absolute differences were composed. To test if the participants who received intervention (i.e. speech therapy) had a more positive HRQoL evolution after one year, a linear mixed model was used to compare the groups (i.e. intervention ($n = 17$) vs. no intervention ($n = 18$)) over time, using a scaled identity covariance structure. The factors time, intervention and group-by-time interaction were specified as fixed effects, and a random intercept was included. Model assumptions were checked by inspecting the distribution of the residuals. Only responses on the parent report were included in the analyses, as the number of participants of the youth report was too small (no intervention ($n = 6$), intervention ($n = 2$)).

2.3.3. Construct validity and internal consistency

The correlation between the age of the participant and the VELO scores was evaluated using Spearman's rank order correlation (r_s). Finally, to re-evaluate the internal consistency of the Dutch version of the VELO questionnaire, Cronbach's α was calculated for the total questionnaire (parent report and youth report) and the subscales. A value between 0.70 and 0.95 was considered good [20].

3. Results

3.1.Reproducibility

Tables 2 and 3 provide an overview of the scores at the first and second time point for the parent report and the youth report respectively. At the second time point (T2), the median VELO score for the parent report was 79.3. This did not significantly differ from the median score of 77.4 at the first time point (T1) ($Z = 1.072$, $p = 0.284$, $r = 0.11$). This non-significant difference between the two time points was also found for the youth report ($Z = -0.400$, $p = 0.689$, $r = -0.08$). Subtraction of the scores on the parent report on T1 from those on T2 resulted in a mean difference of +0.4 (standard deviation (SD) = 5.28) and a median difference of +1.4. For the youth report a mean difference of -0.85 ($SD = 5.93$) and a median difference of 0.0 was obtained. Computation of a cumulative table with the absolute differences between the time points, showed that for the total score on both the parent report and the youth report, the difference was equal or less than 3 in 50% or more (26/50; 7/14) of the cases (table 4). ICC values of the total parent report and youth report, and their subscales, indicated excellent relative consistency (table 5). Moreover, the SEM was 3.58 for the parent report, and 4.00 for the youth report, resulting in MDD's of 9.92 and 11.09 for the parent report and the youth report respectively.

3.2.Responsiveness

The median value (Median (Mdn) = 77.9) of the parent report at the second time point, i.e. after one year, did not significantly differ ($Z = 1.146$, $p = 0.252$, $r = 0.14$) from the first response ($Mdn = 81.7$) (table 6). Subtraction of the VELO scores at T1 from those at the T2 resulted in positive mean and median differences (table 7). Using the MDD determined in 3.1. as a cut-off, a difference in VELO scores of 10 or more was found in 13 (37%, 13/35) participants (table 7). Ten of these 13 patients (77%, 10/13) had a positive evolution in HRQoL, of which four (40%, 4/10) received speech therapy. Of the three patients with a

Table 2: median, quartiles and range of scores on the parent report at the first time point and the second time point (mean time interval of 18.8 days) for the evaluation of the questionnaire's reproducibility

	First response (<i>n</i> =50)		Second response (<i>n</i> =50)	
	Median (Q1-Q3)	Min. – Max.	Median (Q1-Q3)	Min. – Max.
VELO Parent – Total	77.4 (66.35 - 91.35)	12 - 100	79.3 (64.42 – 92.79)	8– 100
VELO Parent – Speech	73.2 (49.11 - 83.04)	21 - 100	67.9 (50.00 – 90.18)	7 - 100
VELO Parent – Swallowing	100.0 (75.00 - 100.00)	8 - 100	91.7 (72.92 – 100.00)	8 – 100
VELO Parent – Situation	75.0 (48.75 - 95.00)	0 - 100	75.0 (43.75 – 95.00)	0 - 100
VELO Parent – Emotional	78.1 (60.94 - 100.00)	6 - 100	87.5 (68.75 – 100.00)	6 - 100
VELO Parent – Perception	93.8 (75.00 - 100.00)	25 – 100	100.0 (75.00 – 100.00)	6 – 100
VELO Parent – Caregiver	79.2 (66.67 – 100.00)	0 - 100	83.3 (66.67 – 100.00)	0 - 100

Table 3: median, quartiles and range of scores on the youth report at the first time point and the second time point (mean time interval of 17.6 days) for the evaluation of the questionnaire's reproducibility

	First response (n=14)		Second response (n=14)	
	Median (Q1-Q3)	Min. – Max.	Median (Q1-Q3)	Min. – Max.
VELO Youth – Total	90.8 (82.34 – 100.00)	34 – 100	90.2 (81.25 – 99.18)	35 – 100
VELO Youth – Speech	82.1 (71.43 – 100.00)	17 – 100	94.6 (74.11 – 100.00)	21 – 100
VELO Youth – Swallowing	100.0 (91.67 – 100.00)	50 – 100	100.0 (81.25 – 100.00)	42 – 100
VELO Youth – Situation	97.5 (77.50 – 100.00)	35 – 100	95.0 (75.00 – 100.00)	40 – 100
VELO Youth – Emotional	96.9 (81.25 – 100.00)	31 - 100	90.6 (67.19 – 100.00)	25 – 100
VELO Youth – Perception	100.0 (89.06 – 100.00)	50 - 100	100.0 (92.19 – 100.00)	50 – 100

Table 4: Cumulative frequencies (raw/%) for the differences on the VELO questionnaire between the first and second time point (i.e. two weeks later) of the parent report and the youth report for the evaluation of the reproducibility

VELO time point 2 – VELO time point 1	Parent report (<i>n</i> = 50)	Youth report total (<i>n</i> = 14)
0	4 / 8	3 / 21
≤ 1	12 / 24	6 / 43
≤ 2	20 / 40	6 / 43
≤ 3	26 / 52	7 / 50
≤ 4	33 / 66	9 / 64
≤ 5	36 / 72	9 / 64
≤ 6	38 / 76	9 / 64
≤ 7	41 / 82	10 / 71
≤ 8	44 / 88	11 / 79
≤ 9	46 / 92	12 / 86
≤ 10	48 / 96	13 / 93
≤ 11	48 / 96	13 / 93
≤ 12	49 / 98	14 / 100
≤ 13	49 / 98	
≤ 14	49 / 98	
≤ 15	49 / 98	
≤ 16	50 / 100	

Table 5: ICC (one-way random, single measures) values for the total parent report and youth report and their subscales

	ICC	95% CI ICC
Parent report (<i>n</i> = 50)		
VELO Parent - Total	0.967	0.943 - 0.981
VELO Parent - Speech	0.930	0.880 - 0.959
VELO Parent - Swallowing	0.851	0.753 - 0.913
VELO Parent - Situation	0.923	0.869 - 0.956
VELO Parent - Emotional	0.923	0.868 - 0.955
VELO Parent - Perception	0.844	0.741 - 0.908
VELO Parent – Caregiver	0.904	0.837 - 0.944
Youth report (<i>n</i> = 14)		
VELO Child – Total	0.951	0.859 - 0.984
VELO Child – Speech	0.941	0.833 - 0.981
VELO Child – Swallowing	0.748	0.395 - 0.911
VELO Child – Situation	0.846	0.589 - 0.947
VELO Child – Emotional	0.813	0.527 - 0.935
VELO Child – Perception	0.941	0.832 - 0.980

negative difference of 10 or more (23%, 3/13), 2 (67%, 2/3) received speech therapy. When observing all VELO scores differences of the parent reports ($n = 35$), 14 participants (40%, 14/35) had a negative difference (intervention in 7/14), 20 participants (57%, 20/35) a positive difference (intervention in 9/20), and one participant (3%, 1/35) had no difference.

The intervention group had a mean VELO score of 73.0 ($SD = 14.22$, 95% CI = 65.20 – 80.85) at T1 and 74.5 ($SD = 11.16$, 95% CI = 66.70 – 83.6) at T2, whereas the non-intervention group presented with higher scores of 82.3 ($SD = 18.97$, 95% CI = 74.66 – 89.87) and 85.09 ($SD = 17.95$, 95% CI = 77.48 – 92.69) at T1 and T2 respectively. Comparison between groups revealed a non-significant time-by-group interaction effect ($p = 0.699$), implicating no differences in the evolution of VELO scores between the intervention group and the non-intervention group.

3.3. Internal consistency and construct validity

Cronbach's α was 0.958 and 0.951 for the parent report and the youth report respectively (table 8). Furthermore, there was a significant positive correlation between the age of the participants and the results on the parent report ($r_s(71) = +0.275$; $p = 0.019$), indicating a more positive perception of HRQoL as indicated by the parents with increasing age. In contrast, a borderline significant negative correlation was found between age and the results on the youth report ($r_s(22) = -0.403$; $p = 0.051$).

4. Discussion

In an overall aim to provide evidence for the validity and reliability of the Dutch version of the VELO questionnaire, two consecutive studies were set up. In the current study, following

Table 6: median, quartiles and range of scores on the parent report at the first time point and the second time point (mean time interval of 11.9 months) for the evaluation of the questionnaire's responsiveness

	First response (<i>n</i>=35)		Second response (<i>n</i>=35)	
	Median (Q1-Q3)	Min. – Max.	Median (Q1-Q3)	Min. – Max.
VELO Parent – Total	81.7 (64.42 – 92.31)	23 – 100	77.9 (69.23 – 95.00)	39 – 100
VELO Parent – Speech	67.9 (53.57 – 85.71)	21 – 100	75.0 (60.71 – 85.71)	21 – 100
VELO Parent – Swallowing	100.0 (83.33 – 100.00)	17 – 100	100.0 (83.33 – 100.00)	50 – 100
VELO Parent – Situation	70.0 (50.00 – 95.00)	10 – 100	75.0 (55.00 – 100.00)	20 – 100
VELO Parent – Emotional	93.8 (75.00 – 100.00)	25 – 100	87.5 (68.75 – 100.00)	31 – 100
VELO Parent – Perception	100.0 (81.25 – 100.00)	19 – 100	100.0 (81.25 – 100.00)	50 – 100
VELO Parent – Caregiver	91.7 (66.67 – 100.00)	50 - 100	91.7 (75.00 – 100.00)	50 - 100

Table 7: Mean and median differences of the scores on the VELO questionnaire between time point 1 and 2 (i.e. one year later) and cumulative frequencies (raw/%) of the absolute differences for the evaluation of the questionnaire's responsiveness

	Parent report total (<i>n</i> = 35)	Youth report total (<i>n</i> = 8)
	Mean difference: +2.2 (<i>SD</i> = 9.82)	Mean difference: +4.4 (<i>SD</i> = 4.35)
	Median difference: 0.96	Median difference: 3.26
VELO time point 2 – VELO time point 1		
0	1 / 3	2 / 25
≤ 1	6 / 17	3 / 38
≤ 2	9 / 26	4 / 50
≤ 3	11 / 31	4 / 50
≤ 4	12 / 34	5 / 63
≤ 5	13 / 37	5 / 63
≤ 6	16 / 46	5 / 63
≤ 7	18 / 51	5 / 63
≤ 8	21 / 60	7 / 88
≤ 9	22 / 63	7 / 88
≤ 10	23 / 66	7 / 88
≤ 11	23 / 66	7 / 88
≤ 12	25 / 66	8 / 100
≤ 13	27 / 77	
≤ 14	29 / 83	
≤ 15	30 / 86	
≤ 16	32 / 91	
≤ 17	33 / 94	
≤ 18	34 / 97	
≤ 21	35 / 100	

Table 8: Internal consistency (Cronbach's α) of the parent report and the youth report of the Dutch VELO questionnaire

	Parent report (<i>n</i> = 73)	report (<i>n</i> = 24)
Total score	0.958	0.951
Speech	0.819	0.864
Swallowing	0.836	0.767
Situational difficulties	0.942	0.858
Emotional impact	0.858	0.887
Perception by others	0.887	0.773
Caregiver impact	0.870	N.A.

the study by Bruneel et al. [19], the questionnaire's reproducibility after a two-week interval, responsiveness, construct validity and internal consistency were (re-)evaluated in patients with an isolated cleft (lip and) palate.

For the evaluation of the reproducibility, several analyses were performed. The questionnaire showed excellent relative consistency, as all ICC's exceeded the 0.70 benchmark [20]. In combination with the descriptive data (mean and median differences between the two time points), and the results of the Wilcoxon signed-rank test, these ICC values indicate good reproducibility of the Dutch version of the VELO questionnaire. Regarding the absolute consistency, Terwee et al. [20] stated that the MDD should be smaller than the minimal important change (MIC). However, a limitation of this study was that no MIC was predefined. Skirko et al. [15] described an MIC of 15, calculated using an anchor-based approach based on the recompletion of the VELO questionnaire and a global rating of change in QoL by 37 patients after one year. Comparison of the MDD's obtained in our study (9.92 and 11.09) with this MIC of 15, suggests good absolute consistency of the questionnaire. Nevertheless, we do acknowledge the need to determine MIC's, and thus normative (Dutch) VELO scores for different groups of interest, e.g. after one year of treatment and for different age groups.

The underlying hypothesis for the evaluation of the responsiveness and the construct validity was that HRQoL is perceived more positively with increasing age, as a result of maturation on one hand and intervention on the other hand [19]. The positive correlation between age and the parent report provided evidence for this hypothesis and thus the construct validity. In contrast, a borderline significant, yet negative correlation was found between age and the youth report, strengthening the findings by Bruneel et al. [19]. This finding might be explained by an increasing awareness about the impact of speech and/or swallowing difficulties on functioning and well-being, as Hall et al. [4] pointed out based on qualitative interviews with children and adolescents with cleft lip and/or palate. Interestingly, these

authors also described the influence of intervention on this gradual awareness. However, responses at only one time point were included to determine the relationship between age and the youth report, and the responsiveness of the youth report was not analysed due to the small sample size, limiting the generalisability of these results. Analysis of the participants' evolution after one year at group level revealed no significant change in HRQoL, neither when the analysis was repeated for the intervention group nor for the non-intervention group. Observation of VELO score differences in both groups, suggest the influence of additional factors other than intervention and maturation alone. The VELO questionnaire qualifies several aspects of the ICF-model: body functions, activities and participation, and to a smaller extent, environmental factors [19, 24]. Using this ICF-framework, some other influencing factors can be identified. The parent report assesses the child's HRQoL as estimated by their parents. Hence, the parent's perspective, influenced by their context, beliefs, expectations, motivation, coping style and experiences, will hypothetically influence results as well [4, 25, 26]. The same factors associated with the patients may also play a role in the evolution in HRQoL [4, 27-29]. As such, identification of contextual factors that influence the patient's functioning and well-being, and specifically their evolution of HRQoL, is subject for further research. Furthermore, it takes perhaps more than one year, or secondary speech surgery [15, 16] to detect a significant evolution in HRQoL perception.

In this study, only patients with an isolated cleft (lip and) palate were included, and no indications of VPI were taken into account, as the authors argued the VELO questionnaire to be applicable for all patients with cleft palate, based on the results by Bruneel et al. and the content of the items. As only patients with an isolated cleft (lip and) palate were included, the influence of an additional condition such as a specific language impairment on HRQoL results was minimized. However, as pointed out by Feragen et al. [30], excluding patients with syndromes or additional conditions might cloud our general understanding of the cleft palate

population. Future studies providing normative data for patients with cleft palate with or without VPI, and patients with additional conditions, will provide insight in the overall population of patients with cleft palate, and the influence of additional conditions on HRQoL. To compose such normative data, the inclusion of greater sample sizes is needed. In the future, we aim to set up multicentre studies. This not only allows for greater sample sizes, but also reduces the bias of including participants of only one centre, given that e.g. the experiences of the parents and the patients can be coloured by the approach of one specific centre. Nevertheless, the inclusion of a greater sample size than the previous study by Bruneel et al. [19], enabled a more reliable determination of the questionnaire's internal consistency using Cronbach's α . Results confirmed the conclusion of this previous study, namely excellent internal consistency of both the total questionnaire and the subscales of the Dutch version of the VELO questionnaire.

A valid and reliable instrument that assesses the patient's HRQoL, and more specifically for the VELO questionnaire, the influence of speech and swallowing difficulties on functioning and well-being, can lead the way to a more patient-centred approach. By incorporating the VELO questionnaire as part of the routine assessment of patients with cleft palate in the SLP practice, a more holistic perspective instead of a symptomatic approach can be achieved. This can eventually aid clinical decision-making, e.g. taking into account the patient's perspective when deciding on the continuation of speech therapy, and improvement of (therapeutic) outcomes, e.g. evaluating whether speech therapy or secondary speech surgery improves the patient's HRQoL.

5. Conclusion

The VELO questionnaire shows good reproducibility and internal consistency. Furthermore, there was a positive correlation between age and the parent report, and a negative correlation between age and the youth report. After one year, no significant VELO score differences were

found, neither when the analysis was repeated for the intervention group and the non-intervention group separately. Based on these results, the identification of additional factors influencing the evolution of HRQoL is subject for future research. The aim of using a valid and reliable instrument to assess HRQoL in clinical practice, and to evaluate outcomes and service delivery, is a patient-centered approach, improving the patient's HRQoL and quality of care.

6. Acknowledgments

This work was supported by a PhD fellowship of the Research Foundation Flanders (L.B.).

7. References

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