

Universiteit Utrecht Opleiding MSc Logopediewetenschap

Clinical Language, Speech, and Hearing Sciences

Master's Thesis

The Value of Videofluoroscopy in the Clinical Assessment of Velopharyngeal Insufficiency

A.K. Miglo 3966887

Supervision: Dr. C.C. Breugem Dr. D.M.A. Kamalski

30 september 2014

Abstract

Introduction. Velopharyngeal insufficiency (VPI) is a complex abnormality and should be assessed carefully. Well-executed visualizing assessments such as nasendoscopy and videofluoroscopy can provide valuable information for further treatment decisions, if the four goals of velopharyngeal assessment are met (structure, movement, extent of closure, and timing). Nasendoscopy is the reference standard of visualizing instruments in VPI diagnostics, although it cannot provide objective values. One crucial advantage of videofluoroscopy is the possibility of obtaining measurement ratios and reaching objective and comparable assessment outcomes. However, in clinical practice it is not clear what value videofluoroscopy has in VPI assessment next to nasendoscopy, in terms of usability, the four goals and decisions of further treatment.

Methods. In three meetings specialists of the CLPT analyzed retrospective material, videofluoroscopic recordings (n=22), on usability, the four goals and further treatment decisions. Inclusion of participants was based on availability of videofluoroscopies. Paired nasendoscopies (n=15) were rated based on the same criteria. Recordings declared as usable were graded on the four goals based on severity scales. Treatment decisions were also made based on patients' history, speech components and recordings. The ratings were summarized and represented in percentages. In subgroup analyses usable videofluoroscopies and nasendoscopies were set in relation to the four goals and treatment decisions.

Results. Based on the quality of the recording 12/22 videofluoroscopies and 4/15 nasendoscopies were not to moderately usable. In videofluoroscopies the majority is not to mildly abnormal but proportionally more surgical treatment decisions were made.

Discussion. Recordings included in this study did not have an added value in the VPI assessment. They may even result in unnecessary burdening surgeries. However, to support this statement, more research is needed. For prevention of unusable recordings research and clinical practice should concentrate on standardization of videofluoroscopic assessment.

Introduction

The term velopharyngeal insufficiency (VPI) describes an incomplete closure of the velopharyngeal sphincter due to an inadequate movement of the velum. The lateral and posterior pharyngeal walls cannot compensate a velum that is too short or not strong enough (1,2).

As a result, the nasal cavity cannot be separated completely from the oral cavity and patients' speech can be influenced by 'stigmatizing speech abnormalities' (2), such as hypernasality, nasal emission, nasal turbulence and facial grimacing (3).

VPI is commonly based on anatomical, iatrogenic or neuromuscular factors (4). In most cases VPI occurs in patients with repaired cleft palates (incidence of 15-20%) but can also be found in other conditions such as neuromuscular dysfunctions or syndromes, e.g. velocardiofacial syndrome (VCF; 2, 5, 6, 7). Despite the cause, VPI should be evaluated and managed carefully for effective intervention. Before and after treatment, measurement of VPI should be used to determine the severity of VPI and to monitor effectiveness of the treatment. However, assessment of velopharyngeal closure can be complex and challenging due to the highly complex mechanisms in the velopharynx and a large variance between patients (8).

Perceptual assessment is essential to determine the severity of VPI. There is a significant positive correlation between the velopharyngeal gap size and the severity of the hypernasality, enabling experienced speech and language therapists (SLTs) to predict the presence and approximate size of the gap based on auditory perception (3, 9). However, perceptual assessment does not provide objective outcomes and results may differ between different SLTs (9). An objective instrument that is often used by SLTs is therefore the nasometer (11). This instrument is used to detect hypernasality by measuring the nasal and oral airflow during speaking and singing (11, 12). The nasometer is consequently a valuable assessing instrument but should not be the only objective instrument used. Although it can give an objective impression of hypernasality, it cannot show the structures of the velopharynx and its outcome depends on the child's cooperation (12).

For this reason velopharyngeal imaging instruments are used in order to objectify the diagnostic procedure. Several diagnostic instruments are used to measure the size and characteristics of the velopharyngeal gap objectively. The most common instruments are videofluoroscopy, naseondoscopy and the latest development, Magnetic Resonance Imaging (MRI).

VIDEOFLUOROSCOPY

The movement of the velopharynx during speech can be analyzed with a radiographic study. A high-contrasting liquid injected into the velum or applied through the nostrils makes the patterns of movement visible during a radiographic examination (8, 9, 10). This assessment is usually performed in lateral and anteroposterior view (9).

A considerable advantage of videofluoroscopy over other assessing instruments is that it provides an objective view of the velopharynx and makes quantitative measurement of the velopharyngeal gap size possible. Mehlendale et al.

(2004) reproduced a possible way of objectifying the velopharyngeal gap size by measuring and calculating the gap size area (13). A well-executed videofluoroscopy can therefore provide objective ratio values. In the anteroposterior view an assessment of lateral wall movement and the detection of submuceus gaps is possible (16).

In many cases, specialists prefer videofluoroscopy over nasendoscopy as videofluoroscopy is less invasive and requires less cooperation from the child than nasendoscopy and magnetic resonance imaging (MRI; 2, 9).

A disadvantage of this technique is that, videofluoroscopy only provides a two-dimensional view. An analysis of closure patterns is harder to make (2, 9, 14). Also, the exposure of radiation in children should be avoided as much as possible because they react more sensitively than adults (8).

NASENDOSCOPY

A flexible endoscope is used to visualize the inside of the velopharyngeal cavity. By inserting the scope through the nasal passage to the pharynx, a view of the velum, the sagittal and lateral pharynx is provided. This allows a direct visualization of the structures and movement of the velopharynx (2, 9).

Closure patterns and direction of motion of the velopharynx in speech can be established by executing a nasendoscopic assessment. In addition, an estimation of the defect size can be made (9).

In comparison with videofluoroscopy, nasendoscopy enables an analysis of the velopharyngeal structures and shows a slightly higher correlation with the perceptual assessment than the videofluoroscopy (2).

On the other hand, a nasendoscopy is an unpleasant assessment and the child's cooperation is necessary (9). The quantitative assessment of velar length and depth of the pharynx can be affected negatively by distortion and changes in perspective because of movements of the scope (2, 9). Additionally, the measurement of structures and sizes is restricted due to distortions (8). It is expected that this instrument cannot be used in all cases because it is quite uncomfortable and could possibly be counter-indicated in several children due to anatomical or physiological problems such as a narrow nasal passage.

MAGNETIC RESONANCE IMAGING (MRI)

The MRI scan provides high resolution and good quality images at different times of nasopharyngeal closure, resulting in a quantitative measurement of the velopharyngeal gap size (1). It is preferred over the nasal endoscopy by children and is less invasive than videofluoroscopy and nasendoscopy (1).

In recent years it has become possible to realize dynamic visualization during audio recording but there are still significant technical problems, which challenge the synchronization of pictures and sound (1, 4, 15).

The current technical problems have a significant impact on the sensitivity and specificity of the instrument. Moreover, the high cost and limited time-efficiency are reasons to exclude MRI from the analysis (1, 4, 15).

In summary, nasendoscopy and videofluoroscopy are both dynamic visualization instruments, giving complementary information. Both quantitative data and closure patterns can be assessed (2, 10, 16).

Both instruments have advantages and disadvantages in application and interpretation. Sphrintzen & Kushner-Golding (1989) suggested the use of *both* assessing instruments to combine all information needed for an indication of further therapy (17). However, there are few medical centers which implemented this advice (15).

Since Pigott et al. (1969) introduced nasendoscopy into the diagnostic procedure of velopharyngeal insufficiency, it became the so-called 'golden standard' and most medical centers extract their information from nasendoscopy alone (15, 16). Currently, it is not clear whether it is possible, let alone necessary, to perform both examinations. A study by Havstam et al. (2005) even alleges that a complete picture of the VPI is not required for further clinical decisions but rather an individualized instrumental assessing procedure (8). This claim is based on the statement that instrumental measurement should provide information about the four basic parameters or goals of velopharyngeal measurement: structure, movement, extent of closure and timing (8,18). The structure of the velopharynx varies between individuals with and without VPI. This variability can have an influence on the outcome of surgical treatment. Velopharyngeal movements have a substantial influence on the decision of treatment and should be taken into account during assessments. The extent of closure correlates with perceived velopharyngeal insufficiency but incomplete closure does not guarantee symptoms such as hypernasality (3,18). The last component, which should be taken into account, is the timing. All velopharyngeal structures should move in a coordinated fashion and reach closure at the same time to prevent VPI. When analyzing visualizing assessing instruments, those four goals can be used to obtain sufficient information for further therapy decisions. In this decision-making process, objective values such as ratio calculations could be very valuable (2,13,14). These calculations however depend on the quality of videofluoroscopic recordings. Therefore, it is important to investigate which role videofluoroscopy plays in the clinical reality of diagnostic procedures of VPI next to the 'golden standard' nasendoscopy and to compare the two assessments not only in terms of assessing quality but also with regard to diverse patient populations and clinical decisions that follow. Based on this goal the following question will be answered: What value does the videofluoroscopy have in addition to nasendoscopy in the assessment and clinical decision-making process of velopharyngeal insufficiency?

Methods

A retrospective observational medical files study was performed and recordings of patients' videofluoroscopic and nasoendoscopic assessments were analyzed with regard to usability, reaching the goals of velopharyngeal assessment and decisions for further treatment. Anonymous assessment was performed by a plastic surgeon, an otolaryngologist and speech and language therapists.

PARTICIPANTS

Medical files of children with VPI collected by the Cleft Lip and Palate Team (CLPT) of the Academic Medical Center Utrecht (UMC Utrecht) between November 2010 and February 2014 were analyzed. All available recordings of lateral videofluoroscopic assessment based on VPI were evaluated in this study (n=22). The policy in the institution was that nasoendoscopy is performed in patients with VPI. In patients where an inconclusive or unsatisfactory result is seen after nasendoscopy, a videofluoroscopy was performed. Children included in this study were between the ages of 3 and 18 with VPI and registered in the digital dossier of the UMC Utrecht. As VPI cannot be diagnosed before the age of 3, children under the age of 3 have not been assessed (19). Exclusion was based on missing videofluoroscopic recordings.

AVAILABLE DATA

The following patient historic data were analyzed: age, primary diagnosis and previous surgeries. Speech components assessed by 1 out of 3 SLTs contained data about hypernasality, intelligibility and phonetic speech abnormalities (modified speech sounds, nasal emission, nasal turbulence, compensatory articulation), which were assessed following a standardized assessment of cleft lip and palate (Nederlandstalig Diagnostisch Schisisonderzoek, (20)).

ASESSMENT PROCEDURE

As the initial grading of nasendoscopies and videofluoroscopies was not based on standardized criteria and executed by variable disciplines, an expert team of the CLPT reanalyzed all available recordings. The usability of each recording was judged as usable, moderately usable, or not usable for this analysis of the four goals and the decision of treatment (see also table 2.). When usable or moderately usable, the specialists were asked to find consensus about participants' abnormality rating on the four goals (none, mild, moderate, or severe abnormality) and to make decisions about further treatment and patients' history without information about previous nasendoscopies or videofluoroscopies. The possible treatment decisions were: expectancy, speech therapy, surgery or, in case of doubt, no decision, based on this assessment. The evaluation form of this assessment can be found in appendix 1.

Videofluoroscopy

In order to recreate the clinical situation an inter-disciplinary consult was conducted by the following specialists; a plastic surgeon, an otolaryngologist and two speech therapists. During these meetings (n=3 over a period of seven weeks), all available videofluorscopic recordings (n=22) were reviewed and judged on usability, the four goals based on the word level part of the recording. Additionally, they were asked to find consensus about further therapy decisions based on the whole recording (word and sentence level).

During the first session, after executing one randomly chosen pilot instructed by the researcher, all disciplines were asked to assess the available videofluoroscopies and to find consensus. The pilot recording was randomly reincluded into the analysis.

Nasendoscopy

Nasenodscopic recordings contained visualizations of repeated words or spontaneous speech. In one session, the otolaryngologist of the CLPT analyzed all available nasendoscopic recordings (n=14) following the same criteria on usability, the four goals and further therapy decisions based on complete recordings and patients' history. To prevent possible biasing factors, this otolaryngologist was not present during the analysis of videofluoroscopic recordings.

ANALYSIS

This thesis is observational and descriptive. All data were collected and summarized in order to calculate percentages of usability, the four goals and treatment decisions. In a subgroup analysis of participants who had undergone both assessments (n=10), the agreement of all four goals was calculated. For this calculation the outcome was ranked in two groups; the number of children, whose VPI was classified as not or mildly abnormal in structure, movement and closure, was added to the number of children without timing abnormalities. The same procedure was executed in participants with moderate and severe abnormalities in structure, movement and closure and present timing abnormalities. Thus, two comparable groups of both assessments were created.

The decision of treatment was also analyzed in a subgroup analysis (n=12). The inclusion into this calculation was based on availability of nasendoscopies and on the possibility of grading the recordings.

Results

PARTICIPANTS

Participants' clinical history is represented in table 1. Children participating (n = 22) ranged between the age of 3,5 and 16,10 years with an average of 7,7 years.

Besides the primary diagnosis (see table 1) 18 out of 22 children had comorbidities such as developmental delay or multiple syndromes. All participants underwent a videofluoroscopic assessment based on uni- or bilateral cleft palate (n=7), submuceus cleft (n=5), VCF (n=7), developmental delay (n=1) and hypernasality (n=2).

Of those who underwent VPI surgery, 4 out of 7 also had VPI-related speech therapy.

Previous VPI surgeries included intravelar veloplasties (n=2), augmentations (n=2), lipofillings (n=1), levatorplasties (n=1) and

Table 1. Participants' history							
Particip	Participants (n = 22)						
Age							
average	7,7						
range	3,5-16,10						
Primary diagnosis							
uni/bilateral cleft palate	7						
submuceus cleft	5						
VCF	7						
VPI + developmental delay	1						
hypernasality (NOS)	2						
Previous speech related treatments							
none	7						
previous speech therapy	8						
surgeries							
no surgeries	11						
primary cleft surgeries	9(x=18)						
VPI surgeries	7 (x=9)						
other speech related	4 (x=5)						
surgeries							

NOS = Not Otherwise Specified x: number of surgical procedures

T 11 1 D

...

pharyngoplasties (n=4). Other surgeries with an influence on speech (speech related surgeries) included two tonsillectomies, one frenulectomy and one adenotomy.

Speech components

Speech components were extracted out of SLTs reports, written during the previous diagnostic procedure. The majority of participants showed no to mild hypernasality (13 out of 20 impressions of hypernasality), whereas 4 participants were not classified as hypernasal.

Parents (n=13) and SLTs (n=21) rated intelligibility of participants (see also (20)). These data are ranged on an ordinal scale from 1 (no audible abnormalities in speech) to 5 (not intelligible). The median of the parental impression of intelligibility is 2.5 (SD=0.8), which could be interpreted as 'the speech differs from other children's speech, sometimes evokes comments but is intelligible'. SLTs judgment met on the median of 3 (SD=0.6), which is slightly higher than the parents' and can be interpreted as 'audible disorder, to some extent intelligible'.

Phonetic speech abnormality is reported in 20 out of 22 children. One participant without hypernasality and phonetic speech abnormalities was included in

the videofluoroscopic assessment for unknown reasons. In appendix 2 more detailed information about speech components is reproduced.

ASSESSMENTS

Fifteen out of 22 participants had undergone a nasendoscopy as well as a lateral videofluoroscopy. An otolaryngologist assessed these 15 nasendoscopies. The 22 lateral videofluoroscopies were assessed by the CLPT team. The results will be described in more detail below.

USABILITY

In table 2 the usability of the videofluoroscopy and nasendoscopy is shown.

Videofluoroscopy

Four out of 22 videofluoroscopies (18%) were not usable in order to analyze the four goals. In 2 cases, the specialists were able to make a decision about further treatment based on their impression on word and sentence level. Reasons for non-usability were technical problems (during word level recording), agility of the head, rotated head positions and chin down positions during the recordings. Videofluoroscopies were classified as moderately usable (n=8; 36%) when there were head movements or technical problems such as dark recordings, but the specialists could analyze the recordings based on the four goals and make decisions about further treatment. Ten out of 22 recordings (46%) were usable. However, during the assessment of all recordings the specialists missed reference recordings of normally functioning veli in children to be able to grade abnormalities. Based on technical problems, recordings were not usable for objectifying calculations as presented by Lipira (2011) et al., Lam et al. (2006) or Mehendale et al. (2004) (2, 13, 14).

Nasendoscopy

Three nasendoscopies (21%) could not be used for this assessment because of technical problems during the recording or limited length of the recordings. The otolaryngologist graded recordings as moderately usable (n=1; 7%) when impaired by the same factors but when an analysis of recordings based on the four goals and decision of further treatment was possible. Ten nasendoscopies (72% out of the 14 available) were classified as usable; this provides 46% usable nasendoscopies from a total of 22 participants.

		Table 2. Out	come assessmei	nt videoflour	oscopy and	d nasendosc	ору				
		Videofluoros	сору					Nasendoscopy			
	n (%)						n (%)				
Usability		-	±	+				-	±	+	
		4 (18)	8 (36)	10 (46)				3 (21)	1 (7)	10 (72)	
	n=18						n=11				
Goals											
		None	Mild	Moderate	Severe	Not reliable		None	Mild	Moderate	Severe
Structure		7 (39)	6 (33)	1 (6)	3 (17)	1 (5)		3 (28)	2 (18)	4 (36)	2 (18)
Movement		6 (33)	3 (17)	6 (33)	3 (17)	-		-	6 (55)	4 (55)	1 (9)
Closure		5 (28)	4 (22)	6 (33)	3 (17)	-		-	6 (55)	4 (36)	1 (9)
		No abnormality	Abnormality	Not reliable				No abnormality	Abnormality		
Timing		9 (50)	6 (33)	3 (17)				4 (36)	7 (64)		
Treatment	n=20						n=13				
		SLT	Surgery	No decision				SLT	Surgery	No decision	
		6 (30)	7 (35)	7 (35)				5 (38.5)	5 (38.5)	3 (23)	

THE FOUR GOALS

The outcome of the assessment of the four goals using videofluoroscopy and nasendoscopy is reproduced in table 2.

Structures

The nasendoscopic recordings were almost evenly distributed in no to mild (46%) and moderate to severe abnormalities (54%). In contrast, the majority of videofluoroscopies structural abnormalities were classified as none to mild abnormalities (72%). In one videofluoroscopy the disciplines could not reliably assess the recording because structures were affected by previous surgeries.

Movement

Outcomes of videofluoroscopic moving assessments were equally distributed in no to mild and moderate to severe abnormalities. In 6 nasendoscopic assessments the classification of movement was graded in no to mild abnormalities (55%) whereas all 6 recording were mildly abnormal. In contrast, the CLPT graded the movement in 6 videofluoroscopies (33%) as normal. When comparing the classifications of severe abnormal movements, the following findings are noted; 3 cases were rated as severely abnormal (17%) in videofluoroscopy compared to 1 rating in nasendoscopic recordings (9%).

Closure

The degree of closure was almost equally distributed in no to mild and moderate to severe abnormality within the videofluoroscopy (50/50%) and the nasendoscopy (55/45%).

In three videofluoroscopies, the CLPT team detected compensatory movements from the posterior pharynx. In 2 out of these 3 recordings participants reached complete closure because of compensatory movement; semicolon one recording was classified as moderately abnormal.

According to the specialists, movement and closure correlate strongly which could be an explanation for these equivalent outcomes.

Timing

The timing could not be classified on the same scale that has been used for structure, movement and closure. Therefore, the scale of timing is rated as abnormality – no abnormality. In 9 videofluoroscopic recordings (50%), the timing was graded as normal, whereas the group with normal timing in nasendoscopic recordings was much smaller (n=4; 36%). The amount of participants with an abnormal timing was comparable in both groups.

Three videofluoroscopic recordings (17%) could not reliably be classified because of abnormal structures in two cases and the quality of the videofluoroscopic assessment in one case. During this recording, the participant read out the words quickly, which made judgment of timing on word level impossible.

Table 3. Subgroup analysis on the four goals									
	Videof	luoroscop	y	Nasendoscopy					
n=10 (%)									
	None	Mild	Moderate	Severe	n.r.	None	Mild	Moderate	Severe
Structure	4 (40)	2 (20)	1 (10)	2 (20)	1	2 (20)	2 (20)	4 (40)	2 (20)
Movement	3 (30)	3 (20)	2 (20)	2 (20)	-	_	6 (60)	3 (30)	1 (10)
Closure	3 (30)	3 (30)	2 (20)	2 (20)	-	_	6 (60)	3 (30)	1 (10)
	No abn	ormality	Abnormali	ty	-	No abn	ormality	Abnormalit	у
Timing	5 (50)		5 (50)		-	3 (30)		7 (70)	

n.r.: not reliable

Agreement of the four goals

The four goals were analyzed in a subgroup of children who had undergone videofluoroscopy and nasendoscopy and both assessments were classified as usable (n=10). In table 3 the outcome of this subgroup is reproduced. The specialists of the CLPT could not rank the timing of one child included into this analysis because of the length of the recording on word level. This participant was nevertheless included into the subgroup analysis in order to preserve all available data.

Outcomes of videofluoroscopy and nasendoscopy were ranked into no abnormality and abnormality and reproduced as percentages (table 4). During the analysis of the four goals, videofluoroscopy classified the majority of participants as having no velopharyngeal abnormality whereas nasendoscopy finds more abnormalities in participants' velar structures, movement, closure and timing.

Table 4. Subgroup analysis outcome goals						
	No abnormality	Abnormality	Not reliable			
n=10						
Videofluoroscopy	58%	40%	2%			
Nasendoscopy	47,5%	52,5%	-			

DECISION OF TREATMENT

During this analysis, whole recordings combined with patients' history decisions were used in order to make a decision about further treatments.

Videofluoroscopy

It is noticeable that the CLPT could not make a decision about further treatment in 35% of all cases (n=7). In 6 participants instrumental reevaluation was recommended using nasendoscopy (n=4), videofluoroscopy (n=1) or a combination of both instruments (n=1). The specialists made the treatment decisions based on the wishes of the participant's parents due to the child's young age and unawareness of its VPI. The majority of recordings however led to a decision of SLT (n=6; 30%) and surgery (n=7; 35%).

Nasendoscopy

The otolaryngologist of the CLPT could make a treatment decision in 77% (n=10) of all recordings. Recordings were equally distributed in surgical and SLT interventions (n=5, 38,5% in both groups).

In 3 nasendoscopies (23%) no decision could be made. In one case, the specialist chose not to decide without consulting with the surgeon of the CLPT on possible surgical options and in two recordings, she asked for an instrumental assessment, one nasendoscopy and one videofluoroscopy based on the lack of cooperation of the child. This participant has also been excluded from the assessment of the four goals.

Agreement of treatment decisions

In two children the specialists could not give a statement about the four goals but based on the whole recording they could find consensus about further clinical treatments. For this reason, 12 children were included into the subgroup analysis of treatment decisions of both instruments (table 5.). Although the videofluoroscopy led to a smaller number of clinical decisions than the nasendoscopy, it also tends to lead to more surgical decisions than to SLT.

Table 5. Subgroup analysis on the decision of treatment								
	Video	fluoroscopy	Nasendsocopy					
n=12 (%)								
	SLT	Surgery	No decision	SLT	Surgery	No decision		
	2	4 (33)	6 (50)	5 (41,5)	5 (41,5)	2 (17)		
	(17)							

Discussion

In this paper the value of videofluoroscopy in addition to nasendoscopy was evaluated. The results of this study showed that a substantial part of videofluoroscopies was not usable and the scoring of the four goals presented by Karnell & Seaver (1990) was graded less severely in the videofluoroscopy than in the nasendoscopy, but lead to more surgical decisions (18). Therefore, at this stage of clinical practice, the videofluoroscopy has no added value in the assessment of VPI.

In videofluoroscopies, the group with no to mild abnormalities in structures, movement, degree of closure and timing prevailed. However, even though the distribution between no to mild abnormality and moderate to severe abnormality was more equal in the group of nasendoscopies, the majority of nasendoscopic recordings were graded as moderately to severely abnormal. This is in conflict with the study of Lipira et al. (2011) who found a higher percentage of closure in videofluoroscopy than in nasendoscopy (2).

Due to a great diversity between participants and qualitative deficiencies based on the lack of standardization of videofluoroscopic assessments a substantial part of videofluoroscopies was graded as not usable (18%) or only moderately usable (36%).

The decision of treatment in both assessments was equally distributed between SLT and surgery but subgroup data show that the CLPT advises more surgical than speech and language treatment unlike the study of Havstam et al. (2005), which proved agreement of both assessments irrespectively the amount of information (8). It is conceivable that missing reference recordings led to the impression of more structural abnormalities and therefore more surgical decisions.

This outcome is based on a multidisciplinary grading of all available videofluoroscopies made in the UMC Utrecht. All disciplines were experienced specialists. Biasing factors such as previous treatment decisions based on medical files or present specialists during both examinations were eliminated. The grading system was based on standardized point scales, which were used during the analysis throughout. The inclusion of information about usability and further treatment decisions in combination with the rating of the four goals made the outcome of this study suitable for clinical specialists in daily practice of videofluoroscopic assessment of VPI.

Nonetheless, the outcomes of this study should be interpreted with caution because retrospective data have been analyzed and the assessed groups were small (n=22) and particularly diverse. Children followed different assessing procedures, which led to different group sizes. For these reasons significant information could have been missed or influenced by factors not included. Another missing factor was the anteroposterior view of videofluoroscopic recordings. Henningsson et al. (1991) advise both, the anteroposterior and the lateral view for an optimal diagnostic result (16). The lack of anteroposterior viewing direction could have altered the outcome of this study because of missing information about lateral wall movements (12).

The analysis procedure could have been affected by the fact that the otolaryngologist who assessed the nasendoscopies did not have the input of other specialists. During the analysis of videofluoroscopies, the specialists were cautious in

grading severity rates without having reference recordings of normally functioning veli in children. Another factor was the lack of experience of the CLPT in grading videofluoroscopies compared to the nasendoscopies, the otolaryngologist executed on a daily basis. Based on the quality of videofluoroscopic recordings it is not possible to measure the velopharyngeal gap size and calculate ratios, which makes objectifying the outcome impossible. Furthermore, because of missing references, in 7 out of 18 assessments, the CPLT team could not make a clinical decision based on videofluoroscopic recordings and the patients' history.

Another interesting limitation of this study was that 4 children without resonance abnormalities were included in a videofluoroscopic assessment, which is, amongst others, intended to extract causes of hypernasality.

Despite the limitations of this study, the implementation of the videofluoroscopy should be explored because it can provide objective values, is less invasive and requires less cooperation from children than the nasendoscopy (2, 9, 13, 14).

In order to prevent the unnecessary burden of assessments and treatments, research should therefore not only be concentrated on the standardization of videofluoroscopic calculations but also the recording procedures and assessment protocols. Based on a new protocol a prospective cross-sectional study should be executed by multiple medical centers to evaluate norm data of videofluoroscopic recordings and a standardized assessing procedure. Recommendations for the implementation of videofluoroscopy can be found below.

CONCLUSION

On the basis of this study the application of videofluoroscopy does not seem to have an added value in the diagnostic procedure of VPI. A standardization of videofluoroscopic assessment and the presence of reference recordings could provide great improvement in the clinical diagnostic procedures of VPI.

RECOMMENDATIONS

For the production of reference recordings and the assessment in the clinical practice the following aspects are recommended:

1. Prevent head movements and adverse head positions (rotated or chin up/down position)

Havstam et al. (2005) used a cephalostat in an upright position in order to immobilize the head and to prevent chin up or down positions (8).

Another option, which could immobilize the head movement, is the ViewmasterTM because children look through glasses and see pictures of the target words (2, 22). This way the assessment could be interpreted as a game thus allowing the child to remain focused.

2. Consider posteroanterior videofluoroscopic recordings

Even though the lateral view of videofluoroscopic recording is more common, posteroanterior recordings are advised by Henningsson et al. (16) This view can provide further insights over pharyngeal wall moving patterns which should be taken into account when making treatment decisions.

3. Use a standardized word and sentence sequence

Not only to provide comparable reference recordings of normal velar movement but also to be able to compare in clinical practice, standardized word lists should be consequently used during the videofluoroscopic assessment. An English criteria list, which is used in a moderated form by Lipira et al. (2011) is the Pittsburgh Weighted Speech Scale (PWSS), and could be a suggested list (2, 21). The Dutch criteria list, which is already used in the clinical practice is another option (20).

4. Prevent technical problems

In 3 out of 22 videofluoroscopic recordings, no assessment could be conducted because of technical problems as dark or diffuse visualizations. If possible radiologists should prevent these technical problems.

Acknowledgement

Using this opportunity, I would like to express my gratitude to everybody supporting me in this project.

I would like to thank Drs H de Wilde, A.M.B. van der Heul, MSc, Dr. D.M.A. Kamalski and Dr.C.C. Breugem for their time and inspiration during three long afternoons scoring the videofluoroscopies. Dr. K.P.Q. Oomen was a great help when scoring the nasendoscopies.

I am very thankful for their welcoming and inspirational guidance during this project and their support with the realization throughout.

References

- 1. Maturo, S., Silver, A., Nimkin, K., Sagar, P., Ashland, J., Kouwe, van der, A. J. W., Hartnick, C. (2012). MRI With Synchronized Audio to Evaluate Velopharyngeal Insufficiency, *The Cleft Palate Craniofacial Journal*, 49(6), 761-763.
- Lipira, A. B., Grames, L. M., Molter, D., Govier, D., Kane, A. A. & Woo, A. S. (2011). Videoflouroscopic and Nasendoscopic Correlates of Speech in Velopharyngeal Dysfunction. *The Cleft Palate Craniofacial Journal*, 48 (5) 550-560.
- 3. Kummer, A. W., Briggs, M. & Lee, L. (2003). The Relationship Between the Characteristics of Speech and Velopharyngeal Gap Size. *Cleft Palate-Craniofacial Journal*, 40 (6), 590-596.
- 4. Ruda, J. M., Krakovitz, P. & Rose, A. S. (2012). A Review of the Evaluation and Management of Velopharyngeal Insufficiency in Children. *Otolaryngologic Clinics of North America*, 45, 653-669.
- 5. Ma., L., Shi, B., Li, Y. Zheng, Q. (2013). Velopharyngeal Function Assessment in Patients with Cleft Palate: Perceptual Speech Assessment Versus Nasopharyngoscopy. *The Journal of Craniofacial Surgery*, 24(4), 1229-1231.
- 6. Rudnick, E. F. & Sie, K. C. (2007). Velopharyngeal Insufficiency: Current Concepts in Diagnosis and Management. *Current Opinion in Otolaryngology & Head Neck Surgery*, 48(5), 530-535.
- 7. Losken, A., Williams, J. K., Burstein, F. D., Malick, D. N., & Riski, J. E. (2006). Surgical Correction of Velopharyngeal Insufficiency in Children with Velocardiofacial Syndrome. *Plastic and Reconstructive Surgery*, 117, 1493-1498.
- 8. Havstam, C., Lohmander, A., Persson, C., Dotevall, H., Lith, A. & Lilja, J. (2005). Evaluation of VPI-Assessment with Videofluoroscopy and Nasoendoscopy. *British Journal of Plastic Surgery*, 58, 922-931.
- 9. Woo, A. S. (2012). Velopharyngeal Dysfunction. *Seminars in Plastic Surgery*, 26 (4), 170-177.
- 10. Dalston, R. M., Warren, D. W. & Dalston, E. T. (1991). Use of Nasometry as a Diagnostic Tool for Identifying Patients with Velopharyngeal Impairment. *Cleft Palate Craniofacial Journal*, 28 (2), 184-189.

- 11. Heijden, van der, P., Hobbel, H. H. F., Laan, van der, B. F. A. M., Korsten-Meijer, A. G. W. & Goorhuis-Brouwer, S. M. (2011). Nasometry normative data for young Dutch children. *International Journal of Pediatric Otorhinolaryngology*, 75, 420-424.
- 12. Mehendale, F. V., Birch, M. J., Birkett, L., Sell, D. & Sommerlad, B. C. (2004). Surgical Management of Velopharyngeal Incompetence in Velocardiofacial Syndrome. *Cleft Palate-Craniofacial Journal*, 41 (2).
- 13. Lam, D., Starr, J. R., Perkins, J. A., Lewis, C. W., Eblen, L., E., Dunlap, J. & Sie, K. C. Y. (2006). A Comparison of Nasendoscopy and Multiview Videofluoroscopy in Assessing Velopharyngeal Insufficiency. *Otolaryngology Head and Neck Surgery*, 134, 394-402.
- 14. Shprintzen, R. J. & Marrinan, E. (2009). Velopharyngeal Insufficiency: Diagnosis and Management. *Current Opinion in Otolaryngology & Head and Neck Surgery*, 17(4), 302-307.
- 15. Pigott, R. W. & Makepeace, A.P. (1982). Some Characteristics of Endoscopic and Radiological Systems used in Elaboration of the Diagnosis of Velopharyngeal Incompetence. *British Journal of Plastic Surgery*, 35, 19–32.
- 16. Henningsson, G. & Isberg, A. (1991). Comparison between Multiview Videofluoroscopy and Nasendoscopy of Velopharyngeal Movements. *The Cleft Palate-Craniofacial Journal*, 28, 413–417.
- 17. Shprintzen, R. J, & Golding-Kushner K. (1989). Evaluation of velopharyngeal insufficiency. *Otolaryngologic Clinics of North America*, 22, 519–536.
- 18. Karnell, M.P., Seaver, E.J. (1990). Measurement Problems in Estimating Velopharyngeal Function. Chapter in *Multidisciplinary Management of Cleft Lip and Palate*. (776-786) Editors: Bardach, J. & Morris, H. L., Philadelphia: W.B. Saunders Co.
- 19. Brigger, M. T., Ashland, J. & Hartnick, C, J. (2010). Diagnosis and Treatment of Velopharyngeal Insufficiency. Chapter in *Clinical Management of Children's Voice Disorders*. Editors: Hartnick, J. & Boseley, M. E., San Diego: Plural Publishing Inc.
- 20. Meijer, M. (2003). Het Logopedisch Onderzoek van Kinderen, geboren met een Schisis. Werkgroep Logopedie van de Nederlandse Vereniging voor Schisis en Craniofaciale Afwijkingen; intern publication.
- 21. McWilliams, B. J. & Phillips B. J. (1979). Velopharyngeal Incompetence. *Audio Seminars in Speech Pathology*. Philadelphia: W.B. Saunders, Inc.

22. Sommerlad, B., Rowland, N., Harland, K. (1994). Lateral videofluoroscopy: a modification to aid in velopharyngeal assessment and measurement. *Cleft Palate Craniofacial Journal*, 13, 134-135.

Appendix 1. Assessment form

Casusnummer:

1. Analyse per doel op woordniveau

	Bruikbaarheid	Ernstgraad van de afwijking
	opname	
Structuren van de pharynx (lengte		
van het velum, diepte van de pharynx)		
Bewegingen		
(contact tussen het velum en de		
posteriore pharynxachterwand, mediale		
bewegingen van de laterale pharyngeale		
wanden)		
Sluitingsgraad		
Timing		

Bruikbaarheid opname in beoordeling doelen:

+ bruikbaar

+/- bewogen maar bruikbaar

- niet bruikbaar/afgekeurd

- niet afwijkend, velum raakt meestal de
pahrynxachterwand

- niet afwijkend, velum raakt meestal de
pahrynxachterwand

- niet afwijkend, velum raakt meestal de
pahrynxachterwand

- a ernstig afwijkend, velum raakt niet/een
enkele keer de pharynxachterwand

- Bijzonderheden
- a. Pharynxachterwand trekt naar velum toe

2. <u>Besluit verder beleid (gebaseerd op kindsgebonden achtergrond en volledige opname pharyngogram):</u>

0 = afwachten

1 = logopedische behandeling

2 = operatie:_____

Table A 2. Speech compon	ents				
Hypernasality (n=20)					
No hypernasality	4				
Mild	9				
Moderate	4				
Severe	3				
Mirror test (n=17)					
Positive	7				
Negative	3				
Variable	7				
Nasometry (n=9)					
Normal	2				
Increased (>+ 2SD) 7				
Phonetic speech abnormalit	ies (n=20)				
Modified vowels	1				
Modified consonar	nts 6				
Nasal turbulence	12				
Nasal emission	5				
Compensatory artic	culation				
Glottal st	cops 6				
Nasal sub	ostitution 4				
Grimacin	ng 3				
Intelligibility					
Parents (n=13)					
Median	2.5				
SD	0.8				
SLT (n=21)					
Median	3.0				
SD	0.7				

Appendix 2. Speech components