



ARTICLE



On the role of the frontal projection in videoradiography of velopharynx in decision-making for a velopharyngeal flap plasty in patients with cleft palate

Frida Appelros^a, Magnus Becker^{a,b}, Hanna Salé^c and Henry Svensson^{a,b}

^aDepartment of Clinical Sciences in Malmö, Lund University, Lund, Sweden; ^bDepartment of Plastic and Reconstructive Surgery, Skåne University Hospital Malmö, Malmö, Sweden; ^cDepartment for Neuroradiology, Center of Medical Imaging and Physiology, Skåne University Hospital, Lund, Sweden

ABSTRACT

Despite uneventful primary surgery, patients with cleft palate may experience velopharyngeal insufficiency (VPI) and hypernasal speech. Videoradiography of velopharynx is a commonly used method to visualize velopharyngeal function and a velopharyngeal flap is often used to counteract VPI. The aim of this study was to investigate whether the frontal projection on videoradiography plays a role in the decision-making about velopharyngeal flap surgery, or possibly the width and orientation of the flap. A secondary aim was to evaluate the effect of the flap in improving velopharyngeal function. Between 2007 and 2016, 75 patients had received a flap at our department. During the same period of time, 41 patients who had undergone videoradiography did not receive a flap. Medical records, particularly regarding speech assessments, videoradiography statements and operating records, were scrutinised to seek information about the factors leading up to the decision about whether or not to perform a flap. In only one instance, reduced lateral pharyngeal wall movement found on the frontal projection was clearly taken into account when deciding to refrain from performing a velopharyngeal flap. Only a slight agreement was found between pre-operative speech assessment and findings in videoradiography. Hypernasality was reduced by flap surgery in 97% of the patients. We conclude the frontal projection of the videoradiographic examination seems to have no crucial role in the decision-making on performing a velopharyngeal flap or not in patients with cleft palate. Even with reduced lateral pharyngeal wall movement, a velopharyngeal flap effectively reduces hypernasality and VPI.

ARTICLE HISTORY

Received 30 December 2019
Revised 26 March 2020
Accepted 23 April 2020

KEYWORDS

Cleft lip and palate;
videoradiography;
pharyngoplasty

Introduction



Patients operated on due to cleft palate are at risk of developing velopharyngeal insufficiency (VPI) and hypernasal speech. In the case of prominent symptoms and documented velopharyngeal insufficiency, a complementary surgical procedure is indicated. The classical and widely used operation is to perform a velopharyngeal flap [1]. By movements of the lateral pharyngeal walls against the flap, the idea is to reduce resonance to the nose, and thereby reduce the hypernasality. Hence, a velopharyngeal flap narrows the velopharyngeal space. In some cases, this may introduce unfortunate side effects such as the occurrence of hyponasality and snoring.

After clinical evaluation with perceptual speech analysis by a speech and language pathologist, videoradiography can be applied to visualize the performance of the soft palate in relation to the pharyngeal walls [2]. A lateral projection visualises the configuration and activity of the soft palate and posterior pharyngeal wall. It also allows for the assessment of a possible Passavant's pad. The frontal projection shows the lateral pharyngeal wall movement and its contributions to the closure of the velopharynx. By using both a frontal and a lateral projection, a three dimensional and dynamic conception of the velopharyngeal morphology can be achieved. However, the frontal projection,

focusing on the movement of the lateral pharyngeal walls, might not be mandatory for the decision making to perform a velopharyngeal flap. This issue was highlighted in 2005 by Havstam and co-workers who investigated the clinical role of the frontal projection for the decision making preceding velopharyngeal flap surgery at the cleft palate center in Gothenburg, Sweden. They concluded that lateral projection in videoradiography should be the first step when evaluating velopharyngeal function. They suggest nasendoscopy to be the next step when more information is needed [3].

A drawback of videoradiography is the ionising radiation. The effective dose of ionising radiation for one videoradiography session at our institution, with both lateral and frontal projection, is presently less than 0.1 mSv (range: 26–77 µSv). This corresponds to a little more radiation than for a pulmonary X-ray, or the background radiation of less than one month. The lateral and frontal projections contribute about the same amount of radiation.

As we have used both the frontal and lateral projection since decades, the primary aim of the present study was to review our own experiences regarding the significance of the lateral projection in the perspective of velopharyngeal flap surgery. A secondary aim was to assess the efficacy of the velopharyngeal flap in reducing hypernasality in our cohort of patients.

CONTACT Frida Appelros  frida.appelros@gmail.com  Department of Clinical Sciences in Malmö, Lund University, Lund, Sweden
This article has been republished with minor changes. These changes do not impact the academic content of the article.

© 2020 The Author(s). Published by Informa UK Limited, trading as Taylor & Francis Group.
This is an Open Access article distributed under the terms of the Creative Commons Attribution-NonCommercial-NoDerivatives License (<http://creativecommons.org/licenses/by-nc-nd/4.0/>), which permits non-commercial re-use, distribution, and reproduction in any medium, provided the original work is properly cited, and is not altered, transformed, or built upon in any way.

Materials and methods

Subjects in group A – velopharyngeal flap

The inclusion criteria in group A were: cleft palate, pre- and post-operative speech evaluation, frontal and lateral projection on videoradiography and having a velopharyngeal flap. To identify patients who had a velopharyngeal flap, we used the patient administrative programme of our hospital and the search limit was set to 2007–2016. One hundred and twenty-four surgeries were retrieved. Twenty-eight patients were excluded due to not meeting the inclusion criterion of having a cleft palate. Two patients had syndromes affecting cognition and their ability to cooperate. Three patients had no frontal projection on videoradiography. Two patients had undergone surgery without pre-operative videoradiography. One patient's videoradiography report was ambiguous, and one patient's videoradiography report was missing. Eight had a velopharyngeal flap done before 2007. Their surgery reflected an adjustment of an already existing velopharyngeal flap. Four patients had two flaps during the time period for this study. Data from the first surgery was used in these instances. Taken together, 75 surgeries in the 75 patients constitute group A.

Subjects in group B – no velopharyngeal flap

The inclusion criteria in group B were: cleft palate, a speech evaluation, frontal and lateral projection on videoradiography, but no velopharyngeal flap. A videoradiography of the velopharynx does not necessarily lead to a velopharyngeal flap. Thus, we were also interested in those who were deemed unsuitable for surgery, and the reason why. For this purpose, we used the local registry containing information on all videoradiographic investigations performed in the period 2007–2016. We retrieved a list of referrals for videoradiography and 41 patients were eventually identified for group B. Seventy-one patients had then been excluded as they did not meet the inclusion criterion of having a cleft palate. Seven patients already had a velopharyngeal flap. Eleven patients underwent another type of surgery with the aim of improving speech. Five patients went through a velopharyngeal flap surgery after 2016, and consequently there was no post-operative speech evaluation in their cases. One patient went through a videoradiography for other reason. One patient had no frontal projection performed. One patient received a flap by a cleft team elsewhere. One patient had missing data in the medical records. One patient had moved abroad, and the forthcoming remained unknown.

Data collection

For all patients, information was collected from the electronic medical records. The following statements were particularly scrutinised: evaluation by the speech and language pathologist, the interpretation of the videoradiography by the radiologist, and the conclusion drawn at the cleft team conference at our department. The latter is an interdisciplinary meeting held once a month when videoradiographic findings are displayed and discussed by the cleft team consisting of plastic surgeons, radiologists, ENT specialists, and speech and language pathologists. All participants are specially trained in, and dedicated for, cleft care. For group A, the operation records and the one-year post-operative follow-up evaluations by the speech and language pathologist were also scrutinised. Additional information that was considered relevant for interpretation of the results was also noted.

Table 1. Distribution of sex and cleft type in groups A and B (numbers and percentages).

| | A | B |
|--------|-------------|-------------|
| Female | 25 (33.3%) | 14 (34.1%) |
| Male | 50 (66.7%) | 27 (65.9%) |
| Total | 75 (100.0%) | 41 (100.0%) |
| CP | 37 (49.3%) | 18 (43.9%) |
| CLP | 38 (50.7%) | 23 (56.1%) |
| Total | 75 (100.0%) | 41 (100.0%) |

A: patients with a flap. B: patients without a flap. CP: cleft palate only. CLP: cleft lip and palate.

Three levels of hypernasality could be distinguished based on the statements made by the speech and language pathologist: mild to moderate, moderate, and moderate to severe. In cases of a velopharyngeal flap, pre- and postoperative statements were compared with one another. Two levels of lateral pharyngeal wall movement in the frontal projection of videoradiography could be distinguished based on the statements made by the radiologist: appearing normal and reduced. Three levels of gap between the soft palate and the posterior pharyngeal wall in the lateral projection could be distinguished based on the statements made by the radiologist: no or almost no gap, moderate gap, and severe gap. If the concluding statement of the videoradiography was not completely clear, the entire report was looked into again by the radiologist. This occurred in 34 instances in group A, and six in group B. Some patients had gone through more than one videoradiography. In group A, the videoradiography preceding the flap was the one we paid attention to. In group B, we chose the most recent one as it was considered to provide the most relevant information to consider for surgery. Based on these findings, a database was created also including demographic information such as age, sex and cleft type.

Statistical methods and calculations

The statistical analyses were performed using SPSS Statistics (version 25, IBM Corporation, NY, USA). Fisher's exact method was used for calculating 95% confidence intervals (CI).

An unweighted kappa-analysis was performed to investigate the agreement between the pre-operative speech evaluation and the gap between the soft palate and posterior pharyngeal wall on the lateral projection of videoradiography. The unweighted kappa-analysis has five levels of increasing agreement: 0–0.20 indicates a slight agreement, 0.21–0.40 fair, 0.41–0.60 moderate, 0.61–0.80 substantial; and 0.81–1.00 almost perfect. Information from the Swedish National Quality Registry for Cleft Lip and Palate was used to calculate the risk ratio for hypernasality and VPI in relation to cleft type and sex. It was also used to calculate the risk ratio for receiving a flap.

Ethics

The project was approved by the Regional Ethical Review Board in Lund, Sweden (Dnr 2018/425).

Results

Group A

The distribution of sex and cleft type is shown in Table 1. The median age at surgery was six years (range: 3–26). Thirteen patients (17.3%) were classified by a speech and language pathologist as having low to moderate hypernasality, 26 moderate (34.7%), and 36 moderate to severe (48.0%). Videoradiography was performed eight months (median) before the operation

(range: 0–67). The gap between the soft palate and the posterior pharyngeal wall was deemed as none or almost none in 10 cases (13.0%), moderate in 37 (49.3%), and severe in 28 (37.3%). In 21 cases (28.0%) the lateral pharyngeal wall movement was judged as reduced. In no case was documentation found stating that a reduced lateral wall movement was considered during the treatment decision of a flap. Neither was it documented that the lateral wall movement affected the design of the flap. The post-operative speech evaluation was done after one year according to departmental routines. A reduction in hypernasality was noted in 71 out of 73 cases (97.3%; 95% CI 90.5–99.7%) as judged from post-operative speech evaluations. In two cases, no post-operative speech evaluation was documented.

Group B

The distribution of sex and cleft type is shown in Table 1. Nineteen (46.3%) patients were classified by a speech and language pathologist as having low to moderate hypernasality, 12 moderate (29.3%), and 10 moderate to severe (24.4%). The median age at videoradiography was five years (range: 3–21). The gap between the soft palate and the posterior pharyngeal wall was deemed as none or almost none in 29 cases (70.7%), moderate in nine (22.0%), and severe in three (7.3%). In four cases (9.8%), the lateral pharyngeal wall movement was judged as reduced. In only one case, documentation was found regarding the reduced lateral wall movement that had made the surgeon hesitate about making a flap. This patient underwent a second videoradiography three years later when the frontal projection still showed reduced lateral wall movement. Despite this finding, the patient was at that stage offered a flap surgery, but the family chose to not follow through.

Agreement between pre-operative speech assessment and assessment of the lateral projection in videoradiography

The unweighted kappa value was 0.14 (95% CI 0.01–0.27) showing a slight agreement between the two methods.

Risk ratios for hypernasality and VPI

For cleft palate only (CP) compared with cleft lip and palate (CLP), the risk ratio was 0.92 (95% CI; 0.69–1.19; *p*-value 0.57) for hypernasality and VPI. The risk ratio for having a flap was 0.95 (95% CI; 0.67–1.30; *p*-value 0.83).

For girls compared with boys, the risk ratio was 0.76 (95% CI 0.54–1.05, *p*-value 0.10) for hypernasality and VPI. The risk ratio for having a flap was 0.76 (95% CI 0.49–1.12, *p*-value 0.18).

Discussion

We consider the sample representative as all patients included had undergone videoradiography during a defined period of time. Some of them received a flap (group A), while some of them did not (group B). The time period of the recent 10 years was chosen as the results would thus be representative of the present clinical setting. The time period ended in 2016 to allow for a one-year post-operative follow-up. Since there is only one operational registry code for velopharyngeal flap surgery, and only two operational registry codes for CP and CLP, the risk of misclassification is very low. However, a patient with an undiagnosed inconspicuous cleft palate going through videoradiography might not have entered study group B owing to mild symptoms

and discreet findings on perceptual speech analysis and videoradiography. Without a reason for an appointment with a plastic surgeon, the diagnosis of cleft palate was never made.

Two children with syndromes reducing cognitive function had to be excluded as they were not able to adequately participate in the pre-operative videoradiography or the post-operative follow-up. Children with other syndromes or additional malformations, for instance the Pierre Robin sequence, Klippel–Feil and hemifacial microsomia, were included as these conditions have no negative impact on cognitive functions. 22q11 deletion syndrome and Dubowitz syndrome may affect cognitive function but, in our study, patients with these syndromes were able to participate in an adequate way and could therefore be included.

Regarding the speech assessments, we trusted the statements by the speech and language pathologist in the medical records. These statements follow a standardised model, making it possible to categorise the degree of hypernasality into three levels. Reduction of hypernasality by surgery varied somewhat, but for evaluating the effect of the flap we considered it sufficient to categorise it into two levels; reduced hypernasality or not. In scientific research in the field of logopaedics, speech evaluation is often carried out by having two or three independent speech and language pathologists listen to the speech recordings. However, such an approach would be beyond the scope of this study as it focuses on the implication of the perceptual speech analysis in an ordinary clinical setting. The same can be said regarding the statements concerning the videoradiographies. One option would be to reassess all the images. The statements were, however, in most cases easy to interpret although clarifying comments by the radiologist had to be obtained in 40 instances before categorisation was commenced. The medical records regarding the surgeries were also easy to interpret as they were written in a consistent manner. We also scrutinised the statements from the cleft team conferences and statements from the out-patient visits to seek information on factors important for the decision about surgery. On no occasion was it clearly documented that a reduced movement of the lateral pharyngeal walls was considered an obstacle to performing a velopharyngeal flap.

An important finding of this study is the weak agreement (kappa value of 0.14) between the speech assessment and videoradiography findings in the lateral projection pre-operatively. One reason for this might be the nature of the different grading systems where one relates to perception and the other to morphology. With this in mind, both perceptual speech analysis and videoradiography seem to be necessary to make a confident evaluation of whether a velopharyngeal flap is advisable or not. Patients with poorer scores both in perceptual speech analysis and lateral projection of videoradiography were those who were more likely to receive a flap. However, the use of the frontal projection is uncertain as in no case did we find it documented that the findings concerning the frontal projection were substantially considered in the final decision-making about flap surgery. In only one instance, reduced lateral pharyngeal wall movement was clearly taken into account when deciding to refrain from performing a velopharyngeal flap.

Patients with clefts go through several radiographic examinations for evaluation of jaw relationship and dental occlusion. They are thereby exposed to an increased amount of radiation during their childhood and teens compared with non-cleft patients and are at a higher risk of developing radiation-related cancers [4]. By refraining from the frontal projection on videoradiography, the exposure of ionising radiation will be reduced, but by less than 35 μ Sv. This reduction is of minor clinical significance compared with the total amount of radiation that these patients are exposed

to in relation to their cleft. However, all investigations should adhere to the concept of trying to restrict the radiation dose and thereby minimising the exposure in the total population.

Studies have shown that children with clefts, particularly CP, have some cognitive difficulties when viewed as a whole group [5–7]. We hypothesized that some of these difficulties may be due to the speech and investigated whether CP was overrepresented among patients going through investigation for VPI and among patients receiving a flap. According to the national registry for clefts, the expected number of patients with CP in our cohort would be 60, whereas we had 55. A significantly increased risk for VPI in the subgroup CP compared with CLP was thus not found. Neither was there a significantly increased risk for having a flap.

It has also been found that educational outcomes for girls with clefts are lower compared with boys with clefts [8]. To this, speech might be a contributing factor. According to the national registry the expected number of girls in our cohort would be 51, whereas we had 39. A significantly increased risk for VPI in the subgroup of girls compared with boys was thus not found. Neither was there a significantly increased risk for having a flap.

Previous studies have shown a correlation between the severity and extent of the cleft, and the degree of deviance of the speech and VPI [9–11]. The distribution of CP and CLP in our cohort was 47.4% and 52.6%, respectively. The corresponding figures in the national registry are the opposite, namely 52% for CP and 48% for CLP. The reason for this slight discrepancy might be that bilateral clefts were over-represented in our cohort. In our study, 34.4% of the CLP patients actually had bilateral clefts whereas bilateral clefts in the national registry are encountered in 30.6% of the CLP patients. Another possible contributing factor to a high proportion of CLP is the number of adopted children from foreign countries with special needs coming to Sweden in 2009 and for a couple of years afterwards. These adopted children with clefts tend to have more severe clefts and the primary surgery performed in their native countries failed in many instances.

Seventy-one out of 73 patients had reduced hypernasality after flap surgery. This is in line with previous reported data showing that the velopharyngeal flap plasty is an effective treatment for VPI [12,13]. Middle ear problems in children with cleft palate are common and are most significant during early childhood. These problems are treated alongside the treatment of the cleft. Hearing has an impact on speech and an improvement in hearing might lead to an improvement in speech over time. However, the velopharyngeal flap is a very robust measure and the possible additional contribution of improved hearing to the improved speech ought to be limited.

The statements by the plastic surgeon in the medical records might not cover everything that was brought up with the cleft team when deciding about whether or not to proceed with surgery. Although we acknowledge the quality of the medical documentation, retrospective investigations based on reviews will always come with some limitations due to inconsistency and missing data. However, all relevant factors preceding surgery are mandatory to document and the very sparse documentation regarding the lateral pharyngeal wall movement clearly points to the fact that it was a factor of definite subordinate importance for the decision about whether to perform a flap or not. For all patients, rather the combined information from speech analysis and the lateral projection stood out as decisive.

Although the frontal projection seems less crucial for the decision about performing a flap or not, it might be motivated for other reasons. One such reason is to visualise the configuration of the soft palate, rather than the pharyngeal walls. The frontal

projection can for instance reveal a central dip of the soft palate, reflecting a poor continuity of the levator veli palatine muscle in the midline. In such cases an attempt to a re-repair is an option for a velopharyngeal flap. Buccal flaps used to augment the soft palate is another option in cases of very pronounced tissue deficiencies of the soft palate. Both options have been used with increased frequency over the last years. Moreover, an asymmetrical closing of the velopharyngeal space can sometimes be seen on the frontal projection. This asymmetrical closing of the velopharyngeal space could motivate a different positioning of a velopharyngeal flap. Albeit a questionable role in connection with velopharyngeal flap surgery, the frontal projection may still be an important investigative tool used in the individualized surgical treatment of velopharyngeal insufficiency.

Conclusion

The frontal projection of the videoradiographic examination showing the movements of the lateral pharyngeal walls was found to have no crucial role in the decision-making on performing a velopharyngeal flap or not in patients with cleft palate. Even with reduced pharyngeal wall movement, our findings indicate that a velopharyngeal flap effectively reduces hypernasality and VPI.

Acknowledgements

We thank statistician Jan-Åke Nilsson for his help with the statistical analyses. We are also indebted to speech and language pathologists Maria Sporre and Kristina Klintö and phoniatician Beatriz Arenaz Búa for their valuable contributions to this work.

Disclosure statement

No potential conflict of interest was reported by the author(s).

References

- [1] Hogan VM. A clarification of the surgical goals in cleft palate speech and the introduction of the lateral port control (l.p.c.) pharyngeal flap. *Cleft Palate J.* 1973;10:331–345.
- [2] Skolnick ML. Videofluoroscopic examination of the velopharyngeal portal during phonation in lateral and base projections – a new technique for studying the mechanics of closure. *Cleft Palate J.* 1970;7:803–816.
- [3] Havstam C, Lohmander A, Persson C, et al. Evaluation of VPI-assessment with videofluoroscopy and nasoendoscopy. *Br J Plast Surg.* 2005;58(7):922–931.
- [4] Jacobs R, Pauwels R, Scarfe WC, et al. Pediatric cleft palate patients show a 3- to 5-fold increase in cumulative radiation exposure from dental radiology compared with an age- and gender-matched population: a retrospective cohort study. *Clin Oral Invest.* 2018;22(4):1783–1793.
- [5] Persson M, Becker M, Svensson H. General intellectual capacity of young men with cleft lip with or without cleft palate and cleft palate alone. *Scand J Plast Reconstr Surg Hand Surg.* 2008;42(1):14–16.
- [6] Persson M, Becker M, Svensson H. Academic achievement in individuals with cleft: a population-based register study. *Cleft Palate Craniofac J.* 2012;49(2):153–159.
- [7] Persson M, Sandy J, Kilpatrick N, et al. Educational achievements in Pierre Robin Sequence. *J Plast Surg Hand Surg.* 2013;47(1):36–39.

- [8] Persson M, Becker M, Conrad AL, et al. Female and male differences in academic achievement in individuals with cleft: a population-based register study. *Cleft Palate Craniofac J.* 2018;55(2):196–203.
- [9] Sullivan SR, Marrinan EM, LaBrie RA, et al. Palatoplasty outcomes in nonsyndromic patients with cleft palate: a 29-year assessment of one surgeon's experience. *J Craniofac Surg.* 2009;20 (Suppl 1):612–616.
- [10] Mahoney MH, Swan MC, Fisher DM. Prospective analysis of presurgical risk factors for outcomes in primary palatoplasty. *Plast Reconstr Surg.* 2013;132(1):165–171.
- [11] Klinto K, Falk E, Wilhelmsson S, et al. Speech in 5-year-olds with cleft palate with or without cleft lip treated with primary palatal surgery with muscle reconstruction according to Sommerlad. *Cleft Palate Craniofac J.* 2018;55(10):1399–1408.
- [12] Sullivan SR, Marrinan EM, Mulliken JB. Pharyngeal flap outcomes in nonsyndromic children with repaired cleft palate and velopharyngeal insufficiency. *Plast Reconstr Surg.* 2010; 25(1):290–298.
- [13] Yamaguchi K, Lonic D, Lee CH, et al. A treatment protocol for velopharyngeal insufficiency and the outcome. *Plast Reconstr Surg.* 2016;138(2):290e–299e.